The superiorly based pharyngeal flap with Z-plasty.

Surgery and Speech.

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Surgery and speech

De craniaal gesteelde pharynxplastiek met Z-plastiek.

Chirurgie en spraak

(met een samenvatting in het Nederlands)

Proefschrift

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Chapter 1

Introduction and aims of the study.

Introduction

Cleft of the lip, the alveolair proces and palate (CLP) is the most common congenital malformation of the head. According to the literature, CLP has a birth prevalence ranging from 1 in 1000 to 2.69 in 1000 newborns in different parts of the world (16). In more detail: the prevalence of CLP differs in Asians from 1 in 400 to 1 in 500 newborns and in African Americans from 1 in 1500 to 1 in 2000 newborns. The prevalence in Caucasians is 1 in 750 to 1 in 900 newborns; this corresponds to the prevalence of 1 in 800 newborns in the Netherlands.

CLP occurs more frequently in males than in females. For reasons that are not yet clearly understood, clefts on the left hand side are more frequent than on the right. The causes of CLP are multifactorial: both genetic and environmental factors play a role in the development of the cleft (14).

Whereas the cleft lip is a malformation that primarily affects aesthetics, cleft palate is a malformation that primarily affects function (12). Primary and secondary functional disorders caused by the cleft of the palate can be distinguished. Primary functional disorders include problems of nutrition, swallowing, breathing and mimic disorders e.g. grimacing. After repair of the cleft palate, most of these primary disorders are corrected. Speech and voice disorders as well as conductive hearing loss can occur as secondary disorders. These secondary disorders may persist after primary repair of the palatal cleft (14).

Primary repair of the palatal cleft is generally performed in early childhood. There has been considerable debate over the appropriate timing of repair of the cleft palate (5). There are a few CLP-centers that advocate repair of the palate in the first week after birth but this is rather controversial. In contrast, two succeeding Schweckendiek generations (21, 22, 23, 24) consistently recommended early repair of the soft palate, while the cleft of the hard palate was occluded with a dental appliance until its repair at the age of 12 to 15 years. However, most centers can be distinguished according to their philosophy: early or late repair. Early repair is defined as between 6 months and 15 months of age, whereas late repair is defined as between 15 months and 2 years of age. In general, there is agreement that the earlier the repair of the cleft palate is performed, the lower the incidence will be of velopharyngeal insufficiency and consequently the development of compensatory misarticulations (2, 6). The main reason for repair of the palate is to provide an adequate structure for functions such as breathing, swallowing and speech. However, the early repair of the hard palate has raised concerns about the potential effect on growth of the maxilla, which affects the midfacial appearence (1, 10), whilst the delayed repair of the hard palate may be related to unfavourable and dysfunctional speech (4, 29).

The major morbidity of a cleft palate, and often even of a cleft palate after repair, is defective speech and as its consequence communication impairment. Despite undergoing reconstructive surgery of the palate, 20 to 30% of the children with a repaired cleft palate will have some degree of velopharyngeal dysfunction, resulting in abnormal resonance in speech (12). A substantial number of children will develop a complex speech disorder related to the disturbed function of the velopharyngeal mechanism such as severe resonance disorders, compensatory misarticulations and a decrease of voice quality, especially hoarseness.

Normal velopharyngeal function is achieved by the synchronized movement of the velopharygeal mechanism. The velopharyngeal mechanism is composed of the soft palate and the lateral and posterior pharyngeal walls. The synchronized movement of the velopharyngeal mechanism plays a fundamental role in the production of speech, since it is responsible for the distribution of the expiratory airflow and acoustic vibrations to the oral cavity, during the production of oral sounds, and to the nasal cavity during the production of nasal sounds (10, 26)

Oral resonance, as contrasted to nasal resonance, is obtained by velopharyngeal closure. Typically this closure is accomplished by elevation of the velum and approximation of the lateral pharyngeal walls in order to separate the oropharynx from the nasopharynx.

The undermentioned terms are frequently used to denote an improperly functioning velopharyngeal mechanism.

Velopharyngeal inadequacy is a generic term used to denote any type of pathological velopharyngeal function (27), but some authors (10, 11) also use the term velopharyngeal dysfunction for describing an abnormal velopharyngeal function, regardless of its cause.

The term velopharyngeal insufficiency (VPI) includes any structural defect of the velum or pharyngeal wall at the level of the nasopharynx. Most often these defects are congenital (27).

VPD and/or VPI may affect speech in different manners. The most common speech symptoms are increased nasal resonance, nasal air emission (whether audible or not) and compensatory misarticulations (9, 10, 26, 30).

Increased nasal resonance (interchangeable: hypernasality) is one of the most remarkable clinical manifestations of VPD and may be described as a deviant change in speech resulting from an abnormal open communication between nasal and oral cavities. It corresponds to an excessive nasal resonance of normally non-nasal sounds. From a physiological point of view, increased nasal resonance results from the inability of the velopharyngeal mechanism to close adequately and to separate the orophaynx from the nasopharynx (10, 18). Nasal air emission is also a characteristic. It corresponds to the improper emission of air through the nose during the production of pressure consonants (27). The nasal emission is noticed during the production of plosive, fricative and affricate pressure consonants and may vary from non-audible emissions to a more severe form of audible emission, called nasal turbulence (9, 10). The lack of velopharyngeal closure further leads to the development of compensatory articulations, which may be considered as strategies to compensate for the inability to create pressure in the oral cavity. These efforts to compensate are inadequate in most cases as they correspond to deviant motoric patterns (misarticulations) and usually generate undesired sounds and/or noises that reduce intelligibility. From an aerodynamic point of view, the primary effect of the failure in the articulation performance of the velopharyngeal structures is the development of a weak intra-oral air pressure during production of plosive, fricative and affricate consonants, associated with nasal air emission (26). Thus, individuals with VPD or VPI frequently replace orally articulated sounds by sounds articulated at points behind the area of impairment, in an often unconscious attempt to approximate the acoustic output to a normal sound as much as possible. The most frequent compensatory misarticulations secondary to VPD and/or VPI are: glottal stop, pharyngeal fricative, pharyngeal stop, velar fricative, mid-dorsum palatal stop and posterior nasal fricative (10, 18).

Individuals with a history of VPD or VPI may demonstrate any combination of speech sound errors, increased nasal resonance and nasal air emission. Speech sound distortion can also occur due to other structural malformations, including malocclusion (13). However, Whitaker et al. (28) failed to reveal an association between the severity of lisping and the severity of malocclusion in children with operated unilateral cleft lip and palate.

Sell & al. (25) have presented a global overview of minor and major articulation errors in cleft palate speech.

Articulation errors in cleft palate speech	
Minor errors	**Explanation**
Lateralization	tongue is in a lateral position between the teeth during articulation
Palatalization	tongue is touching the palate during articulation
Interdentalization	tongue is between the front teeth during articulation
Major errors	**Example**
Pharyngealization	tongue is shifted backwards towards the pharynx during articulation
Glottal articulation	the closure of the plosives is done in a glottal manner instead of a labial. This is also called laryngeal replacement.
Backing to uvular	the tongue is shifted backwards towards the uvula
Backing to velar	the tongue is shifted backwards towards the soft palate
Active nasal fricatives	air is emitted though the nose during the articulation of fricatives
Absent pressure consonants	plosives are not formed or weakened during the articulation
Nasal realizations	the nasal air flow is persistent throughout the whole word
Weak nasalized consonants	the consonants are nasalized, i.e., air is emitted during the articulation of the consonants

Whenever structural malformations such as a palatal cleft or an incompetent velopharyngeal mechanism are present, speech can be affected by obligatory distortions such as increased nasal resonance and air emission as well as compensatory misarticulations. In case of these structural malformations, surgery or other forms of physical management are needed to provide a competent and dynamically

functional velopharyngeal mechanism. In contrast, speech therapy is indicated for compensatory misarticulations.
If increased nasal resonance persists after repair of the palate, pharyngeal flap surgery can be performed to improve nasal resonance. The posterior pharyngeal wall flap creates a static obstruction central in the nasopharynx and leaves lateral openings or ports which should remain patent during breathing and production of nasal consonants in speech and should be closed by the medial movement of the lateral pharyngeal walls against the flap during the production of oral consonants. Two types of posterior pharyngeal wall flaps can be distinguished: the inferiorly based and the superiorly based posterior pharyngeal wall flap (19, 20).
Instead of pharyngeal flap surgery, some authors recommend the re-repair of the soft palate with the intention to reposition the muscles anatomically and thereby to lengthen the soft palate. Recently a historic controversy has been revived (3, 7, 15). Chen et al., Hill et al., and Mann et al. advocated the pushback of the soft palate. The pushback of the palate is combined with a flap composed of buccal mucosa and buccinator muscle of the left and the right side. Both flaps are interposed in the gap between the posterior border of the hard palate and the anterior border of the 'pushed back' soft palate, as a sandwich to close the nasal side and the oral side with a double layer.

Other procedures instead of pharyngeal flap surgery may be the sphincter pharyngoplasty (8, 17) or augmentation of the posterior pharyngeal wall. These procedures may be considered in selected cases.

However, the large number of operations available to treat increased nasal resonance in speech due to VPI perhaps best reflects that no one technique is ideal and all techniques have their series of surgical complications and unsatisfactory functional results.

For the sake of completeness obturation of the gap between the posterior pharyngeal wall and the posterior border of the velum by a dental appliance with a speech bulb must be mentioned as a non-surgical treatment to decrease nasal resonance.

All this points out the absolute need for adequate methods for assessing the functional results, and particularly the speech outcome. This is the way to guide surgical decision making, and to improve technical details of surgical techniques.

Aims of the study

The first aim of this research concerns the critical analysis of the results in 130 patients treated with a superiorly based pharyngeal flap with a Z-plasty to cover the raw surface of the oral side of the flap, as performed in the Department of Oral and Maxillofacial Surgery of the University Medical Center Utrecht.

Particular attention is paid to the incidence of revisional surgery, as revisional surgery accounts for either surgical complications or unsatisfactory functional results (Chapter 3).

The limitations in the quantification of the functional outcomes gave rise to the search for a valid assesment protocol suitable for daily practice. As no standard for the perceptual evaluation exists, the development of a method that can reliably assess speech is essential. So, the second aim of this study is the selection of a 'material and method' for a reliable perceptual judgement with respect to differentiated aspects of cleft palate speech. Therefore, intra- and interrater reliability of different types of judges and different types of speech samples are investigated (Chapter 4).

In addition to these subjective perceptual measures, objective measures by means of acoustic nasometry by the Nasometer® are applied. The third aim of this research is to investigate the relation between the subjective perceptual measures and the objective instrumental measures.

Therefore, the correlation between the mean nasalance scores and differentiated perceptual ratings of two types of speech samples are studied. The first type of speech samples consisted of a standard text with a normal distribution of phonemes in the Dutch language, the second type of speech samples consisted of a standard denasal text (Chapter 5).

Because the correlation between mean nasalance scores and the perceptual ratings appeared to be moderate, it is investigated whether higher correlation coefficients may be found when composite measures derived from mean nasalance scores are used. By using the difference between or the quotient of the mean nasalance scores computed for speech samples with a normal distribution of phonemes and speech samples free of nasal consonants, normalization of the nasalance scores to the performance of the speaker is obtained. By reducing patient-dependent effects on the instrumental measure better correlation coefficients may be obtained (Chapter 6).

References

1. Blijdorp P, Egyedi P. The influence of age at operation for clefts on the development of the jaws. J Maxillofac Surg 1984; 12: 193-200.

2. Blijdorp P, Müller H. The influence of the age at which the palate is closed on speech in the adult cleft patient. J Maxillofac Surg 1984; 12: 239-246.

3. Chen GF, Zhong LP. A bilateral musculomucosal buccal flap method for cleft palate surgery. J Oral Maxillofac Surg 2003; 61: 1399-1404.

4. Cosman B, Falk AS. Delayed hard palate repair and speech deficiencies: a cautionary report. Cleft Palate J 1980; 17: 27-33.

5. Egyedi P. Timing of palatal closure. J Maxillofac Surg 1985; 13: 177-182.

6. Hardin-Jones MA, Jones DL. Speech production patterns of preschoolers with cleft palate. Cleft Palate Craniofac J 2005; 42: 7-13.

7. Hill C, Haydan C, Riaz M, Leonard AG. Buccinator Sandwich pushback: A new technique for treatment of secondary velopharyngeal incompetence. Cleft Palate-Craniofac J 2004; 41: 230-237.

8. Hynes W. Pharyngoplasty by muscle transplantation. Br J Plast Surg 1950; 3: 128-135.

9. Johns DF, Rohrich RJ, Awada M. Velopharyngeal incompetence: a guide for clinical evaluation. Plast Reconstr Surg 2003; 112: 1890-1897.

10. Kummer AW. Cleft palate and Craniofacial Anomalies. Effects on speech and resonance. 2nd edition Clifton Park, NY: Delmar Cengage Learning 2008.

11. Kummer AW. Types and causes of velopharyngeal dysfunction. Semin Speech Lang 2011; 32: 150-158.

12. Kummer AW. Perceptual assessment of resonance and velopharyngeal function. Semin Speech Lang. 2011; 32: 159-167.

13. Kummer AW. Speech therapy for errors secondary to cleft palate and velopharyngeal dysfunction. Semin Speech Lang 2011; 32: 191-198.

14. Maier A, Speech of children with Cleft Lip and Palate: Automatic assessment. PhD-thesis. Technische Facultät der Universität Erlangen-Nürnberg 2009.

15. Mann RJ, Neaman KC, Armstrong SD, Ebner B, Bajnrauh R, Naum S. The double-opposing buccal flap procedure for palatal lenghtening. Plast Reconstr Surg 2011; 127: 2413-2418.

16. McLeod NMH, Arana-Urioste ML, Saeed NR. Birth prevalence of cleft lip and palate in Sucre, Bolivia. Cleft Palate-Craniofac J 2004; 41: 195-198.

17. Orticochea M. Construction of a dynamic muscle sphincter in cleft palates. Plast Reconstr Surg 1968; 41: 323-327.

18. Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP. Communication disorders associated with cleft palate. In: Peterson-Falzone SJ, Hardin-Jones MA, Karnell MP. Cleft palate speech. Saint Louis: Mosby. 2001; 7: 162-198.

19. Schoenborn KWEJ. Ueber eine neue Methode der Staphylorrhaphie. Verh Dtsch Ges Chir 1875: 4: 235-239; Arch Klin Chir 1876; 19: 527-531.

20. Schoenborn KWEJ. Vorstellung eines Falles von Staphyloplastik. Verh Dtsch Ges Chir 1886; 15: 57-62.

21. Schweckendiek H. Zur Frage der Früh- und Spätoperationen der angeboren Lippen-Kiefer-Gaumenspalten. Z Laryng 1951; 30: 51-56.

22. Schweckendiek H. Zur zweiphasigen Gaumenspaltenoperation bei primären Velumverschluss. Fortschr Kiefer GesichtsChir 1955; 1: 73-76.

23. Schweckendiek W. Die Technik der primären Veloplastik und ihre Ergebnisse. Acta Chir Plast 1966; 8: 188-194.

24. Schweckendiek W. Primary veloplasty: Long-term results without maxillary deformity. A twenty-five year report. Cleft Palate J 1978; 15: 268-274.

25. Sell D, Grunwell P, Mildinhall S, Murphy T, Cornish TA, Bearn D, Shaw WC, Murray JJ, Williams AC, Sandy JR. Cleft Lip and Palate Care in the United Kingdom— The Clinical Standards Advisory Group (CSAG) Study. Part 3: Speech Outcomes. Cleft Palate Craniofac J 2001; 38: 30-37.

26. Trindade IEK, Genaro KF, Yamashita RP; Miguel HC; Fukushiro AP. Proposal for velopharyngeal function rating in a speech perceptual assessment. Pró-Fono R Atual Cient 2005; 17(2): 259-262.

27. Van Lierde KM. Nasalance and nasality in clinical practice. PhD-thesis Ghent University 2001.

28. Whitaker ME, De Souza Freitas JA, Pegoraro-Krook MI. Relationship between occlusion and lisping in children with cleft lip and palate. Cleft Palate Craniofac J 2012; 49: 96-103.

29. Witzel MA, Salyer KE, Ross RB. Delayed hard palate closure: the philosophy revisited. Cleft Palate J 1984; 21: 263-269.

30. Zuiani TBB, Trindade IEK, Yamashita RP, Trindade Junior AS. The pharyngeal flap surgery in patients with velopharyngeal

insufficiency: perceptual and nasometric speech assessment. Braz J Dysmorphol Speech Dis 1998; 2: 31-42.

Chapter 2

Velopharyngoplasty according to Sanvenero Rosselli

Historical review and contemporary judgement

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Louise M.T.N. Crisi

This chapter is based on Velopharyngoplasty according to Sanvenero Rosselli in *Mund Kiefer GesichtsChir 2000; 4: 95-98* and Letter to the Editor in *Br J Oral Maxillofacial Surg 1998: 36: 157*

Summary

The history of the pharyngeal flap and the contribution of Schoenborn, Sanvenero Rosselli, Padgett, Sercer and Rosenthal are described. The summary 'Divisione palatina e sua cura chirurgica' in the Italian/ French language is translated into English.

Introduction

In this paper an English translation of the original Italian/ French summary of a lecture given by Gustavo Sanvenero Rosselli in 1935 is presented. In addition, we review the relevant literature.

This summary is referred to in many papers on pharyngoplasty and it therefore can be considered a 'citation classic', although obviously it is indeed just a summary of a lecture and not strictly a scientific article.

The problem with translating a publication like this one is that one should adhere to the original text as closely as possible. However, since we had to deal with Italian phraseology unfamiliar to non-Italians, it was decided to present the text in such a way that the reader can easily grasp its essentials. Thus, any embellishments or repetitions are reproduced, but are put in brackets {} and can therefore be skipped over. We were able to obtain only the first part of the discussion at the end of the lecture, but it was thought appropriate enough to present it to the reader also.

Cleft palate and its surgical treatment

Prof. Gustavo Sanvenero Rosselli,

Milan, Italy

Transactions of the Second International

Congress on Stomatology, Bologna, 1935-XIII,

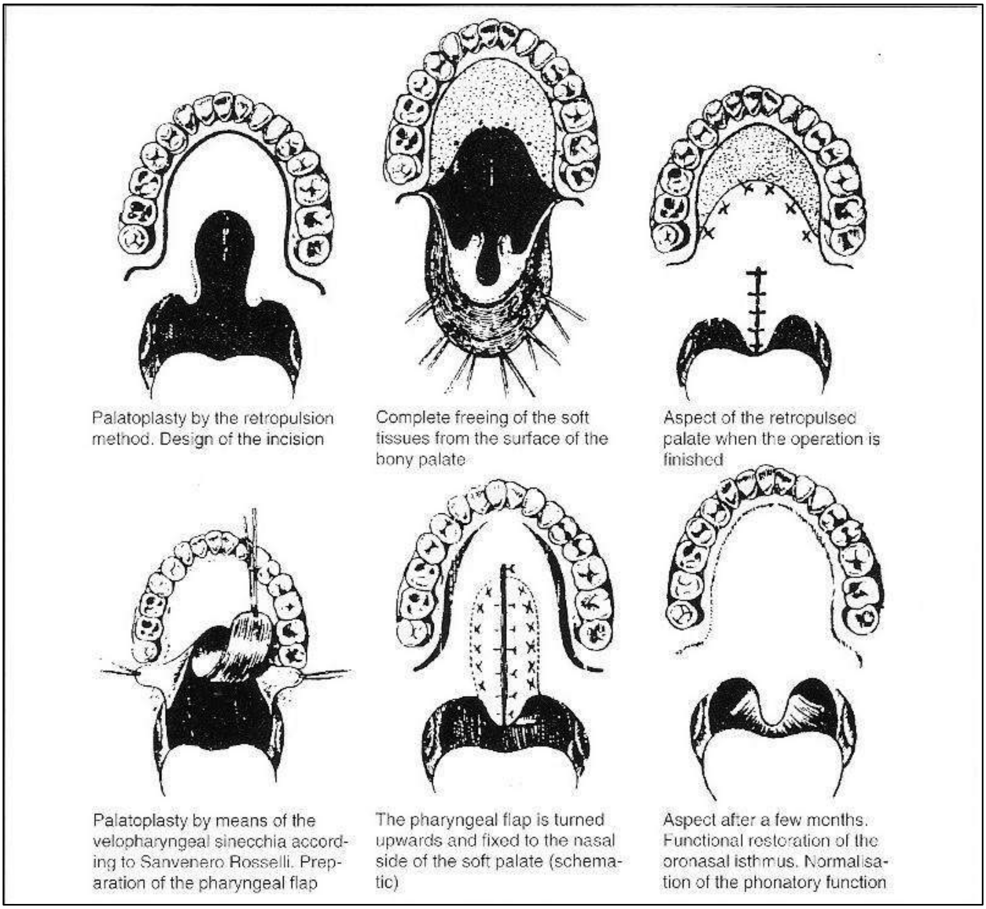
14-19 April p. 391-392.

Under the gracious patronage of H. M. the

King of Italy

President of honour: H.E. Benito Mussolini

Abstract



Palatoplasty by the retropulsion method. Design of the incision

Complete freeing of the soft tissues from the surface of the bony palate

Aspect of the retropulsed palate when the operation is finished

Palatoplasty by means of the velopharyngeal sinecchia according to Sanvenero Rosselli. Preparation of the pharyngeal flap

The pharyngeal flap is turned upwards and fixed to the nasal side of the soft palate (schematic)

Aspect after a few months. Functional restoration of the oronasal isthmus. Normalisation of the phonatory function

The cleft palate is a frequent and serious malformation, that compromises one of the most important functions of social life: speech. The palatoplasty operation aims especially at functional restoration and must therefore, within certain limits, be adjusted to the degree and the variety of the deformity. As one of the disadvantages resulting from the classical operation is {precisely} an excessively short soft palate, preventing contact during phonation and therefore resulting in a voice that still has an unpleasant nasal tonality (hyper-rhinolalia), recent techniques have been inspired by criteria of a more satisfactory functional repair. Two quite new and interesting methods of plastic restoration of the cleft palate, both in anatomical and also functional terms {of the cleft palate}, velopharyngeal adhesion and push-back of the soft palate, deserve special attention and should be preferred as the intervention of choice in suitable cases. The former involves pulling the soft palate backwards,

ensuring a sufficient functional closure of the oro-nasal isthmus, while the latter produces a similar effect through different surgical procedures. The conclusions are based on the surgical and phonetical results obtained in 186 patients I treated personally with the different methods.

Discussion

A discussion followed the Sanvenero Rosselli report, during which participants asked about the functional results and risk of infection. One person warned against the dangers of anesthesia performed with Avertin, while another asked whether it had been necessary to de-epitheliaze the margins of the cleft, because that moment in surgery was not shown on the film. The author replied that the functional results could generally be considered good and some excellent, provided that the operation was performed at a not advanced age and particularly if the type of intervention had been chosen in accordance with the degree of deformity. One sole intervention for all the different forms of cleft palate cannot be proposed; the procedure chosen must be dictated by the type and degree of deformity. The lateral incision must be performed very close to the cervix of the teeth to avoid lesion of the artery and to obtain proportionally greater flaps. The author never experienced problems with the well controlled use of Avertin. He employed very low doses, just sufficient to obtain a basal narcosis, free of dangers even if one has to add an inhalation of ether to obtain the necessary complete relaxation of the patient.

Comment

Gustavo Sanvenero Rosselli (died in 1974) trained as an otolaryngologist [10, 15]. There is some confusion about his date of birth: the British journal of Plastic and Reconstructive Surgery, in his obituary of Sanvenero Rosselli, gives 1887, whereas the Journal of Plastic and Reconstructive Surgery mentions 1897 as his year of birth. In the years following World War I, he became interested in facial reconstructive surgery. In this era, specialised centers were established in most countries, especially those that had been involved in the war [4]. He was also a member of the editorial board of the Revue de Chirurgie Plastique, the first international journal on plastic and reconstructive surgery. In the first issue, published in 1931, a paper of his was included on the 'Padiglione Mutilati del Viso' (Pavilion for the facially mutilated) in Milan, of which he was the director. In this hospital, casualties of the war, with their maxillofacial defects, and patients with congenital malformations were treated [17].

The superiorly based posterior pharyngeal wall flap is usually attributed to him. He gave a new impetus to its current widespread use, after a period in which the pharyngeal flap operation had been regarded as an unphysiological procedure. In Germany, this view was verbalized by Ernst [6] and Kirschner [9], who propagated instead the 'push-back' or 'retropulsion' procedure. In this controvery, the opponents were Sercer [25], who like Sanvenero Rosselli propagated the superiorly based pharyngeal flap, and Wassmund [30], who adopted the inferiorly based pharyngeal flap. Sanvenero Rosselli published a series of papers on the pharyngeal flap. Usually, authors refer to: 'Divisione palatina e sua cura chirurgica' [19]. This citation classic is in fact the summary of a lecture, with discussion afterwards, as presented at the Second International Congress on Stomatology, held April 14-19, 1935, in Bologna, Italy. This summary deals with the above mentioned controversy. As it is written in Italian and French and is rather difficult to obtain, we present here a translation into English. A more extensive and illustrated description of this type of flap surgery and its application can be found in the 1932 issue of Archivi Italiani di Laringologia entitled: 'Chirurgia plastica ed otorinolaringoiatria' [18]. The illustration of the surgical technique (Fig. 1) is derived from a paper published by Sanvenero Rosselli in Plastica Chirurgica, entitled: 'Scienza ed arte di chirurgia plastica. Stato attuale ed avvenire' [21] (Science and art of plastic surgery. Present state and future).

His first publication in English on the pharyngeal flap can be found in 1936 in the Revue de Chirurgie Structive, formerly Revue de Chirurgie Plastique entitled: 'De palati congenita fissura chirurgice restituenda' [20] (About the surgically reconstructed congenital cleft palate). This paper has an English text with a Latin title and subheadings. His first publication in the German literature can be found in 1955 in the Fortschritte der Kiefer-und Gesichtschirurgie: 'Verschluss von Gaumenspalten unter Verwendung von Pharynxlappen' [22] (Closure of cleft palates by using pharyngeal flaps). The name of Sanvenero Rosselli is usually associated with the superiorly based pharyngeal flap.

However, the first description of the inferiorly based pharyngea1 flap operation can be found in German literature. Dr Karl W.E.J. Schoenborn, who was Professor and chief of the surgica1 department at the University of Koenigsberg (East Prussia), presented a lecture 'Ueber eine neue Methode der Staphylorrhaphie' on the fourth congress of the German Society for Surgery in Berlin in 1875. This lecture was published in Archiv für Klinische Chirurgie 1876 and in Verhandlungen der Deutschen Gesellschaft für Chirurgie 1875 [23]. Schoenborn refers to Passavant, who described a decade before a method to reduce velopharyngeal aperture, in 'Ueber die Beseitigung der naeselnden Sprache bei angeborenen Spalten des harten und weichen Gaumens' in Archiv für Klinische Chirurgie 1865 [12].

Moreover, Schoenborn introduced the superiorly based pharyngea1 flap in 'Vorstellung eines Falles von Staphyloplastik' on the fifteenth congress of the German Society for Surgery in Berlin in 1886. The presentation was published in Verhandlungen der Deutschen Gesellschaft für Chirurgie 1886 [24].

These publications by Schoenborn have been translated in English by R. Stellmach and have been published in Plastic and Reconstructive Surgery 1972 as: 'The classic reprint. On a new method of staphylorraphy' and 'Presentation of a patient after staphyloplasty' [26]. The pharyngeal flap operation seems to fall into disuse until Wolfgang Rosenthal reintroduced Schoenborn's inferiorly based pharyngeal flap in 'Zur Frage der Gaumenplastik' in Zentralblatt für Chirurgie 1924 [16]. The reintroduction of the superiorly based pharyngeal flap is as early as 1935 by Sercer in 'Beitrag zur Technik der operativen Therapie der Rhinolalia aperta' in Revue de Chirurgie Structive 1935 [25] and Sanvenero-Rosselli in 'De palati congenita fissura chirurgice restituenda' in Rev Chir Struct 1936 [20], but have been described earlier by Sanvenero-Rosselli in Archivi Italiani di Laringologia 1932 [18] in 'Chirurgia Plastica ed Otorinolaringoiatria'.

In the United States the pharyngeal flap operation was introduced in 1929 by Padgett in 'The repair of cleft palates after unsuccessful operations, with special reference to cases with an extensive loss of palatal tissue' in Archives of Surgery 20 [13]. Padgett described the inferiorly based as well as the superiorly based posterior pharyngeal wall flap.

Not all cleft lip and palate surgeons became convinced of the merits of the flap operation. Veau in Paris [29] used the superiorly based posterior pharyngeal wall flap in 3 cases, but the flap did not live up to his expectations, and he renounced this practice.

Complications have been reduced considerably since the first description of the pharyngeal flap. In 1886 Schoenborn reported one death caused by septic pneumonia and 3 flap failures as a result of necrosis in a series of 20 patients treated with the superiorly based flap. Wassmund (Berlin) [30] and Rosenthal (Berlin, Thallwitz) have seen deaths in children, while Pichler (Vienna) and Trauner (Graz) [14] and also Padgett (USA) observed fatal complications in adults. Kindler (Graz) [8] described the fatal course of a mediastinitis as a complication of an inferiorly based pharyngeal flap. In 1990, in a series of 31 patients treated with the superiorly based pharyngeal flap, Tharanon et al. [27] reported no immediate postoperative life-threatening complications but 4 patients with late postoperative complications, including 3 flap dehiscences. Improved surgical techniques but most of all anesthetic and antimicrobial management and postoperative care contributed to this amelioration. Regarding anesthetic techniques, some surgeons performed the inferiorly based pharyngeal flap operation under local anesthesia [16], to avoid the risk of general anesthesia. Schoenborn performed the inferiorly as well as the superiorly based pharyngeal flap operation after a tracheostomy with introduction of a cuffed tube and package of the trachea had been performed. Chloroform was used as the anesthetic agent. This general anesthesia method was described by Trendelenburg in 1871 [28]. As early as 1903, a workable flexible tube for peroral use became available in Germany [7]. Avertin (Tribomethanol) was synthesized in 1923 and first employed clinically in 1926 [1, 3]. In Europe, it has been used as a basal anesthetic agent in cleft lip and palate surgery for a number of years [2, 5, 11]. It can be used in combination with ether (as Sanvenero Rosselli did) and with local anesthesia. Many surgeons have been involved in the development of pharyngeal flap surgery and, although Sanvenero Rosselli's contribution has definitely been of major importance for the acceptance of pharyngeal flap surgery, we thought it appropriate to highlight two aspects: (1) Schoenborn's contribution to pharyngeal flap surgery which in fact preceded Sanvenero Rosselli's work by almost 50 years, and (2) Sanvenero Rosselli's original publication on which the references in the literature are based.

References

1. Adriani J. The chemistry and physics of anesthesia. 2nd edn. Charles C Thomas, Springfield, 1962; pp 321-323.

2. Axhausen G. Die allgemeine Chirurgie in der Zahn-, Mund- und Kieferheilkunde. JF Lehmanns, Muenchen, 1940; pp 178- 180

3. Collins VJ Principles of anesthesiology. 2nd edn. Lea & Febiger, Philadelphia, 1976; pp 526-528.

4. Converse JM. Plastic surgery: the twentieth century. The period of growth (1914-1939). Surg Clin North Am 1968; 47: 261-278.

5. Eckstein A. Rückblick auf das 1. Internationale Hamburger Symposium über Lippen-Kiefer-Gaumenspalten 1954. In: Lippen-Kiefer-Gaumenspalten. Behandlungskonzepte- Spätergebnisse, Teamwork und Fürsorge-Teratologie. 3. Internationales Symposium, Hamburg. Thieme, Stuttgart, 1982: pp 4-9.

6. Ernst F. Zur Frage der Gaumenplastik. Zugleich kritische Bemerkungen zur gleichnamigen Arbeit W. Rosenthal's in Nr 30, 1924 dieser Zeitschrift. Zbl Chir 1925; 9: 464-470.

7. Hoffmann-Axthelm W. Die Geschichte der Mund-, Kiefer- und Gesichtschirurgie. Quintessence, Berlin, 1995; pp 89-90, 115,207.

8. Kindler W. Zur Gefaehrlichkeit der Gaumenplastik nach Schoenborn-Rosenthal sowie nach Ernst-Halle und zur Moeglichkeit, ihr vorzubeugen. Passow-Schaefer Beitraege zur Anatomie, Physiologie, Pathologie und Therapie des Ohres, der Nase und des Halses 1929; 27: 187-192.

9. Kirschner M. Zur Operation der Gaumenspalte. Arch Klin Chir 1925; 138: 515-533.

10. Matthews D. Obituary Gustavo Sanvenero Rosselli. Br J Plast Surg 1974; 27: 297-298.

11. Oettle E. Avertin und Evipan in der chirurgischen Praxis. Zbl Chir 1935; 19: 1104-1110.

12. Passavant G. Ueber die Beseitigung der naeselnden Sprache bei angeborenen Spalten des harten und weichen Gaumens. Arch Klin Chir 1865; 6: 333-349.

13. Padgett EC. The repair of cleft palates after unsuccessful operations with special reference to cases with an extensive loss of palatal tissue. Arch Surg 1929; 20: 453-472.

14. Pichler H, Trauner R. Mund- und Kieferchirurgie. Urban & Schwarzenberg, Vienna, 1948 p 835.

15. Rogers BO. Obituary Gustavo Sanvenero Rosselli. Plast Reconstr Surg 1974; 54: 513-515.

16. Rosenthal W. Zur Frage der Gaumenplastik. Zbl Chir 1924; 30: 1621-1627.

17. Sanvenero Rosselli G. Une clinique de chirurgie plastique. Le "Pavillon des Mutilés de la Face" à Milan. Rev Chir Plast (Bruxelles) 1931; 1: 22-31.

18. Sanvenero Rosselli G. Chirurgia plastica ed otorinolaringoiatria. Archivi Italiani di Laringologia 1932; 51: 301-324.

19. Sanvenero Rosselli G. Divisione palatina e sua cura chirurgica. Atti di II Congresso Internazionale di Stomatologia, Bologna, April 14-19, 1935; pp 391-392.

20. Sanvenero Rosselli G. De palati congenita fissura chirurgice restituenda. Rev Chir Struct 1936; 6: 413-419.

21. Sanvenero Rosselli G. Scienza ed arte di chirurgia plastica. Stato attuale ed avvenire. Plast Chir 1939; I: 3-28.

22. Sanvenero Rosselli G. Verschluss von Gaumenspalten unter Verwendung von Pharynxlappen. In: Schuchardt K, Wassmund M (eds)

Fortschritte der Kiefer- und Gesichts-Chirurgie. Vol 1. Thieme, Stuttgart, 1955; pp 65-69.

23. Schoenborn KWEJ. Ueber eine neue Methode der Staphylorrhaphie. Verh Dtsch Ges Chir 1875; 4: 235-239; Arch Klin Chir 1876; 19: 527-531.

24. Schoenborn KWEJ. Vorstellung eines Falles von Staphyloplastik. Verh Dtsch Ges Chir 1886; 15: 57-62.

25. Sercer A. Beitrag zur Technik der operativen Therapie der Rhinolalia aperta. Rev Chir Struct (Bruxelles) 1935; 5: 5-18.

26. Stellmach RK. The classic reprint: on a new method of staphylorraphy, and presentation of a patient after staphyloplasty. Plast Reconstr Surg 1972; 49: 558-562.

27. Tharanon W, Stella JP, Epker BN. The modified superior based pharyngeal flap. Part III. A retrospective study. Oral Surg Oral Med Oral Pathol 1990; 70: 256-267.

28. Trendelenburg F. Tamponnade der Trachea. Arch Klin Chir 1871; 12: 121-133.

29. Veau V. Division Palatine. Anatomie- Chirurgie-Phonétique. Masson, Paris, 1931; pp 278-279.

30. Wassmund M. Lehrbuch der praktischen Chirurgie des Mundes und der Kiefer. Vol 2. JA Barth, Leipzig, 1939; pp 475-484.

Chapter 3

Revisional surgery following the superiorly based posterior pharyngeal wall flap.

Historical perspectives and current considerations

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Abstract.

The aim of this study was to describe the surgical and functional complications following superiorly based posterior pharyngeal wall (SBPP) flap surgery. Records of 130 patients with velopharyngeal insufficiency (VPI) who had undergone SBPP flap surgery as a secondary procedure to reduce nasal resonance in speech were reviewed. Complications were defined as the incidence of revisional surgery required to obtain a more satisfactory result. 20 patients (15%) required revisional surgery. In 4 patients (3%) early revisional surgery was indicated to treat surgical complications (1 postoperative bleeding, 3 flap dehiscences). In 16 patients (12%) late revisional surgery was indicated to achieve a better functional result with regard to nasal resonance in speech. The low incidence of surgical complications indicates that SBPP flap surgery is a safe procedure. After SBPP flap surgery, a satisfactory functional result with respect to nasal resonance was obtained in 88% of patients. This result was improved after revisional surgery. The hypothesis that the patients of an experienced surgeon have fewer complications and better functional results than those of a less experienced one was tested. The individual skill of the surgeon rather than their experience led to a better functional result.

The superiorly based posterior pharyngeal wall flap (SBPP flap) is widely used to treat velopharyngeal insufficiency (VPI) in patients with cleft palate, who have undergone primary closure of the palatal cleft and still have perceived nasal resonance in speech[38]. VPI occurs in 20-30% of patients with a repaired cleft palate[19,23,25]. Traditionally, the assessment of nasal resonance and the indication for pharyngeal flap surgery has relied on perceptual judgments, but this is subjective, making its reliability questionable[15]. For diagnosing VPI and evaluating therapy, instruments that measure nasal resonance, such as the Nasometer® (Kay Elemetrics Corp, New Jersey, USA) and the Nasalview® (Tiger Electronics, Seattle, USA), are gaining in popularity, but the correlation between these objective measures and the subjective perceptual rating of nasal resonance is moderate[17,18]. Most authors agree that perceptual evaluation serves as the gold standard against which instrumental measurements must be validated. Until now, the trained ear has been the primary tool for determining the presence and severity of VPI[30] and is the final arbiter on determining the functional result with regard to nasal resonance.

Today, imaging techniques such as (video)fluorography and (video)endoscopy are usually part of the diagnostic process. Encouraging results have been obtained, but the interpretation of such imaging is subjective[30,31]. As an alternative, the incidence of revisional surgery can be used as a measure of the functional outcome of SBPP surgery[8.19,23]. The aim of this study is to evaluate the surgical and functional complications of SBPP flap surgery, by scoring the incidence of revisional surgery needed to obtain a more satisfactory result. The hypothesis that the patients of experienced surgeons have fewer complications and better functional results was tested.

Patients and methods

The medical records of 130 patients with VPI who underwent a SBPP flap operation to improve nasal resonance in speech were reviewed. All patients underwent surgery in the Department of Oral and Maxillofacial Surgery of the University Medical Centre of Utrecht (the Netherlands) from 1985 to 2000.

Physical examination and speech evaluation

Patients were examined before and after the flap operation by an oral and maxillofacial surgeon and a qualified speech-language pathologist, both with expertise in cleft palate speech. Spontaneous speech and standard test passages were recorded on tape and perceptually evaluated on: nasal resonance; compensatory misarticulations; audible nasal emission; intelligibility and overall grade of severity. The /a/-/i/ test[7] was used to determine whether there was nasal resonance. The patients were asked to produce a series of /a/ and /i/ sounds alternately with the nares open and closed. To quantify the nasal emission the fogged-mirror test was used, the degree of condensation on a cold mirror held 0.5 cm under the nose, during phonation of vowels and consonants was assessed. The movement of the lateral pharyngeal wall and the soft palate was assessed by transoral examination. The decision to perform SBPP flap surgery was based on these clinical parameters. In some patients, a pharyngogram was made to reach agreement between surgeon and speech-language pathologist. No other instrumental measures were used routinely.

Surgery

The posterior pharyngeal wall and the soft palate were infiltrated with local anaesthesia with a vasoconstrictor, to minimize blood loss and to allow hydrodissection of the tissue layers. The oral and the nasal mucosa of the soft palate was opened longitudinally along the midline, over a distance of about three-quarters of the length of the velum, starting at the tip of the uvula.

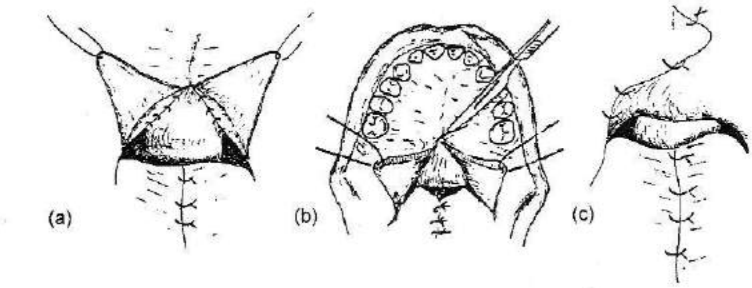


Fig. 1. (a) Sutures placed for antero-lateral traction of the oral mucosal layer of the surgical divided soft palate. SBPP flap sutured in the anterior and the lateral part of the divided nasal mucosal layer. (b) Complete closure at the top of the donor site is not attempted, safeguarding the vascularization of the SBPP flap. The knife is pointed in the direction of the Z-plasty to mobilize the oral mucosal layer to cover the raw surface of the SBPP flap. (c) Operation site in the end stage to illustrate the lateral ports obtained and the Z-plasty to cover most of the raw surface of the SBPP flap.

The oral and nasal layers of the velum were separated with scissors. The posterior pharyngeal arches were opened in the longitudinal direction for at least one-half of their length. Sutures were placed bilaterally through the velum for antero-lateral traction (Fig. la). The posterior pharyngeal wall flap was raised as a composite flap just superficial of the prevertebral fascia, containing both mucosa and superior constrictor muscle. The base of the flap was positioned above the level of the hard palate plane, up to the level of the Eustachian tubes. The flap was raised to a width two-thirds of the width of the posterior wall, to a point that yields a flap that can be sutured without tension in the anterior part of the surgically divided velum. After haemostasis the donor site defect at the posterior pharyngeal wall was approximated with resorbable sutures. Complete closure in the cranial part was not attempted to safeguard the vascular flow in the flap. The flap was sutured into the nasal mucosal layer of the velum (Fig. la). To control the diameter of the remaining openings at both sides of the flap, the so-called lateral ports, flexible catheters with a diameter of 4 mm (French 14), were inserted through each nostril and directed into the pharynx.

The flap was sutured around the catheters, securing a postoperative diameter of 4 mm of the lateral ports. The oral mucosa of the velum was lengthened by means of a releasing incision yielding an atypical "Z"-plasty to cover the wound surface of the pharyngeal flap in order to prevent, or decrease, its tubing (Fig. lb, 1c). Both catheters were removed. Immediate postoperative airway obstruction is always a risk[5], so a 1.0 non-resorbable suture was placed through the tongue to allow anterior traction if necessary. Peri-operative antibiotic prophylaxis was used routinely. To evaluate the function of the flap, a suction catheter was inserted into the nose and positioned above the velum (suction test)[2]. By occluding both nostrils an underpressure was generated. The flap was considered to be functional if lifting of the velum was observed. If not, the lateral ports were tightened by additional suturing.

Revisional surgery

The incidence of revisional surgery was used as a measure to describe the surgical and functional complications. In case of surgical complications, such as postoperative bleeding or dehiscence of the flap, early revisional surgery was performed.

If formal speech therapy following the flap operation did not result in functional improvement, late revisional surgery was performed at the earliest 18 months after the SBPP flap operation. In cases of persistent hypernasality, the lateral ports were tightened, and in cases of severe hyponasality, the stem of the flap was divided.

Statistics

The following potential risk factors for revisional surgery were analysed: gender, cleft type, age at the time of the initial cleft palate repair, interval between cleft palate repair and pharyngeal flap operation, and associated congenital syndromes. The patients were classified as unilateral cleft lip and palate (left or right), bilateral cleft lip and palate, isolated cleft palate, or VPI of non-cleft origin.

Descriptive statistics were carried out to determine the distribution of factors as gender, age, type of cleft palate repair, early and late complications of the SBPP flap, revisional surgery, interval between cleft palate repair and pharyngeal flap, and surgeons (SPSS 11.5; SPSS, Chicago, IL, USA). Logistic regression was used to determine factors that influence the number of late complications of the flap operation. The χ^2 -test was used to determine if the occurrence of surgical and functional complications was equally divided over all patient groups (unilateral cleft lip and palate (left or right), bilateral cleft lip and palate, isolated cleft palate and VPI of non-cleft origin).

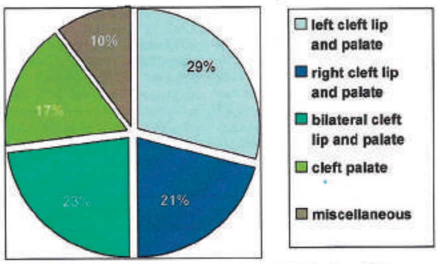


Fig. 2. Distribution of patients in the study sample (n = 130) based on cleft type.

Results

There were 78 male (60%) and 52 female (40%) patients. The mean age at the time of the SBPP flap surgery was 18.8 years (range 4 years 6 months to 61 years 6 months). Of the 130 patients, 65 (50%) had a unilateral cleft, 38 (29%) on the left and 27 (21%) on the right hand side. In 30 patients (23%) a bilateral cleft was found and 22 (17%) patients had an isolated cleft palate (Fig. 2).

A miscellaneous group consisted of 13 patients (10%) in whom VPI existed due to other causes, such as congenital short soft palate, velo-cardiofacial syndrome, or primary VPI without cleft related diagnosis. Logistic regression showed that gender, type of cleft, type of cleft palate repair, age at the time of palate repair, age at the time of SBPP flap surgery, and interval between cleft palate repair and pharyngeal flap had no significant influence on the incidence or the type of complication. The occurrence of surgical and functional complications was equally divided (χ^2 -test; $p= 0.34$) over all patient groups. In 20 of the 130 patients (15%) revisional surgery was performed. Early revisional surgery to treat surgical complications was required in 4 patients: in one (1%) due to postoperative bleeding, and in 3 (2%) due to dehiscence of the flap. In 16 patients (12%) the functional result regarding nasal resonance was considered unsatisfactory after speech therapy for 12-18 months and late revisional surgery was required. Tightening of the lateral ports was carried out to treat persistent hypernasality in 7 patients (5%). In 9 patients (7%) the stem of the flap was divided, in 7 patients due to persistent hyponasality and in 2 patients to treat severe snoring with obstructive symptoms and concomitant hyponasality (Fig. 3).

In most patients (n=88) only a SBPP flap operation was performed. In 32 patients, one additional cleft-related operation was carried out simultaneously including a correction of the external (n= 5) or internal nose (n= 5), the vermillion of the lip (n= 8), closure of a persistent oro-nasal fistula in the anterior part of the palate (n= 10) and repair of the alveolar cleft with a bone graft (n= 4). In 10 patients the SBPP flap operation was combined with two additional operations. Snoring was only scored if mentioned by the patient (n= 18; 14%).

No statistical difference was observed between the number of operations and the occurrence of a functional complication.

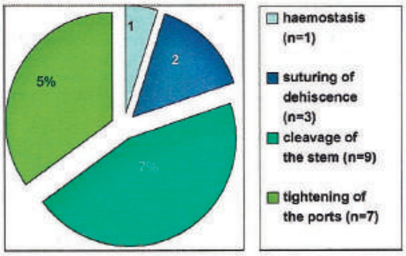


Fig. 3. Distribution of revisional surgery (n = 20) due to early (n = 4) and late complications (n = 16) in 130 patients.

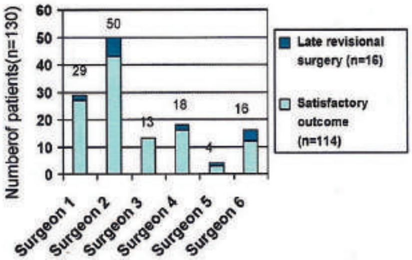


Fig. 4. Number of flap operations and revisional surgery per surgeon.

In Fig. 4 the occurrence of functional complications is related to the number of pharyngeal flap operations performed per surgeon. The most experienced surgeons (surgeons 1, 2) performed 79 pharyngeal flap operations of which 9 (11%) needed revisional surgery. The least experienced surgeons (surgeons 3, 4, 5, 6) performed 51 pharyngeal flaps, of which 7 (14%) needed revisional surgery. The patients of surgeon 3, in the latter group, had no complications.

Discussion

In the nineteenth century and at the beginning of the twentieth century, the pharyngeal flap operation caused severe complications[16] owing to primitive anaesthetic techniques and postoperative care, as well as the lack of antimicrobial management. Schoenborn[28] reported one death from pneumonia and 3 flap failures as a result of necrosis in a series of 20 patients treated with SBPP flap surgery. Owsley and

Blackfield[22] reported postoperative bleeding in 8% of their patients, while Nylen and Wahlin[21] reported it in 14% of their patients. Graham et al.[6], in a study comprising 109 superiorly based and 98 inferiorly based pharyngeal wall flaps, reported postoperative bleeding in 7 patients, major airway obstruction in 8 patients (7 requiring tracheotomy), flap separation in 18 patients, and the death of 1 patient as major complications. Antibiotic prophylaxis was not used routinely in their study. Valnicek et al.[35] reported postoperative bleeding in 8% of patients. In their study, comprising 219 patients, 9% of the patients developed airway obstruction and one patient died. After having optimized their postoperative procedures, Fraulin et al.[5] reported a reduction in postoperative bleeding to 1% and airway obstruction to 3%. In the present study, one patient (1%) had postoperative bleeding following SBPP flap surgery, while bleeding occurred following revisional surgery in another patient. In agreement with the study by Abyholm et al.[1], it may be concluded that the SBPP flap operation is a safe procedure. In cases of bleeding, the donor region of the flap at the posterior pharyngeal wall and the raw surface of the flap are easily accessible due to the design of the SBPP flap. In the present study no patients with airway obstruction were observed, but optimal postoperative surveillance remains mandatory. In the present study, revisional surgery to treat functional complications was indicated in 16 patients (12%). Barot et al.[3] reported similar results, while Witt et al.[40] reported a slightly higher percentage (20%) of revisional surgery. Three causes can be identified for the unsatisfactory functional outcome with respect to nasal resonance. The first two causes, the width of the flap at the time of insertion and the subsequent transformation of the flap into a tube-like structure as a result of shrinkage, are interrelated. The width of the flap is decreased by secondary healing. It seems logical to prepare the flap as wide as possible to counteract the effect of shrinkage[37], but it is impossible to predict the amount of shrinkage and the ultimate width of the flap, so unfavourable effects in either direction (too wide or too narrow) can be expected. If the flap is too wide, hyponasality and obstructed nasal breathing, is induced. In cases of severe tubing of the flap, it is too narrow to achieve sufficient velopharyngeal closure during phonation, thereby causing persistent VPI. Tubing of the flap may be prevented by covering its raw surface, thereby inducing primary healing. To achieve this, several modifications have been proposed. Isshiki and Morimoto[12] folded the flap on itself with the mucosa outside, while Owsley and Blackfield[22] used 'turn back flaps' obtained from the nasal side of the velum to cover the raw surface of the flap. Alternatively, the oral side of the velar mucosa may be lengthened by a releasing incision[4], as was performed in this study. By lining the raw surface of the flap and approximating the mucosa of the posterior pharyngeal wall at the donor site, the operative field is converted into a closed wound. Thereby, primary wound healing is enhanced and the risk of postoperative bleeding is reduced. It may be expected that the lined flap will maintain its size and that the shape of the lateral ports, as designed during the operation, will be stable. The fact that 16 patients (12%) in this study required revisional surgery, underscores that this approach also has its limitations. The third and most decisive parameter for improved speech outcome after flap surgery is the extent of lateral pharyngeal wall movement. In most patients, contraction of the superior constrictor pharyngeus muscle is able to effectively close lateral ports of up to $10mm^2$ on both sides of the flap during phonation. As stated by Hogan[10], who introduced the concept of lateral port control (LPC), this mechanism eliminates nasal resonance during conversational speech. There is no consensus on the concept of LPC. Shprintzen et al.[29] advocated 'tailormade' flaps, suggesting that the width of the flap is determined by the extent of the medial movement of the lateral pharyngeal wall assessed during preoperative videofluoroscopy and nasopharyngoscopy. Swanson et al.[34] reported changes in velopharyngeal valving patterns after pharyngeal flap surgery, suggesting that preoperative lateral wall motion may not always be predictive for postoperative movement. Hall et al.[9] emphasized that pharyngeal flap surgery frustrated the movements of the lateral pharyngeal walls. Vandevoort et al.[36] rejected the concept of 'tailor-made' flaps by arguing that the degree of tubing and subsequent shrinkage is not predictable but a random process. Consequently, tailoring the flap and controlling the size of the ports is at best a 'hit and miss' procedure. They hypothesize, that the success of the velopharyngeal flap procedure, with respect to nasal resonance in speech, suggests that the degree of shrinkage may be less crucial, as long as, due to medial movement, the lateral pharyngeal walls move well enough to close against the flap. This phenomenon of tubing and shrinkage also explains that various surgical modifications show no difference in speech outcome. For example, with respect to the width of the flap at rest and speech outcome, there is no difference between a superiorly based flap merged into a transversely split velum compared with a superiorly based flap merged into a velum split into the midline[14]. Although the SBPP flap is regarded as a well established procedure, the most appropriate surgery for the management of VPI is debated. Alternative procedures to avoid pharyngeal flap operations have been proposed to treat persistent VPI after primary cleft palate repair. Some authors[33] perform a palate re-repair, aiming to restore normal anatomy. By dissection and retro-positioning of the velar muscles the velum is lengthened as a more physiological procedure. After palate re-repair, hyponasality was found in 5% and persistent hypernasality in 12% of patients. In contrast, the authors noted 7% hyponasality and 5% hypernasality following the SBPP flap operation. Sommerlad et al.[33] performed a sphincterpharyngoplasty[11,13] to treat persistent hypernasality after palate re-repair. Successful outcome with respect to nasal resonance of the sphincter-pharyngoplasty ranged from 67% to 85%[26]. Both sphincterpharyngoplasty and pharyngeal flap operations require revisional surgery to treat unsatisfactory functional results in about 15-20%[40]. In the present study, 20 patients (15%) mentioned persistent snoring following SBPP flap surgery. In 2 of these patients the stem of the flap was divided because of obstructive symptoms, such as severe snoring and concomitant hyponasality. The outcome regarding snoring may be underscored in view of a previous study[20] in which frequent or occasional snoring was reported in 89% of the patients. Except for these two patients in whom the stem of the flap was divided, the authors did not specifically pay attention to snoring at that time. Nowadays, however, snoring is regarded as a serious sign, as clinicians are more aware of obstructive sleep apnoea syndrome (OSAS). Snoring and OSAS are different sleep-related breathing disorders, although there is only a quantitative difference between them[27]. According to current opinion, polysomnographic investigations should have been considered to rule out or confirm OSAS. Obstructive sleep symptoms are more frequently associated with posterior pharyngeal flap surgery[27], but many authors consider this flap to be effective in reducing nasal resonance due to VPI, particularly in severe cases[32].

In the literature, whether preoperative measuring of the nasal resonance provides objective information is discussed. Many authors argue that these examinations are mandatory because they provide objective and reliable information on which operative choices can be based. Others state that the correlation between these objective measures and the subjective perceptual rating of nasal resonance is only moderate[17,18].

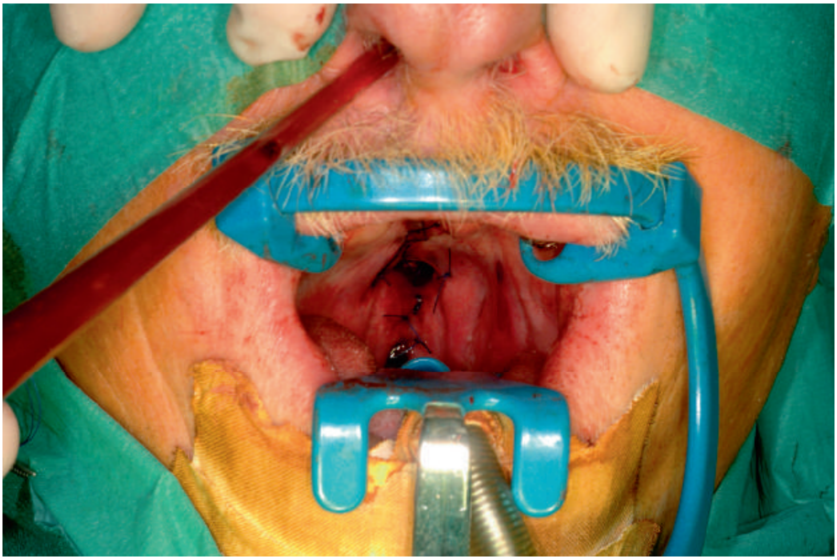
The interpretation of imaging techniques, such as (video) fluorography and (video)endoscopy is subjective[30,31]. Skolnick and Cohn[31] state that the conduct of videofluoroscopic studies are as much an art as a science. Sie et al.[30] argue that in the absence of an objective, quantifiable measure of VP function, it is important to understand the reliability of clinical tools used in the treatment of patients with VPI. In their study on reliability in the endoscopic evaluation of VPI, they found that the inter-observer reliability of rating qualitative characteristics is variable and is too low to be used for comparing subjects across centres.

Preoperative diagnostic imaging was not performed in the patients in the present study, but the functional results are comparable with studies in which such imaging was used[3,26,41]. This indicates that factors other than preoperative information about the movement of the pharyngeal walls and the velum, such as unpredictable wound healing, are important in obtaining a successful outcome. In view of the present concept of a multidimensional diagnostic process, these examinations should be included in the work-up, provided that clinicians are aware of the limitations and value of the information obtained.

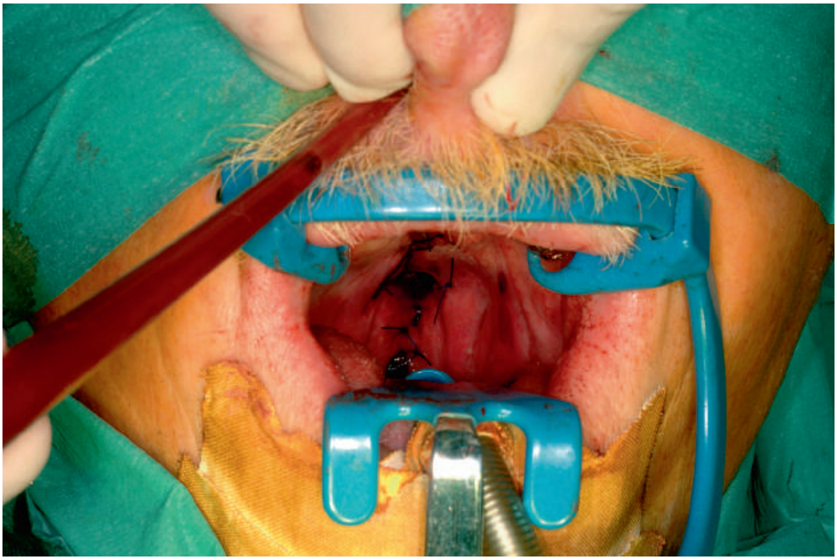
It is recommended that the intraoperative suction test is performed to mimic the dynamics of the structures involved in speech production. The easier the velum lifts during the suction test, the more improvement regarding hypernasality can be expected[33].

Surgical inexperience as a factor responsible for failure has been identified. Despite a 'learning curve', the surgeon's experience plateaus quickly[39]. The learning curve is responsible for a disproportional number of failures early in the surgeon's career; its effect soon disappears after a steady state of experience is reached. In the present study, experienced surgeons (Fig. 4; surgeons 1, 2) showed slightly fewer patients with a functional complication than less experienced ones (Fig. 4; surgeons 3, 4, 5, 6). Surgeon 3 in the less experienced group had no complications, while the most experienced surgeon had a rate of 14%. The individual skill of the surgeon as well as the experience is important in the prevention of complications. This finding is in agreement with a previous study[24].

In conclusion, the SBPP flap operation is an established procedure to treat nasal resonance in speech of patients with VPI. It is a straightforward and safe procedure. The functional results, as described in this study, are in accordance with the current literature. The outcome with respect to nasal resonance is not always predictable, mainly due to unpredictable wound healing; the lateral ports were either too wide or too tight in 12% of patients. After revisional surgery a more satisfactory functional result regarding nasal resonance was achieved. The findings suggest that the individual skills of the surgeon, rather than the experience, yields a better functional outcome.



Suction test, suction catheter inserted into the nose



Suction test, by occluding both nostrils an underpressure is generated. Lifting of the velum may be observed.

1. Abyholm F, D'Antonio L, Davidson Ward SL, Kjøll L, Saeed M, Shaw W, Sloan G, Whitby D, Worhington H, Wyatt R. VPI Surgical Group. Pharyngeal flap and sphincterplasty for velopharyngeal insufficiency have equal outcome at 1 year postoperatively: results of a randomized trial. Cleft Palate Craniofac J 2005; 42: 501-511.

2. Baker S, Millard DR. Intraoperative suction test as a predictor of velopharyngeal competence. Cleft Palate Craniofac J 1993; 30: 452-453.

3. Barot LR, Cohen MA, Larossa D. Surgical indications and techniques for posterior pharyngeal flap revision. Ann Plast Surg 1986; 16: 527-531.

4. Blijdorp P, Müller H. The influence of the age at which the palate is closed on speech in the adult cleft patient. J Max Fac Surg 1984; 12: 239-246.

5. Fraulin FOG, Valnicek SM, Zuker RM. Decreasing the perioperative complications associated with the superior pharyngeal flap operation. Plast Reconstr Surg 1998; 102: 10-18.

6.Graham 3rd WP, Hamilton R, Randall P, Winchester R, Stool S. Complications following posterior pharyngeal flap surgery. Cleft Palate J 1973; 10: 176-180.

7. Gutzmann H. Untersuchungen ueber das Wesen der Nasalitaet. Arch Laryngol Rhinol 1913; 27: 59-125.

8. Härtel J, Gundlach KKH, Ruickoldt K. Incidence of velopharyngoplasty following various techniques of palatoplasty. J Craniomaxillofac Surg 1994; 22: 272-275.

9. Hall CD, Golding-Kushner KJ, Argamaso RV, Strauch B. Pharyngeal flap surgery in adults. Cleft Palate Craniofac Surg 1991; 28: 179-182.

10. 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.

11. Hynes W. Pharyngoplasty by muscle transplantation. Br J Plast Surg 1951; 3: 128-135.

12. Isshiki N, Morimoto M. A new folded pharyngeal flap. Preliminary report. Plast Reconstr Surg 1975; 55: 461-465.

13. Jackson IT. Sphincter pharyngoplasty. Clin Plast Surg 1985; 12: 711-717.

14. Karling J, Henningsson G, Larson 0, Isberg A. Comparison between two types of pharyngeal flap with regard to configuration at rest and function and speech outcome. Cleft Palate Craniofac J 1999; 36: 154-165.

15. Keuning KH, Wieneke GH, Dejonckere PH. The intra-judge reliability of the perceptual rating of cleft palate speech before and after pharyngeal flap surgery: The effect of judges and speech samples. Cleft Palate Craniofac J 1999; 36: 328-333.

16. Keuning KH, Crisi LM. Velopharyngoplasty according to Sanvenero Rosselli. Historical review and contemporary judgement. Mund Kiefer Gesichtschir 2000; 4: 95-98.

17. Keuning KH, Wieneke GH, Van Wijngaarden HA, Dejonckere PH. The correlation between nasalance and a differentiated perceptual rating of speech in Dutch patients with velopharyngeal insufficiency. Cleft Palate Craniofac J 2002; 39: 277-284.

18. Keuning KH, Wieneke.GH, Dejonckere PH. Correlation between the perceptual rating of speech in Dutch patients with velopharyngeal insufficiency and composite measures derived from mean nasalance scores. Folia Phoniatr Logop 2004; 56: 157-164.

19. Morris HL. Velopharyngeal competence and primary cleft palate surgery, 1960-1971: A critical review. Cleft Palate J 1973; 10: 62-71.

20. Morris HL, Bardach J, Jones D, Christiansen JL, Gray SD. Clinical results of pharyngeal flap surgery. Plast Reconstr Surg 1995; 95: 652-662.

21. Nylen B, Wahlin A. Post-operative complications in pharyngeal flap surgery. Cleft Palate J 1966; 3: 347-356.

22. Owsley Jr JQ, Blackfield HM. The technique and complications of pharyngeal flap surgery: A continuing report. Plast Reconstr Surg 1965; 35: 531-539.

23. Pryor LS, Lehman J, Parker MG, Schmidt A, Fox LMA, Murthy AS. Outcomes in pharyngoplasty: a 10-year experience. Cleft Palate Craniofac J 2006; 43: 222-225.

24. Rintala AE, Haapanen M-L. The correlation between training and skill of the surgeon and reoperation rate for persistent cleft palate speech. Br J Oral Maxillofac Surg 1995; 33: 295-298.

25. Riski JE. Articulation skills and oral-nasal resonance in children with pharyngeal flaps. Cleft Palate J 1979; 16: 421-428.

26. Riski JE, Rruff GL, Georgiade GS, Barwick WJ, Edwards PD. Evaluation of the sphincter pharyngoplasty. Cleft Palate Craniofac J 1992; 29: 254-261.

27. Rustemeyer J, Thieme V, Bremerich A. Snoring in cleft patients with velopharyngoplasty. Int J Oral Maxillofac Surg 2008; 37: 17-20.

28. Schoenborn KWEJ. Vorstellung eines Falles von Staphyloplastik. Verh Dtsch Ges Chir 1886; 15: 57-62.

29. Shprintzen RJ, Lewin ML, Croft CB, Daniller AI, Argamaso RV, Ship AG, Strauch B. A comprehensive study of pharyngeal flap surgery: tailor made flaps. Cleft Palate J 1979; 16: 46-55.

30. Sie KCY, Starr JR, Bloom DC, Cunningham M, De Serres LM, Drake AF, Elluru RG, Haddad J, Hartnick C, MacArthur C, Milczuk HA, Muntz HR, Perkins JA, Senders C, Smith ME, Tollefson T, Willging JP, Zdanski CJ. Multicenter interrater and intrarater reliability in the endoscopic evaluation of velopharyngeal insufficiency. Arch Otolaryngol Head Neck Surg 2008; 134: 757-763.

31. Skolnick ML, Cohn ER. Videofluoroscopic studies of speech in patients with cleft palate. New York, Berlin. Springer Verlag 1989; Preface.

32. Sloan GM. Posterior pharyngeal and sphincter pharyngoplasty: The state of the art. Cleft Palate Craniofacial J 2000; 37: 112-122.

33. Sommerlad BC, Mehendale FV, Birch MJ, Sell D, Hattee C, Harland K. Palate re-repair revisited. Cleft Palate Craniofac J 2002; 39: 295-307.

34. Swanson E, Witzel MA, Posnick JC. The effect of pharyngeal flap surgery on velopharyngeal closure. Nasendoscopic findings in 40 patients. Plastic Surg Forum XI, 142-144.

35. Valnicek SM, Zuker RM, Halpern LM, Roy WL. Perioperative complications of superior pharyngeal flap surgery in children. Plast Reconstr Surg 1994; 93: 954-958.

36. Vandevoort MJ, Mercer NS, Albery EH. Superiorly based flap pharyngoplasty: the degree of postoperative "tubing" and its effect on speech. Br J Plast Surg 2001; 54: 192-196.

37. Webster RC, Coffey RJ, Russell JA, Quigley LF. Methods of surgical correction of velopharyngeal sphincter incompetency using palatal and posterior pharyngeal tissues: proposed system of c1assification. Plast Reconstr Surg 1956; 18: 474-489.

38. Witt PD, D'Antonio L. Velopharyngeal insufficiency and secondary palatal management. A new look at an old problem. Clin Plast Surg 1993; 20: 707-721.

39. Witt PD, Marsh JL, Grames LM, Muntz HR. Revision of the failed sphincter pharyngoplasty: an outcome assessment. Plast Reconstr Surg 1995; 96: 129-138.

40. Witt PD, Myckatyn T, Marsh JL. Salvaging the failed pharyngoplasty: Intervention outcome. Cleft Palate Craniofac J 1998; 35: 447-453.

Chapter 4

The intrajudge reliability of the perceptual rating of cleft palate speech, before and after pharyngeal flap surgery. The effect of judges and speech samples.

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Abstract

Objective There is no agreement in the literature on the method nor the type of speech samples with which cleft palate speech should be perceptually evaluated. In addition, speech-language pathologists differ in the way they assess different aspects of cleft palate speech. In this pilot study, the reliabilities of the perceptual ratings of four types of speech samples by six judges, with and without expertise in evaluating cleft palate speech, were studied.

Design The pre- and postoperative tape-recordings of 15 patients with cleft lip and palate who had undergone a superiorly based pharyngeal flap operation were selected.

Five speech-language pathologists and one oral and maxillofacial surgeon perceptually rated the following variables on separate 100 mm visual analog scales:

1. hypernasality
2. audible nasal emission
3. intelligibility
4. misarticulations associated with velopharyngeal insufficiency
5. voice quality, and
6. the presence or absence of hyponasality

These six variables were rated in four types of speech samples: a. reading of three sentences; b. repeating after the speech pathologist of three sentences; c. ten sentences containing the afore-mentioned material; and d. the same ten sentences in paired comparison. All speech samples were re-rated, after 3 months, by the same judges.

Results Judges differ largely in the range they use in their rating. Intrajudge reliability of .56 to .78 was found for ratings of hypernasality. No significant differences in intrajudge reliability were found for the ratings with the different types of speech samples. The intrajudge reliability of a judge with expertise is not necessarily higher than of a judge without this expertise.

Conclusions The improvement in speech is most reliably assessed with speech samples in paired comparison. A speech-language pathologist with expertise in evaluating cleft palate speech does not guarantee a high intrajudge reliability of the rating.

Speech of patients with cleft palate is primarily characterized by abnormalities in nasal resonance. This is a direct result of velopharyngeal insufficiency (VPI). In addition, there may be articulation errors including compensatory misarticulations and a reduced voice quality. The final result is a reduction in intelligibility of speech.

The principal aim of physical or behavioral management in patients with cleft lip and palate (CLP) is to obtain intelligible speech of a pleasing quality. The perceived speech quality is the main criterion for management decisions, and the human ear is the final arbiter in determining whether a satisfactory result has been achieved.

Instrumental means for diagnosis and therapy evaluation in VPI are gradually being perfected and are gaining in popularity, because they may be more objective than perceptual evaluation. To date, no single instrument has proven to be an alternative for perceptual evaluation in clinical practice (McWilliams et al. 1990). Also, the examination should be practical and non-invasive, especially for use in children. Thus, it is generally accepted that perceptual evaluation is still the most appropriate standard against which instrumental measures must be validated (McWilliams et al. 1981, Dalston and Warren 1986, Haapanen 1991, Hirschberg and Van Demark, 1997).

Although most authors agree on the role of perceptual evaluation of cleft palate speech in diagnosis and therapy evaluation, there appears to be no agreement in the literature on the methodology to be used for a reliable rating. Methodological differences are found in the literature regarding the consistency of speech samples (Van Demark 1970, Carney and Sherman 1971), the expertise of judges (Fletcher 1976, Croatto 1984, Dalston and Warren 1986, Schmelzeisen 1992) and the grading of the severity of hypernasality in speech (Pommez and Rebufy 1987, Bzoch 1989, Karling et al. 1993). In the recent literature on perceptual (auditory) evaluation, a number of methodological issues have been addressed. These include the influence of the type of rating scales on the reliability of the judgment and statistical analysis of the data (Kreiman et al. 1993).

As a standardized perceptual description of cleft palate speech is important for clinical consideration and criteria validation as well as in research, further investigation of the factors that may affect the assessment is necessary.

This study was performed to determine a protocol for the rating of speech of a large series of CLP patients who had undergone a superiorly based posterior pharyngeal wall flap, according to Sanvenero Rosselli, modified by Tjebbes (Blijdorp and Müller, 1984).

The following parameters were investigated: (1) the effect of different types of speech samples and (2) the effect of the experience of the judges in evaluating cleft palate speech on the reliability of the rating. The reliability measures reflect the ratio between the variance of interest and the variance related to judgment inconsistencies.

The judges were chosen from a group of clinicians who were involved in the treatment of patients with CLP. The judges did not receive special training for this study. In the Netherlands, in most cleft palate teams, decisions for therapy are based on the assessment of speech by one speech-language pathologist. Therefore, the study will focus on the <u>intra</u>judge reliability determined for the different types of speech samples and the different judges. Of course, for comparison of results from different institutes, the <u>inter</u>judge reliability is an important aspect, but it was not the subject of this study.

Method

Judges

Six judges, 5 speech-language pathologists and 1 oral and maxillofacial surgeon, participated in this experiment. Two of the speech-language pathologists (judges 3 and 4) were general practitioners without special expertise in evaluating cleft-palate speech. Three were current members (judges 2, 5 and 6) of different cleft palate teams. The surgeon (judge 1) was trained in all aspects of cleft surgery, both primary and secondary.

Speech sample

From a library of tape recordings of CLP patients, pre- and postoperative recordings of 15 patients (6 boys, mean age 10y, 5m and 9 girls, mean age 13y at pharyngeal flap) were selected. These patients had received a superiorly based posterior pharyngeal wall flap, according to Sanvenero Rosselli, modified by Tjebbes, at the Department of Oral and Maxillofacial Surgery of the Utrecht University Hospital. Patients with palatal fistulas, those having non-CLP related speech disorders, those speaking a dialect, and those who had been subjected to other operations related to CLP, simultaneously to the pharyngeal flap operation, were excluded from this study. The speech was recorded using an AKG 190 E microphone, connected to an Uher SG 561 Royal tape recorder. Tape recordings had been made approximately six weeks before and 4-6 months after surgery. The hearing of all 15 patients was normal or close to normal.

Both pre- and postoperative tape recordings consisted of (1) seven sentences read by the patient, (2) three sentences repeated by the patient after the speech pathologist and (3) conversational speech. The phonemes in the sentences are representative of the distribution of phonemes in the Dutch language. The duration of the ten sentences was approximately one minute. A recording and its copy of the same sentences spoken by a girl (age: 11y, 1m) without a cleft and with normal speech and a girl (age: 16y, 6m) without a cleft and with a lateral lisp were included as controls. The selected speech recordings were digitized (40 kHz, 12-bit) by the Computerized Speech Laboratory (CSL, Kay Elemetrics Corp., type 4300), connected to a personal computer system (486 DX2, 66 MHz, 16 MB RAM). The sample frequency of 40 kHz was necessary to preserve all acoustic characteristics of the speech. After digitizing, the 'Edit'-function of CSL was used to remove all the text not spoken by the patient.

In order to study the effect of speech material presented to the judges, three different speech fragments were selected from each digitized recording:

Type I: the first 3 sentences, <u>read</u>

Type II: the 3 sentences, <u>repeated</u> after the speech pathologist

Type III: all 10 sentences, 7 read and 3 repeated

These three speech fragments selected from the pre- as well as the postoperative recordings were stored on the hard disk of the computer. This provided 34 samples of each type (15 patients and 2 controls both pre- and postoperatively).

Rating

The speech samples were presented to each judge separately in a sound-treated room. The speech was reproduced by the audio output of the CSL. No visual information of the waveforms was presented to the judges. Speech samples could be listened to by the judges as often as desired. Listening levels could be chosen as desired by the judges.

The 34 samples of one type were presented in a randomized order to each judge. By using pre- and postoperative samples, the whole range of abnormalities encountered in this field was represented. In addition to the presentation of fragments of the Types I, II and III (called speech samples type A, B and C), the pre- and postoperative fragments Type III were presented in pairs (called speech samples type D). No information as to which sample was the pre- or postoperative one was given (see Table 1.). Both samples had to be rated separately.

TABLE 1 Method of Presentation of the Speech Fragments

Speech Sample Type	*Speech Fragment Used*	*Speech Fragments Presented*
A	I	individually
B	II	individually
C	III	individually
D	III	pre–post pairs

Before the rating of a sample type, the speech samples were randomized by a computer program for each judge to eliminate any order effects. The program chose at random, without replacement, a sample and put it into a presentation sequence. Care was taken to avoid that a pre- or postoperative sample was followed by a post- or pre-operative sample of the same patient.

The rating of the four types of speech samples took place on two days, with an interval of one week. This was done because each session lasted approximately three hours. Each session was started with a rating of either type A or type B and was followed by a rating of either type C or type D. This procedure was repeated in the same way 3 months later.

Scoring

During the listening, each speech sample was rated on a score sheet with 5 undifferentiated lines of 100 mm (visual analog scales) below each other. Endpoints of the line were labeled 'normal' on the left and 'extremely deviant' on the right side. The speech samples were rated on: (1) hypernasality, (2) audible nasal emission, (3) intelligibility, (4) misarticulations associated with velopharyngeal insufficiency, and (5) voice quality. The judges were asked to make a mark on the line to indicate to what extent the speech sample demonstrated the given characteristic, using the criteria for rating they normally apply.

Hyponasality was rated as 'sometimes' or 'always present' or 'absent'. Immediately prior to the rating sessions, all judges were given oral and written instructions to familiarize them with their task.

Analysis

To measure the intrajudge reliability of the ratings, the intraclass correlation coefficient of reliability (ICC) was computed (Ebel 1951, Shrout and Fleiss 1979). The reliability was based on the pre- and postoperative speech samples of the patients (30 ratings for each type and each judge).

The reliability is defined as: $\rho = \sigma_{pp}^2 / (\sigma_{pp}^2+\sigma_\varepsilon^2)$, where σ_{pp}^2 is the variance among speech samples, and σ_ε^2 is the residual variance. The variances were estimated by an analysis of variance with patients and sessions as factors. Systematic differences between the two sessions which were found in some judges were not taken into account. Extreme values (values more than 3 box-lengths from 75th, or from 25th percentile) were limited to the maximal non-extreme value.

For analysis of the improvement in speech, the judgments of the post- and preoperative samples within the same session were subtracted. This provided 15 samples of each type. As the improvement may be nearly equal in all patients, the reliability of the improvement in speech was characterized by the coefficient of variance, defined as: $CV = \sigma_\varepsilon / MD$, where σ_ε is the standard deviation among the patients and MD the mean difference for the corresponding type of speech samples and judge. The residual variance (σ_ε^2) was computed as before, but with the difference of the pre- and postoperative judgments as variable. The differences in range of judgments among the judges were taken into account by dividing by the mean difference (MD). The values were limited to the value 2.0. In contrast to the intraclass correlation coefficient of reliability, a low value for the coefficient of variance represents a high reliability.

Statistics

Differences in the ICC and CV between types of speech samples and judges were tested with an analysis of variance.

Differences in the rating of hyponasality between sessions and between the pre- and postoperative samples were assessed by a χ^2-test. For each type of speech samples and for each judge, the changes between session 1 and session 2 were computed.

All analyses were performed with SPSS/PC+ (SPSS/Inc., Chicago, Il., USA.).

Results

Figure 1 shows the distribution of the pre-operative judgements for hypernasality among the patients for each judge and each type of speech sample.

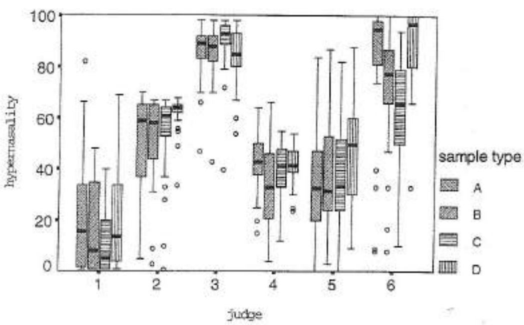

FIGURE 1 Distribution of the preoperative judgments for hypernasality by six judges for four types of speech samples. Vertical axis: 0 = normal, 100 = extremely deviant (values in millimeters). Fifty percent of cases have values within the box. o = values >1.5 × box length from 25th or 75th percentile. ⊤ ⊥ = largest observed value that is not an outlier.

The main characteristics of the results are evident. Judges differ widely in the range they use in their rating. Judge 1 used only the beginning of the rating scale and the distribution was positively skewed. Judge 2 and judge 6 used the right end of the scale when judging the speech samples, whereas the other judges used more or less the entire scale.

Spearman rank correlations between the ratings of pairs of judges ranged from .05 to .81, with a mean value of .60 between the speech pathologists. In contrast to the differences between the judges, differences between the four types of speech samples were not obvious.

An overall analysis of variance of the scores revealed a highly significant effect for the different patients and for the differences between the pre- and postoperative ratings. The difference between judges was also highly significant. Less significant differences between the judgements for the types of speech samples were found.

As the distribution of the scores of judge 1 deviates clearly from normal, the reliabilities of this judge were computed after a logarithmic transformation was applied. The results are shown in the tables, but are not used in the tests.

The nasality and audible nasal emission of the subjects with normal speech and with lateral lisp were judged as normal or nearly normal (median values below 3 mm on the 100 mm scale). These samples were not used in the analysis.

Reliability of the rating

Table 2 shows the values and mean values of the intrajudge reliability for the 4 types of speech samples and the 6 judges. The individual values ranged from .09 to .89. Hypernasality and audible nasal emission were assessed to be slightly more reliable than intelligibility, misarticulations and voice quality (means .68, .64, .61, .56, .63, $p < .01$). The reliability did not significantly differ between the types of

Table 2. Reliability (ICC) of the assessment of speech samples.

HYPERNASALITY

type \ judge	1	2	3	4	5	6	mean
A	.70	.62	.76	.76	.68	.63	.69
B	.77	.83	.69	.62	.79	.66	.73
C	.66	.82	.72	.71	.44	.39	.62
D	.65	.86	.62	.78	.61	.56	.68
mean	.70	.78	.70	.72	.63	.56	.68

AUDIBLE NASAL EMISSION

type \ judge	1	2	3	4	5	6	mean
A	.50	.61	.88	.44	.57	.62	.60
B	.53	.82	.70	.52	.59	.75	.65
C	.50	.79	.78	.64	.54	.51	.62
D	.50	.88	.81	.79	.62	.61	.70
mean	.51	.78	.79	.60	.58	.62	.64

INTELLIGIBILITY

type \ judge	1	2	3	4	5	6	mean
A	.62	.49	.49	.40	.70	.47	.53
B	.40	.72	.60	.50	.81	.74	.63
C	.67	.72	.71	.53	.60	.61	.64
D	.61	.77	.83	.58	.63	.47	.65
mean	.58	.68	.66	.50	.69	.57	.61

MISARTICULATIONS

type \ judge	1	2	3	4	5	6	mean
A	.67	.74	.46	.59	.60	.26	.55
B	.56	.62	.72	.09	.59	.63	.53
C	.77	.62	.58	.68	.50	.70	.64
D	.70	.78	.72	.45	.12	.23	.50
mean	.68	.69	.62	.45	.45	.46	.56

VOICE QUALITY

type \ judge	1	2	3	4	5	6	mean
A	.68	.58	.67	.18	.70	.51	.55
B	.47	.75	.75	.49	.74	.68	.65
C	.61	.89	.73	.65	.62	.84	.72
D	.56	.80	.76	.18	.60	.65	.59
mean	.58	.76	.73	.38	.67	.67	.63

Mean reliablity of the assessment of speech samples.

Variable	mean reliability
Hypernasality	.68
Emission	.67
Intelligibility	.62
Misarticulations	.53
Voice Quality	.64

Judge	mean reliability
1	.61*
2	.74
3	.70
4	.53
5	.60
6	.58

Type of samples	mean reliability
A	.58
B	.65
C	.66
D	.63

*Scores of Judge I were not used in computation of overall mean reliability statistics

speech samples. However, significant differences between the judges were found ($p < .001$). The last effect is mainly due to the differences for audible nasal emission and voice quality. The analysis of the reliability of only hypernasality and audible nasal emission revealed significant interactions ($p < .01$) between the judges and type of speech samples as well as between judges and these variables. Three judges rated hypernasality more reliably than audible nasal emission, whereas the reverse was true for two judges. For one judge no difference was found.

An analysis of variance showed a significant difference between the reliabilities found for the members of cleft palate teams and the general practitioners ($p < .001$). Generally, the former were more reliable than the latter (see

Table 2), but the differences were small. The effect was mainly found in the assessment of the audible nasal emission and voice quality and strongly dependent on the high reliability of one of the members of a cleft palate team. One of the general practitioners was generally more reliable than the other members of a cleft palate team.

For some judges (1, 3, 5) systematic differences between sessions were found, suggesting a shift in their internal standards for rating.

The improvement in speech

Figure 2 shows for each judge and each type of speech samples the distribution of the improvement in hypernasality. The improvement in speech was, on the average, about 23 mm on the visual analog scale for hypernasality and audible nasal emission, 15 mm for intelligibility and about 10 mm for articulation and voice quality.

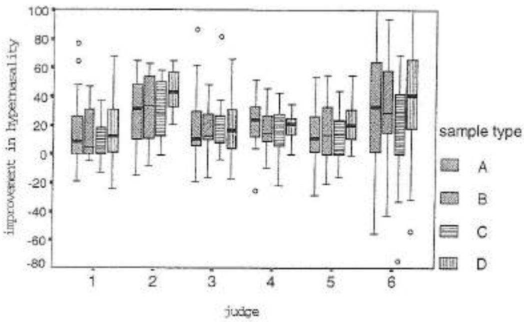

FIGURE 2 Distribution of the improvement for hypernasility by 6 judges and for 4 types of speech samples. Vertial axis = computed improvement (values in millimeters, 0 = normal). Fifty percent of cases have values within the box. o = values >1.5 × box length from 25th or 75th percentile. ┬ ┴ = largest observed value that is not an outlier.

The mean improvement was largest when pre- and postoperative speech samples were presented in pairs (type D). Surprisingly, the smallest improvement was generally found for the stimulus with 10 sentences (type C). However, the differences in mean improvement between the types of speech samples were maximally ± 20%.

TABLE 3 Coefficient of Variation of the Improvement in Speech

	CV
Variable	
Hypernasality	0.73
Emission	0.68
Intelligibility	0.82
Misarticulations	1.37
Voice quality	1.24
Judge	
1	1.26*
2	0.62
3	0.87
4	1.03
5	1.08
6	1.23
Type of samples	
A	1.06
B	1.06
C	1.01
D	0.74

* Scores of Judge 1 were not used in computation of overall statistics.

The reliabilities expressed in the ratio of the random standard deviation of the assessment to the mean difference of a panel's judgement (CV) are shown in table 3. An analysis of variance revealed a significant effect for the type of speech samples and for judges (both $p< .001$). The mean CV of the presentation in pairs (type D) was lowest (.74); for the stimulus types A, B and C values of 1.06, 1.06 and 1.01 were found. There were no significant differences found between the type of speech samples A, B, and C. The improvements in nasality, audible nasal emission and intelligibility were generally more reliably scored than the improvements in misarticulations and voice quality ($p< .001$). The mean CV varied among the judges from .62 to 1.26. This was statistically significant ($p< .001$). The intrajudge reliability found for the members of cleft palate teams was somewhat higher than for the general practitioners, but these differences were not significant. For the improvement in speech, interaction between judges and type of speech samples and between judges and variables were found ($p < .01$).

Only a clear improvement can be reliably determined. From a statistical point of view, the improvement had to be at least 1.65 * CV * MD for a reliable detection ($p < .05$). In this study, the lowest values for such an improvement were 15 mm on the visual analog scale.

Reliability of hyponasality

The difference in judgment of hyponasality between the pre- and postoperative speech samples is shown in table 4.

TABLE 4 Presence of Hyponasality (n = 599)

	Always	*Sometimes*	*Not*
Pre	35	190	374
Post	43	259	297

A χ^2-test shows there is a significant increase in the assessed hyponasality ($\chi^2 = 20.54$; $p < .0001$).

The difference in judgement of hyponasality between session 1 and 2 is shown in table 5. In the two sessions, 62% of the speech samples were equally assessed, 35% were assessed with a difference of one step on the scale backwards or forwards, 3% were assessed with two steps on the scale backwards or forwards. Differences between type of samples were not significant ($\chi^2 = 1.17$). Significant differences between judges were found ($\chi^2 = 50.59$, $p < .001$).

TABLE 5 Difference in Hyponasality Between Sessions 1 and 2 (Number of judgements)

Type of stimulus	*No Difference*	*$\Delta \pm 1$*	*$\Delta \pm 2$*
A	92	53	5
B	90	56	4
C	93	54	3
D	98	47	4

Discussion

The present findings demonstrate wide variation in listener judgments of speech samples and confirm the opinion that judges use internal standards which differ from judge to judge. Significant interactions related to the factor judge were found, suggesting that even the acoustical features used in the assessment might vary from judge to judge. Moreover, the systematic difference in listener assessment between session 1 and session 2 found for some judges, suggests that this standard can be unstable over time. These findings are consistent with recent literature (Kreiman et al. 1993).

Our results demonstrate that speech-language pathologists with expertise in the assessment of cleft palate speech were slightly more consistent in rating the speech variables than those with less expertise. Within the limits of this study, these findings do not support the opinion that speech-language pathologists with expertise are obligatory for reliable rating of cleft palate speech. The surgeon in this study was not less reliable than the speech-language pathologists, but used a totally different standard.

The type of speech sample appeared to be of minor importance. The significant difference found in an overall analysis of variance is mainly due to the large degrees of freedom. It appeared that the presentation of 10 sentences did not significantly enhance the intrajudge reliability of the ratings, so 3 sentences permitted a reliable judgement. The improvement of speech is most reliably assessed by paired comparison.

In the literature about perceptual rating of cleft palate speech, a preference can be found for samples obtained from spontaneous speech. Van Demark (1964) found a high correlation between a task of sentence repetition and spontaneous speech.

The same author (1970) reported better articulatory skills on the repeated sentence test than for the reading sentences test. In the present study, no significant differences were found between speech samples which were read or which were repeated by the patient, but the presentation of speech samples in paired comparison makes differences between speech samples more distinct. An explanation for this finding can be that judges refer to a previous rating when judging a succeeding sample. This effect disappears in the analysis of ratings of randomized samples. In addition, systematic differences between independent pre- and postoperative assessments are avoided in this type of presentation.

Bradford et al. (1964) reported for the rating of hypernasality in spontaneous speech on a scale with equal appearing intervals, by individual judges, a value for reliability (ICC) of .14 for experienced and .25 for inexperienced raters. In the present study, the intrajudge reliability for individual judges rating hypernasality in speech samples in paired comparison, using visual analog scales, ranges from .56 to .86 (mean .68). The reliability for misarticulations in repeated speech samples and in reading sentence test ranges from .09 to .72 (mean .53) and .26 to .74 (mean .55).

In the present population that is primarily characterized by abnormalities in resonance, the full range of deviances in intelligibility, misarticulations and voice quality cannot be expected. For these 3 variables, the variance of the ratings among the speech samples was about two thirds of that for nasality and audible nasal escape. This may explain the lower values for the reliability of the ratings of intelligibility, articulation and voice quality compared to the other two variables. According to Fleiss (1986), the range of values of ρ (ICC) can be roughly divided in 3 parts; below 0.4, from 0.4

to 0.75 and 0.75 to 1.0 representing a poor, fair to good and excellent reliability respectively. In this study, the results indicated fair to good intrajudge reliability.

Conclusions

The results of this study indicate the following:

A. Ratings of cleft palate speech by different judges are generally not comparable. Without special training of judges, use only ratings within one judge for assessing difference between pre- and postoperative samples.

B. There is little difference in intrajudge reliability between the ratings of speech samples which are read or speech samples which are repeated by the patient and there is only a slight difference in reliability between the ratings of 3 or 10 sentences.

C. The assessment of the improvement in speech is most reliable when rated in paired comparison.

D. Generally, speech-language pathologists experienced in evaluating cleft palate speech are slightly more reliable in rating the speech characteristics of cleft palate speech than speech-language pathologists without this experience. However, experience with cleft palate speech does not guarantee a higher intrajudge reliability.

References

Blijdorp P, Müller H, The influence of the age at which the palate is closed on speech in the adult cleft patient. J Max.fac.Surg. 1984; 12: 239-246.

Bradford LJ, Brooks AR, Shelton RL. Clinical judgement of hypernasality in cleft palate children. Cleft Palate J 1964; 1: 329-335.

Bzoch KR. Measurement and assessment of categorical aspects of cleft palate speech. In: Bzoch KR, ed. Communicative Disorders related to Cleft Lip and Palate. 2nd edition. Boston: Little, Brown & Co. 1989; 137-173.

Carney PJ, Sherman D. Severity of nasality in three selected speech tasks. J Speech Hear Res. 1971; 14: 396-407.

Croatto L. L'évaluation phoniatrique de l'insuffisance vélo-pharyngienne. Bulletin d'Audiophonologie 1984; 17: 163-175.

Dalston RM, Warren D. Comparison of Tonar II, pressure-flow, and listener judgements of hypernasality in the assessment of velopharyngeal function. Cleft Palate J 1986; 23: 108-115.

Ebel RL. Estimation of reliability of ratings. Psychometrika 1951; 16: 407-424.

Fleiss JL. The design and analysis of clinical experiments. New York: John Wiley & Sons, 1986.

Fletcher SG. Nasalance vs listener judgements of nasality. Cleft Palate J 1976; 13: 31-44.

Haapanen ML. A simple clinical method of evaluating perceived hypernasality. Folia Phoniatr 1991; 43: 122-132.

Hirschberg J, Van Demark DR. A proposal for standardization of speech and hearing evaluations to assess velopharyngeal function. Folia Phoniatr Logop 1997; 49: 158-167.

Karling J, Larson O, Leanderson R, Henningsson G. Speech in unilateral and bilateral cleft palate patients from Stockholm. Cleft Palate-Craniofacial J 1993; 30: 73-77.

Kreiman J, Gerratt BR, Kempster GB, Erman A, Berke GS. Perceptual evaluation of voice quality: review, tutorial, and a framework for future research. J Speech Hear Res 1993; 36: 21-40.

McWilliams BJ, Glaser ER, Philips BJ, Lawrence C, Lavorato AS, Beery QC, Skolnick ML. A comparative study of four methods of evaluating velopharyngeal adequacy. Plastic and Reconstructive Surgery 1981; 68: 1-9.

McWilliams BJ, Morris HL, Shelton RJ. Cleft Palate Speech. Philadelphia, Toronto: BC Decker Inc. 1990; 247-268.

Pommez J, Rebufy M. L'examen phoniatrique de l'insuffisance vélaire congénitale. Rev. Laryngol. 1987; 108: 299-305.

Schmelzeisen R, Hausamen JE, Löbell E, Hacki T. Long-term results following velopharyngoplasty with a cranially based pharyngeal flap. Plast Reconstr Surg 1992; 90: 774-778.

Shrout PE, Fleiss JL. Intraclass correlations: uses in assessing rater reliability. Psychological Bulletin 1979; 86(2): 420-428.

Van Demark DR. Misarticulations and listener judgements of the speech of individuals with cleft palates. Cleft Palate J 1964; 1: 232-245.

Van Demark DR. A comparison of the results of pressure articulation testing in various contexts for subjects with cleft palates. J Speech Hear Res 1970; 13: 741-754.

Chapter 5

The correlation between nasalance and a differentiated perceptual rating of speech in Dutch patients with velopharyngeal insufficiency

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Abstract

Objective: The correlation between the nasalance score and the perceptual rating of several aspects of speech of speakers with velopharyngeal insufficiency (VPI) by six speech-language pathologists was evaluated.
Procedure: The overall grade of severity, hypernasality, audible nasal emission, misarticulations, and intelligibility were rated on visual analog scales. Speech samples with a normal distribution of phonemes (normal text [NT]) and those free of nasal consonants (denasal text [DT]) of 43 patients with VPI were used. Mean nasalance scores were computed for the speech samples, and Spearman correlation coefficients were computed between the mean nasalance score and the five parameters of the differentiated rating.
Setting: The Institute of Phoniatrics, Utrecht University Hospital, the Netherlands.
Results: The correlation coefficient between the mean nasalance and the perceptual rating of hypernasality ranged among judges from .31 to .56 for NT speech samples and .36 to .60 for DT speech samples. Only small differences were found between speech pathologists with and without expertise in cleft palate speech. The rating of the overall grade of severity appeared to correlate quite well with the rating of the intelligibility (r_{NT}= .77, r_{DT} = .79). Lower correlation coefficients, ranging from .34 to .71, were found between overall grade of severity and hypernasality, audible nasal emission, and misarticulations.
Conclusions: A low correlation between the nasalance and the perceptual rating of hypernasality was found. The parameter overall grade of severity appeared to be determined mainly by the parameter intelligibility. Expertise in rating of cleft palate speech does not guarantee a high correlation between instrumental measurement and perceptual rating.

The speech of individuals with repaired palatal clefts and velopharyngeal insufficiency (VPI), hereafter referred to as cleft palate speech (CPS), is commonly characterized by increased nasal resonance. There may be compensatory misarticulations and distorsion of consonants as a result of audible nasal emission.

Speech intelligibility and voice quality may also be affected. Traditionally, the assessment of CPS and therapy outcome has relied heavily on perceptual judgments (Croatto, 1984). The perceptual judgment of hypernasality has proven to be difficult and subjective in nature. Because of the different aspects of CPS, there is no single scale or measure that can be used to describe all aspects of the abnormal speech.

The methodology used for perceptual rating of CPS differs with respect to the type of speech samples (Vandemark, 1970 Carney and Sherman, 1971), phonetic context (Fletcher et al, 1989; Watterson et al., 1996), expertise of judges (Fletcher, 1976; Croatto, 1984; Dalston and Warren, 1986; Schmelzeisen et al, 1992), and scale terms used for grading the severity (Pommez and Rebufy, 1987; Bzoch, 1989; Karling et al., 1993). The computed reliability of the rating is influenced by the type of rating scales used and the statistical analysis of the data (Kreiman et al. 1993). Moreover, it appears that the internal standards of judges against which their judgments are made may vary with time (Kreiman et al., 1993). In addition, the presence of other symptoms, such as misarticulations, may mask or enhance the perception of hypernasality (Fletcher et al., 1989; McWilliams et al., 1990).

To avoid these uncertain factors, instruments to measure the extent of nasal resonance have been developed. An example of such an instrument is the Nasometer (Kay Elemetrics Corp., Pine Brook, NJ), the computer-based successor of Tonar II. Acoustic nasometry is regarded to be an attractive technique because it claims to yield reliable data. As a nonintrusive procedure, it is suitable for use in children. The essence of

the procedure is that, during speech, the emitted nasal and oral acoustic energy (within a specified frequency band) are measured separately. The ratio of acoustic energy from the nasal cavity and the sum of acoustic energy from nasal and oral cavity are used as a measure of nasal resonance. This ratio, defined by Fletcher (1976) as nasalance, reflects the relative amount of nasal acoustic energy in speech and is expressed as a percentage. High nasalance scores can be expected in patients with incomplete velopharyngeal closure and in speech samples loaded with a high percentage of nasal consonants (Fletcher et al., 1989). Low nasalance scores can be expected in patients with obstruction in the nasopharynx or in the nose and in speech samples free of nasal consonants. The measurement appears to be sensitive to the phonetic composition of the speech samples (Fletcher et al., 1989; Dalston and Seaver, 1992; Watterson et al., 1996). The normative values appear to be dependent on native language (Santos-Terron et al., 1991) or regional dialect (Seaver et al., 1991). However, Kavanagh et al. (1994) did not find differences in normative nasalance scores for three Canadian regional dialects. Differences because of age (Seaver et al., 1991) and sex (Fletcher, 1978; Seaver et al., 1991) have also been identified.
Because the distribution of phonemes is different in each language, standard passages for each language should be developed. The corresponding normative nasalance scores should be computed for each language or regional dialect (Seaver et al., 1991) because vowels are intentionally nasalized in some languages (e.g., French) and some regional dialects. For instance, English phoneticians often describe the vowels in American English dialects as more nasalized than the same vowels in Queen's English (Wise, 1957). For American English (Fletcher et al., 1989), Australian English (Van Doorn and Purcell, 1998), German (Heppt et al., 1991, Castilian Spanish (Santos-Terron et al., 1991, Finnish et al., 1991), (Haapanen,

1991) and midwest Japanese (Tachimura et al., 2000), these normative nasalance scores have been computed.
According to the nasometer manufacturer's manual, the normative nasalance scores for American English-speaking children are 15.53% (SD 4.86) for the Zoo passage (speech sample free of nasal consonants) and 35.69% (SD 5.20) for the Rainbow passage (speech sample with a normal distribution of nasal consonants). However, it should be noted that these values have been computed for a Southern American-speaking population (Fletcher et al., 1989; Van Doorn and Purcell, 1998). For standard passages in the Dutch language, Van De Weyer and Slis (1991) computed normative nasalance scores. A mean nasalance of 11.75% (SD 4.23) for speech samples free of nasal consonants and a mean nasalance of 31.95% (SD 5.24) for speech samples with a normal distribution of phonemes were established. It appeared that these normative scores did not differ between male and female speakers or between children and adults. Although the nasometer manufacturer's manual indicates normative nasalance scores, it does not indicate the values at which the nasalance scores should be considered abnormal. Assuming that the extent of hypernasality is normally distributed among a healthy population, the upper limit of the 95% confidence interval (mean nasalance score + 2 SDs) may be considered as the cut-off point for abnormality. On average, 2.5% of the subjects will be misclassified. Moreover, the real distribution may deviate from the mathematically defined normal distribution. Therefore, the cut-off point should be based on the distribution of the nasalance in a normal population and a population with VPI. Because hypernasality is a perceptual phenomenon, most authors agree that the usefulness of the instrumental measure essentially depends on its positive correlation with the perceptual rating by judges with expertise in cleft palate speech. This correlation has been the subject of several studies. Dalston et al. (1991) examined the correlation

between the nasalance scores of 117 patients, computed during the production of the Zoo passage and the perceptual rating of hypernasality in conversational speech. In that experiment, all ratings were performed by one experienced judge on a six-point rating scale.

They reported a Pearson correlation coefficient of 0.82 between the nasalance score and the perceptual rating of hypernasality. A cross-dialect and cross-culture study was completed by Dalston et al (1993). A total of 514 patients were studied in two cleft lip and palate centers in the United States and one center in Spain. Nasalance scores were computed for a standard passage free of nasal consonants. In both American centers, the Zoo passage was used, and in the Spanish center, the Texto El Bosque was used. The perceptual ratings of hypernasality were performed on conversational speech. An overall Spearman correlation coefficient of .73 and an overall Pearson correlation coefficient of .78 was computed. Hardin et al. (1992) studied 22 patients with a repaired cleft palate, 29 patients with a repaired cleft palate and velopharyngeal flap, and 23 noncleft controls. Nasometry scores were computed for the Zoo passage, and perceptual ratings were performed by three certified speech language pathologists on test sentences considered to be representative of conversational speech, constructed previously by Van Demark (1964). One of their conclusions was that the association between the nasalance scores and the perceptual ratings of hypernasality increased when patients with a pharyngeal flap were excluded from the study.

Nellis et al. (1992) did not find a significant correlation between mean nasalance scores and mean judge's rating of hypernasality in 16 patients who had received a pharyngeal flap. In that experiment, the perceptual ratings were performed by 10 graduate students. Because of the differences in methodological design among these studies, the results are difficult to compare.

Another source of variation among studies might be the confounding effect of audible nasal emission on the computed nasalance score (Watterson et al., 1993; Karnell, 1995). A reason for this variation might be that the nasometer cannot discriminate between the energy in resonance and the energy in nasal emission. The aim of the present experiment was to investigate the correlation between the mean nasalance score for two standard passages in the Dutch language (one with a normal distribution of phonemes and the other free of nasal consonants) and the parameters of a differentiated perceptual rating (overall grade of severity, hypernasality, audible nasal emission, misarticulations associated with VPI, and intelligibility) of the same passages in patients with VPI.

MATERIAL AND METHOD

Subjects

Subjects for this study were selected from the patients who visited the outpatient clinic at the Institute of Phoniatrics of the Utrecht University Hospital between March 1995 and March 1997 for a medical examination related to their perceived hypernasality. From this group, 43 subjects (26 boys and men and 17 girls and women, median 8 years, range 4 to 83 years) whose clinical records and tape recordings were complete were included (Table 1). None reported a handicapping hearing loss. The patients read two standard passages in the Dutch language. Patients who were too young or unable to read the speech samples easily were asked to repeat the sentences after the examiner.

TABLE 1 Primary Diagnosis of the 43 Subjects Employed in the Present Study

Diagnosis	*Frequency*	*Percentage*
Congenital palatopharyngeal incompetence (short velum, deep pharynx, or both)	22	51
Unilateral cleft of primary and secondary palate (right)	6	14
Unilateral cleft of primary and secondary palate (left)	3	7
Bilateral complete cleft of primary and secondary palate	4	9
Partial resection of velum for malignancy	2	5
Robin sequence	1	2
CHARGE* association with short velum	1	2
Cleft of secondary palate involving hard palate and velum	2	5
Dysarthric involvement of velopharyngeal mechanism	2	5
Total	43	100

*The diagnosis CHARGE-syndrome refers to children with a specific set of birth defects and is based on finding several of these and possibly other features. The birth defects are coloboma, heart defects, atresia of the choanae, retardation of growth and development, genital and urinary abnormalities, ear abnormalities and hearing loss.

Passages

The phonemes in the text of the first passage (normal text [NT]) are representative of the distribution of phonemes in the Dutch language (11.67% of the consonants are nasal). The second passage is a text free of nasal consonants (denasal text [DT]). Each passage contained approximately 70 words, which is comparable to the Zoo passage.

Instrumentation

Nasalance was measured with the Nasometer, model 6200 (Kay Elemetrics Corp.). Calibration was performed according to the instructions provided by the manufacturer's manual. The nasometer's headgear was adjusted in accordance with the instructions provided by the manual. During testing the speech samples were recorded simultaneously on tape for the perceptual rating. Attention was paid to an appropriate placement of the microphone of the tape recorder (at a distance of 70 to 90 cm, in front of the subject's face), to ensure that the face mask did not affect the taped speech signal. In cases in which the patient repeated the sentences after the examiner, the nasometer was activated only when the patient was speaking. The mean nasalance was ca1culated between the onset and offset of the data displayed for each sample separately, using the nasometer software (version 3.22). All speech samples were recorded in a sound-treated room. The above-mentioned procedure is a part of the standard evaluation protocol. Taped speech samples (NT and DT) were digitized at 40 kHz using the Computerized Speech Laboratory (CSL, type 4300, Kay Elemetrics Corp.). All text not spoken by the patient was removed from the files using the CSL program.

Judges

Six speech-language pathologists (SLPs) rated the speech samples independently. Three were general practitioners without special expertise in CPS rating and three were members of different cleft palate teams.

Rating

Digitized speech samples were presented to two judges simultaneously in a sound-treated room at a comfortable listening level, both being equidistant from the audiospeaker of the CSL system. The speech samples were presented three times and randomized before each rating session to eliminate any order effects. No information about the patients was provided. Ratings of the speech samples took place on one day, with a break of 30 minutes between sessions. Each rating session lasted approximately 2 hours 15 minutes. Speech samples were rated on the overall grade of severity, hypernasality (hyperrhinophonia), audible nasal emission, misarticulations associated with VPI, and intelligibility (Hirschberg and Van Demark, 1997). Each speech sample was rated on a score sheet with five visual analog scales (VAS) of 100 mm underneath one another. End points of the scales were

defined as 'normal' (left side) and 'extremely deviant' (right side).
The judges were asked to indicate by a mark on the VAS to what extent the speech samples demonstrated the given characteristic using the criteria for rating they normally apply. In addition, hyponasality was rated as 'absent', 'sometimes present', or 'always present'; voice quality was rated as 'good', 'moderate', or 'bad'. Just prior to the rating session, a task-specific listening orientation was conducted during which the listeners were provided with oral and written instructions and definitions of scale terms. Three preselected speech samples, each presenting a clear example of hypernasality, audible nasal emission, or misarticulations associated with VPI, were presented to the judges. The ratings of these examples by two experienced judges (not participating in this experiment) were given and discussed to ensure that all judges were listening to the same percept. These samples were not included among the samples to be rated. The task-specific orientation lasted approximately 30 minutes.

DATA ANALYSIS

The data analysis was performed using the statistical package SPSS 8.0 for Windows (SPSS Inc., Chicago, IL). Because the values of the perceptually rated parameters did not appear to be normally distributed, nonparametric statistics were performed.
For this reason, the Spearman correlation coefficient was used to reflect the relationship among variables. To test the differences between two groups, Wilcoxon matched pairs signed-ranked tests were performed. Interjudge variability was computed as the mean Spearman correlation coefficient of all possible bivariate correlations between judges.

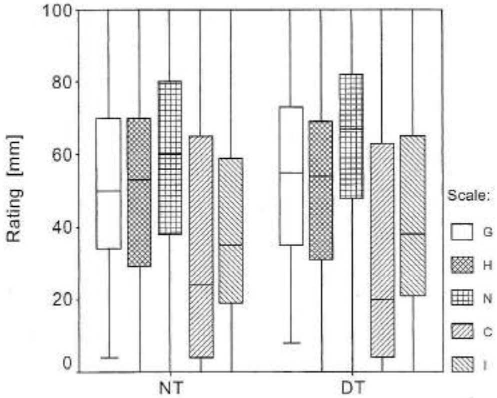

FIGURE 1 Box-Whiskerplot distribution of the mean perceptual ratings of the speech of patients with velopharyngeal insufficiency for speech samples with a normal distribution of phonemes (NT) and speech samples free of nasal consonants (DT). G overall grade of severity; H hypernasality; N audible nasal emission; C compensatory misarticulations; I intelligibility. For each patient, the mean of six judges on a visual analog scale was used. □ 25th to 75th percentile; — median value; ⊤⊥ maximum/minimum extreme values.

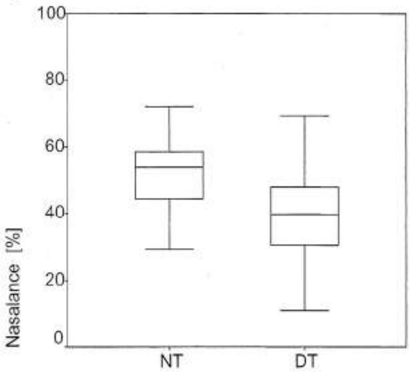

FIGURE 2 Box-Whiskerplot distribution of the nasalance scores of the speech samples with a normal distribution of phonemes (NT) and the speech samples free of nasal consonants (DT).

RESULTS

Speech Samples

The distribution of the values rated by the 6 SLPs for the NT and DT speech samples is shown in Figure 1. The average score for the mean nasalance computed for the NT speech samples was 51.5% (SD 10.7) and 40.1% (SD 14.0) for the DT speech samples (Fig. 2).

Correlation among Perceptual Ratings

Table 2 shows the Spearman correlation coefficients among the perceptual ratings, computed separately for the NT and DT speech samples. The ratings of overall grade of severity, hypernasality, audible nasal emission, intelligibility, and

TABLE 2 Spearman Correlation Coefficients Between the Parameters of the Perceptual Rating for Speech Samples With a Normal Distribution of Phonemes (NT) and Speech Samples Free of Nasal Consonants (DT)

	Hyper-nasality	*Nasal Emission*	*Misarticu-lations*	*Intelligi-bility*
NT				
Overall grade of severity	.61	.60	.55	.77
Hypernasality	—	.60	.46	.63
Audible nasal emission	—	—	.37	.53
Compensatory misarticulations	—	—	—	.65
DT				
Overall grade of severity	.54	.53	.59	.79
Hypernasality	—	.51	.58	.60
Audible nasal emission	—	—	.34	.49
Compensatory misarticulations	—	—	—	.71

misarticulations were found to be correlated. Only small differences were found among judges. It appeared that the rating of the overall grade of severity was determined mainly by the rating of the intelligibility (r_{NT}= .77, r_{DT} = .79) and less by the rating of hypernasality (r_{NT}= .61, r_{DT} = .54). The stronger correlation between grade and intelligibility than between grade and hypernasality was found for all judges. Differences between general practitioner SLPs and SLPs with expertise were found, but none of these differences was significant.

Differences among Types of Speech Samples

The perceptual ratings of overall grade of severity, audible nasal emission and intelligibility were, on average, more severe for DT speech samples than for NT speech samples. The differences on the VAS were 3.1 mm (p = .001), 4.5 mm (p= .000) and 3.6 mm (p = .001), respectively. Although these differences are statistically significant, it is unlikely that they are of clinical importance. No significant differences for the rating of hyponasality and voice quality were found between NT and DT speech samples.

Correlation among Judges

The mean of the Spearman correlation coefficients among judges is shown in Table3.

TABLE 3 Mean Spearman Correlation Coefficients and SD Among Judges for Differentiated Parameters

Parameter	*Mean (SD)*
Overall grade of severity	.65 (.08)
Hypernasality	.49 (.08)
Audible nasal emission	.58 (.10)
Compensatory misarticulations	.59 (.13)
Intelligibility	.70 (.07)

The strongest correlations were found for the rating of intelligibility (r =.70) and the overall grade of severity (r = .65). A weaker correlation was found for hypernasality (r = .49) and audible nasal emission (r = .58). The correlation for hypernasality was stronger among the SLPs with expertise (r = .55) than among general practitioner SLPs (r = .46), but the reverse was found for audible nasal emission (r = .46 and r = .60). In general, general practitioner SLPs rated the abnormalities significantly more severe than SLPs with expertise (overall test NT: manova F{5, 175} = 19.41, *P* = .000 and overall test DT: manova F{5, 172} = 15.69, *P* = .000.) Overall, the average rating of the general practitioners was 13 mm higher on the VAS.

Differences between SLPs with and without expertise were less distinct for ratings of misarticulations. The SLPs agreed on 47% of the ratings of voice quality and on 48.3% of the ratings of hyponasality.

Correlation between Nasalance and Perceptual Rating

Figure 3 shows the relationship between nasalance scores and the mean perceptual ratings of hypernasality for NT and DT speech samples.

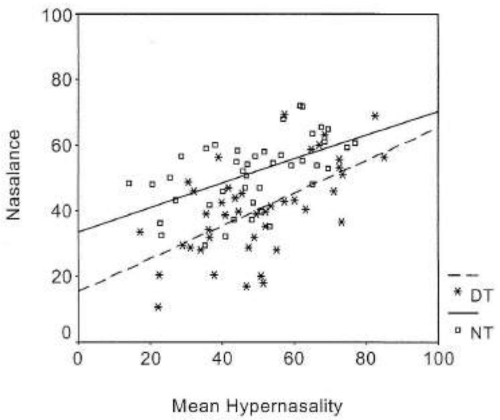

FIGURE 3 **The relationship between the nasalance scores and perceptual ratings of hypernasality in speech samples with a normal distribution of phonemes (NT) and speech samples free of nasal consonants (DT).**

The nasalance score for the DT speech samples was lower than for the NT speech samples (Wilcoxon, $p < .001$). This difference was not found for the perceptual rating. In general, the correlation was slightly higher for the NT speech samples than for the DT samples, but the differences were not significant. To demonstrate the variability among the judges, correlation coefficients between the perceptual rating and nasalance of the NT and DT speech samples for each judge are shown in Table 4. The strongest correlation coefficient was found between hypernasality and nasalance. This confirms the notion that nasalance represents the perceptual impression of hypernasality.

The correlation coefficients between the rating of hypernasality and nasalance ranged from $r_{NT}= .31$ and $r_{DT}= .36$ (judge C) to $r_{NT}= .56$ and $r_{DT}= .60$ (judge B), with an average value of $r= .43$ for general practitioner SLPs (judges A, B, and C) and r= .42 for SLPs with expertise (judges D, E, and F). The correlation between the audible nasal emission and nasalance was unexpectedly low and not significant. These correlation coefficients ranged from r_{NT}= .15 and r_{NT}= .34 to r_{DT} = - .03 and r_{DT}= .50, with an average value of r = .24 for general practitioner SLPs and r = .18 for SLPs with expertise in CPS.

The Spearman correlation coefficient between the rating of hypernasality in NT speech samples and the nasalance score for the DT speech sample was .43.

TABLE 4 Spearman Correlation Coefficients Between the Nasalance and Differentiated Perceptual Rating for Speech Samples With a Normal Distribution of Phonemes (NT) and Those Free of Nasal Consonants (DT)

*Judge**	*Overall Grade of Severity*	*Hyper-nasal-ity*	*Audible Nasal Emission*	*Compen-satory Misartic-ulations*	*Intellig-ibility*
NT					
Mean	.41†	.57†	.28	.41†	.37‡
A	.32‡	.36‡	.25	.10	.32‡
B	.37‡	.56†	.34‡	.25	.38‡
C	.26	.31‡	.18	.35‡	.27
D	.46†	.48†	.15	.33‡	.26
E	.46†	.33‡	.25	.41†	.38‡
F	.36‡	.43†	.28	.24	.27
DT					
Mean	.40†	.54†	.18	.23	.34‡
A	.35‡	.40†	.18	.22	.22
B	.44†	.60†	.33‡	.25	.44‡
C	.15	.36‡	.18	.06	.18
D	.41†	.41†	−.03	.29	.37‡
E	.41†	.41†	.50†	.30‡	.47‡
F	.32‡	.47†	−.05	.22	.32‡

* Judges A, B, and C are speech-pathologist general practitioners; judges D, E, and F are speech-pathologists with expertise in cleft palate speech.
† $p < .01$.
‡ $.05 > p > .01$.

DISCUSSION

Speech of patients with VPI is primarily characterized by increased hypernasality. One would expect the parameter "overall grade of severity" to be characterizedby the same percept as well. However, this parameter appeared to be determined mainly by the parameter of intelligibility

(r_{NT}= ,77, r_{DT}= ,79) and less by "hypernasality" (r_{NT}= .61, r_{DT}= .54).

Moreover, the parameters of the differentiated perceptual rating appeared to be mutually correlated. This might be a factor in the finding that hypernasality was rated more severely when other symptoms, such as misarticulations, are present (McWilliams et al., 1990).

The results of the current experiment do not support the supposition that the nasalance score is higher in the presence of a more severe rating of audible nasal emission. The low correlation coefficient between the nasalance score and the rating of audible nasal emission (r_{NT}= .28, r_{DT} = .18) makes this supposition unlikely.

Speech pathologists with expertise in CPS rating do not distinguish themselves from their general practitioner colleagues by a higher correlation coefficient between the perceptual rating and the instrumental measure.

In an earlier experiment (Keuning et al., 1999), speech pathologists with expertise were only slightly more consistent in the rating of CPS than general practitioner speech pathologists. Both findings do not support the opinion that speech pathologists with expertise in evaluating CPS are obligatory for a reliable rating with a strong correlation with the instrumental measure.

A relatively high correlation coefficient among judges (r= .65) was found for the parameter of overall grade of severity. A comparable correlation coefficient was found for the parameter grade, the first parameter of the GRBAS scale, used for the perceptual rating of voice pathology (Dejonckere et al., 1993). A relatively high correlation coefficient among judges may be a characteristic of overall perceptual judgments (Dejonckere et al., 1993, 1996).

To be a useful tool in daily clinical practice, acoustic nasometry has to be suitable for pre- and posttherapy assessment. For that reason we did not exclude from this study speech samples of three patients who had been subjected to a cranially based pharyngeal flap. However, the inclusion of these speech samples may have slightly influenced the results of the current experiment (Hardin et al., 1992; Nellis et al., 1992).

To compare the results of the present study with the results of Dalston et al. (1993), the Spearman correlation coefficient between the rating of hypernasality in speech samples with a normal distribution of phonemes (regarded to be representative of conversational speech) and the nasalance score for the speech samples free of nasal consonants was computed. This correlation coefficient (.43) is considerably lower than the overall correlation coefficient (.73) found by Dalston et al. (1993). However, in that study, pronounced differences in correlation coefficients were found among the participating centers. The correlation coefficient in the current study (.43) is comparable to the correlation coefficient of the Spanish cleft lip and palate center (.52) and one of the two American centers (.55). The high correlation found in the second American center (.80) may be the result of random variability, as suggested by Dalston et al. (1993). The stronger correlation between the nasalance and the rating of hypernasality, reported by Dalston et al. (1991, 1993), might also be explained by the fact that the hypernasality of the patients participating in their experiments ranged from mild to severe. The higher values for the Pearson correlation coefficient than for the Spearman correlation coefficient in that study might indicate that the severe cases had a clear effect on the correlation. In the current study, no severe cases of hypernasality were present.

Extensive training to tune the judges is regarded to be a method to enhance reliability of the rating. Offering anchored stimuli to judges may enhance reliability of judges as well. (Kreiman et al., 1993).If it is possible to tune judges by training, then it is reasonable to assume that judges can be tuned to the instrumental measure by training as well. It is likely that the judge from the better-scoring American center had extensive experience with the nasometer. In the current experiment, no training session was provided and none of the speech pathologists had extensive experience with the nasometer.

The fairly low correlation coefficient between nasalance and perceptual rating may also be the result of random errors in measures and ratings. An upper limit of this effect was estimated by using the two speech samples (NT and DT) as repeated measures. Two analyses of variance were used to estimate the random error and variance among the patients for the nasalance and rating of hypernasality, leaving out systematic differences between the NT and DT samples. Taking the estimates of these errors into account, the correlation coefficient between nasalance and the rating of hypernasality increased only slightly. Therefore, this effect cannot explain the low correlation coefficient found in this experiment. Clearly, the nasometer measures other phenomena than those perceptually rated by the judges. The nasometer computes the nasalance from the difference in the oral-nasal intensity at 500 Hz (± 150 Hz; Fletcher et al., 1989). The acoustic effects of hypernasality are not restricted to this frequency range (Watterson et al., 1993). Thus, the nasalance score does not inc1ude all of the acoustic information that is available to the listeners and may characterize only a part of the phenomenon of hypernasal speech. Therefore, the question of whether perceptual rating or acoustic nasometry provides the most valuable result remains unanswered. Nasometry is not a substitute for the perceptual rating of hypernasality, but it may be used to quantify hypernasality and the outcome of therapy on hypernasality as part of a multidimensional diagnostic process.

Conclusions

The results of this study indicate the following:

1. The fairly low correlation between the nasalance score and the perceptual rating of hypernasality indicates that nasalance scores should be interpreted cautiously. Nasometry should not be used as a substitute for perceptual rating.
2. There are no significant difference among speech speech pathologists with or without expertise in CPS with regard to the correlation coefficient between perceptual rating and nasalance.
3. The rating of the parameters 'overall grade of severity' appears to be determined mainly by the parameter 'intelligibility'.

References

Bzoch KR. Measurement and assessment of categorical aspects of cleft palate speech. In: Bzoch KR, ed. Communicative Disorders Related to Cleft Lip and Palate. 2nd ed. Boston: Little, Brown; 1989: 137-173.

Carney PJ, Sherman D. Severity of nasality in three selected speech tasks. J Speech Hear Res. 1971; 14: 396-407.

Croatto L. L'évaluation phoniatrique de l'insuffisance vélopharyngienne. Bull Audiophonol. 1984; 17: 163-175.

Dalston RM, Neiman GS, Gonzalez-Landa G. Nasometric sensitivity and specificity: a cross-dialect and cross-culture study. Cleft Palate Craniofac J. 1993; 30: 285-291.

Dalston RM, Seaver EJ. Relative values of various standardized passages in the nasometric assessment of patients with velopharyngeal impairment. Cleft Palate Craniofac J. 1992; 29: 17-21.

Dalston RM, Warren D. Comparison of Tonar II, pressure-flow, and listener judgments of hypernasality in the assessment of velopharyngeal function. Cleft Palate J. 1986; 23: 108-115.

Dalston RM, Warren DW, Dalston ET. Use of nasometry as a diagnostic tool for identifying patients with velopharyngeal impairment. Cleft Palate Craniofac J. 1991; 28: 184-188.

Dejonckere PH, Obbens C, De Moor GM, Wieneke GH. Perceptual evaluation of dysphonia: reliability and relevance. Folia Phoniatr. 1993; 45: 76-83.

Dejonckere PH, Remacle M, Fresnel-Elbaz E, Woisard V, Crevier-Buchman L, Millet B. Differentiated perceptual evaluation of voice quality: reliability and correlations with acoustic measurements. Rev Laryngol Otol Rhinol (Bord.). 1996; 117: 219-224.

Fletcher SG. Diagnosing Speech Disorders From Cleft Palate. New York: Grune and Stratton; 1978: 92-157.

Fletcher SG. 'Nasalance' vs listener judgements of nasality. Cleft Palate J. 1976; 13: 31-44.

Fletcher SG, Adams LE, McCutcheon MJ. Cleft palate speech assessment through oral-nasal acoustic measures. In: Bzoch KR, ed. Communicative Disorders Related to Cleft Lip and Palate. Boston: Little, Brown; 1989: 246-257.

Haapanen M-L. Nasalance scores in normal Finnish speech. Folia Phoniatr. 1991; 43: 197-203.

Hardin MA, Van Demark DR, Morris HL, Payne MM. Correspondence between nasalance scores and listener judgments of hypernasality and hyponasality. Cleft Palate Craniofac J. 1992; 29: 346-351.

Heppt W, Westrich M, Strate B, Moering L. Nasalanz: Ein neuer Begriff der objectiven Nasalitaetsanalyse Laryngo-Rhino-Otol. 1991; 70: 208-213.

Hirschberg J, van Demark DR. A proposal for standardization of speech and hearing evaluations to assess velopharyngeal function. Folia Phoniatr Logop. 1997; 49: 158-167.

Karling J, Larson O, Leanderson R, Henningsson G. Speech in unilateral and bilateral cleft palate patients from Stockholm. Cleft Palate Craniofac J. 1993; 30: 73-77.

Karnell MP. Nasometric discrimination of hypernasality and turbulent nasal airflow. Cleft Palate Craniofac J. 1995; 32: 145-148.

Kavanagh JL, Fee EJ, Kalinowski J, Doyle PC, Leeper HA. Nasometric values for three dialectal groups within the Atlantic provinces of Canada. J Speech Lang Pathol Audiol. 1994; 18: 7-13.

Keuning KHD, Wieneke GH, Dejonckere PH. The intrajudge reliability of the perceptual rating of cleft palate speech before and after pharyngeal flap surgery: the effect of judges and speech samples. Cleft Palate Craniofac J. 1999; 36: 328-333.

Kreiman J, Gerratt BR, Kempster GB, Erman A, Berke GS. Perceptual evaluation of voice quality: review, tutorial, and a framework for future research. J Speech Hear Res. 1993; 36: 21-40.

McWilliams BJ, Morris HL, Shelton RJ. Cleft Palate Speech. Philadelphia and Toronto: BC Decker; 1990.

Nellis JL, Neiman GS, Lehman JA. Comparison of nasometer and listener judgements of nasality in the assessment of velopharyngeal function after pharyngeal flap surgery. Cleft Palate Craniofac J. 1992; 29: 157-163.

Pommez J, Rebufy M. L'examen phoniatrique de l'insuffisance vélaire congénitale. Rev Laryngol. 1987; 108: 299-305.

Santos-Terron MJ, Gonzalez-Landa G, Sanchez-Ruiz I. Patrones normales del nasometer en ninos de habla castellana [nasometric patterns in the speech of normal child speakers of Castilian Spanish]. Rev Esp Foniatr. 1991; 4: 71-75.

Schmelzeisen R, Hausamen JE, Löbell E, Hacki T. Long-term results following velopharyngoplasty with a cranially based pharyngeal flap. Plast Reconstr Surg. 1992; 90: 774-778.

Seaver EJ, Dalston RM, Leeper HA, Adams LE. A study of nasometric values for normal resonance. J Speech Hear Res. 1991; 34: 715-721.

Tachimura T, Mori C, Hirata S, Wada T. Nasalance score variation in normal adult Japanese speakers of mid-west Japanese dialect. Cleft Palate Craniofac J. 2000; 37: 463-467.

Van Demark DR. A comparison of the results of pressure articulation testing in various contexts for subjects with cleft palates. J Speech Hear Res. 1970; 13: 741-754.

Van Demark DR. Misarticulations and listener judgements of the speech of individuals with cleft palates. Cleft Palate J. 1964; 1: 232-245.

van de Weyer JC, Slis IH. Nasaliteitsmeting met de nasometer. Logoped Foniatr. 1991; 63: 97-101.

Van Doorn J, Purcell A. Nasalance levels in the speech of normal Australian children. Cleft Palate Craniofac J. 1998; 35: 287-292.

Watterson T, Hinton J, McFarlane SC. Novel stimuli for obtaining nasalance measures from young children. Cleft Palate Craniofac J. 1996; 33: 67-73.

Watterson T, McFarlane SC, Wright DS. The relationship between nasalance and nasality in children with cleft palate. J Commun Disord. 1993; 26: 13-28.

Wise CM. Applied Phonetics. Englewood Cliffs, NJ: Prentice Hall; 1957.

Chapter 6

Correlation between the Perceptual Rating of Speech in Dutch Patients with Velopharyngeal Insufficiency and Composite Measures Derived from Mean Nasalance Scores

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Abstract

Objective: To evaluate the potential clinical use of composite measures derived from mean nasalance scores.

Procedure: Speech samples with a normal distribution of phonemes (normal text, NT) and speech samples free of nasal consonants (denasal text, DT) of 43 patients with perceived hypernasality were used. The overall grade of severity, hyperrhinophonia, audible nasal emission, misarticulations associated with velopharyngeal insufficiency and intelligibility were perceptually rated on separate visual analog scales. Mean nasalance scores were computed by the Nasometer for the same speech samples on which the perceptual ratings were performed. From the mean nasalance scores computed for the NT and DT passages the difference and the quotient were calculated. The advantage could be that the derived measures provide some normalization with regard to the performance of the individual speaker. Spearman correlation coefficients were computed between these composite measures and the perceptually rated parameters. The results were compared with the correlation coefficients between the mean nasalance scores and the ratings.

Setting: The Institute of Phoniatrics, Utrecht University Hospital, the Netherlands.

Results: The correlations between the composite measures and the perceptual ratings were generally lower than the correlations between mean nasalance scores and the ratings.

Conclusion: Normalization of the nasalance scores did not enhance the correlation with the perceptual ratings in this study.

Perceptual rating is widely used to document the speech of speakers with velopharyngeal insufficiency (VPI) and to assess the outcome of behavioral or surgical therapy. The rating of hypernasality has proven to be difficult and subjective in nature [1-6]. To avoid the uncertain factors inherent in perceptual rating, instruments intending to quantify the extent of nasality have been developed. Examples of such instruments are the Nasometer® (Kay Elemetrics Corp., N.J., USA) and the NasalView® (Tiger Electronics, Wash., USA). Acoustic nasometry is regarded as an attractive technique because the measurement claims to provide reliable data. Most authors agree that perceptual evaluation serves as the gold standard against which instrumental measures must be validated. Unfortunately, the literature dealing with the extent to which acoustic measurements correspond with perceptual ratings of nasal resonance is not unequivocal. For example, Dalston et al. (7] reported a Spearman correlation coefficient of 0.73 between measure and rating of hypernasality. In that study, nasalance scores were computed for a standard passage free of nasal consonants and the ratings of hypernasality were performed on conversational speech. Keuning et al. [8] reported a Spearman correlation coefficient of 0.57 between the mean nasalance score and the rating of hypernasality of a standard passage in which the occurrence of phonemes is similar to their occurrence in Dutch conversational speech, and a Spearman correlation coefficient of 0.54 for a standard passage free of nasal consonants. In that study, mean nasalance scores were computed for the same passages that were used to perform the perceptual ratings. Nellis et al. [9] did not find a significant correlation between the mean nasalance score and the rating of hypernasality. The inclusion in their study of speakers with a pharyngeal flap and the rating of both hypernasality and hyponasality in the test sentences, confounded by the inability of some of the judges to distinguish between different nasal parameters, were considered to be factors for the poor correlation. In most studies, the mean nasalance score is used as instrumental measure.

However, other measures derived from acoustic nasometry may provide better correlations with the perceptual ratings of hypernasality. There have been few attempts to investigate other measures derived from nasometry. The standard deviation of the mean nasalance score was explored (10, 11) to evaluate its potential clinical use.

It was concluded that the standard deviation has no general clinical utility other than enabling a gross distinction between normal and abnormal resonance. Therefore, it cannot be used to distinguish between speakers with varying degrees of hypernasality. A reason for the unsatisfactory correlation between nasometric measures and ratings of nasal resonance may be the considerable variability of mean nasalance scores computed for speakers with perceptually normal speech. The variability may be explained by an individual variation as well as dialectal aspects of speech [10]. In addition, grading the severity of impairment based on nasalance scores may not agree with perceptual ratings.

Bressmann et al. (12] reasoned that the perceptual impression of nasal speech may not be attributed solely to excessive nasal energy but rather to a lack of oral-nasal sound balance. They assumed that every speaker has an individual range of nasality in speech and that this range may be measured more accurately in terms of maximum and minimum rather than in mean nasalance scores. They introduced two composite measures obtained from acoustic nasometry: nasalance distance and nasalance ratio. Nasalance distance was defined as the difference between the mean nasalance scores computed for sentences loaded with nasal consonants and sentences free of nasal consonants. Nasalance ratio was defined as the quotient of these two nasalance scores. They reported that these measures, obtained by using the NasalView, provide a good discrimination between speakers with and without nasal resonance and that the validity of the measures is confirmed by the favorable results regarding sensitivity and specificity. They did not give an explanation for the outcome. However, the outcome may be explained by a decrease of the intersubject variability in the composite measures as compared to the mean nasalance scores. The use of a difference between or a quotient of the mean nasalance scores computed for varying passages may reduce patient-dependent effects on the instrumental measure. If this is true, the correlation between the composite measures and the perceptual ratings of hypernasality will increase and will be higher than the correlation between the mean nasalance scores and the perceptual ratings. Because we found only moderate correlation coefficients for the latter relation in a previous study [8], we were interested in whether higher correlation coefficients could be found when composite measures were used. The results will be compared with the correlation between the perceptual rating and the mean nasalance score.

Material and Method

Subjects

Subjects for this study were selected from the patients with perceived hypernasality who visited the outpatient clinic at the Institute of Phoniatrics of Utrecht University Hospital between March 1995 and March 1997. From this group, 43 subjects (26 boys and men and 17 girls and women, median 8 years, range 4-83 years) whose clinical records and tape recordings were complete were included. There were no severe

cases of nasal resonance in the subjects with congenital defects. In order to get a distribution over the whole range from mild to severe, 4 patients (table 1) with severe hyperrhinophonia with another origin than congenital defects were added.
The patients read two standard passages in Dutch. Patients who were too young or unable to read the speech samples easily were asked to repeat the sentences after the examiner.

Passages

The phonemes in the text of the first passage are representative of the distribution of phonemes in the Dutch language (normal text, NT). In the Dutch language, 11.67% of the consonants are nasal. The second passage is a text free of nasal consonants (denasal text, DT). Each passage contained approximately 70 words, which is comparable to the ZOO passage.

Instrumentation

Nasalance was measured with the Nasometer, model 6200 (Kay Elemetrics Corp., N.J., USA). During testing, the speech samples were recorded simultaneously on tape for the perceptual rating. The microphone of the tape recorder was placed at a distance of 50 cm. Mean nasalance was ca1culated between the onset and offset of the nasometric signal displayed for each speech sample using the nasometer software (version 3.22). All speech samples were recorded in a sound-treated room. Tape-recorded speech samples (NT and DT) were digitized at 40 kHz using the Computerized Speech Laboratory (CSL, type 4300, Kay Elemetrics Corp., N.J., USA). All text not spoken by the patient was removed from the files using the CSL software.

Judges

Six speech-language pathologists (SLPs) rated the speech samples independently.

Table 1. Primary diagnosis of the 43 subjects employed in the present study

Diagnosis	Frequency	Percentage
Congenital palatopharyngeal incompetence (short velum, deep pharynx, or both)	22	51
Unilateral cleft of primary and secondary palate	9	21
Bilateral complete cleft of primary and secondary palate	4	9
Iatrogenic (resection of velum)	2	5
Robin sequence	1	2
CHARGE[1] syndrome with short velum	1	2
Cleft of secondary palate involving hard palate and velum	2	5
Dysarthric involvement of velopharyngeal mechanism	2	5
Total	43	100

[1] The diagnosis CHARGE syndrome refers to children with a specific set of birth defects. These defects are coloboma, heart defects, atresia of the choanae, retardation of growth and development, genital and urinary abnormalities, ear abnormalities and hearing loss.

Rating

Digitized speech samples were presented to 2 SLPs simultaneously in a sound-treated room at a comfortable listening level, both being equidistant from the audiospeaker of the CSL system. The speech samples were presented 3 times and randomized before each rating session in order to eliminate any order effects. No information about the patients was provided.

Ratings of the speech samples took place on 1 day, with a break of 30 min between sessions. Each rating session lasted approximately 2 h and 15 min. Speech samples were rated on (a) the overall grade of severity, (b) degree of excess in nasal resonance (hyperrhinophonia), (c) audible nasal emission, (d) misarticulations associated with VPI, and (e) intelligibility

[13]. Each speech sample was rated on a score sheet with five visual analog scales (VAS) of 100 mm underneath one another. The endpoints of the scales were defined as 'normal' (left side) and 'extremely deviant' (right side). The judges were asked to indicate by a mark on the VAS to what extent the speech samples demonstrated the given characteristic. Just prior to the rating session, a task-specific listening orientation was conducted during which the listeners were provided with oral and written instructions and definitions of scale terms. Three preselected speech samples, each presenting a clear example of hyperrhinophonia, audible nasal emission, or misarticulations associated with VPI, were presented to the judges. The ratings of these examples by 2 experienced judges (not participating in this experiment) were given and discussed to ensure that the judges were listening to the same percept. These samples were not included among the samples to be rated. The task-specific orientation lasted approximately 30 min.

Data Analysis

The nasalance scores were normalized by subtracting the mean nasalance scores computed for the DT passages from the mean nasalance scores computed for the NT passages and by dividing the mean nasalance scores computed for the NT passages by the mean nasalance scores computed for the DT passages. The correlation was computed between the arithmetic means of the ratings by the 6 judges and the difference between the mean nasalance scores computed for NT and DT passages. In addition, the correlation was computed between the arithmetic means of the ratings by the 6 judges and the quotient of both mean nasalance scores. As the values of the perceptually rated parameters appeared to be nonnormally distributed, Spearman correlation coefficients were computed. Data analysis was performed using the Statistical Package for Social Sciences, version 8.0 for Windows (SPSS Inc., Chicago, Ill., USA).

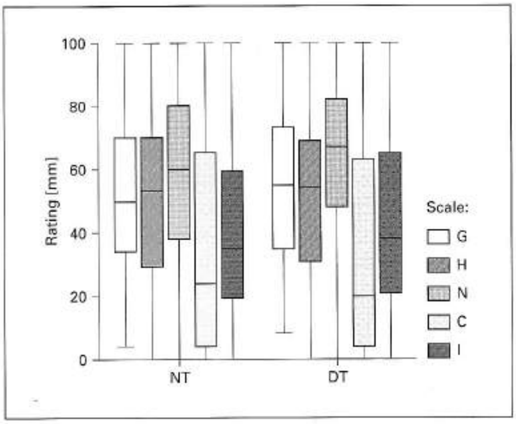


Fig. 1. Box-whisker plot distribution of the mean perceptual ratings of the speech of patients with VPI for speech samples with a normal distribution of phonemes (NT) and speech samples free of nasal consonants (DT). G = Overall grade of severity; H = hypernasality; N = audible nasal emission; C = compensatory misarticulations; I = intelligibility. For each patient the mean of 6 judges on a VAS was used. 25–75 percentiles (boxes), median values (horizontal lines), and maximum–minimum values (whiskers) are given.

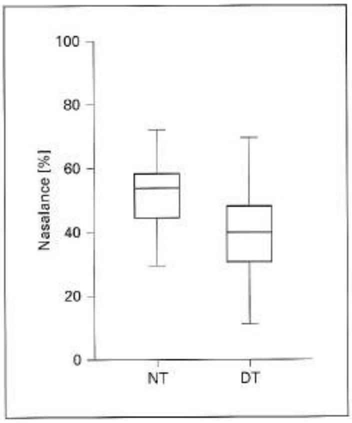


Fig. 2. Box-whisker plot distribution of the nasalance scores of the speech samples with a normal distribution of phonemes (NT) and the speech samples free of nasal consonants (DT).

Results

The distribution of the values rated by the 6 SLPs for the NT and DT speech samples is shown in figure 1. The average score for the mean nasalance computed for the NT speech samples was 51.5% (SD 10.7) and 40.1% (SD 14.0) for the DT speech samples (fig. 2). Table 2 shows the Spearman correlation coefficients between the composite measures derived from mean nasalance scores and the parameters of the differentiated perceptual rating for NT and DT speech samples separately. The correlation coefficients between the mean nasalance scores and the parameters of the differentiated perceptual rating are given to compare the present with previously reported results [8].

As the mean nasalance computed for a NT passage and a DT passage will become similar in case of severe hypernasality, the value for (NT-DT) will decrease with increasing hypernasality. Therefore, the correlation coefficient of this measure with the perceptual rating will become negative.

The correlation between the parameters of the perceptual rating and the mean nasalance is generally higher than between these parameters and the normalized nasalance measures, although this difference is not significant.

Specifically, the correlation between the rating and the difference between NT and DT is distinctly lower than the correlation between the rating and the mean nasalance score.

Table 2. Spearman correlation coefficients between composite measures/mean nasalance scores and the perceptual rating for speech samples with a normal distribution of phonemes (NT) and speech samples free of nasal consonants (DT) (n = 43)

	Mean nasalance		
	NT–DT	NT/DT	NT
Perceptual rating for NT passages			
Overall grade of severity	–0.26	0.33*	0.41**
Hyperrhinophonia	–0.35*	0.49**	0.57**
Audible nasal emission	0.01	0.08	0.28
Compensatory misarticulations	–0.25	0.32*	0.41**
Intelligibility	–0.33*	0.37*	0.37*
Perceptual rating for DT passages			
Overall grade of severity	–0.32*	0.39**	0.40**
Hyperrhinophonia	–0.33*	0.49**	0.54**
Audible nasal emission	0.04	0.07	0.18
Compensatory misarticulations	–0.27	0.27	0.23
Intelligibility	–0.31*	0.35*	0.34*

* Correlation is significant at the 0.05 level (two-tailed).
** Correlation is significant at the 0.01 level (two-tailed).

Discussion

The principal aim of physical or behavioral therapy in speakers with VPI is to obtain intelligible speech of a pleasing quality. Consequently, the perceptual evaluation is the main criterion for management decisions and the human ear is the final arbiter in determining whether a satisfactory therapeutic result has been achieved.

Acoustic nasometry must not be considered as a substitute for the perceptual evaluation, but as a tool to quantify nasal resonance before and after therapy as part of a multidimensional diagnostic process.

Even though most authors agree that the usefulness of the instrumental measure essentially depends on its positive correlation with the perceptual rating by judges with expertise in cleft palate speech [4, 13, 14], the results of our earlier experiments [6, 8] did not support the opinion that SLPs with expertise in the assessment of cleft palate speech are obligatory for a reliable rating with a strong correlation with the instrumental measure. However, it is necessary to continue the search for measures with an optimal correlation with the perceptual rating.

In the present study, the use of composite measures derived from mean nasalance scores did not yield a higher correlation with the perceptual rating than the mean nasalance score itself. For the parameter hyperrhinophonia, the correlation coefficient decreased from 0.57 (for the mean nasalance score) to 0.49 and 0.35 (for composite measures derived from the mean nasalance score) for the NT passage and from 0.54 to 0.49 and 0.33 for the DT passage. Consequently, the use of our composite measures has little clinical value in the sense that they do not improve the correlation between measure and rating.

In the literature [12] it is reported that composite measures improve the discrimination between hypernasal speech and normal speech. An explanation for that finding could be that the nasalance score is influenced by a patient-dependent error, independent of the number of nasal consonants. If, due to this error, the nasalance scores for passages with a different number of nasal consonants will increase or decrease, the difference between both passages may be a better measure than the mean nasalance score. However, if such an error does not exist, a composite measure will have a higher variance than the mean nasalance score, because in that case the variances of both nasalance scores will add up. Consequently, the correlation between measure and rating will decrease. Since the Nasometer measures only the acoustic aspect of nasalization in speech, there is no theoretical basis for another correlation than that between nasometric measures and the perceptual rating of hyperrhinophonia. Specifically, the correlation between the nasalance scores and the rating of misarticulations associated with VPI is curious and may be a factor in the finding that hyperrhinophonia is rated more severely in the presence of other symptoms, such as misarticulations [4]. Moreover, all parameters of the differentiated perceptual rating appeared to be mutually correlated.

Limitations of the present study have also to be considered. The outcome of this study cannot be compared with the literature. Only data on sensitivity and specificity of composite measures derived from mean nasalance scores have been reported [12], providing information that is different from a correlation analysis. Considering the reported data on sensitivity and specificity of the composite measures and assuming that our explanation is correct, the outcome of the present correlation study is disappointing.

However, the measuring equipment, the composition of the measures and the language may have influenced the outcome.

A. Acoustic nasometry was performed by using the Nasometer. Nasalance scores computed with the NasalView tend to be higher for passages free of nasal consonants and for passages with a normal distribution of phonemes than for those computed by the Nasometer.

Moreover, the nasalance scores computed with the NasalView tend to be lower for passages loaded with nasal consonants than for those computed by the Nasometer [15].

B. A passage with a normal distribution of phonemes and a passage free of nasal consonants were used to compute the difference and the quotient. Because the mean nasalance score for a passage loaded with nasal consonants is higher than that for a passage with a normal distribution of phonemes, the difference between nasalance scores for (NT-DT) is smaller than the difference between a passage loaded with nasal consonants and a passage free of nasal consonants. However, it must be noticed that passages loaded with nasal consonants are apparently better suited for perceptual evaluation of speech of speakers with perceived hyponasality [9, 16-18].

C. The normative nasalance scores computed by the Nasometer for the German language appeared to be slightly higher than the normative nasalance scores computed for the Dutch language. For the German language, normative nasalance scores of 13.0% for passages free of nasal consonants, 67.2% for passages loaded with nasal consonants and 33-41% for mixed (phonetically nonbalanced) passages were computed [19-21]. For the Dutch language, normative nasalance scores of 11.75% for passages free of nasal consonants, 52.33% for passages loaded with nasal consonants and 31.95% for passages with a normal distribution of phonemes were computed [22, 23].

If we had used the NasalView and passages loaded with nasal consonants [12] higher correlation coefficients might have been computed, although the results of this study do not suggest that.

Conclusion

Normalization of the nasalance scores to the performance of the speaker, by using the difference between or the quotient of the mean nasalance scores computed for speech samples with a normal distribution of phonemes (NT passages) and speech samples free of nasal consonants (DT passages), did not enhance the correlation with the perceptual ratings of hyperrhinophonia. On the contrary, as a consequence of the computation of the difference and the ratio, the variance increased and therefore the correlations decreased as compared to the correlations with the mean nasalance scores. Consequently, the use of these composite measures must not be encouraged.

References

1 Counihan DT, Cullinan WL: Reliability and dispersion of nasality ratings. Cleft Palate J 1970; 7: 261-270.

2 Carney PJ, Shennan D: Severity of nasality in three selected speech tasks. J Speech Hear Res 1971; 14: 396-407.

3 Moore WH, Sommers RK: Phonetic contexts: Their effects on perceived nasality in c1eft palate speakers. Cleft Palate J 1973; 10: 72-83.

4 McWilliams BJ, Moms HL, Shelton Rl: Cleft Palate Speech, Philadelphia, Decker, 1990.

5 Shprintzen Rl, Bardach J: Cleft Palate Speech Management: A Multidisciplinary Approach. St Louis, Mosby-YearBook, 1995.

6 Keuning KHD, Wieneke GH, Dejonckere PH: The intrajudge reliability of the perceptual rating of c1eft palate speech before and after pharyngeal flap surgery: The effect of judges and speech samples. Cleft Palate Craniofac J 1999; 36: 328-333.

7 Dalston RM, Neiman GS, Gonzalez-Landa G: Nasometric sensitivity and specificity: A cross-dialect and cross-culture study. Cleft Palate Craniofac J 1993; 30: 285-291.

8 Keuning KHD, Wieneke GH, Van Wijngaarden HA, Dejonckere PH: The correlation between nasalance and a differentiated perceptual rating of speech in Dutch patients with velopharyngeal insufficiency. Cleft Palate Craniofac J 2002; 39: 277-284.

9 Nellis JL, Neiman GS, Lehman JA: Comparison of nasometer and listener judgements of nasality in the assessment of

velopharyngeal function after pharyngeal flap surgery. Cleft Palate Craniofac J 1992; 29: 157-163.

10 Seaver EJ, Dalston RM, Leeper HA, Adams LE: A study of nasometric values for normal resonance. J Speech Hear Res 1991; 34: 715-721.

11 Vallino-Napoli LD, Montgomery AA: Examination of the standard deviation of mean nasalance scores in subjects with cleft palate: Implications for clinical use. Cleft Palate Craniofac J 1997; 34: 512-519.

12 Bressmann T, Sader R, Whitehall TL, Awan SA, Zeilhofer HF, Horch HH: Nasalance distance and ratio: Two new measures. Cleft Palate Craniofac J 2000; 37: 248-256.

13 Hirschberg J, Van Demark DR: A proposal for standardization of speech and hearing evaluations to assess velopharyngeal function. Folia Phoniatr Logop 1997; 49: 158-167.

14 Haapanen M-L: Nasalance scores in normal Finnish speech. Folia Phoniatr (Basel) 1991; 43: 197-203.

15 Awan SN: Analysis of nasalance: NasalView; in Ziegler W, Deger K (eds): Clinical Phonetics and Linguistics. London, Whurr, 1997, pp 518-527.

16 Dalston RM, Warren DW, Dalston ET: A preliminary investigation concerning the use of nasometry in identifying patients with hyponasality and/or nasal airway impairment. J Speech Hear Res 1991; 34: 11-18.

17 Dalston RM, Seaver EJ: Relative va1ues of various standardized passages in the nasometrlc assessment of patients with velopharyngeal impairment. Cleft Palate Craniofac J 1992; 29: 17-21.

18 Watterson T, McFarlane SC, Wright DS: The relationship between nasalance and nasality in children with cleft palate. J Commun Disord 1993; 26: 13-28.

19 Heppt W, Westrich M, Strate B, Moehring L: Nasalanz: Ein neuer Begriff der objektiven Nasalitätsanalyse. Laryngorhinootologie 1991; 70: 208-213.

20 Reuter W, Eichler M, Gross M: Objektive Nasalitätsmessung: Bestimmung der Normalschwelle; in Gross M, Eysholdt K (eds): Aktuelle phoniatrisch-pädaudiologische Aspekte. Heidelberg, Median- Verlag, 1997, vol 5, pp 193-196.

21 Mueller R, Beleites T, Hloucal U, Kuehn M: Objektive Messung der normalen Nasalanz im sächsischen Sprachraum. HNO 2000; 48: 937-942.

22 Van De Weyer JC, Slis IH: Nasaliteitsmeting met de nasometer. Logopedie Foniatrie 1991; 63: 97-101.

23 Van Zundert M, Rietveld T, Mugge AM: Normgegevens voor het gebruik van de nasometer bij jonge kinderen. Stem- Spraak Taalpathol 1998; 7: 237-247.

Chapter 7

General discussion, address to the aims and future perspectives

1. The surgical aspect

Since the posterior pharyngeal wall flap has been introduced in 1875 (23) favourable and unfavourable outcome of this type of surgery has been discussed. Initially, some surgeons advocated the use of the pharyngeal flap to correct hypernasality in speech while others rejected this type of flap surgery. This controversy was mainly because the pharyngeal flap was considered to be unphysiological. It was argued that by 'pushback' or 'retropulse' the mucosa of the hard palate, more lenght of the soft palate could be obtained without compromising the physiology or the anatomy (13). Today most authors accept the important role of the pharyngeal flap to treat persistent hypernasality after primary cleft palate repair (16), especially in severe cases. However, there is a revival of the historic controversy. Recently, the use of the pushback operation has been re-advocated (4, 7, 17).

The pharyngeal flap provides a static obstruction in the midline of the nasopharynx and leaves two lateral openings or ports which should remain patent during breathing and nasal consonant speech production and should be closed by the medial movement of the lateral pharyngeal walls against the flap during the production of oral consonants.

Two types of posterior pharyngeal wall flaps can be distinguished: the inferiorly based or the superiorly based posterior pharyngeal wall flap. A long-standing discussion is the decision to perform the inferiorly based or the superiorly based flap. Both types of flaps have advantages and disadvantages and as a consequence their supporters and their opponents. In brief the advantages of the superiorly based flap are the easier control of postoperative bleeding and the easier way to raise a flap with greater lenght that can bridge a larger gap. On the other hand the inferiorly based flap is easier to attach to the soft palate. A disadvantage of the inferiorly based pharygeal flap may be the inferior tethering of the flap below the palatal plane and in opposite direction of the motion need to effect velopharyneal closure.

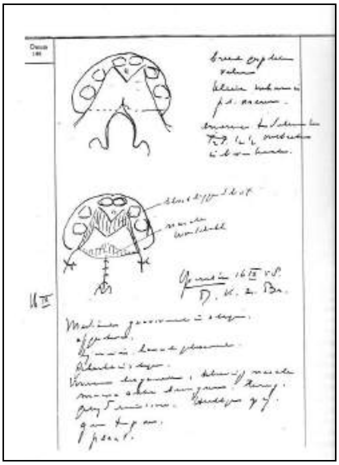
Inferiorly based posterior pharyngeal wall flap tethering the velum inferiorly and below the palatal plane.

However, Whitaker et al. (31) did not find any difference in speech outcome, or complication rate comparing the superiorly based with the inferiorly based posterior pharyngeal wall flap. Moreover, Skoog (26) compared both types of flaps and was unable to demonstrate any significant difference in either short or long-term effect and found it difficult to conclude which flap was used after examination of the oropharynx.

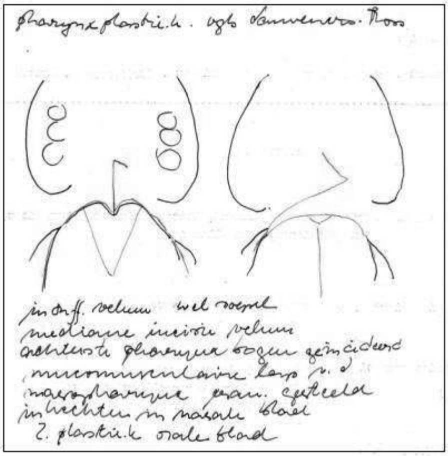
Several modifications of the originally described technique of the superiorly based pharyngeal flap have been proposed. These modifications are intended to increase woundhealing and to decrease scar contraction, thinking that the width of the flap and the diameter of the lateral ports, as designed during the operation, will be stable.

There is still a dispute on the proper place of insertion of the superiorly based flap into the velum. Two main streams oppose: the soft palate can be split along the midline (2, 14) or can be split transversely. The transverse split can be combined with a pushback of the palate, following a releasing division of the nasal mucosa and the velar muscles from the posterior edge of the hard palate (9, 20, 32). Subsequently, the pharyngeal flap may be inserted either into the velum that is surgically divided in the midline or that is divided transversely. Although both surgical designs intend to provide normal resonance in speech, the theoretical basis of these designs is different. The former design is based on obturation by the pharyngeal flap and the relying upon the dynamic separation of the oropharynx from the nasopharynx by the medial movement of the lateral pharyngeal walls against the flap, whereas the latter design is based on the relying of the velar movement for the dynamic separation of the oropharynx from the nasopharynx. In the latter design the pharyngeal flap is not expected to serve as an obturator and should not block soft palate motion but should serve as the lining for the nasal pushback raw area between hard and soft palate and to keep the velum from being pulled forward by scar contraction (10).

In a comparison of different pharyngeal flap insertions, with or without pushback, Millard (19), citating a lecture by Lewin et al. (1977), reported that the unlined pharyngeal flap used to cover the defect in the nasal mucosa after the pushback of the palate provides the least obturation because the pedicle of the flap contracts into a tube. The pharyngeal flap inserted into the longitudinal split and sutured to the nasal side of the velum, with the uvula being sutured to the base of the pharyngeal flap to provide lining, is suited for the majority of patients with VPI. Its failures seem to be limited to patients with absence of lateral pharyngeal wall movement.



Surgeon's report. superiorly based pharyngeal flap with pushback of the palate.

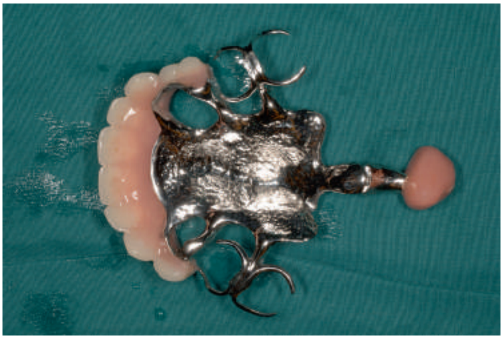


Surgeon's report. superiorly based pharyngeal flap without pushback of the palate.

Despite strongly held views, Karling et al. (11) could not find difference in speech outcome between both types of flaps.

In an international multicenter randomized trial (1) to compare the outcome of superiorly based pharyngeal flap surgery with the outcome of sphincter pharyngoplasty no significant differences in outcome were detected for nasal resonance, nasalance, endoscopic outcomes, or surgical complications at 1 year postsurgery. Moreover, no difference was found between the two procedures in the long-term incidence of sleep apnea.

A dental appliance with a speech bulb appeared to be as effective as a superiorly based pharyngeal flap regarding speech outcome in the treatment of patients with VPI. However, about 30% of the prosthetic treatments failed in consequence of patient's non-compliance. Therefore, use of a speech bulb to solve the problem of VPI is only indicated in selected cases (18).



Partial dental prosthesis (‘chromes’) with a speech bulb. Lingual view

Partial dental prosthesis with a speech bulb. Palatal view

Since the introduction of the pharyngeal flap operation, surgical complications, such as bleeding, flap dehiscence, or even death have been reduced considerably. The introduction of antibiotics and more sophisticated anaesthesiologic procedures contributed to this amelioration. Although the rate of surgical complications is reduced, the nature of the complications is still the same. Moreover, new complications have been recognized and defined. Snoring, an unwanted outcome after pharyngeal flap surgery, was considered as no more than an inconvenience during many decades, these last years it has gained considerable importance due to its relation with the sleep apnea syndrome.

An advantage of the posterior pharyngeal wall flap comparing with the 'pushback' operation is that the dynamics of the closure of the velopharyngeal valve mainly depend on the contraction of the lateral pharyngeal walls (constrictor pharyngis muscle), and not on the velar movement. The constrictor pharyngis muscle is not affected in cleft palate patients, and well-fitted for active training with speech therapy. From this point of view, the anatomical change induced by the surgical procedure and the acquired compensation technique acts as complementary mechanisms.

When relying upon the low complication rate and upon the proportion of necessary re-interventions, the SBPP-flap with a Z-plasty to cover the raw surface of the oral side of the flap can, on the base of our experience, be considered as the intervention of first choice to improve the velopharyngeal function after primary cleft palate repair.

The ultimate goal of pharyngeal flap surgery is to create a velopharyngeal mechanism that separates the oropharynx from the nasopharynx if adequate, and that improves nasal resonance but avoids upper airway obstruction. Velopharyngeal surgery is still more an art than a science (21).

2. The functional aspect

Perceptual evaluation and nasometry

Why is perceptual evaluation of cleft palate speech of limited value?
Nasality is a perceptual phenomenon, mostly related to a change in velopharyngeal anatomy and/or function: so for decades it has been considered that the human ear is the final detector and arbiter in determining whether there is nasal resonance in speech and whether a satisfactory result after

therapy has been achieved (12). However, considering the limited reliability of perceptual evaluation, its clinical value has been frequently questioned in the past, even by highly experienced clinicians. Bzoch (3) considered that only the presence or the absence of nasality could be estimated in a reliable way.

If the quality being rated is multidimensional in nature but is rated on a unidimensional scale, listeners may selectively focus on one dimension or another, reducing apparent agreement levels. For example, deviant voice quality also comprises different dimensions as breathiness and roughness. Some investigators (15) demonstrated that listeners' differential attention to various aspects of each component is a significant source of interrater unreliability in quality ratings. That is, listeners frequently agree about what constitutes normal speech or severe pathology, but disagree more about the extent of mild-to-moderate behaviors.
If the stimulus set includes a large number of (sub)normal samples, the levels of agreement may be misleadingly high. Experts in speech evaluation, such as speech-language pathologists or phoniatricians, can judge many different details of the speech of a test subject, while holistic features, e.g. the speech intelligibility, have the advantage that they often can be quantified easily even by naïve listeners, and give a much better insight on the type and extent of the speech disorder.

Speech is the cornerstone of social integration and peer acceptance. To evaluate resonance in speech and/or to evaluate velopharyngeal function of a patient with a palatal cleft perceptual evaluation of speech must be performed by an expert. The goal of the perceptual evaluation is to determine whether an abnormality in speech exists and if so, the type and severity is assessed. As nasality is basically an acoustic

phenomenon, one would like to have at disposal purely acoustic criteria – as formant shifts- for supporting perception (29), but there is to date no valid and uniform diagnostic method which makes possible the analysis, documentation and comparison of changes in timbre (6).
Considering the complexity of disordered speech in VPD and the related adaptation and compensation mechanisms, our results in comparing nasometry and perception seem to point out an erroneous hypothesized relationship between the acoustic measures and their proposed perceptual consequences.

What kind of instrumental approach can validly be used beside perception?

Nasometry is obviously a physiologically based measurement; it has been found reliable (28) and discriminates accurately normal subjects from patients with VPI (8). However, it specifically accounts for a selected aspect of the distribution of the (filtered) acoustic energy, which is not comparable with the complexity of the central auditive pathways and their cognitive webwork.
Nevertheless, one may consider that an improvement in nasometric scores indicates an improvement of velopharyngeal function. If in a particular case this is not on a par with perceptual rating, the clinician needs to investigate and understand the underlying reason, e.g. a dyspractic articulation pattern.

3. The importance of a multidimensional approach for increased nasal resonance.

The statement of Kent (12) that "auditory-perceptual judgments are typically the final arbiter in clinical decision-making and often provide the standards against which instrumental, so-called 'objective' measures are evaluated", is in our opinion questionable, at least for the second part of the sentence. Our research rather points out that there is no single measure or rating that can account for all aspects of nasality.

The appropriate method for measuring what listeners hear when they listen to voices remains an unresolved issue and providing accurate, replicable, valid measures of speech quality presents significant challenges. However, as soon as it is pertinent to the investigated phenomenon, a physiologically based and clinically validated objective measurement keeps its intrinsic value, independently of perceptual rating.

In our opinion it is plausible that results of different dimensions are only weakly correlated with each other (if there was a high correlation, there would be no need for several dimensions!)

Discrepancies between outcomes of different dimensions help to better understand how treatments really are working, and need individual critical examination.

4. A Proposal for clinical routine assessment, suitable for applied research.

As a result of our findings, discussions and conclusions, proposals can be made for a multidimensional diagnostic protocol of cleft palate speech in patients (children and adults): An important issue is that such a protocol is suitable for routine clinical work, but also that it is built up in such a manner that its data can be used a posteriori for research purposes, particularly applied research on efficacy of treatments. Also recent new developments, originated from advanced Information and Communication Technology (ICT), such as Automatic Speech Recognition systems (ASR) will be considered, particularly from the point of view of their potential clinical relevance.

We propose a 5-dimensional protocol, including perception, endoscopic morphodynamics, acoustic nasometry, aerodynamics and self-evaluation (speech related quality-of-life)

*Perception

It is suggested to use a standard rating scale comprising differentiated aspects: the nasal resonance, the audible nasal air emission and the compensatory misarticulations during speech. Furthermore the intelligibility and the voice quality need to be taken in account separately. All these parameters can be scored on visual analog scales (cf. Chapter 4).
It is suggested to use the same speech material (a phonetically balanced text containing the proportion of nasal phonemes that is characteristic for the language) for perceptual rating as for nasometry. For young children unable to read, a set of short adapted sentences of 3-4 words is required: the child then repeats sentence by sentence.
Additionally, the speech-language therapist can perform a systematic articulation test, in order to make a detailed inventory and transcription of the altered phonemes.
Essential is to record the reading (or repeating) of the patient, in order to make possible a blind rating a posteriori for research purposes. In case of a young child, the voice of the parent or the speech-language therapist can easily be removed. Paired comparisons (e.g.) pre-/post treatment are – according to our findings in Chapter 4 – to be recommended.

*Endoscopy (rating morphodynamics)

Videonasopharyngoscopy has become the basic procedure for morphodynamic evaluation of the velopharynx, and is an indispensable tool for any clinical examination. A dedicated protocol for velopharyngeal dysfunction has still to be worked out and validated (types and degrees of closure), in order to obtain an optimal reproducibility of ratings, but it seems that it should include the observation of different graded conditions: rest – sustained denasal phoneme at comfortable loudness - sustained denasal phoneme loud – sustained fricative – short denasal sentence. This grading provides relevant information about the plasticity of the velopharyngeal mechanism. In the same way as for perception, the endoscopy needs to be recorded, for possible blind evaluation a posteriori.

*Aerodynamic test (rating nasal air escape)

The mirror-fogging test from Glatzell cannot be avoided, as it is extremely easy to perform and instantaneously provides totally reliable and physiologically based information about presence or absence of nasal airflow, which is distinct information from the other dimensions. Nasal air escape is assessed by evaluating the degree of condensation on a cold metallic mirror held 0,5 cm under the nose in the same test conditions as described for endoscopy. The test result is either (+) = air escape or (-) = no air escape. Van Lierde and collegues (28) have proposed a more precise scale (four concentric circles on the mirror), but this requires a temperature control of the mirror.

*Acoustic Nasometry

For routine clinical work, the standard text as used in chapter 4 is recommended, as it is representative for the language of the patient. Adapted material, as proposed by Hogen Esch and Dejonckere (8), is available for children, and it demonstrated excellent discrimination between normal and hypernasal children.

*Self-evaluation by patient/ caregiver (speech related quality-of-life)

Vide infra in 'Future research'.

5. Future research

Two aspects are currently in development and obviously require applied research in the field of nasal speech assessment, particularly in effect studies of treatments (surgical, functional or prosthetic):

(1) The quantification of intelligibility by using dedicated Automatic Speech Recognition systems.

Our research has demonstrated that the rating of the parameter 'Overall grade of severity' appears to be mainly determined by the parameter 'Intelligibility' which emphasizes the crucial importance of intelligibility in VPI. An important breakthrough is occurring currently due to new developments in ICT. Intelligibility of the speech as influenced by cleft palate can be quantified objectively and numerically by means of automatic speech recognition technology in speech recordings (30). If an automatic speech recognition (ASR) system is trained with normal speakers' speech data (adults or children), the system learns the correct utterance and if it is tested with hypernasal speech data, it may clearly indicate the speech impairment rating. Based on these aspects, researchers have focused on utilizing an ASR system for the assessment of hypernasal speech. The automatic speech evaluation can focus on the word accuracy which represents the percentage of correctly recognized words (24). The technique avoids subjective influences from human raters with different experience and is therefore of high clinical and scientific value. Tracking of the children's therapy progress is also within the reach of the system. Automatic evaluation in real-time will avoid long evaluation proceedings by human experts. However, the method is language-dependent, and a standardized protocol is mandatory for each language. Furthermore, it must be suitable for children as well as adults.

(2) Self-evaluation by patient/ caregiver of the speech related quality of life.

The introduction in clinical practice of patient's self-evaluation tools of speech related quality of life, in the same way as the Voice Handicap Index for dysphonia. Recently a Speech Handicap Index (SHI) has been developed; aiming to the speech related quality of life. It has been tried out in treated head and neck cancer patients, and a psychometric study indicates that the SHI is a reliable and valid questionnaire for assessing speech problems (22). Further research is needed for evaluating the adequacy in the field of nasality, as well as the suitability of a care-givers evaluation in case of young children.

References

1. Abyholm F, D'Antonio L, Davidson Ward SL, Kjøll L, Saeed M, Shaw W, Sloan G, Whitby D, Worhington H, Wyatt R. VPI Surgical Group. Pharyngeal flap and sphincterplasty for velopharyngeal insufficiency have equal outcome at 1 year postoperatively: results of a randomized trial. Cleft Palate Craniofac J 2005; 42: 501-511.

2. Blijdorp P, Müller H. The influence of the age at which the palate is closed on the speech in the adult cleft patient. J Maxillofac Surg 1984; 12: 239-246.

3. Bzoch KR Measurement and assessment of categorical aspects of cleft palate speech. In: Communicative disorders related to cleft lip and palate. Little, Brown & Company, Boston 1989: 137-173.

4. Chen GF, Zhong LP. A bilateral musculomucosal buccal flap method for cleft palate surgery. J Oral Maxillofac Surg 2003; 61: 1399-1404.

5. Dwivedi RC, St Rose S, Roe JW, Chisholm E, Elmiyeh B, Nutting CM, Clarke PM, Kerawala CJ, Rhys-Evans PH, Harrington KJ, Kazi R. First report on the reliability and validity of speech handicap index in native English-speaking patients with head and neck cancer. Head Neck Surg 2011: 33: 3: 341-348.

6. Gugsch C, Dannhauer KH, Fuchs M. Evaluation of the progress of therapy in patients with cleft lip, jaw and palate, using voice analysis-a pilot study. J Orofac Orthop 2008; 69: 257-267.

7. Hill C, Haydan C, Riaz M, Leonard AG. Buccinator Sandwich pushback: A new technique for treatment of secondary velopharyngeal incompetence. Cleft Palate-Craniofac J 2004; 41: 230-237.

8. Hogen Esch TT, Dejonckere PhH. Objectivating nasality in healthy and velopharyngeal insufficient children with the nasalance Acquisition system (Nasalview®). Defining minimal required speech task assessing normative values for Dutch language. Int J Ped Otol 2004: 68: 1039-1046.

9. Honig CA. Over pharyngoplastiek. PhD-thesis Utrecht University 1963.

10. Jobe R. The pharyngeal pushback. In: Operative techniques in Plastic and Reconstructive surgery. Elsevier Inc 1995: Vol2 245-249.

11. Karling J, Henningsson G, Larson O, Isberg A. Comparison between two types of pharyngeal flap with regard to configuration at rest and function and speech outcome. Cleft Palate Craniofac J 1999: 36: 154-165.

12. Kent RD. Hearing and believing: some limits to the auditory-perceptual assessment of speech and voice disorders Am J Speech Lang Pathol 1996: 55: 7–23.

13. Keuning KHDM, Crisi LMTN. Velopharyngoplasty according to Sanvenero Rosselli. Historical review and contempory judgement. Mund Kiefer GesichtsChir 2000: 95-98.

14. Keuning KHDM, Meijer GJ, van der Bilt A, Koole R. Revisional surgery following the superiorly based posterior pharyngeal wall flap. Historical perspectives and current considerations. Int J Oral Maxillofac Surg 2009:38: 1137-1142.

15. Kreiman J, Gerratt BR, Berke GS. The multidimensional nature of pathologic vocal quality. J Acoust Soc Am 1994: 96: 3: 1291-1302.

16. Kummer AW, Clark SL, Redle EE, Thomsen LL, Billmire DA. Current practice in assessing and reporting speech outcomes of cleft palate and velopharyngeal surgery: a survey of cleft palate/craniofacial professionals. Cleft Palate Craniofac J 2012: 49: 146-152.

17. Mann RJ, Neaman KC, Armstrong SD, Ebner B, Bajnrauh R, Naum S. The double-opposing buccal flap procedure for palatal lenghtening. Plast Reconstr Surg 2011; 127: 2413-2418.

18. Marsh JL, Wray RC. Speech prosthesis versus pharyngeal flap: a randomized evaluation of the management of velopharyngeal incompetency. Plast Reconstr Surg 1980: 65; 592-594.

19. Millard DR. Cleft craft. The evolution of its surgery. III Alveolar and palatal deformities. Little, Brown and comp Boston 1980: 685-687.

20. Mink van der Molen AB, Janssen K, Specken TF, Stubenitsky BM. The modified Honig velopharyngoplasty- A new technique to treat hypernasality by palatal lenghtening. J Plast Reconstr Aesth Surg 2009: 62: 646-649.

21. Patel PK, Grasseschi MF, McGraw EK, O'Gara MM, Ramaswamy R, Witt PD. Surgical treatment of velopharyngeal dysfunction. 2011 Medscape reference©.

22. Rinkel RN, Verdonck-de Leeuw IM, Van Reij EJ, Aaronson NK, Leemans CR. Speech handicap index in patients with oral and pharyngeal cancer: better understanding of patients’ complaints. Head Neck Surg 2008: 30: 7: 868-874.

23. Schoenborn KWEJ. Ueber eine neue Methode der Staphylorrhaphie. Verh Dtsch Ges Chir 1875: 4: 235-239; Arch Klin Chir 1876: 19: 527-531.

24. Schuster M, Maier A, Haderlein T, Nkenke E, Wohlleben U, Rosanowski F, Eysholdt U, Nöth E. Evaluation of speech intelligibility for children with cleft lip and palate by means of automatic speech recognition. Int J Pediatr Otorhinolaryngol 2006: 70: 10: 1741-1747.

25. Sell D, Grunwell P, Mildinhall S, Murphy T, Cornish T, Bearn D, Shaw W, Murray J, Williams A, Sandy J. Cleft Lip and Palate Care in the United Kingdom — The Clinical Standards Advisory Group (CSAG) Study. Part 3: Speech Outcomes. Cleft Palate Craniofac J 2001: 38: 1: 30–37.

26. Skoog T. The pharyngeal flap operation in cleft palate. Br J Plast Surg 1965; 18: 265.

27. Van Lierde KM. Nasalance and nasality in clinical practice. PhD-thesis. Ghent University 2001.

28. Van Lierde KM, Schepers S, Timmermans L, Verhoye I, Cauwenberge P The impact of mandibular advancement on articulation, resonance and voice characteristics in Flemish speaking adults: a pilot study. Int J Oral Maxillofac Surg 2006: 35(2): 137-144.

29. Vogel AP, Ibrahim HM, Reilly S, Kilpatrick N. A comparative study of two acoustic measures of hypernasality. J Speech Lang Hear Res 2009: 52: 6: 1640-1651.

30. Vogt B, Maier A, Batliner A, Nöth E, Nkenke E, Eysholdt U, Schuster M. Numeric quantification of intelligibility in schoolchildren with isolated and combined cleft palate. HNO 2007: 55: 11: 891-898.

31. Whitaker LA, Randall P, Graham WP, Hamilton RW, Winchester R. A prospective and randomized series comparing superiorly and inferiorly based posterior pharyngeal flaps. Cleft Palate J 1972: 9: 304-311.

32. Winters HJP. Het aangeboren te korte verhemelte. PhD-thesis Utrecht University 1975.

Chapter 8

Summary

Chapter 1. In this chapter the consequences on speech of structural malformations such as a palatal cleft and/or velopharyngeal insufficiency are outlined. Whenever a palatal cleft or incompetent velopharyngeal mechanism is present, speech is generally affected by obligatory distortions such as increased nasal resonance and air emission as well as compensatory misarticulations. In case of these structural malformations, surgery or other forms of physical management are needed for a functional and competent velopharyngeal mechanism. An introduction to cleft palate speech and various operations that are available to treat velopharyngeal insufficiency is given.

Chapter 2. This chapter is devoted to an analysis of Sanvenero Rosselli's original papers and a historic discussion of the alternatives: the inferiorly based pharyngeal flap and the "pushback" or retropulsion procedure. The history of the pharyngeal flap is hunt out. The superiorly based pharyngeal flap operation has been mainly advocated by Gustavo Sanvenero Rosselli. However, this type of pharyngeal flap has originally been described by Schoenborn from Germany as early as 1886.

Chapter 3. This chapter reflects the long term experience of the department of oral and maxillofacial surgery with the superiorly based posterior pharyngeal wall flap with a Z-plasty to cover the raw surface of the flap. Records of 130 patients with velopharyngeal insufficiency who had undergone this type of flap surgery as a secondary procedure to reduce nasal resonance in speech are reviewed. Special emphasis is put on the incidence of revisional surgery, as this accounts for either surgical complications or unsatisfactory functional results. Potential risk factors for revisional surgery are analyzed. In case of a too wide flap, hyponasality and obstructed nasal breathing is induced, as well as snoring. In cases of severe tubing of the flap, it is too narrow to achieve sufficient velopharyngeal closure during speech, thereby causing persisting velopharyngeal insufficiency. Tailoring of the flap is discussed, and individual skills of the surgeon seem – rather than experience – to yield a better functional outcome.

Chapter 4. The perceptual rating of speech of 15 patients before and after pharyngeal flap surgery is analyzed. Perceptual assessment of speech should logically be the main indicator of the clinical significance of VPD-related symptoms and as such, it should be an essential part in the diagnosis of VPD, along with physical examination and clinical history. However, despite its well recognized importance, the perceptual assessment presents some limitations due to its subjectivity. A protocol is worked out, comprising differentiated aspects: the nasal resonance, the turbulent nasal air emission and the compensatory articulation during speech. Furthermore, intelligibility and the voice quality are taken into account separately. All these parameters are scored on visual analog scales. Inter-, and intrajudge reliability is quantified. The experience levels of the raters and different types of speech materials are compared. This research clearly indicates the limitations of perceptual assessment: Judges – even highly experienced ones – differ largely in the range they use in their rating, which points out that their internal standards differ from judge to judge. Even intrajudge reliability is moderate. The conclusion is that there is a need for an objective reference that – if noninvasive and based on physiology – can support the perceptual evaluation.

Chapter 5. In this chapter an objective, noninvasive instrumental technique: nasometry is introduced. The essence of nasometry is that, during speech, the emitted nasal and oral acoustic energy (within a specific frequency band: 500 +/- 150 Hz) is registered and quantified separately. The ratio of averaged acoustic nasal energy/ averaged total acoustic energy has been defined as 'nasalance': it reflects the relative amount of nasal acoustic energy in speech, and is expressed as a percentage. High nasalance scores can be expected in patients with velopharyngeal insufficiency, but also in speech samples comprising a high percentage of nasal consonants (phonetic selection). Low nasalance scores will occur in patients with nasopharyngeal or nasal obstruction, but also in speech samples free of nasal consonants. This means that nasometry is really a physiology-based measure, but that it is dependent on standardization of the speech sample (and thus language-dependent) and on normative scores obtained by testing normal speakers. Nasometry is performed together with detailed perceptual rating in 43 VPI-patients (children and adults) by a panel comprising as well highly experienced speech pathologists as general practitioners. The outcome and particularly the fairly low correlation clearly demonstrate that the nasometry measures other phenomena than those perceptually rated by the judges, particularly for cases that are not extremely deviant. This already suggests that nasometry may not be considered as a substitute for the perceptual rating.

Chapter 6. There is a considerable variability of mean nasalance scores computed for speakers with perceptually normal speech. The variability may be explained by an individual variation as well as dialectal aspects of speech. It is reasoned in literature, that the perceptual impression of nasal speech may not be attributed solely to excessive nasal energy but rather to a lack of oral-nasal sound balance. Moreover, every speaker has an individual range of nasality in speech and this range may be measured more accurately in terms of maximum and minimum rather than in mean nasalance scores. For that reason two composite measures obtained from acoustic nasometry can be used. It is reported in literature that these composite measures provide a good discrimination between speakers with and without nasal resonance and that the validity of the measures is confirmed by the favorable results regarding sensitivity and specificity. This can be ascribed to a decrease of the intersubject variability in the composite measures as compared to the mean nasalance scores. The use of a difference between or a quotient of the mean nasalance scores computed for varying passages may reduce patient-dependent effects on the instrumental measure. The question is if normalization of the nasalance scores to the performance of the speaker (43 VPI-patients, children and adults), by using the difference between or the quotient of the mean nasalance scores computed for speech samples with a normal distribution of phonemes and speech samples free of nasal consonants succeeds in enhancing the correlation with the perceptual ratings of nasality.

Chapter 7. In the general discussion, as a result of our findings and conclusions, proposals are made for a multidimensional diagnostic protocol of cleft palate speech in patients (children and adults): An important issue is that such a protocol is suitable for routine clinical work, but also that it is built up in such a manner that its data can be used a posteriori for research purposes, particularly applied research on efficacy of treatments. Also recent new developments, originated from advanced Information and

Communication Technology (ICT) –such as automatic speech recognition systems (ASR) are considered, particularly from the point of view of their potential clinical relevance.

Hoofdstuk 9

Samenvatting

Hoofdstuk 1. Een verhemeltespleet en/of een velopharyngeale insufficiëntie leidt meestal tot misvorming van de spraak. Dit wordt veroorzaakt door een toename van zowel de resonantie en de ontsnappende neuslucht als door het optreden van articulatie stoornissen. Om de onvoldoende afsluiting van het velopharyngeale mechanisme te verbeteren is een chirurgische, of een andere geneeskundige behandeling nodig. Voor de chirurgische behandeling zijn diverse operaties beschreven. In dit hoofdstuk wordt een introductie gegeven van de afwijkingen in de spraak die kunnen optreden bij de patiënt met velopharyngeale insufficiëntie en van de operaties die gebruikt worden om tot een vermindering van de insufficientie en tot een verbetering van de spraak te komen.

Hoofdstuk 2. De oorspronkelijke wetenschappelijke publicatie van Sanvenero Rosselli over de craniaal gesteelde pharynxplastiek is uit het Italiaans in het Engels vertaald. De destijds gevoerde discussie over de pharynxplastiek en de alternatieven zoals de caudaal gesteelde pharynxplastiek of de pushback operatie worden in een historische context geplaatst. De craniaal gesteelde pharynxplastiek is populair geworden in de eerste helft van de twintigste eeuw maar is al eerder beschreven in 1886 door Schoenborn in Duitsland.

Hoofdstuk 3. De craniaal gesteelde lap uit de pharynxachterwand, met een Z-plastiek uit de mucosa van het velum wordt al vele tientallen jaren op de afdeling Mondziekten, Kaak-, en Aangezichtschirurgie van het UMCU gebruikt om velopharyngeale insufficiëntie te behandelen.

De medische gegevens van 130 patienten met velopharyngeale insufficiëntie, waarbij deze pharynxplastiek is verricht als een secundaire ingreep om de resonantie in de spraak te verminderen, zijn onderzocht. In het bijzonder is aandacht besteed aan de incidentie van heringrepen om een onbevredigend functioneel resultaat en/of ongewenste gevolgen van de genoemde operatie te behandelen. Mogelijke risicofactoren die tot heringrepen leiden zijn onderzocht. Als de steel van de flap te breed is, leidt dit tot hyponasaliteit en snurken. Als de steel van de flap zich oprolt tot een buis, is er onvoldoende afsluiting van de oropharynx van de nasopharynx gedurende het spreken, met als gevolg persisterende velopharyngeale insufficiëntie. Op maat gemaakte pharynxlappen, die een oplossing voor dit onbevredigende resultaat kunnen zijn, worden besproken, evenals de individuele vaardigheden van de operateur die meer dan zijn ervaring tot een beter functioneel resultaat van de operatie blijken te leiden.

Hoofdstuk 4. In dit hoofdstuk wordt de perceptieve beoordeling van de spraak van 15 patienten met velopharyngeale insufficientie, voor en na de pharynxplastiek onderzocht. Het perceptieve oordeel is een belangrijk klinisch instrument om velopharyngeale dysfunctie (en/of insufficiëntie) te beoordelen. Als zodanig is het perceptieve oordeel een essentieel onderdeel in de diagnostiek, samen met het lichamelijk onderzoek en de ziekte geschiedenis. Ondanks het klinische belang van de perceptieve beoordeling, heeft het oordeel beperkingen vooral als gevolg van de subjectiviteit. Een protocol wordt beschreven, waarin de verschillende aspecten zoals de resonantie, de ontsnappende neuslucht en de articulatie stoornissen aan de orde komen. Bovendien wordt er aandacht besteed aan de verstaanbaarheid en aan de stemkwaliteit. Alle genoemde parameters worden beoordeeld op een visueel analoge schaal. De inter-, en de intra beoordelaars betrouwbaarheid wordt bepaald, al dan niet voor een gepaarde beoordeling van de testzinnen. De invloed van ervaring van de beoordelaars met de beoordeling van spraak van schisispatienten en de invloed van verschillende types testzinnen zijn onderzocht. Het resultaat van het onderzoek toont de beperkingen van de perceptieve beoordeling aan: de beoordelaars, zelfs degenen met langdurige ervaring beoordelen de spraak verschillend. Het onderscheid in de beoordeling geeft aan dat de interne standaard van de beoordelaars varieert zowel in de tijd als tussen beoordelaars onderling. De conclusie is dat er behoefte is aan een objectieve meting die de perceptieve beoordeling van de spraak kan ondersteunen. Bij voorkeur is deze meting niet invasief en gebaseerd op de fysiologie en geschikt voor het gebruik in de dagelijkse praktijk.

In hoofdstuk 5 wordt een objectieve, niet-invasieve instrumentele meting besproken: de nasometrie. De essentie van de nasometrie is dat gedurende de spraak de geproduceerde acoustische nasale en orale energie (binnen een gegeven frequentie gebied: 500 Hz +/- 150 Hz) gescheiden wordt geregistreerd en gequantificeerd. De gemiddelde nasale acoustische energie gedeeld door de gemiddelde totale energie wordt gedefinieerd als “nasalance”. De nasalance geeft de gemiddelde nasale acoustische energie in de spraak weer, en wordt uitgedrukt als een percentage. Een hoge nasalance waarde kan men verwachten bij patiënten met velopharyngeale insufficiëntie, maar ook bij het gebruik van testzinnen met meer nasale consonanten dan voor de gebruikte taal berekend is. Lage nasalance scores komen voor bij patiënten met een afsluiting in de neus of in de nasopharynx. Maar ook bij het gebruik van testzinnen met minder of geen nasale consonanten. Nasometrie is dus een instrumentele meting die gebaseerd is op de fysiologie. Wel blijkt de uitkomst van de nasometrie afhankelijk te zijn van de standaardisatie van de aangeboden testzinnen en blijkt bovendien afhankelijk te zijn van de gebruikte taal.

Bij 43 patienten met velopharyngeale insufficientie (kinderen en volwassenen) is nasometrie verricht evenals een gedifferentieerde perceptieve beoordeling. De perceptieve beoordeling is verricht zowel door logopedisten met ervaring in de beoordeling van de spraak van schisis patiënten als door logopedisten die werkzaam zijn in de algemene praktijk. De uitkomst van de nasometrie en in het bijzonder de matige correlatie tussen nasometrie en de perceptieve beoordeling toont aan dat nasometrie andere eigenschappen meet dan die worden beoordeeld door de beoordelaars. Dit geldt in het bijzonder voor de spraak die niet heel erg afwijkend is. Dit toont aan dat de nasometrie niet kan worden beschouwd als een vervanging voor de perceptieve beoordeling

Hoofdstuk 6. Er is een aanzienlijke spreiding van nasalance scores voor sprekers die perceptief een normale spraak hebben. Deze spreiding kan worden verklaard door de individuele variatie en door het spreken van een dialect. In de literatuur wordt aangevoerd dat de perceptieve indruk van nasaliteit niet alleen door teveel nasale energie wordt veroorzaakt maar eerder door een gebrek aan oro-nasale balans. Er wordt bovendien gesteld dat iedere spreker een individueel bepaald bereik van nasaliteit heeft en dat dit bereik beter kan worden weergegeven in termen van maximum en minimum dan in gemiddelde nasalance scores. Daarom zijn twee samengestelde maten geïntroduceerd die een goed onderscheid kunnen maken tussen sprekers met en zonder nasale resonantie en waarvan de validiteit wordt bevestigd door de sensitiviteit en specificiteit. Dit wordt toegeschreven aan een afname van de intersubject variabiliteit in deze samengestelde maten vergeleken met de gemiddelde nasalance scores. De onderzoeksvraag is of normalisatie van de nasalance score van 43 patienten (beschreven in hoofdstuk 5) tot een betere correlatie leidt met de perceptuele beoordeling.

Hoofdstuk 7. In de algemene discussie worden de beschikbare chirurgische technieken om de velopharyngeale insufficiëntie en de hypernasaliteit te verminderen met elkaar vergeleken. Op basis van de literatuur kan er niet een voorkeur worden uitgesproken voor een bepaalde techniek.

Er worden een voorstel gedaan voor het gebruik van een multidimensioneel protocol voor de diagnostiek van patiënten met velopharyngeale insufficiëntie (zowel kinderen als volwassenen). Een belangrijke voorwaarde is dat het protocol geschikt is voor gebruik in de dagelijkse praktijk. Bovendien is het protocol idealiter geschikt voor wetenschappelijk onderzoek, in het bijzonder voor onderzoek naar de effectiviteit van de behandeling. Een nieuwe ontwikkeling vanuit de ICT, namelijk de automatische spraak herkenning wordt besproken, in het bijzonder vanuit het perspectief van de mogelijke klinische toepassing.

List of abbreviations.

ASR	Automatic Speech Recognition
CLP	Cleft Lip and Palate
CPS	Cleft Palate Speech
CSL	Computerized Speech Laboratory
DT	Denasal Text (meaning a text without nasal consonants)
ICT	Information and Communication Technology
NT	Normal Text (meaning a text with a normal distribution of phonemes)
OSAS	Obstructive Sleep Apnea Syndrome
SBPP-flap	Superiorly Based Posterior Pharyngeal wall flap
SHI	Speech Handicap Index
SLP	Speech Language Pathologist
SPSS	Statistical Package for the Social Sciences
VAS	Visual Analogue Scale
VPD	Velopharyngeal Dysfunction
VPI	Velopharyngeal Insufficiency

Appendix: reading stimuli

Oronasal text

Papa en Marloes staan op het station.

Ze wachten op de trein.

Eerst hebben ze een kaartje gekocht.

Er stond een hele lange rij, dus dat duurde wel even.

Nu wachten ze tot de trein eraan komt.

Het is al vijf over drie, dus het duurt nog vier minuten.

Er staan nog veel meer mensen te wachten.

Marloes kijkt naar links, in de verte ziet ze de trein al aankomen.

Oral text

Het is zaterdag.

Els heeft vrij.

Ze loopt door de stad.

Het is prachtig weer, de lucht is blauw.

Op straat ziet ze Bart op de fiets.

Hij wacht voor het rode licht.

Als Bart haar ziet, zwaait hij.

Els loopt weer verder.

Bij de bakker koopt ze brood, bij de slager koopt ze vlees.

Als het vijf uur is, gaat ze terug, zodat ze op tijd weer thuis is.

Curriculum Vitae

Kornelis Keuning was born on April 8^{th}, 1954 in The Hague.

In April 1981 he graduated as a dentist and in April 1987 he graduated as a physician from the Rijksuniversiteit Groningen. He worked as a dentist in general practice from April 1981 until October 1989.

He was trained in Oral and Maxillofacial Surgery (OMFS) from November 1989 till November 1993 at the University Medical Center Utrecht (Head of Department: dr. H.Mueller, Prof.dr. P.Egyedi, dr. R.Koole) and after that in Head and Neck Surgery and Oncology. He worked until the middle of 2011 as an OMF-Surgeon, with the focus on the area of Head and Neck Surgery/ Oncology and on the area of Cleft Lip and Palate.

At the moment he fulfills a part-time position as a locum tenens in OMFS.

Gaarne wil ik een ieder bedanken die heeft bijgedragen aan het verschijnen van dit proefschrift.

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