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Velopharyngeal Insufficiency and Cleft

Guest Editors: Travis T. Tollefson, David White, James Brookes, and Steven Goudy

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Editorial

Velopharyngeal Insufficiency and Cleft

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Management of orofacial clefts requires a comprehensive approach to address the multifaceted aspects of possible aesthetic and functional deficits. Many children with cleft palate with or without cleft lip continue to have speech abnormalities that can affect socialization, education, and childhood development. An interdisciplinary cleft team ideally manages most orofacial clefts and can include members from the fields of facial plastic surgery, otolaryngology, speech and language pathology, pediatrics, nursing, audiology, genetics, oral surgery, orthodontics, dentistry, and social work. These teams follow a relatively established timeline and algorithm for each of the discrete steps of orofacial cleft treatment. Cleft lip repair is often performed after 3–5 months of age. Palatoplasty is delayed after 8–14 months of age. Speech and language pathology assessment and therapy are targeted as the vocabulary develops (2–5 years old).

This special issue deals with the cleft surgeon and speech therapists role in treatment of velopharyngeal dysfunction (VPD), which is often related to velopharyngeal insufficiency (VPI). This lack of coordinated closure of the soft palate to the posterior pharyngeal wall is often a sequela of cleft palate, even after primary repair. One paper from this issue titled "*Nonlinear dynamic analysis of vowels in cleft palate children with or without hypernasality*" reports on cephalometric differences between the skull base and velopharyngeal anatomy in patients with unilateral cleft lip and palate compared to normal controls. These data contribute to the existing literature of abnormal skull base anatomy and inclination in children with velocardiofacial syndrome, which often associated with VPD.

The next paper titled "*Noncleft velopharyngeal insufficiency: etiology and need for surgical treatment*" analyzes those children with VPD without obvious orofacial clefting, and assesses the etiologies and potential treatment options. This allows for comparison to the traditional orofacial cleft-related VPD management.

The velopharyngeal workup is often a coordinated assessment by the cleft surgeon and speech pathologists. It can include subjective assessment, which is the subject of the next two papers. First, the next paper titled "*Nonlinear dynamic analysis of vowels in cleft palate children with or without hypernasality*" assesses the relationship between two speech assessment tools, Lyapunov exponents, and Nasalance scores in the presence of hypernasal speech. Furthermore, an additional paper titled "*Assessment of single-word production for children under three years of age: comparison of children with and without cleft plate*" presents a case-control testing of a single word assessment tool in children less than 3 years of age with cleft palate, which indentified speech differences in error patterns, consonants, articulation, and accuracy between children with and with clefts.

Objective assessment of VPD also can include either fluoroscopic speech examination or nasopharyngoscopy to characterize the pattern of VPD and target a surgical goal. The next paper titled "*Objective assessment of hypernasality in patients with cleft lip and palate with the nasalView system: a clinical validation study*" assesses the validity and reliability of a new tool for hypernasality testing called NasalView. Speech and language therapy is instituted to address typical compensatory speech patterns seen in children with cleft palate (e.g., glottal stops).

If hypernasal speech is not responsive to speech and language therapy, secondary speech surgery is warranted to address hypernasality in this time period. Surgical options

may include a superiorly based pharyngeal flap, dynamic sphincter pharyngoplasty, or posterior pharyngeal wall augmentation. Occasionally, a palate-lengthening procedure (Furlow double-opposing Z-plasty) is performed. These procedures can limit nasal air emissions, but potential obstructive sleep apnea from excessive nasal obstruction is a risk.

The risk of sleep apnea after surgically treating the velopharynx may be decreased if the tonsils and adenoids are addressed preemptively. Before pharyngeal flap surgery, some surgeons perform tonsillectomy to limit the potential variability of speech and airway changes that comes with fluctuating tonsillar hypertrophy, tonsillitis, or upper respiratory infections. The use of tonsillectomy in the face of VPI in children with cleft palate is somewhat controversial and is the topic of the last paper "*Speech outcomes after tonsillectomy in patients with known velopharyngeal insufficiency*".

The guest editors are pleased to present a collection of papers dealing with management of the speech-related difficulties in children with orofacial clefts. In this era of the expanding role of evidenced-based medicine, the effectiveness of treatments can only be monitored and improved by assessment with valid and consistent outcome measures, which are reviewed in this issue.

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Research Article

Assessment of Single-Word Production for Children under Three Years of Age: Comparison of Children with and without Cleft Palate

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Background. This study reports comparative phonological assessment results for children with cleft lip and/or palate (CLP) to typically developing peers using an evaluation tool for early phonological skills. *Methods*. Children without clefts (NC = noncleft) and 24 children with CLP, ages of 18–36 months, were evaluated using the *Profile of Early Expressive Phonological* Skills (PEEPSs) [1]. Children interacted with toy manipulatives to elicit a representative sample of target English consonants and syllable structures that are typically acquired by children between 18 and 27 months of age. *Results*. Results revealed significant differences between the two groups with regard to measures of consonant inventory, place of articulation, manner of production, accuracy, and error patterns. Syllable structure did not indicate differences, with the exception of initial consonant clusters. *Conclusions*. findings provide support for PEEPS as a viable option for single-word assessment of children with CLP prior to 3 years of age.

1. Introduction

Early intervention based on appropriate and thorough analyses of articulatory and phonological skills is essential; largely due to the role phonology plays in early linguistic development [1–9]. Evidence across a number of studies indicates that phonological abilities, in addition to lexical development, are the two distinguishing characteristics in identifying children as late talkers at an early age [2, 6–8, 10]. For example, Paul and Jennings [7] reported that toddlers who were identified as late talkers had a smaller consonant repertoire and a lower percentage of consonant accuracy compared to typically developing age peers. Based on their findings, Paul and Jennings recommended that the overall number of different consonants in a young child's inventory be used as a sensitive indicator of development and delay and that syllable complexity serves as an effective means for monitoring phonological development. In an investigation of late talker development, Williams and Elbert [9] examined a number of phonetic and phonological measures, including phonetic inventory, syllable structure, syllable diversity, sound variability, percentage of consonants correct, and error patterns. They reported that both quantitative and qualitative phonetic and phonological measures were important in determining recovery from late talking or persistence of a phonological delay.

In addition to phonology's role in early identification, a number of phonetic and phonologic measures have been reported to predict future performance with regard to persistence versus recovery of delayed speech and language development in young children. Carson et al. [2] summarized evidence across several studies that indicate that typically developing children 24–31 months of age had significantly larger and more diverse phonetic inventories for vowels and

consonants across all word positions compared to children with SLI-E [10], produced significantly more complex syllables, and were more accurate in their productions than late-talking toddlers [7]. Similarly, Thal et al. [11] reported that children identified as late talkers made greater gains in lexical development if they had larger phonetic inventories and 10 or more words in their vocabularies than children who had smaller phonetic inventories and an expressive lexicon with less than 10 words. Paul and Jennings [7] concluded that global measures of phonological ability, such as syllable structure complexity, phonetic inventory size, and percentage of consonants, can be used as prognostic indicators of development for children identified as late talkers. In a longitudinal study of recovery or persistence of delay, Carson et al. [2] examined 20 different phonological measures, including the number of different consonants (overall and in initial and final positions), number of different consonant clusters (in initial and final positions), and percentage of syllable structures with final consonants. Similar to the quantitative findings reported by Williams and Elbert [9], they found that the greater the delay in phonological development at two years, the more at risk the child was for continuing delays at three years.

Despite the role of phonology in the early identification and prediction of language delay, assessment tools of early phonological skills have not been available [12]. The importance of early phonological assessment is especially important for children with cleft lip and/or palate (CLP) who are at risk for early speech and language delays due to the presence of the cleft during the first year of life [13–18]. For some children, this delay persists through the preschool period and may impact early literacy skills [19–24]. It is thought that early structural deficits restrict the speech sound inventories of young children with CLP, which in turn limits vocabulary growth. It has been established that young children with CLP produce more words beginning with nasals, vowels, glides, and fewer words beginning with oral stop consonants than children without clefts [25]. It is also apparent that they show an early preference for sounds made at the labial, velar, and glottal place of articulation [25].

The recent focus on early intervention for children with CLP attempts to change the trajectory of early speech and language development; however, limitations in speech assessment materials for children under three years restrict the ease and accuracy of speech sound assessment that, in turn, drives goal selection for treatment [18]. Assessment of phonological development for children with CLP, as well as all young children, has been based primarily on parent report (e.g., McArthur Communication Development Inventories) [26] or a brief subtest within a larger standardized test (e.g., *Sequenced Inventory of Communicative Development*; [27]). These measures, however, are extremely limited and cannot provide a representative or even sufficient evaluation of the key phonetic and phonemic characteristics which are required for an age-appropriate clinical assessment. The Parameters of Care from the American Cleft Palate-Craniofacial Association [28] recommend a speech-language evaluation twice in the first two years of life and again at three years of age. A single-word measure of speech production specifically designed to assess the major components of phonological development in young children would be helpful in identifying children who would benefit from early intervention.

Assessment of speech sound development in children less than three years of age is challenging due to the inherent variability in early language and phonological development [29, 30]. This variability is due in part to the strategies young children use to select words from their vocabulary and to the absence of phonological rules that govern the more stable sound system observed in older children [1]. We know that the phonological characteristics of the words play an important role in determining which words a child includes in their early vocabularies for typically developing children, as well as children with cleft lip and palate [25, 31–35]. Children choose words with phonological characteristics that are consistent with their developing sound systems and avoid words with characteristics that are outside their sound system. A second feature of early word learning is that words are produced as an unanalyzed whole. This learning strategy results in significant variability in children's production of speech sounds and poor association between the child and adult model. It is important, therefore, to assess sufficient exemplars with representative phonetic characteristics to check for consistency and variability and to sample words with sounds that do not occur in routine conversation in addition to sounds that the child uses frequently [36, 37].

The Profiles of Early Expressive Phonological Skills (PEEPSs; [1]) was developed to assess consonant inventory, place of articulation, manner of production, syllable structure, accuracy, and error patterns of children between 18–36 months of age. PEEPS was constructed to represent the diversity of place, manner, and voicing of English consonant production, as well as different syllable structures. Finally, multiple exemplars of the different phonetic characteristics were included across a number of words in order to check consistency and variability of children's early phonological skills. The purpose of this study was to compare the phonologic development of a group of children with CLP between 18–36 months to a group of children without clefts (NC) using the PEEPS assessment tool.

2. Method

2.1. Participants. This study was approved by the East Tennessee State University (ETSU) and Vanderbilt University Institutional Review Boards and the study was conducted with the understanding and consent of the parents of the participants. Forty-two children without cleft lip and/or palate and 26 children with cleft lip and/or palate between the ages of 18 and 36 months of age participated in this study. Mean age of the NC group was 25.5 months. (SD = 7.4 months) and mean age of the children with CLP was 27.4 months (SD = 6.5 months). The NC group had 16 males and 26 females and the children with CLP had 16 males and 10 females. No significant gender differences were identified for the phonological components analyzed. The NC children were recruited from the university childcare

centers at ETSU. They had no cognitive, speech-language, or hearing impairments, as reported by their parents. The children with cleft lip and palate were part of an ongoing longitudinal study of early intervention at two sites: ETSU in Johnson City, TN, and Vanderbilt University in Nashville, TN. The children with CLP had not received any intervention at the time of this study. All of the children with CLP met the following inclusion criteria: (1) palate repair by 12 months of age; (2) absence of dysmorphology associated with a genetic syndrome according to a geneticist, cognitive delay, or sensorineural hearing loss; (3) monolingual English speaking family. The children with CLP had PE tubes placed bilaterally between the time of lip and/or palate repair. All the children passed a hearing screening at the time of evaluation although 75% of the children had reports of 3–5 middle ear infections since birth. Five children had bilateral CLP, 16 had unilateral CLP, and 5 had cleft palate. The children with CLP had a mean receptive and expressive language standard score of 103 (SD = 7) and 89 (SD = 10), respectively, on the Preschool Language Scale-4 [38].

2.2. Assessment Procedures. Each child was assessed individually using the PEEPS [1] either in a clinic setting or in their childcare center by a speech-language pathologist or graduate student in speech-language pathology trained in its administration. The testing took approximately 15–20 minutes and was recorded with a digital video recorder. The assessment was transcribed by a speech-language pathologist or graduate student in speech-language pathology trained in phonetic transcription.

PEEPS consists of a total of 60 words divided into two sections: a Basic Word List of 40 words for younger children (18–24 months) and an Expanded Word List of an additional 20 words for older children (24–36 months). The words were selected on the basis of two criteria: (1) age of acquisition (AOA) based on vocabulary words from the MacArthur Communicative Development Inventories [26]; (2) phonetic characteristics to elicit target English consonants across all places, voice, and manner categories of production, as well as in different syllable structures. Specifically, words were selected that are typically acquired by children between 18 and 27 months of age, as determined by Dale and Fenson [39]. The AOA for the total test words was 20.5 months (range 18–27 months) with the AOA for the Basic Word List at 19.4 months (range 18–21 months) and 22.7 months for the Expanded Word List (range 21–27 months). Thus, all but two of the total test words (96.7%) were acquired by 24 months of age. Additionally, words were selected to represent the diversity of place, manner, and voicing of English consonant production, as well as syllable structure. The Basic Word List included words to elicit production of target consonants in all seven places of English consonant articulation, all five manner classes, voiced and voiceless consonants, and simple syllable structures (CV, CVC, CVCV), along with some multisyllabic words ("peek-a-boo" and "belly button") and a word-final nasal cluster (e.g., "hand"). The Expanded Word List builds on the Basic Word List with additional words that sample more polysyllabic words (e.g., "dinosaur"), complex syllable structures that elicit clusters in all three word positions (e.g., "truck," "monkey," and "drink"), and includes additional consonants in word positions that were not examined in the Basic Word List. Finally, multiple exemplars of the different phonetic characteristics were included across a number of words in order to check consistency and variability of children's early phonological skills.

The children were seated at a table or on the floor. A wireless microphone was worn in an adapted shirt and a video camera was placed on a tripod with full-face view of the child. An assistant was available to reposition the camera to maintain full-face view. A brief 5-minute warm-up activity with novel toys was available if the children did not immediately engage with toy manipulatives. The words were elicited in any order through interaction with toy manipulatives. The manipulatives were placed in a soft sack that was used to playfully engage the child in pulling the items out of the bag and naming each one as it was pulled out. Only a few items were placed in the sack at a time and the child was asked to pull out one toy at a time. A hierarchy of elicitation strategies was employed to facilitate naming of the target words. At the first level, *spontaneous elicitation* was attempted by simply asking "What is that?" If the child did not label the toy, the child was prompted by the clinician and then asked them again "What is this?" If the child still did not spontaneously label the item, then *sentence completion* was used; for example, "The baby is sleepy. Put the baby in the…(bed)." The third level of elicitation involved *modeling* the word by saying "It's a fish!" Look at this pretty fish. Would you like this? What do you want?" If the child gave a different label for a target word, then the clinician recast it with a model and then asked a question to elicit the word. For example, "That's right, you can call this a "baby," but I sometimes call it a "doll." What do I call it?" If the child still did not label the item after the model, then the toy was put back in the sack and the child had the opportunity to label the toy later. Following the assessment, the children's responses were phonetically transcribed using the International Phonetic Alphabet [40].

2.3. Measures of Phonological Development. Phonologic components assessed included phonetic inventory (word initial, medial, and final), including segmental characteristics of *place* (labial, alveolar, and velar), *voice*, and *manner* of consonant production, syllable structure (production of two and three syllable words, initial consonant clusters); accuracy (Percentage of Consonants Correct: PCC; [41, 42]); error patterns (presence of common phonological processes of substitution or deletion; compensatory error use for children with CLP); nasal emission (percent of words with nasal emission, for CLP children).

2.4. Procedural Fidelity and Reliability. The procedural fidelity of the PEEPS administration was assessed through coding of 50% of the assessment session for each child. Videotapes of the testing sessions were reviewed and coded by a graduate student in speech-language pathology not

Table 1: Percent agreement for procedural components of the PEEPS assessments for the children with CLP and NC children.

Procedure	CLP	NC
Correctly presents stimulus	98%	94%
Follows correct prompt sequence	96%	94%
Responds to child's questions/requests	92%	93%
Praises for engagement rather than correct response	91%	92%

associated with the assessments of the children. Table 1 displays the average percent agreement for the children with CLP and without clefts for the five major components of the administration. All components for both groups achieved a reliability score of ≥91% agreement.

Transcription reliability was performed on 25% or more of each child's assessment by a speech-language pathologist or graduate student familiar with the phonetic transcription of young children with phonological disorders. In order to be considered an agreement, the consonant needed to be transcribed identically for place, manner, and voicing. Interjudge reliability ranged from 78% to 94% (mean 85%) for children with CLP and 90%–96% (mean 93%) for the children without clefts. A weighted κ was calculated to examine transcription agreement based on the frequency of occurrence of different manner classes. A κ of .76 for the children with CLP and .85 for the noncleft children was obtained indicating excellent agreement for both groups [43]. However, since many of the disagreements that did occur involved compensatory articulation errors, these disagreements were examined further. Forty-five percent (32/72) of the disagreements included compensatory errors of glottal stops (i.e., /k in duck), pharyngeal fricatives (i.e., /in shoe), and posterior nasal fricatives (i.e., /s in sock). All words with disagreements were retranscribed by two senior investigators to reach consensus.

3. Results and Discussion

3.1. Phonetic Inventory. Table 2 shows the mean, standard deviation, t-test, probability, and effect size for three measures describing the children's phonetic inventory. The total number of consonants correct on the PEEPS showed significantly fewer correct consonants for the children with CLP when compared to the NC children ($t = 9.88$, $P \leq 0.0001$, $d = 2.91$). When initial and medial/final consonants were examined separately, both variables showed significantly fewer consonants than the NC group ($t = 8.66$, $P \leq 0.0001$, $d = 2.65$; $t = 11.37$, $P \leq 0.0001$, $d = 3.47$). Effect sizes (Cohen's d [44]) were calculated using G*Power (version 3.0.10; http://www.psycho.uni-duesseldorf.de/aap/projects/gpower). Values ranged from −4.72 to 4.72 for the statistically significant comparisons, indicating large effect sizes (see Table 2).

The growth in acquisition of initial and medial/final consonants is shown in Figures 1 and 2. Figure 1 shows the number of correct initial consonants used on the PEEPS for children with CLP (circles) and NC children (squares) across the ages of 18 to 36 months. The linear (CLP: dotted line) and curvilinear (NC: solid line) polynomials and means (filled symbols) are displayed for the groups. For the NC children, there is substantial growth in consonant production between 18 and 24 months of age. This corresponds with the rapid growth that typically occurs as children move from the unanalyzed whole-word stage to a rule-governed stage of phonological acquisition. The children with CLP showed a gradual improvement in consonant production over time, as indicated by the slope of the linear regression. Figure 2 illustrates the number of correct medial/final consonants on the PEEPS for children with CLP (circles) and NC children (squares) across the 18 to 36 month ages. The children with CLP produced fewer medial/final consonants correct than initial consonants (shown in Figure 1) whereas the NC children produced similar numbers of medial/final and initial consonants correct. The pattern of variability in early sound production for NC children at 18–24 months of age appears for medial/final consonants whereas the children with CLP show gradual increase in correct sound production through 36 months of age.

The presence or absence of three places of articulation features (labial, alveolar, velar) was examined for both groups. The children with CLP showed the presence of fewer place contrasts than the NC children. Particularly, the children with CLP produced fewer alveolar-velar contrasts ($\chi^2 = 11.28$, $P = 0.001$), labial-velar contrasts ($\chi^2 = 15.9$, $P = 0.001$), and presence of all three contrasts ($\chi^2 = 18.8$, $P = 0.001$) than did the children with NC. The groups did not differ in the presence of labial-alveolar contrasts ($\chi^2 = 3.42$, $P = 0.064$).

3.2. Syllable Structure. The production of consonants correct in two and three syllable words on the PEEPS was examined for both groups and showed no significant differences between the groups ($\chi^2 = 2.25$, $P = 0.13$; $\chi^2 = 0.05$, $P = 0.81$). The consonants correct within initial consonant clusters, however, showed significantly fewer correct productions for the children with CLP than the NC children ($t = 4.58$, $P \leq 0.0001$, $d = 1.38$). Final consonant clusters could not be compared because so few children in either group produced them.

3.3. Accuracy. The Percent Consonants Correct (PCC; [41]) was calculated for the total PEEPS sample and separately by manner class (see Table 2). The mean total PCC for the children with CLP was 34.9% compared to the NC children with 86.5% ($t = 9.83$, $P \leq 0.0001$, $d = 2.89$). When the manner class PCCs were compared, nasals, stops, fricatives, affricates, and liquids showed significantly poorer scores for the children with CLP than the NC children. The glides did not show a difference between the groups. The effect sizes for all comparisons were high, ranging from a d of 1.67 to 4.72.

Figure 3 summarizes PCC for the total PEEPS sample for children with CLP (circles) and NC children (squares) from 18 to 36 months of age. The growth in PCC was greatest between 18 and 24 months of age for the NC children.

Table 2: Mean and standard deviation for PEEPS measures for children with and without cleft palate and statistical comparisons.

Measure	NC		Cleft		t-test	P	d*
	Mean	SD*	Mean	SD*			
Phonetic Inventory							
No. Consonants	65.9	11.5	25.8	15.7	9.88	≤0.001	2.91
No. Initial Cons	14.6	2.5	6.9	3.3	8.66	≤0.001	2.65
No. Medial/Final Cons	14.0	2.7	4.0	3.0	11.37	≤0.001	3.47
Syllable Structure							
Initial Clusters	1.3	0.7	0.4	0.7	4.58	≤0.001	1.38
Accuracy							
PCC-Total	86.5	15.1	34.9	20.2	9.83	≤0.001	2.89
PCC-Nasals	97.9	5.2	66.9	25.7	5.05	≤0.001	1.67
PCC-Glides	78.1	42.0	69.2	48.0	0.58	0.567	0.19
PCC-Stops	96.0	6.0	47.3	25.2	8.05	≤0.001	2.65
PCC-Fricative	94.5	8.0	29.6	11.8	14.76	≤0.001	4.72
PCC-Affricates	79.7	33.3	11.8	28.1	7.54	≤0.001	2.20
PCC-Liquids	84.3	16.1	24.1	18.8	11.44	≤0.001	3.43
Error Analysis							
% Word Errors	10.7	9.8	76.3	17.0	−15.69	≤0.001	−4.72
% Substitutions	3.5	3.7	23.9	13.9	−6.44	≤0.001	−2.00
% Omissions	2.3	3.5	16.6	13.9	−4.54	≤0.001	−1.42

*Note. SD: Standard deviation, d: Cohen's.

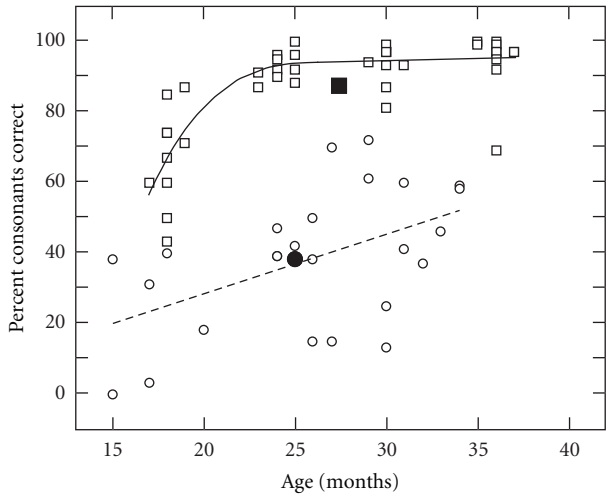

Figure 1: The Percentage of Consonants Correct (PCC) used on the PEEPS for children with CLP (circles) and NC children (squares) across the ages of 18 to 36 months. The linear (CLP: dotted line) and curvilinear (NC: solid line) polynomials and means (filled symbols) are displayed for the groups.

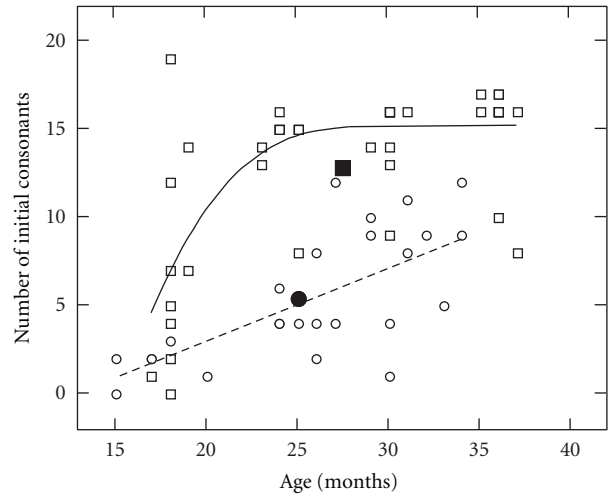

Figure 2: Number of initial consonants in the phonetic inventories of the children with and without CLP across age determined from the PEEPS. The number of correct initial consonants used on the PEEPS for children with CLP (circles) and NC children (squares) across the ages of 18 to 36 months. The linear (CLP: dotted line) and curvilinear (NC: solid line) polynomials and means (filled symbols) are displayed for the groups.

In contrast, the children with CLP showed slow, gradual acquisition of consonant production over time.

3.4. Error Analysis. The percent of words with sound errors on the PEEPS indicated a large difference between the groups, with the children with CLP significantly higher (Mean = 76.3%, SD = 17) than the NC children (Mean = 10.7%, SD 9.8; $t = -15.69$, $P \leq 0.0001$, $d = -4.72$). An analysis of the error types indicated that children with CLP used more substitutions and omissions than did the NC children ($t = -6.44$, $P \leq 0.0001$, $d = -2.0$; $t = -4.54$, $P \leq 0.0001$, $d = -1.42$).

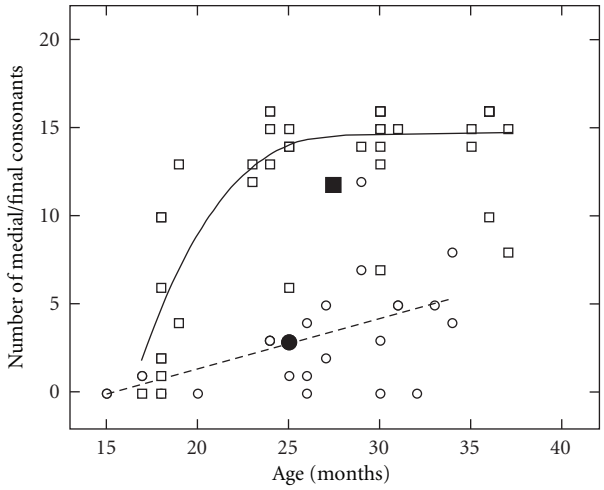

Figure 3: Number of medial and final consonants in the phonetic inventory of children with CLP (circles) and NC children (squares) across the ages of 18 to 36 months. The linear (CLP: dotted line) and curvilinear (NC: solid line) polynomials and means (filled symbols) are displayed for the groups.

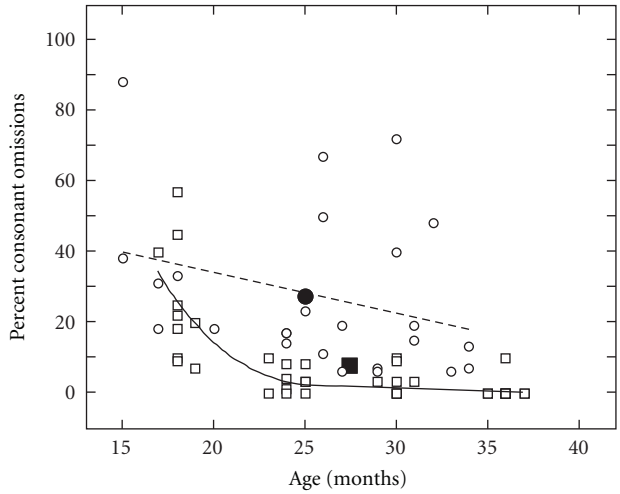

Figure 4: Percent of consonant omissions determined from the PEEPS for the children with CLP (circles) and NC children (squares) across the ages of 18 to 36 months. The linear (CLP: dotted line) and curvilinear (NC: solid line) polynomials and means (filled symbols) are displayed for the groups.

Figures 4 and 5 represent the percent of consonant omissions and substitutions on the PEEPS for children with CLP (circles) and NC children (squares) across ages from 15 to 37 months of age. The NC children showed a rapid decrease in their consonant omission and substitution patterns from 18 to 24 months of age whereas the children with CLP demonstrated a gradual reduction in omission and substitution patterns across the 18–36 month range.

3.5. Compensatory Errors. The children with CLP used a mean percent of compensatory errors on the PEEPS of 7.0, SD 6.8; the NC group used none. Fifteen of the 26 children with CLP used compensatory errors at least twice during the sample. Thirteen of the 15 children had fewer than 10 (3–13% of the words in their sample) compensatory errors in the sample. The remaining two children had 13 and 14 errors, respectively (18% of the words in their sample). Glottal stops were the predominant compensatory error used, with pharyngeal fricative the second most used error pattern. Posterior nasal fricatives were used 1-2 times by three children.

3.6. Nasal Emission. Nasal emission was coded on consonant production of the PEEPS. Three of the 26 children with CLP used nasal emission on more than 2 occurrences. The three children used nasal emission on an average of eight words on the PEEPS. Additionally, these three children also used 1-2 occurrences of posterior nasal fricative substitutions. No direct measurements of velopharyngeal function were conducted on these young children.

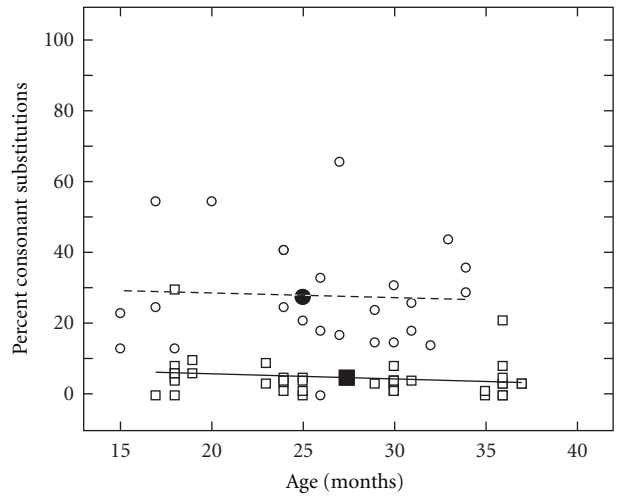

Figure 5: Percent of consonant substitutions determined from the PEEPS for the children with CLP (circles) and NC children (squares) across the ages of 18 to 36 months. The linear (CLP: dotted line) and curvilinear (NC: solid line) polynomials and means (filled symbols) are displayed for the groups.

4. Conclusions and Discussion

The PEEPS protocol provided a systematic and developmentally appropriate means of assessing differences between a group of children with and without early speech delays. PEEPS provided a profile of early phonological development that captured consonant inventory, syllable structure, accuracy, and error analysis from 18 to 36 months of age. From the results from this study, PEEPS revealed significant differences between typical phonological development and delays across all four major parameters in comparison to the children with CLP. Additionally, the rate of growth revealed that children with CLP were slower than the NC children in all phonological measures. Finally, qualitative differences in addition to quantitative differences were noted with regard to compensatory errors and nasal emission.

In conclusion, the following observations can be summarized from these results.

- The typical profiles at 18 months reflect what is known about normal phonological acquisition with regard to individual variability and a whole-word strategy.
- The shift to a rule-based strategy at 24 months for typically developing toddlers was observed with variability across children being largely extinguished at that age.
- The CLP children mirrored normal development at a slower pace; in general, reflecting the 18-month NC children at 30 months, which was a full 12 months later.

4.1. Clinical Implications. This first examination of PEEPS [1] suggests that it provides an instrument to assess reliably the phonetic and phonological development of typical and atypical children between the ages of 18 and 36 months. Additionally, the data from the children with CLP could be used to design appropriate intervention strategies. For example, if the phonological profile of a child with CLP reflects a whole-word stage of lexical and phonological acquisition, implementation of a broad-based intervention strategy directed to an emerging sound system might be indicated. This could include a parent-implemented naturalistic language approach rather than a more structured, rule-based phonological approach. PEEPS, therefore, provides a thorough and developmentally appropriate method for assessing the phonological skills of toddlers that can allow for early intervention for phonological disorders.

4.2. Future Research Directions. These results represent a preliminary examination of PEEPS and additional studies with larger samples of typically and atypically developing children are needed. In addition, future studies are needed that compare PEEPS to a reference standard, such as a language sample, to evaluate test specificity and sensitivity.

Acknowledgment

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Clinical Study

Noncleft Velopharyngeal Insufficiency: Etiology and Need For Surgical Treatment

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Objective. Velopharyngeal insufficiency (VPI) occurs frequently in cleft palate patients. VPI also occurs in patients without cleft palate, but little is known about this patient population and this presents a diagnostic dilemma. Our goal is to review the etiology of noncleft VPI and the surgical treatment involved. *Design/Patients*. A retrospective review of VPI patients from 1990 to 2005. Demographic, genetic, speech, and surgical data were collected. We compared the need for surgery and outcomes data between noncleft and cleft VPI patients using a Student's *t*-test. *Results*. We identified 43 patients with noncleft VPI, of which 24 were females and 19 were males. The average age at presentation of noncleft VPI was 9.6 years (range 4.5–21). The average patient age at the time of study was 13.4 years. The etiology of VPI in these noncleft patients was neurologic dysfunction 44%, syndrome-associated 35%, postadenotonsillectomy 7%, and multiple causes 14%. The need for surgical intervention in the noncleft VPI group was 37% (15/43) compared to the cleft palate controls, which was 27% (12/43). There was not a statistical difference between these two groups ($P > 0.5$). *Conclusion*. Noncleft VPI often occurs in patients who have underlying neurologic disorders or have syndromes. The rate of speech surgery to address VPI is similar to that of cleft palate patients. We propose that newly diagnosed noncleft VPI patients should undergo a thorough neurologic and genetic evaluation prior to surgery.

1. Introduction

The velopharyngeal complex closes against the posterior pharyngeal wall, to separate the oral cavity from the nasal cavity. Inability of the velopharyngeal complex to close leads to nasal air emissions and hypernasal speech. The occurrence of velopharyngeal insufficiency (VPI) in the absence of cleft palate formation has been reported many times in the literature. VPI is a recognized complication of adenoidectomy [1–8]. Moreover, patients with velocardiofacial syndrome (VCFS) also have VPI without cleft palate [5, 9, 10].

Individuals with suspected VPI are best evaluated by a team of professionals specializing in VPI and speech characteristics associated with cleft palate. Initially, each of these patients is evaluated by a speech pathologist to assess the patient's speech patterns, nasality, and articulation disorders [11]. From there, they will undergo evaluation of their velopharyngeal port closure using multiview fluoroscopy and/or video nasoendoscopy [12, 13].

Once the diagnosis of VPI has been made a trial of speech therapy is often used to improve the intelligibility of speech. If speech remains unintelligible and/or VPI is a significant component of the speech pattern, surgical intervention is often needed. The type of surgery required is often based on the video nasoendoscopy [14]. The outcomes after speech surgery may vary based on the type of surgery performed and underlying medical diagnosis, especially in patients with VCFS [15, 16]. We hypothesize that patients with noncleft VPI require similar surgical management and have similar speech outcomes compared to cleft palate patients.

2. Materials and Methods

After obtaining IRB approval, a retrospective review of patients charts diagnosed with VPI who did and did not have a cleft palate was performed. Patients with VPI treated at the University of Iowa Hospitals and Clinics from 1990

to 2005 were reviewed. Demographic, genetic, speech, and surgical data were collected. For each noncleft VPI patient we identified an age-and-sex matched control patient with a cleft palate. We compared the outcomes data using a Student's t-test. Briefly, each patient was evaluated according to the protocol by Dailey et al. for hyper and hyponasality (1 = normal, 2 = mild, 3 = mild-moderate, 4 = moderate, 5 = moderate-severe, 6 = severe) and underwent video nasal endoscopy [17]. Based on these findings, each patient was assessed to have a competent, marginally competent, or incompetent velopharyngeal closure. Patients judged to have marginally competent or incompetent velopharyngeal closure underwent surgical management of their VPI based on their port closure characteristics.

3. Results

We identified 43 patients who had noncleft VPI. There were 24 females and 19 males found. The average age at the diagnosis of noncleft VPI was 9.6 years (range 4.5–21). The average age of the patient in the study was 13.4 years. The etiology of VPI in these noncleft patients was postadenotonsillectomy 7%, neurologic dysfunction 44%, syndrome-associated 35%, and unknown causes 14%.

The neurologic dysfunction group comprised the largest portion of the noncleft VPI group (44%). There were a wide spectrum of disorders included in this group detailed in Table 1, which includes posttraumatic brain injury, developmental delay, and cerebral palsy.

The occurrence of a syndrome in the noncleft patients with VPI was 35% and is shown in Table 2, which includes VCFS, Turner syndrome, and VATER syndrome. Of note, a new diagnosis of VCFS was made in 6 of our patients. The remaining group of patients did not have any definable risk factors for VPI.

The need for surgical intervention in the noncleft VPI group was 37% (15/43) compared to the cleft palate controls, which was 27% (12/43). In noncleft VPI patients, 10 pharyngeal flaps, 4 double-opposing Z plasties, and 1 sphincter pharyngoplasty were performed. In the cleft palate control group 11 pharyngeal flaps and 1 double-opposing Z plasty were performed. There was not a statistical difference in the number of VPI surgeries between these two groups ($P > 0.5$).

Table 1: Neurologic causes of noncleft VPI.

Posttraumatic brain injury	$N = 2$
Developmental delay	$N = 2$
Cerebral palsy	$N = 1$
Myasthenia gravis	$N = 1$
Associated speech delay	$N = 3$
Undiagnosed neurologic condition	$N = 10$

Table 2: Syndromic causes of noncleft VPI.

Velocardiofacial syndrome	$N = 7$
Klippel-Feil syndrome	$N = 1$
Epidermal-Nevus syndrome	$N = 1$
Alagille syndrome	$N = 1$
Turner syndrome	$N = 1$
VATER syndrome	$N = 1$
Unrecognized syndrome	$N = 1$

4. Discussion

It is clear that noncleft VPI occurs in an assortment of anatomic, neurologic, postsurgical, and syndromic patients. A standard speech assessment is critical for making the diagnosis. The noncleft VPI patients were identified at a much later age, 9.6 years, than children with cleft palates. This is likely due to the speech screening that occurs in most cleft palate teams that identifies VPI earlier in children with cleft palate.

Surgical intervention was required in 37% of noncleft VPI patients which is lower than what is reported in other studies [2, 3, 6]. The rate of VPI surgery did not statistically differ between noncleft and cleft palate patients. This suggests that noncleft VPI can be treated similar to cleft palate patients with initial speech therapy to improve articulation. However unresolved VPI required surgical intervention in more than a third of patients.

It is important to note that an etiology of the VPI could be specifically identified in 86% of patients. Many of the patients had underlying neurological diagnoses or syndromes suggesting that altered neuromuscular control is associated with noncleft VPI. Newly diagnosed VCFS was identified in 16% of our patients, the importance of genetic evaluation has been highlighted by other authors [5].

5. Conclusion

Noncleft VPI occurs in a diverse group of patients. The majority of these patients will not require surgical intervention. Each patient with noncleft VPI should be carefully screened for the etiology of their VPI because only 14% will have an unknown cause. The need for VPI surgery in noncleft patients is not different than cleft palate patients.

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Research Article

Nasopharyngeal Development in Patients with Cleft Lip and Palate: A Retrospective Case-Control Study

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Introduction. The aim of this paper was to evaluate cephalometrically the nasopharyngeal development of patients with complete unilateral cleft lip and palate. Influencing factors were evaluated and cleft to noncleft subjects were compared to each other. *Material and Methods*. The lateral cephalograms of 66 patients with complete cleft lip and palate were measured and compared retrospectively to the cephalograms of 123 healthy probands. Measurements were derived from a standardized analysis of 56 landmarks. *Results*. We observed significant differences between cleft and control group: the cleft patients showed amaxillary retroposition and a reduced maxillary length; the inclination of the maxilla was significantly more posterior and cranial; the anterior nasopharyngeal height was reduced; the nasopharyngeal growth followed a vertical tendency with reduced sagittal dimensions concerning hard and soft tissue. The velum length was reduced. In the cleft group, an accumulation of mandibular retrognathia and an anterior position of the hyoid were observed. Skeletal configuration and type of growth were predominantly vertical. *Conclusions*. Our data provides a fundamental radiological analysis of the nasopharyngeal development in cleft patients. It confirms the lateral cephalogram as a basic diagnostic device in the analysis of nasopharyngeal and skeletal growth in cleft patients.

1. Introduction

The nasopharyngeal area, with its complex interactions of bony fundament, muscular functionality, and soft tissues, influences not only the aesthetic facial harmony but also provides the anatomical basis of speech and hearing.

In patients with (unilateral) cleft lip and palate, the restoring of the velo- and nasopharyngeal function represents the crucial surgical step to a solid rehabilitation as far a s phonation, articulation, and speech development are concerned.

Many morphometric studies have been performed to analyse the cephalometric characteristics of cleft patients; the causality of the altered configuration of the bony facial structures is discussed controversially.

Chatzistavrou et al. debate the influence of an inherited growth deficiency [1]. The impaired functionality of the cleaved oropharyngeal structures and their lacking potency as growth centres represents another explanation of the altered skeletal development [2–5]. Finally an iatrogenic impact on the development of the facial structures, their position, and function, after complex surgery during the first months of extra uterine development, is described [6–10].

The aim of the present investigation was to analyse the morphologic development of hard and soft tissues in the nasopharyngeal region as a pivotal anatomical region in aspects of functional rehabilitation, that is, speech, breathing, and hearing (aeration of the middle ear cavity via the tympanic tube) in a collective of cleft lip and palate patients after consistent surgical therapy in comparison to a healthy collective.

The analysis of the configuration of the skull base, characteristic growth pattern, and decisive skeletal maxillary and mandibular parameters was of special interest in this retrospective cephalometric study. This static approach does not include the correlation to functional impairment but provides an anatomical basis.

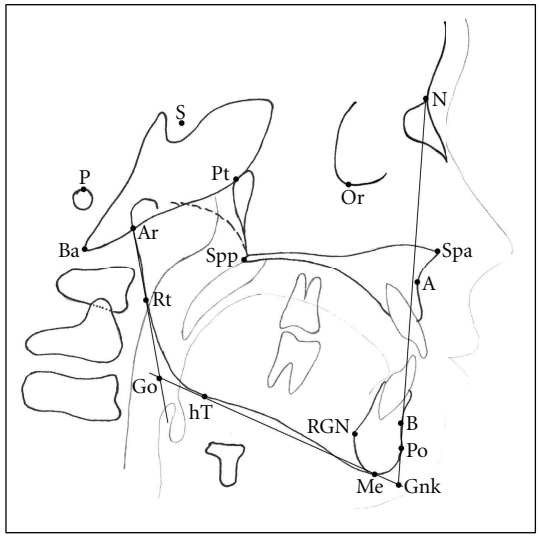


Figure 1: Cephalometric landmarks of the facial skeleton and skull base. (N: nasion, Or: orbitale, S: sella, P: porion, Ba: basion, Ar: articulare, Pt: pterygoid, Spp: posterior nasal spine, Spa: anterior nasal spine, A: subnasal/A-point, B: submentale/B-point, Po: pogonion, Gnk: gnathion, Me: menton, RGN: retrognathia, hT: horizontal mandibular tangential point, Rt: vertical ramus tangential point, Go: gonion).

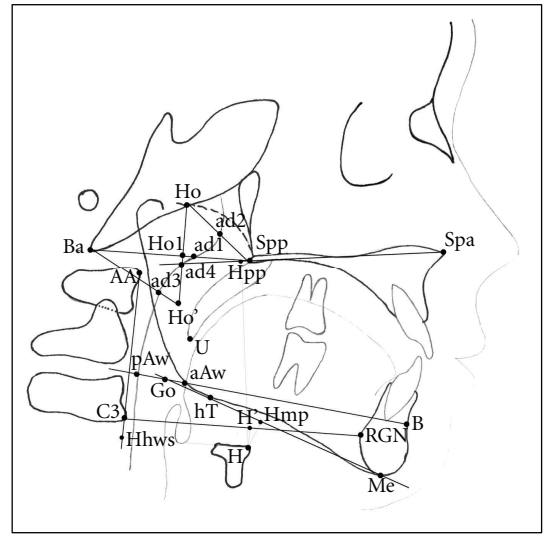


Figure 2: Cephalometric landmarks of the naso- and oropharyngeal area. (Spa: anterior nasal spine, Spp: posterior nasal spine, Ho: hormion, Ba: basion, AA: anterior arcus atlantis (first cervical spine), C3: anterior third cercival spine (C3), H: hyoid, Me: menton, RGN: retrognathia, B: submentale/B-point, aAw: anterior airway, pAw: posterior airway, ad1–4: adenoids 1–4).

This small investigation aims to answer the following questions.

Are there significant differences concerning the configuration of the skull base between cleft and control group?

What are the most meaningful measured values in the analysis of the lateral cephalogram, differentiating cleft and control group?

What are the significant differences in the position of the velopharyngeal position?

Is there an altered position of the hyoid in the cleft group?

2. Material and Methods

2.1. Subjects. In total, we analysed the cephalometric X-rays of 66 patients with complete unilateral cleft lip, alveolus, and palate who had undergone the same operations by the same team (uCLP group). In all patients, primary closure of the lip was performed according to Millards technique [11] at the age of 6 to 8 months, and one-step closure of hard and soft palate was done according to Campbell [12] and Widmaier [13] at the age of 12 to 16 months. Age below 6 years or already performed surgery influencing the anatomy and configuration of naso-, oro-, or velopharynx, for example, velopharyngoplasty or adenoidectomy, leads to exclusion from further analysis.

As controls served 123 healthy patients from our orthodontic department (Control group). Again patients younger than 6 years and syndromes were excluded.

2.2. Cephalometry. Cephalometric X-rays were made under usual standard conditions during inspiration (using cephalostats, film-focus-distance 4.0 meters, and consecutively factor of enlargement 2%, 32 mAs and depending on age and constitution 72 kV to 80 k). Digitalised X-rays showed a minimum solution of 400 dpi; analysis was performed using the software FRWIN "(Computer konkret, Systemhaus Falkenstein, Falkenstein, Germany) or for measurement of areas using Scion Image 4.0.2" (Scion Corporation, Frederick, MD, USA).

Parameters and variables measured in this study were adopted from various previously described cephalometric analyses [14–21], with most attention to the naso-, oro-, and velopharyngeal area. Figures 1 and 2 show the relevant landmarks used in this analysis.

Linear measurements were adjusted to the total length of the skull base (distance N-Ba) as internal reference to overcome confounding by different enlargement factors and to make different age groups more comparable. This technique of adjusting linear measurements to this internal reference line was also described and used by Ross [22], for example, in multicenter studies. Figures 3, 4, 5, and 6 show analysed variables of interest with regard to their anatomical region.

2.3. Statistical Analysis. To determine the measurement errors, 30 X-rays were randomly selected and measured twice within 2 weeks by the same examiner (KW). Randomized error according to Houston [23], combined methodology error according Dahlberg [24], and test on concordance according to the method described by Bland and Altman [25] were calculated.

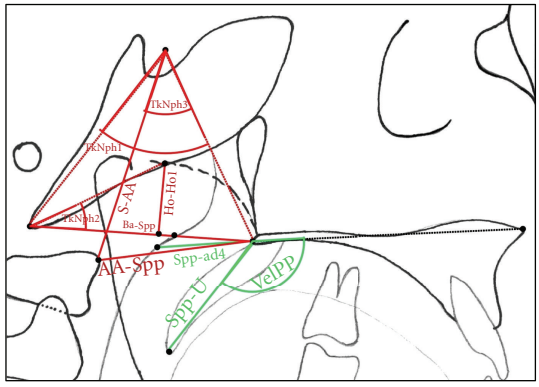


Figure 3: Cephalometric variables of the naso- and oropharyngeal area. (angular measurements: TkNph1: angle Ba-S-Spp, TkNph2: angle Ho-Ba-ad1, TkNph3: angle AA-S-Spp, VelPP: angle Spa-Spp-U; linear measurements are visible in this figure).

Table 1: Definition of subgroups based on age.

Age at taking cephalometric X-ray	Age Subgroup
6–11 years	I
>11–16 years	II
>16 years	III

Table 2: Cleft group (explanation of age-subgroups see Table 1).

Age subgroup	Gender				Total	
	Female		Male			
	n	%	*n*	%	*n*	%
I	4	6.1%	8	12.1%	12	18.2%
II	8	12.1%	10	15.2%	18	27.3%
III	11	16.7%	25	37.9%	36	54.5%
Total	23	34.8%	43	65.2%	66	100.0%

Table 3: Control group (for explanation of age-subgroups, see Table 1).

Age subgroup	Gender				Total	
	Female		Male			
	n	%	*n*	%	*n*	%
I	11	8.9%	14	11.4%	25	20.3%
II	20	16.3%	22	17.9%	42	34.1%
III	44	35.8%	12	9.8%	56	45.5%
Total	75	61.0%	48	39.0%	123	100.0%

To overcome serious confounding by age, for comparison of both collectives (cleft and controls), a division into three subgroups of age as follows was done (Table 1).

Comparison of two groups (e.g., male versus female and cleft versus control) was performed using chi-square test for nominal or ordinal scaled variables; for metric variables *t*-test was used. Comparing more than two groups (e.g., comparing the three subgroups of age) was performed with ANOVA and post hoc Scheffé procedure. Correlations between variables of the maxillary region or the skull base on one side and nasopharyngeal parameters on the other side were tested on significance using Spearman's correlation analysis.

3. Results

3.1. Error Analysis. Error analysis revealed excellent quality of measurement and good reliability. The standard deviation of 0.8 degree error in the measuring of angles and under 0.8 mm error in linear measurements documents precise analysis. Neither random errors nor systematic errors could be found. Using the Bland-Altmann procedure, except for two variables of areas (OphF and LRFV), the differences between two measurements were within the double-standard deviation, which means a good quality of the cephalometric analysis.

3.2. Subjects, Age, and Gender Influence. Tables 2 and 3 give an overview over age and gender distribution of included patients in both collectives. Median age was 16.4 (SD 4.1) years in the uCLP group and 15.4 (SD 6.9) years in the control group, showing no statistical significant differences between both groups concerning median age (*t*-test: $P > 0.05$) or age-subgroups (chi-square test: $P > 0.05$).

Concerning cephalometric measurements, no statistically significant differences could be found in our study population between male and female probands either in the cleft nor in the control group, although in the control group percentage of female patients was significantly higher than in the cleft group. Influence of age was certainly visible concerning linear measurements, but did not influence skeletal configuration of the face, nasopharyngeal configuration, or other relevant regions in both groups. Due to limited space, we hereby do not present the results in detail.

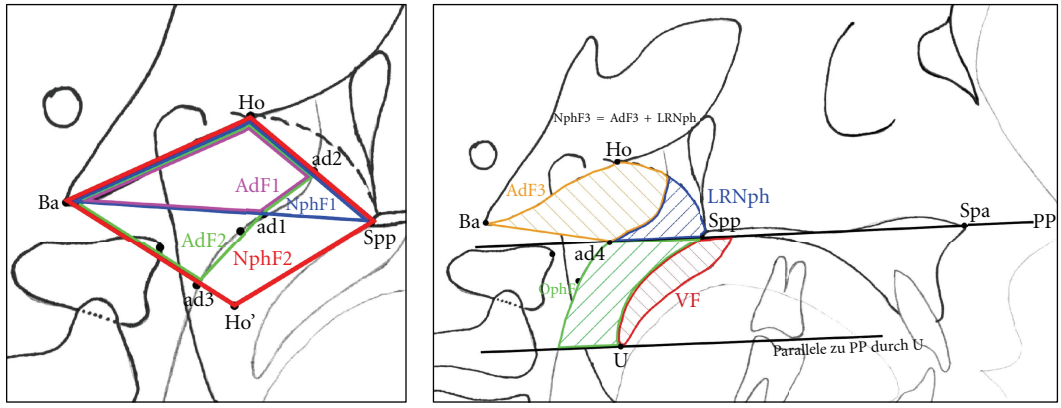
3.3. Correlations between Skull Base and Nasopharyngeal Parameters. A strong and statistical significant correlation ($|r_s| > 0.6$) could be found between flexion of the skull base (angle N-S-Ba) and posterior vertical facial height (distance S-Spp) and also the anterior skull base length (distance N-S). A greater skull base flexion was associated with shorter distances S-Spp and N-S.

Clear significant correlations ($|r_s| > 0.5$) could be determined between the N-S-Ba angle on the one side and angles S-N-B (position of the mandible), Ba-S-Spp (TkNph1, depth of the bony nasopharynx), and S-N-H (hyoid position).

Significant correlations with $0.4 < |r_s| < 0.5$ were observed between skull base flexion N-S-Ba and variables S-Go (total posterior facial height), GSHVER (facial height relation), and Ho-Ba-ad1 angle (TkNph2, depth of the bony nasopharynx II).

Table 4 shows the significant correlations in detail.

3.4. Differences between Cleft and Noncleft Probands. Table 5 shows the various differences between uCLP group and



(a) (b)

Figure 4: (a) (NphF1: area Spp-Ho-Ba-Spp, NphF2: area Spp-Ho-Ba-Ho′-Spp, AdF1: area ad2-Ho-Ba-ad1-ad2, AdF2: area ad2-Ho-Ba-ad3-ad1-ad2). ((a) and (b)) Measurement of areas in the naso- and oropharynx.

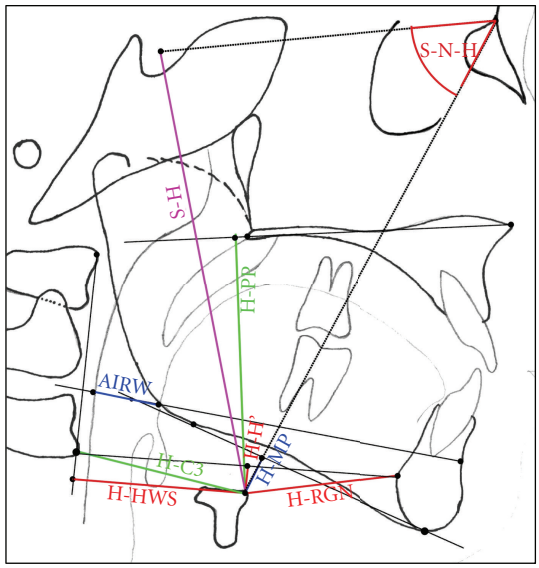


Figure 5: Variables measured to evaluate position of the hyoid. (angel S-N-H: hyoid position, AIRW: airway, that is, distance aAw-pAw).

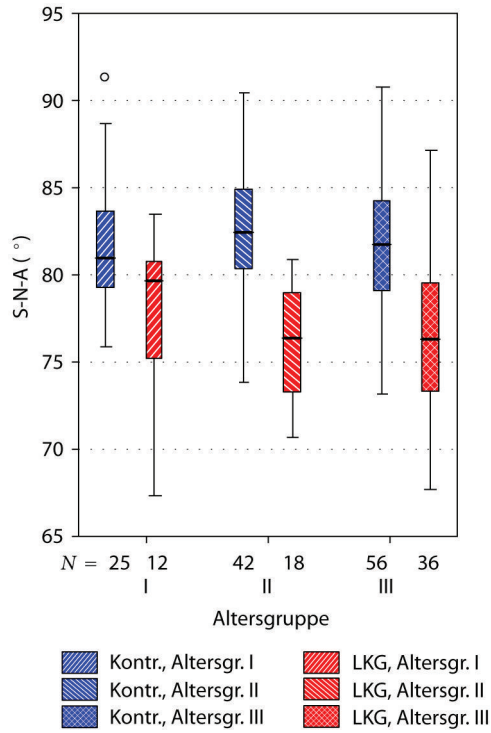


Figure 6: Boxplot illustrating significant differences between uCLP group and controls concerning maxillary position (angle S-N-A). "Altersgruppe": age subgroups, explanation see Table 1. "LKG": uCLP group (cleft). "Kontr.": control group (noncleft).

noncleft probands in detail. Patients with uCLP showed a statistically highly reduced posterior facial height (S-Spp, $P < 0.001$). Furthermore, values for angles S-N-A and Ba-N-A (position of the maxilla) and Conv-A (convexity of Point A) were highly and significantly reduced which can be interpreted as sign of maxillary retrognathia and retroposition. Also mandibular parameters differed significantly (angles S-N-B and N-Go-Me).

In the nasopharyngeal region, vertical height (Ho-Ho1), horizontal depth (Ba-Spp), and depth of the bony nasopharynx (angle AA-S-Spp, TkNph3) were significantly diminished. Patients with uCLP showed also a reduced velar length (Spp-U) and consecutively a more unfavourable need ratio (Spp-ad4/Spp-U) ($P < 0.01$).

Comparisons between cleft and noncleft group subgrouped to three age groups (see Table 1) came to similar results. Highest significance was observed concerning the angles S-N-A (position of the maxilla), AA-S-Spp (TkNph3, depth of th bony nasopharynx), and the linear distance Ba-Spp (sagittal depth of the bony nasopharynx).

The Figures 6, 7, and 8 illustrate these differences clearly.

4. Discussion

Generally a retrospective cephalometric analysis has to face the problem of measuring faults. There are radiographic

Table 4: Significant correlations found between skull base flexion and other cephalometric variables.

Correlation of N-S-Ba to	N-S	N-S	S-Go	GSHVER
Correlation coefficient r_s	−0.645**	−0.645**	−0.401**	0.414**
Correlation of N-S-Ba to	S-N-A	Conv-A	S-N-B	S-AA
Correlation coefficient r_s	−0.344**	0.294**	−0.537**	−0.346**
Correlation of N-S-Ba to	Ho-Ho1	TkNph1	TkNph2	TkNph3
Correlation coefficient r_s	−0.322**	0.518**	−0.472**	0.336**
Correlation of N-S-Ba to	H-S	S-N-H		
Correlation coefficient r_s	−0.342**	−0.535**		

r_s: Spearman's correlation coefficient.
Level of significance: $*P < 0.05$, $**P < 0.01$).
(GSHVER = facial height relation = S-Go/N-Me, Conv-A = convexity of point A = distance of A-point perpendicular to N-Po; other variables see Figures 1–5).

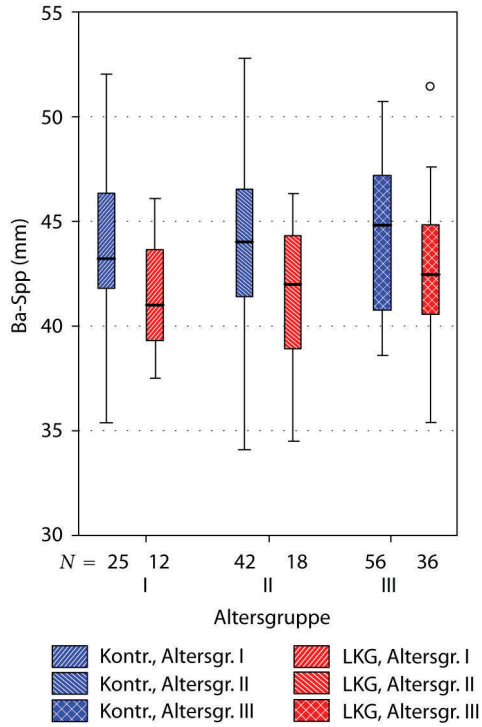


Figure 7: Boxplot illustrating significant differences between uCLP group and controls concerning sagittal nasopharyngeal depth (distance Ba-Spp). "Altersgruppe": age subgroups, explanation see Table 1. "LKG": uCLP group (cleft). "Kontr.": control group (noncleft).

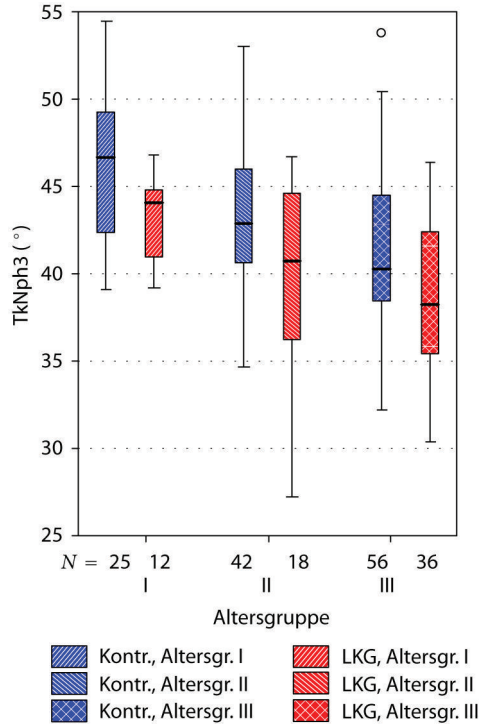


Figure 8: Boxplot illustrating significant differences between uCLP group and controls concerning bony nasopharyngeal depth (angle AA-S-Spp). "Altersgruppe": age subgroups, explanation see Table 1. "LKG": uCLP group (cleft). "Kontr.": control group (noncleft).

faults, caused by the radiological technique, errors in the identification of measuring points, and errors during the appraisal of the measured points, distances, and angles.

To reduce radiological variations, all linear measurements were calibrated to the length of the scull base N-Ba according to the data of the multicentric investigations of Ross [22, 26, 27]. The standard deviation of 0.8 degree in the measuring of angles and under 0.8 mm in linear measurements stands for a high metering precision and corresponds well with the results of comparable investigations [28].

Although both compared groups showed different percentage in their male-to-female ratio, analysis revealed no statistical influence of gender on cephalometric variables as no difference could be established between males and females in this study neither in the uCLP nor in the control group. Even when keeping this missing gender matching in mind, for this reason, we assumed the control group being sufficient enough for this comparative analysis.

We found in our data a constricted development of the midface, a reduced facial depth, a more vertical growth pattern and a reduced sagittal dimension of the nasopharyngeal complex in favour of a more vertical naso-pharyngeal development.

Table 5: Significant differences between uCLP and control group in total.

Variable	Unit		Group uCLP $n = 66$	Group uCLP $n = 123$	Sign.
FACAX	∘	mean	88.1	90.7	**
		SD	6.8	4.7	
N-BA	mm	mean	101.2	97.9	**
		SD	6.2	6.7	
N-S	mm	mean	67.2	68.3	*
		SD	2.3	2.6	
S-Spp	mm	mean	43.9	46.4	***
		SD	3.1	4.8	
A-Me	mm	mean	59.7	58.1	*
		SD	4.7	4.9	
N-Me	mm	mean	113.8	111.6	*
		SD	7.0	7.1	
GSHVER	%	mean	64.0	65.8	*
		SD	5.6	5.3	
Spp-A	mm	mean	43.5	44.9	**
		SD	3.5	2.8	
Spp-Spa	mm	mean	47.2	48.3	*
		SD	3.3	2.9	
S-N-A	∘	mean	76.7	82.0	***
		SD	4.5	3.9	
Ba-N-A	∘	mean	58.2	63.4	***
		SD	4.8	3.6	
Conv-A	mm	mean	−0.6	2.2	***
		SD	4.0	2.9	
S-N-B	∘	mean	76.0	78.5	***
		SD	4.3	4.7	
Ar-Go-Me	∘	mean	129.1	125.9	**
		SD	6.9	7.8	
N-Go-Me	∘	mean	76.5	72.5	***
		SD	6.8	5.3	
FACDEP	∘	mean	83.6	85.6	*
		SD	5.6	4.0	
MANPLA	∘	mean	30.1	26.3	**
		SD	9.0	6.0	
S-AA	mm	mean	51.3	49.1	**
		SD	5.9	4.5	
Ho-Ho1	mm	mean	17.5	16.2	***
		SD	2.2	1.5	
Ba-Spp	mm	mean	41.3	44.8	***
		SD	4.1	3.6	
AA-Spp	mm	mean	33.4	35.3	**
		SD	4.5	3.2	
TkNph1	∘	mean	58.1	61.1	**
		SD	5.8	4.6	
TkNph3	∘	mean	39.8	43.1	***
		SD	5.0	4.6	
NphF1	mm^2	mean	375.4	349.7	*
		SD	69.9	56.7	
NphF2	mm^2	mean	750.8	698.9	*
		SD	139.7	119.0	
AdF1	mm^2	mean	231.2	212.4	*
		SD	44.4	45.0	
AdF2	mm^2	mean	363.2	335.8	*
		SD	76.6	76.7	
Spp-U	mm	mean	29.1	31.8	***
		SD	5.0	3.4	
Spp-ad4	mm	mean	22.9	27.5	***
		SD	3.7	3.3	
NeedRat	%	mean	80.8	87.3	*
		SD	18.8	13.2	
H-C3	mm	mean	34.4	32.4	**
		SD	4.7	4.5	
H-MP	mm	mean	19.0	17.0	*
		SD	6.1	5.6	
H-PP	mm	mean	60.5	58.0	*
		SD	7.3	6.5	
H-HWS	mm	mean	32.9	30.8	**
		SD	4.4	4.2	

Level of significance (t-test): $^*P < 0.05$, $^{**}P < 0.01$, $^{***}P < 0.001$ (FACAX = facial axis = angle between Ba-N and Pt-Gnk, GSHVER = facial height relation = S-Go/N-Me, CONV-A = convexity of point A = distance of A-point perpendicular to N-Po, FACDEP = facial depth = angle between P-Or and N-Po, MANPLA = mandibular plane = angle between P-Or and hT-Me, NEEDRAT = need ratio = Spp-ad4/Spp-U, other variables see Figures 1–5).

These results correspond with comparable findings in the literature [28–32].

The need ratio, defined as quotient of velar length and distance of the velum to the pharyngeal posterior wall, ranges in our data from 79.7% to 81.6% depending on the age of the cleft patients. The results of the control group range from 85.1% to 89.1%.

This significant difference stands for an impaired velopharyngeal closure and underlines the clinical relevance of the cephalometric measuring especially when it comes to decisions concerning surgical interventions, for example, velopharyngoplasty.

But it has to be critically kept in mind, that in our here presented cephalometric study, based on lateral X-rays of the skull, a separate assessment of velopharyngeal function and speech was not performed. Despite other studies showed clear correlations between velopharyngeal insufficiency and distinct changes concerning cephalometric measurements, this study is not able and was not designed to compare VPI and non-VPI patients [9, 31].

Which factors may have influenced the depicted differences of nasopharyngeal development and configuration in

cleft patients compared to a noncleft control group? Of etiological relevance may be an impaired nasal breathing in cleft patients, postoperative scaring especially after palatal closure, and inphysiological or insufficient function of velopharyngeal muscles.

In our data, the cleft patients showed a more caudal and anterior position of the hyoid compared to a healthy collective. In addition, the hyoid position comes with greater distances to the cervical spine and the palatinal and the mandibular plane. Kaduk et al. and Rose et al. report similar results [33, 34]. In many clinical investigations, the caudal position of the hyoid is correlated to a statistical significant accumulation of nightly respiration disorders like snoring or even sleep apnoea. Additionally, these patients present frequently mandibular or maxillary retrognathia, enlarged tonsils and adenoids, and a vertical facial growth pattern. Some authors conclude that the caudal hyoid position represents a habitual adaptation to the narrower pharyngeal area [18, 35–38].

The lateral cephalometry as a two-dimensional analysis provides enough meaningful information for an effectual appraisal not only of the skeletal skull but also of the nasopharyngeal area [39]. It does not include the transversal dimension and neglects, therefore influencing factors like a deviated nasal septum or functional aspects like mobility of the velum and velopharyngeal closure mechanism, but it represents a low price and convenient routine diagnostic device in the analysis of skeletal and soft tissue landmarks.

5. Conclusions

The cephalometric comparison of cleft patients to a healthy control group showed significant differences: due to an impaired ventrocaudal growth tendency of the naso-maxillary area a retroposition of the maxillary complex in combination with a reduced maxillary length and height, especially in the area of the posterior nasal spine, was detected. In the cleft group, a reduced sagittal dimension of the hard and soft tissues of the nasopharyngeal complex in favour of a more accentuated vertical development became obvious. Combined with a reduced velar length, the result is an insufficient velopharyngeal closure.

In the cleft collective, a cumulation of mandibular retrognathia and a more caudal and anterior position of the hyoid was observed.

The results underline the pivotal role of the functional reconstruction of the velo-pharyngeal muscles to allow not only for a physiological speech development but also for a regular growth of the naso-, oro-, and velopharyngeal structures.

Conflict of Interests

The authors declare no conflict of interests.

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Clinical Study

Objective Assessment of Hypernasality in Patients with Cleft Lip and Palate with the NasalView System: A Clinical Validation Study

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Introduction. The objective of this investigation was to evaluate the reliability and validity of the NasalView system as a screening tool for hypernasality within the scope of a routine diagnostic procedure in cleft lip and palate patients. *Material and Methods*. In a collective of 95 patients with cleft and lip palate ranging from 4 to 25 years of age, hypernasality was exploited perceptually, patients were classified in four degrees, and nasalance was measured objectively with the NasalView system. Speech stimuli existed in one nasal and one nonnasal sentence; nasalance ratio and distance were calculated. *Results*. The test-retest error was within a range of 2%. Sensitivity ranged from 83.3% to 91.1% for the nonnasal sentence, from 70% to 78.4% for nasalance ratio and from 68.1% to 81.1% for nasalance distance. Specifity ranged from 87% to 93.1% for the nonnasal sentence, from 69.6% to 97.5% for nasalance ratio, and from 70.7% to 73.9% for nasalance distance. *Conclusions*. With a quick and gentle screening procedure, it is easily possible to identify hypernasal patients by an objective diagnostic tool of hypernasality, the NasalView system, with good reliability and validity.

1. Introduction

The most common speech impairment of patients with cleft lip and palate are resonance disorders [1]. They are characterized as impairments of the balance between oral and nasal acoustic energy and occur in form of hypernasality, hyponasality and mixed forms, for example, the cul-de-sac resonance (Vrticka [2–5], Bressmann and Sader [6]). Hypernasality due to velopharyngeal insufficiency in patients with cleft lip and palate can be caused by structural inadequacy and functional incompetence [6]. In some cases even after anatomically surgical reconstruction of the soft and the hard palate and consequent logopaedic treatment, hypernasality remains. Velopharyngeal morphology and function and perceptual consequence have to be assessed objectively to decide on a surgical intervention, for example, velopharyngoplasty to improve speech capability.

Regular speech and resonance disorders are evaluated by perceptual assessment; the quality of judgement depends on the listeners experience and academic training (Lewis et al. [7]). Because of this interrater variability, it is useful to augme that the subjective assessment by an objective quantitative instrumental analysis of resonance disorders.

An instrumental measurement of hypernasality can be performed by the Kay Nasometer (Kay Elemetrics, Lincoln Park, NJ, USA) and with the NasalView system (Tiger Electronics, Seattle, WA, USA), developed by Awan in 1997 [8]. Both systems measure the nasalance by calculating the proportion of the nasal energy in speech from separate measurements of nasal and oral sound pressure level, nasalance = nasal/[nasal + oral] × 100% (Fletcher [9, 10]).

For the Nasometer already several validation studies exist, demonstrating the good correlation between perceptual judgement of oronasal sound balance and nasalance measurement. In addition, they present solid validation data such as sensitivity, specificity, and efficiency (Dalston et al. [11, 12], Hardin et al. [13], Nellis et al. [14], Watterson et al. [15, 16], and Stellzig et al. [17]).

Bressmann et al. [18–21] established a solid validation for the NasalView System in German speaking cleft

patients. Depending on the measurement, nonnasal sentences, nasalance distance and nasalance ratio, and depending on the differentiation of patients with different levels of hypernasality, in their investigations the sensitivity ranged from 70.7% to 91.1%, the specificity ranged from 73% to 97.1%, and the efficiency ranged from 71.4% to 91.5%.

The objective of this prospective clinical validation study was to evaluate practicability, reliability, and validity of the NasalView system as a noninvasive screening tool for resonance disorders in the course of a routine diagnostic for patients with cleft lip and palate.

2. Material and Methods

2.1. Subjects. 100 patients with cleft lip and palate were examined. Three patients were excluded from the analysis due to reduced cooperation and compliance, in two cases the data had to be rejected due to inaccurate recording.

A total of 95 patients from 4 to 25 years of age (median age 9.25 years ± 4.25) were analysed in this investigation. Of these 52 females and 43 males, 54 had a complete unilateral cleft of the lip, alveolus and palate, 12 had an bilateral cleft lip, alveolus, and palate, 19 a cleft of the palate, and 10 patients suffered from velopharyngeal insufficiency in combination with minor forms of clefting (submucous cleft and uvula bifida).

2.2. Objective Measurement of Nasalance. Nasalance was measured with the NasalView system, version 1.2, Tiger Electronics DRS Inc., Seattle, WA, USA. The measuring was performed following standard procedures in a noiseless room. Before each recording the NasalView was calibrated according to the manufacturer instructions. The test items were recorded at a sampling rate of 22.5 Hz stereo, 16 bit; a generic IBM compatible PC with an Intel Pentium IV Processor and a Soundblaster Audio PCI 128 soundcard (Creative Labs, Singapore) were used.

For each test item the NasalView measures the following parameters of nasalance.

The average nasalance (Ave, i.e., the arithmetic mean), the standard deviation (SD), maximum (max) and minimum (min), the median of nasalance (Median), and mode. Figure 1 illustrates a sample.

In our study the nasalance values Ave and Median were analysed. The other parameters (mode, maximum, minimum, and standard deviation) were analyzed in other studies before and were not reliable enough to allow for conclusive results for there is no sharp distinction between nasal and nonnasal test items (Bressmann et al. [18–21]). Even in the same patient they show similar results in nonnasal and nasal speech stimuli, so a clear distinction between hypernasal and normal speech is clearly impossible.

The nonnasal sentence (nnS-testitem) "Der Peter trinkt die Tasse Kakao" ("The Peter drinks a cup of cocoa") and the nasal sentence (nS-testitem) "Mama und Nina naschen Marmelade" ("Mama and Nina nibble marmalade") served as speech stimuli. Other investigators had underlined the importance of adjusting the level of complexity to the developmental stage of the examined children (Van Denmark and Swickard [22], Baker et al. [23]). The chosen two sentences are simple enough to be recorded easily from children from the age of four years on. In addition, the results of other studies underlined the efficiency of a stimulus length of six syllables or a shortened recording procedure [21, 24].

2.3. Perceptual Analysis. Perceptual assessment of nasality was achieved by samples of spontaneous speech during a semistandardized interview routine by an experienced speech therapist. The degree of hypernasality was then classified as normal (no hypernasality), borderline (mild hypernasality), marked (hypernasality), and severe (impaired understanding). Patients were then grouped according to results of the perceptual assessment.

2.4. Statistical Analysis. Test items were recorded twice with the NasalView system during one examination; the arithmetic means and standard deviations of the difference between the nasalance measuring were calculated to determine the test-retest error for both, the Ave-nasalance value and the Median-nasalance value. To decide which nasalance value was more reliable, Ave or Median, the differentiation between both values was tested on significance by using the Wilcoxon matched pairs signed rank test.

When a sentence was recorded more than once, the arithmetic mean was calculated and valued for this test item. For each patient nasalance distance (maximum nasalance–minimum nasalance) were calculated (Bressmann et al. [21]).

For each group of patients classified according to the perceptual assessment of four degrees of hypernasality, arithmetic means and standard deviation for the various measures were calculated. Differences among the groups were tested on significance using ANOVA and the post hoc Scheffé tests.

In diagnostics high sensitivity, specificity, and efficiency are required. Sensitivity in our case is the percentage of patients correctly identified by the NasalView system as hypernasal in relation to all hypernasal patients. Specificity is defined as percentage of patients with no perceptual hypernasality in relation to all patients with regular nasalance parameters measured by the NasalView system. Efficiency or overall diagnostic accuracy is defined as the number of all correct decisions over the number of all decisions. An apt tool to achieve optimum values are Receiver-Operating Characteristics (ROC) curves that plot the sensitivity and false positive rate [25, 26]. With these ROC curves, validation data was determined for the various discriminations between degrees of hypernasality.

3. Results

3.1. Reliability. A low test error stands for good reliability and is a prerequisite for the routine application of a diagnostic tool. A total sum of 157 pairs of sentences were analysed to determine the test-retest error. Table 1 presents the results.

Because the test-retest error for Ave-nasalance values are statistically significantly lower than the test-retest error for

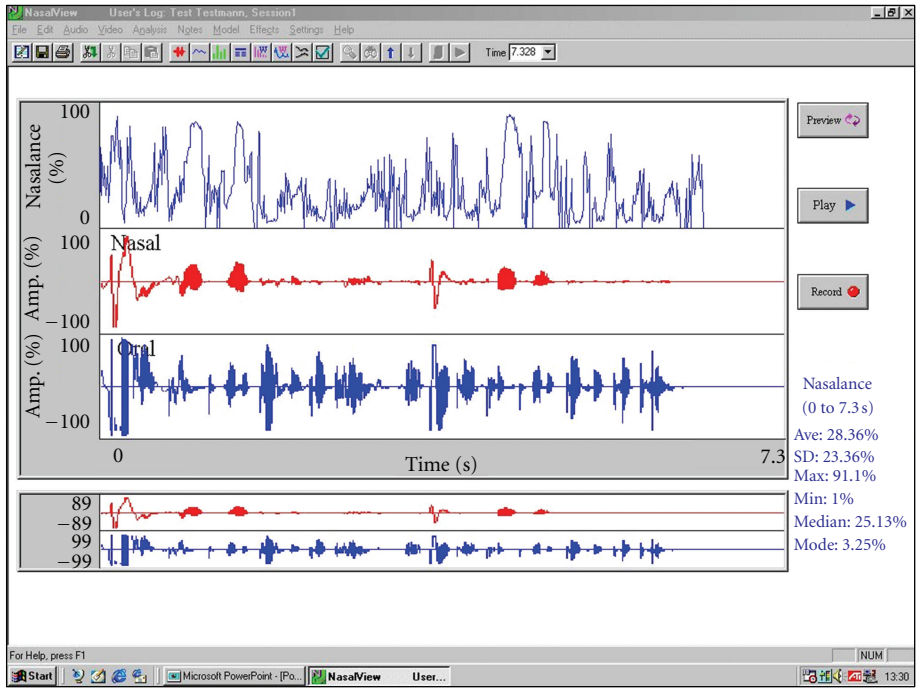

Figure 1: Sample screenshot of the NasalView system showing oszillograms for oral, nasal, and nasalance curves recorded realtime and nasalance statistic for a nonnasal test item.

Table 1: Test-retest errors for ave and median nasalance measurement.

Measurement	Pairs of test items		Total ($n = 157$)
	Nasal sentences ($n = 95$)	Nonnasal sentences ($n = 62$)	
D-Ave [%]			
Mean	1.50	1.37	1.44
SD	0.99	1.15	1.05
D-Median [%]			
Mean	2.84	1.66	2.37
SD	2.31	1.35	2.06

D-ave: difference of ave nasalance values between two test items; D-median: difference of median nasalance values between two test items mean: arithmetic mean; SD: standard deviation.

Median-nasalance values ($P < 0.001$), only Ave values are reported as nasalance value in the following results.

3.2. Group Differences. According to the results of the perceptual analysis, 23 patients were classified normal, 35 patients were borderline cases, 27 patients showed marked hypernasality, and 10 patients had severe hypernasal speech.

Table 2 provides the results of the objective computerized measurements with the NasalView system. With the exception of the nasalance of the nnS-test item (nonnasal sentence) the differences between the single groups were significant at the $P < 0.05$ level. For the nonnasal sentence group, differences were significant with $P < 0.01$.

3.3. Validation Data. The optimum cutoff, sensitivity, specificity, and efficiency were calculated for the various measurements regarding the following discriminations:

(a) differentiation between all patients with regular resonance and all hypernasal patients (Table 3);

(b) differentiation between all patients with regular resonance and hypernasal patients with marked and severe hypernasality excluding borderline cases (Table 4);

(c) differentiation between patients with marked and severe hypernasality (Table 5);

(d) differentiation between patients with no or borderline rhinophonia and hypernasal patients with marked and severe hypernasality (Table 6). This discrimination is based upon the therapeutic consequence: in cases of marked and severe hypernasality, treatment of the speech disorder—speech therapy or surgical intervention is necessary, whereas in borderline cases the further clinical development is scrutinized.

4. Discussion

The application of the NasalView system was easy due to the fact that the measuring of nasalance is entirely noninvasive. In addition to the practicability, this investigation also demonstrates the good reliability of computerized assessment of hypernasality with the NasalView system. In accordance with Awan [8], who accounted for a test-retest

Table 2: Results grouped in different degrees of hypernasality.

Measurement	degree of hypernasality				P
	Normal ($n = 23$)	Borderline ($n = 35$)	Marked ($n = 27$)	Severe ($n = 10$)	
Nasalance of nS [%]					
Mean	44.08	45.06	47.65	55.30	0.048*
SD	4.63	6.12	5.66	5.42	
Nasalance of nnS [%]					
Mean	26.30	29.80	35.36	45.21	0.007**
SD	2.93	3.49	5.21	4.25	
Nasalance ratio					
Mean	0.602	0.671	0.745	0.819	0.036*
SD	0.081	0.139	0.142	0.085	
Nasalance distance [%]					
Mean	17.76	15.26	12.29	10.08	0.033*
SD	4.85	5.94	5.04	3.43	

nS: nasal sentence; nnS: nonnasal sentence; mean: arithmetic mean; SD: standard deviation P: P value of ANOVA (differences between degrees of hypernasality)*: $P < 0.05$ **: $P < 0.01$.

Table 3: Validation data for differentiation between patients with regular resonance and hypernasal patients.

Measurement	Cutoff	Sensitivity [%]	Specificity [%]	Efficiency [%]
Nasalance of nnS	28.5%	83.3	87.0	84.2
Nasalance ratio	0.66	70.8	69.6	70.5
Nasalance distance	15.3%	68.1	73.9	69.5

nnS: nonnasal sentence.

Table 4: Validation data for differentiation between patients with regular resonance and hypernasal patients excluding borderline cases.

Measurement	Cutoff	Sensitivity [%]	Specificity [%]	Efficiency [%]
Nasalance of nnS	28.6%	91.9	91.3	91.7
Nasalance ratio	0.72	78.4	95.7	85.0
Nasalance distance	15.0%	81.1	73.9	78.3

nnS: nonnasal sentence.

Table 5: Validation data for differentiation between patients with marked and severe hypernasality.

Measurement	Cutoff	Sensitivity [%]	Specificity [%]	Efficiency [%]
Nasalance of nnS	40.0%	90.0	88.9	89.2
Nasalance ratio	0.818	70.0	77.8	75.7
Nasalance distance	10.5%	70.0	59.3	62.2

nnS: nonnasal sentence.

error of 2% for the NasalView system and a test-retest error of 3% for the Nasometer in a study with 20 adult subjects, we established a test-retest error under 2% when the average value of the reported nasalance statistic was used.

Table 6: Validation data for differentiation between patients with no and borderline rhinophonia and patients with marked and severe hypernasality.

Measurement	Cutoff	Sensitivity [%]	Specificity [%]	Efficiency [%]
Nasalance of nnS	33.0%	86.5	93.1	90.5
Nasalance ratio	0.72	78.4	84.5	82.1
Nasalance distance	14.02%	75.7	70.7	72.6

nnS: nonnasal sentence.

This small error implicates a good reliability of the measuring technique.

The validation data established in our investigation are comparable to the results of other studies dealing with the NasalView system or the Nasometer.

For the Nasometer, Dalston et al. [11, 12] and Hardin et al. [13] reported an efficiency ranging from 79.2% to 93% depending on whether borderline cases were included or not. In 30 German speaking cleft patients with a mean age of 13.5 years, Stellzig et al. measured in 1994 an overall diagnostic accuracy of 90% for the Nasometer [17]. Nasalance measurement with the NasalView system and the Nasometer differ technically and in appliance. Therefore, a direct comparison of the detailed results is not possible (Lewis and Watterson [27]).

As far as the NasalView system is concerned comparable investigations have been performed by Bressmann, who analysed nasalance measurements of 133 and 140 German speaking patients with cleft lip and palate from 10 to 66 years of age, mean age = 17 years (Bressmann et al. [20, 21]).

When differentiating between healthy subjects and hypernasal patients, we found the same optimum cutoff of 28.5% for the nonnasal test item. Whereas Bressmann reported an efficiency of 73%, we found a higher overall

diagnostic accuracy of 84.2% for this test item and the discrimination. For the nasalance ratio and nasalance distance in our study, we found lower values of 70.5% and 69.5% than Bressmann, who reported an accuracy of ranging from 75.2% to 77.4%. When excluding borderline cases from the analysis, sensitivity, specificity, and efficiency improve considerably. In the present study for the nonnasal test item efficiency improved to 91.7% compared to 81.4% found by Bressmann et al. [20]. Although efficiency for nasalance ratio and distance improved to 85% and 78.3%, the results are lower than the values measured by Bressmann et al. [21].

In our data, optimum cutoffs for the nasalance ratio were higher and cutoffs for the nasalance distance were lower than in the comparable validation study [21]. These discrepancies can be provoked by the influence of a different dialect, as described by Van Lierde et al. [28], or the use of similar but not identical sentences.

Another decision of clinical importance is the differentiation between patients with no and mild hypernasality, normal and borderline cases, and patients with marked and severe rhinophonia. Due to clinical experience, mostly a mild hypernasality does not impair intelligibility of speech and represents no stigma for the patient in his social environment. In most cases of borderline hypernasality, no treatment is mandatory. On the other hand, in patients who suffer from marked or severe hypernasality with impaired speech regularly treatment, that is, surgical intervention with velopharyngoplasty is necessary. Differentiating between patients with the need for intervention and those who do not need therapy as described above, again the best efficiency of 90.5% results from the nonnasal sentence with a cutoff of 33%. When using nasalance ratio and distance, we found a diagnostic accuracy of 82.1% and 72.6%, respectively, for the differentiation between patients with no and borderline hypernasality on the one side and patients with marked and severe hypernasality on the other side. This clear differentiation between hypernasality with the need for treatment and no or mild hypernasality without need of treatment need was not yet described in previous studies.

5. Conclusions

The results of this clinical validation identify the NasalView system as an apt and userfriendly device to identify patients with resonance disorders. A short screening procedure including a nasal and a nonnasal test item is sufficient to differentiate between the perceptual degrees of hypernasality with good reliability and validity.

To come to a sensible decision as far as the further treatment of patients with cleft lip and palate and resulting hypernasality is concerned, it is necessary to assess the velopharyngeal sphincter with regard to its structure, function, and the perceptual consequences. The validation data of our investigation among others indicates that an objective measuring of rhinophonia is helpful to supplement the perceptual assessment of hypernasality by an experienced speech therapist.

We consider the objective computerized diagnostic of nasality with the NasalView system an expedient completion and helpful screening tool for resonance disorders, but not as a replacement of perceptual judgements by experienced speech therapists and listeners.

Conflict of Interests

All authors declare that there exist no conflict of interests.

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Clinical Study

Objective Assessment of Hypernasality in Patients with Cleft Lip and Palate with the NasalView System: A Clinical Validation Study

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Introduction. The objective of this investigation was to evaluate the reliability and validity of the NasalView system as a screening tool for hypernasality within the scope of a routine diagnostic procedure in cleft lip and palate patients. *Material and Methods*. In a collective of 95 patients with cleft and lip palate ranging from 4 to 25 years of age, hypernasality was exploited perceptually, patients were classified in four degrees, and nasalance was measured objectively with the NasalView system. Speech stimuli existed in one nasal and one nonnasal sentence; nasalance ratio and distance were calculated. *Results*. The test-retest error was within a range of 2%. Sensitivity ranged from 83.3% to 91.1% for the nonnasal sentence, from 70% to 78.4% for nasalance ratio and from 68.1% to 81.1% for nasalance distance. Specifity ranged from 87% to 93.1% for the nonnasal sentence, from 69.6% to 97.5% for nasalance ratio, and from 70.7% to 73.9% for nasalance distance. *Conclusions*. With a quick and gentle screening procedure, it is easily possible to identify hypernasal patients by an objective diagnostic tool of hypernasality, the NasalView system, with good reliability and validity.

1. Introduction

The most common speech impairment of patients with cleft lip and palate are resonance disorders [1]. They are characterized as impairments of the balance between oral and nasal acoustic energy and occur in form of hypernasality, hyponasality and mixed forms, for example, the cul-de-sac resonance (Vrticka [2–5], Bressmann and Sader [6]). Hypernasality due to velopharyngeal insufficiency in patients with cleft lip and palate can be caused by structural inadequacy and functional incompetence [6]. In some cases even after anatomically surgical reconstruction of the soft and the hard palate and consequent logopaedic treatment, hypernasality remains. Velopharyngeal morphology and function and perceptual consequence have to be assessed objectively to decide on a surgical intervention, for example, velopharyngoplasty to improve speech capability.

Regular speech and resonance disorders are evaluated by perceptual assessment; the quality of judgement depends on the listeners experience and academic training (Lewis et al. [7]). Because of this interrater variability, it is useful to augme that the subjective assessment by an objective quantitative instrumental analysis of resonance disorders.

An instrumental measurement of hypernasality can be performed by the Kay Nasometer (Kay Elemetrics, Lincoln Park, NJ, USA) and with the NasalView system (Tiger Electronics, Seattle, WA, USA), developed by Awan in 1997 [8]. Both systems measure the nasalance by calculating the proportion of the nasal energy in speech from separate measurements of nasal and oral sound pressure level, nasalance = nasal/[nasal + oral] × 100% (Fletcher [9, 10]).

For the Nasometer already several validation studies exist, demonstrating the good correlation between perceptual judgement of oronasal sound balance and nasalance measurement. In addition, they present solid validation data such as sensitivity, specificity, and efficiency (Dalston et al. [11, 12], Hardin et al. [13], Nellis et al. [14], Watterson et al. [15, 16], and Stellzig et al. [17]).

Bressmann et al. [18–21] established a solid validation for the NasalView System in German speaking cleft

patients. Depending on the measurement, nonnasal sentences, nasalance distance and nasalance ratio, and depending on the differentiation of patients with different levels of hypernasality, in their investigations the sensitivity ranged from 70.7% to 91.1%, the specificity ranged from 73% to 97.1%, and the efficiency ranged from 71.4% to 91.5%.

The objective of this prospective clinical validation study was to evaluate practicability, reliability, and validity of the NasalView system as a noninvasive screening tool for resonance disorders in the course of a routine diagnostic for patients with cleft lip and palate.

2. Material and Methods

2.1. Subjects. 100 patients with cleft lip and palate were examined. Three patients were excluded from the analysis due to reduced cooperation and compliance, in two cases the data had to be rejected due to inaccurate recording.

A total of 95 patients from 4 to 25 years of age (median age 9.25 years ± 4.25) were analysed in this investigation. Of these 52 females and 43 males, 54 had a complete unilateral cleft of the lip, alveolus and palate, 12 had an bilateral cleft lip, alveolus, and palate, 19 a cleft of the palate, and 10 patients suffered from velopharyngeal insufficiency in combination with minor forms of clefting (submucous cleft and uvula bifida).

2.2. Objective Measurement of Nasalance. Nasalance was measured with the NasalView system, version 1.2, Tiger Electronics DRS Inc., Seattle, WA, USA. The measuring was performed following standard procedures in a noiseless room. Before each recording the NasalView was calibrated according to the manufacturer instructions. The test items were recorded at a sampling rate of 22.5 Hz stereo, 16 bit; a generic IBM compatible PC with an Intel Pentium IV Processor and a Soundblaster Audio PCI 128 soundcard (Creative Labs, Singapore) were used.

For each test item the NasalView measures the following parameters of nasalance.

The average nasalance (Ave, i.e., the arithmetic mean), the standard deviation (SD), maximum (max) and minimum (min), the median of nasalance (Median), and mode. Figure 1 illustrates a sample.

In our study the nasalance values Ave and Median were analysed. The other parameters (mode, maximum, minimum, and standard deviation) were analyzed in other studies before and were not reliable enough to allow for conclusive results for there is no sharp distinction between nasal and nonnasal test items (Bressmann et al. [18–21]). Even in the same patient they show similar results in nonnasal and nasal speech stimuli, so a clear distinction between hypernasal and normal speech is clearly impossible.

The nonnasal sentence (nnS-testitem) "Der Peter trinkt die Tasse Kakao" ("The Peter drinks a cup of cocoa") and the nasal sentence (nS-testitem) "Mama und Nina naschen Marmelade" ("Mama and Nina nibble marmalade") served as speech stimuli. Other investigators had underlined the importance of adjusting the level of complexity to the developmental stage of the examined children (Van Denmark and Swickard [22], Baker et al. [23]). The chosen two sentences are simple enough to be recorded easily from children from the age of four years on. In addition, the results of other studies underlined the efficiency of a stimulus length of six syllables or a shortened recording procedure [21, 24].

2.3. Perceptual Analysis. Perceptual assessment of nasality was achieved by samples of spontaneous speech during a semistandardized interview routine by an experienced speech therapist. The degree of hypernasality was then classified as normal (no hypernasality), borderline (mild hypernasality), marked (hypernasality), and severe (impaired understanding). Patients were then grouped according to results of the perceptual assessment.

2.4. Statistical Analysis. Test items were recorded twice with the NasalView system during one examination; the arithmetic means and standard deviations of the difference between the nasalance measuring were calculated to determine the test-retest error for both, the Ave-nasalance value and the Median-nasalance value. To decide which nasalance value was more reliable, Ave or Median, the differentiation between both values was tested on significance by using the Wilcoxon matched pairs signed rank test.

When a sentence was recorded more than once, the arithmetic mean was calculated and valued for this test item. For each patient nasalance distance (maximum nasalance–minimum nasalance) were calculated (Bressmann et al. [21]).

For each group of patients classified according to the perceptual assessment of four degrees of hypernasality, arithmetic means and standard deviation for the various measures were calculated. Differences among the groups were tested on significance using ANOVA and the post hoc Scheffé tests.

In diagnostics high sensitivity, specificity, and efficiency are required. Sensitivity in our case is the percentage of patients correctly identified by the NasalView system as hypernasal in relation to all hypernasal patients. Specificity is defined as percentage of patients with no perceptual hypernasality in relation to all patients with regular nasalance parameters measured by the NasalView system. Efficiency or overall diagnostic accuracy is defined as the number of all correct decisions over the number of all decisions. An apt tool to achieve optimum values are Receiver-Operating Characteristics (ROC) curves that plot the sensitivity and false positive rate [25, 26]. With these ROC curves, validation data was determined for the various discriminations between degrees of hypernasality.

3. Results

3.1. Reliability. A low test error stands for good reliability and is a prerequisite for the routine application of a diagnostic tool. A total sum of 157 pairs of sentences were analysed to determine the test-retest error. Table 1 presents the results.

Because the test-retest error for Ave-nasalance values are statistically significantly lower than the test-retest error for

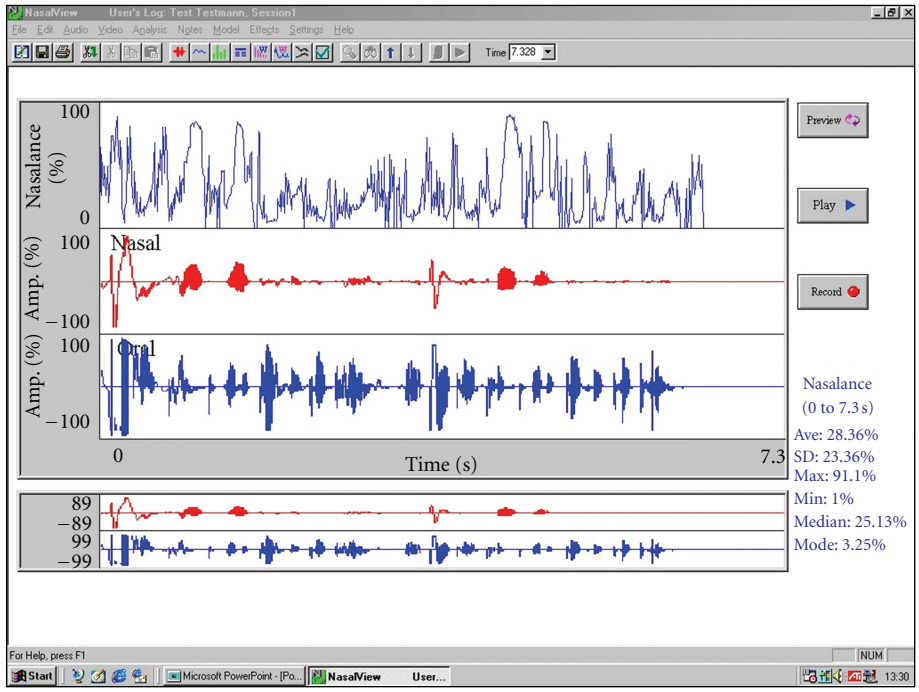

Figure 1: Sample screenshot of the NasalView system showing oszillograms for oral, nasal, and nasalance curves recorded realtime and nasalance statistic for a nonnasal test item.

Table 1: Test-retest errors for ave and median nasalance measurement.

Measurement	Pairs of test items		Total ($n = 157$)
	Nasal sentences ($n = 95$)	Nonnasal sentences ($n = 62$)	
D-Ave [%]			
Mean	1.50	1.37	1.44
SD	0.99	1.15	1.05
D-Median [%]			
Mean	2.84	1.66	2.37
SD	2.31	1.35	2.06

D-ave: difference of ave nasalance values between two test items; D-median: difference of median nasalance values between two test items mean: arithmetic mean; SD: standard deviation.

Median-nasalance values ($P < 0.001$), only Ave values are reported as nasalance value in the following results.

3.2. Group Differences. According to the results of the perceptual analysis, 23 patients were classified normal, 35 patients were borderline cases, 27 patients showed marked hypernasality, and 10 patients had severe hypernasal speech.

Table 2 provides the results of the objective computerized measurements with the NasalView system. With the exception of the nasalance of the nnS-test item (nonnasal sentence) the differences between the single groups were significant at the $P < 0.05$ level. For the nonnasal sentence group, differences were significant with $P < 0.01$.

3.3. Validation Data. The optimum cutoff, sensitivity, specificity, and efficiency were calculated for the various measurements regarding the following discriminations:

(a) differentiation between all patients with regular resonance and all hypernasal patients (Table 3);

(b) differentiation between all patients with regular resonance and hypernasal patients with marked and severe hypernasality excluding borderline cases (Table 4);

(c) differentiation between patients with marked and severe hypernasality (Table 5);

(d) differentiation between patients with no or borderline rhinophonia and hypernasal patients with marked and severe hypernasality (Table 6). This discrimination is based upon the therapeutic consequence: in cases of marked and severe hypernasality, treatment of the speech disorder—speech therapy or surgical intervention is necessary, whereas in borderline cases the further clinical development is scrutinized.

4. Discussion

The application of the NasalView system was easy due to the fact that the measuring of nasalance is entirely noninvasive. In addition to the practicability, this investigation also demonstrates the good reliability of computerized assessment of hypernasality with the NasalView system. In accordance with Awan [8], who accounted for a test-retest

Table 2: Results grouped in different degrees of hypernasality.

Measurement	degree of hypernasality				P
	Normal ($n = 23$)	Borderline ($n = 35$)	Marked ($n = 27$)	Severe ($n = 10$)	
Nasalance of nS [%]					
Mean	44.08	45.06	47.65	55.30	0.048*
SD	4.63	6.12	5.66	5.42	
Nasalance of nnS [%]					
Mean	26.30	29.80	35.36	45.21	0.007**
SD	2.93	3.49	5.21	4.25	
Nasalance ratio					
Mean	0.602	0.671	0.745	0.819	0.036*
SD	0.081	0.139	0.142	0.085	
Nasalance distance [%]					
Mean	17.76	15.26	12.29	10.08	0.033*
SD	4.85	5.94	5.04	3.43	

nS: nasal sentence; nnS: nonnasal sentence; mean: arithmetic mean; SD: standard deviation P: P value of ANOVA (differences between degrees of hypernasality)*: $P < 0.05$ **: $P < 0.01$.

Table 3: Validation data for differentiation between patients with regular resonance and hypernasal patients.

Measurement	Cutoff	Sensitivity [%]	Specificity [%]	Efficiency [%]
Nasalance of nnS	28.5%	83.3	87.0	84.2
Nasalance ratio	0.66	70.8	69.6	70.5
Nasalance distance	15.3%	68.1	73.9	69.5

nnS: nonnasal sentence.

Table 4: Validation data for differentiation between patients with regular resonance and hypernasal patients excluding borderline cases.

Measurement	Cutoff	Sensitivity [%]	Specificity [%]	Efficiency [%]
Nasalance of nnS	28.6%	91.9	91.3	91.7
Nasalance ratio	0.72	78.4	95.7	85.0
Nasalance distance	15.0%	81.1	73.9	78.3

nnS: nonnasal sentence.

Table 5: Validation data for differentiation between patients with marked and severe hypernasality.

Measurement	Cutoff	Sensitivity [%]	Specificity [%]	Efficiency [%]
Nasalance of nnS	40.0%	90.0	88.9	89.2
Nasalance ratio	0.818	70.0	77.8	75.7
Nasalance distance	10.5%	70.0	59.3	62.2

nnS: nonnasal sentence.

error of 2% for the NasalView system and a test-retest error of 3% for the Nasometer in a study with 20 adult subjects, we established a test-retest error under 2% when the average value of the reported nasalance statistic was used.

Table 6: Validation data for differentiation between patients with no and borderline rhinophonia and patients with marked and severe hypernasality.

Measurement	Cutoff	Sensitivity [%]	Specificity [%]	Efficiency [%]
Nasalance of nnS	33.0%	86.5	93.1	90.5
Nasalance ratio	0.72	78.4	84.5	82.1
Nasalance distance	14.02%	75.7	70.7	72.6

nnS: nonnasal sentence.

This small error implicates a good reliability of the measuring technique.

The validation data established in our investigation are comparable to the results of other studies dealing with the NasalView system or the Nasometer.

For the Nasometer, Dalston et al. [11, 12] and Hardin et al. [13] reported an efficiency ranging from 79.2% to 93% depending on whether borderline cases were included or not. In 30 German speaking cleft patients with a mean age of 13.5 years, Stellzig et al. measured in 1994 an overall diagnostic accuracy of 90% for the Nasometer [17]. Nasalance measurement with the NasalView system and the Nasometer differ technically and in appliance. Therefore, a direct comparison of the detailed results is not possible (Lewis and Watterson [27]).

As far as the NasalView system is concerned comparable investigations have been performed by Bressmann, who analysed nasalance measurements of 133 and 140 German speaking patients with cleft lip and palate from 10 to 66 years of age, mean age = 17 years (Bressmann et al. [20, 21]).

When differentiating between healthy subjects and hypernasal patients, we found the same optimum cutoff of 28.5% for the nonnasal test item. Whereas Bressmann reported an efficiency of 73%, we found a higher overall

diagnostic accuracy of 84.2% for this test item and the discrimination. For the nasalance ratio and nasalance distance in our study, we found lower values of 70.5% and 69.5% than Bressmann, who reported an accuracy of ranging from 75.2% to 77.4%. When excluding borderline cases from the analysis, sensitivity, specificity, and efficiency improve considerably. In the present study for the nonnasal test item efficiency improved to 91.7% compared to 81.4% found by Bressmann et al. [20]. Although efficiency for nasalance ratio and distance improved to 85% and 78.3%, the results are lower than the values measured by Bressmann et al. [21].

In our data, optimum cutoffs for the nasalance ratio were higher and cutoffs for the nasalance distance were lower than in the comparable validation study [21]. These discrepancies can be provoked by the influence of a different dialect, as described by Van Lierde et al. [28], or the use of similar but not identical sentences.

Another decision of clinical importance is the differentiation between patients with no and mild hypernasality, normal and borderline cases, and patients with marked and severe rhinophonia. Due to clinical experience, mostly a mild hypernasality does not impair intelligibility of speech and represents no stigma for the patient in his social environment. In most cases of borderline hypernasality, no treatment is mandatory. On the other hand, in patients who suffer from marked or severe hypernasality with impaired speech regularly treatment, that is, surgical intervention with velopharyngoplasty is necessary. Differentiating between patients with the need for intervention and those who do not need therapy as described above, again the best efficiency of 90.5% results from the nonnasal sentence with a cutoff of 33%. When using nasalance ratio and distance, we found a diagnostic accuracy of 82.1% and 72.6%, respectively, for the differentiation between patients with no and borderline hypernasality on the one side and patients with marked and severe hypernasality on the other side. This clear differentiation between hypernasality with the need for treatment and no or mild hypernasality without need of treatment need was not yet described in previous studies.

5. Conclusions

The results of this clinical validation identify the NasalView system as an apt and userfriendly device to identify patients with resonance disorders. A short screening procedure including a nasal and a nonnasal test item is sufficient to differentiate between the perceptual degrees of hypernasality with good reliability and validity.

To come to a sensible decision as far as the further treatment of patients with cleft lip and palate and resulting hypernasality is concerned, it is necessary to assess the velopharyngeal sphincter with regard to its structure, function, and the perceptual consequences. The validation data of our investigation among others indicates that an objective measuring of rhinophonia is helpful to supplement the perceptual assessment of hypernasality by an experienced speech therapist.

We consider the objective computerized diagnostic of nasality with the NasalView system an expedient completion and helpful screening tool for resonance disorders, but not as a replacement of perceptual judgements by experienced speech therapists and listeners.

Conflict of Interests

All authors declare that there exist no conflict of interests.

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Research Article

Speech Outcomes after Tonsillectomy in Patients with Known Velopharyngeal Insufficiency

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Introduction. Controversy exists over whether tonsillectomy will affect speech in patients with known velopharyngeal insufficiency (VPI), particularly in those with cleft palate. *Methods*. All patients seen at the OHSU Doernbecher Children's Hospital VPI clinic between 1997 and 2010 with VPI who underwent tonsillectomy were reviewed. Speech parameters were assessed before and after tonsillectomy. Wilcoxon rank-sum testing was used to evaluate for significance. *Results*. A total of 46 patients with VPI underwent tonsillectomy during this period. Twenty-three had pre- and postoperative speech evaluation sufficient for analysis. The majority (87%) had a history of cleft palate. Indications for tonsillectomy included obstructive sleep apnea in 11 (48%) and staged tonsillectomy prior to pharyngoplasty in 10 (43%). There was no significant difference between pre- and postoperative speech intelligibility or velopharyngeal competency in this population. *Conclusion*. In this study, tonsillectomy in patients with VPI did not significantly alter speech intelligibility or velopharyngeal competence.

1. Introduction

Patients with velopharyngeal insufficiency (VPI) present a unique challenge to the pediatric otolaryngologist. Otherwise straightforward problems such as sleep apnea and chronic tonsillitis become far more complicated in the cleft population due to the risk of VPI, and yet, sleep disordered breathing is increasingly being diagnosed in this group. Several recent studies demonstrated that cleft patients exhibit a significantly higher incidence of sleepdisordered breathing, with syndromic patients carrying an increased risk for obstructive sleep apnea (OSA) [1, 2]. However, OSA tends to be underdiagnosed and undertreated in this population, which may be due to a reluctance to operate in the oropharynx due to risk of velopharyngeal insufficiency.

Adenoidectomy has long been known to carry a risk of worsening velopharyngeal insufficiency in patients with known VPI and may unmask previously undiagnosed VPI, particularly in patients with submucous cleft palate [3–6]. However, controversy exists over whether tonsillectomy alone will affect speech in patients with known velopharyngeal insufficiency, particularly in those with cleft palate.

In this study we hypothesize that, in carefully chosen patients, tonsillectomy can be safely performed on patients with existing VPI without significant adverse effects to their speech.

2. Methods

All patients seen at the Doernbecher Children's Hospital Multidisciplinary Velopharyngeal Insufficiency Clinic between 1997 and 2010 were prospectively entered into a database. The database was screened for all patients undergoing tonsillectomy during this time. Inclusion criteria included a previous diagnosis of velopharyngeal insufficiency, a history of tonsillectomy performed at this institution, and adequate pre- and postoperative speech assessment. Exclusion criteria included concordant adenoidectomy, pharyngoplasty or pharyngeal flap, or inadequate speech evaluations for analysis.

Speech assessments were obtained from routine speech analysis by pediatric speech pathologists, on the scale developed at this institution prior to the acceptance of the universal speech parameters. The analysis was translated to

Table 1: Patient characteristics.

Patient	Sex	Age at tonsil surgery	Tonsillectomy indication	Tonsil size	CP type	Initial cleft repair	Speech intelligibility			VPI severity		
							Preop.	Postop.	Diff	Preop.	Postop.	Diff
1	F	4	OSA	3.5	UC	2 flap	4	2	−2	3	2	−1
2	M	4	OSA	4.0	BC	push back	1	1	0	0	2	2
3	M	5	OSA	4.0	UC	2 flap	2	1	−1	1	1	0
4	F	12	VPI	3.0	SM	Unk.	2	2	0	2	2	0
5	M	6	VPI	3.5	None	None	2	2	0	3	2	−1
6	M	8	VPI	4.0	UC	2 flap	3	2	−1	2	2	0
7	M	5	VPI	3.0	UC	2 flap	3	2	−1	1	2	1
8	M	8	OSA	3.5	UC	Unk.	0	0	0	1	1	0
9	M	6	VPI	3.0	UC	2 flap	3	4	1	4	4	0
10	M	7	VPI	2.5	SM	Furlow	3	1	−2	3	2	−1
11	F	4	OSA + VPI	4.0	None	None	1	4	3	2	3	1
12	F	5	VPI	4.0	UC	2 flap	4	4	0	4	4	0
13	M	7	OSA	4.0	UC	2 flap	3	2	−1	1	1	0
14	F	7	OSA	3.0	BC	Push back	1	1	0	2	1	−1
15	M	7	Strep+VPI	4.0	I	2 flap	2	1	−1	1	2	1
16	F	7	VPI	2.5	None	none	3	3	0	3	3	0
17	M	7	OSA	4.0	UC	2 flap	2	2	0	2	2	0
18	F	12	OSA+VPI	4.0	I	Furlow	3	2	−1	2	1	−1
19	M	4	OSA	4.0	I	Unk.	3	2	−1	0	1	1
20	F	8	Dysphagia	4.0	I	Unk.	0	0	0	0	1	1
21	F	7	OSA	4.0	UC	Unk.	1	1	0	1	1	0
22	F	8	VPI	3.5	SM	None	4	4	0	3	3	0
23	M	4	OSA	3.5	UC	2 flap	2	2	0	2	1	−1

Patient characteristics. Cleft type: UC: unilateral complete, BC: bilateral complete, SM: submucous, I: incomplete.

a nonparametric scale as indicated below in Tables 3 and 4. Those patients who underwent pre- and postoperative nasal endoscopy were graded on the Golding-Kushner scale [7] on palatal motion and lateral pharyngeal wall motion as judged by a single faculty pediatric otolaryngologist (H. A. Milczuk).

Attention was specifically turned to the assessment of two primary parameters for analysis: speech intelligibility and velopharyngeal sufficiency. Pre- and post operative evaluations were compared. For individual patients, a significant change in function was defined to be a change in 2 points on the scale. For the overall group, a two-tailed Wilcoxon rank-sum test was used to evaluate for significance.

3. Results

A total of 46 patients with known VPI underwent tonsillectomy over this time period. Of these, 23 had both pre- and postoperative speech evaluations that were sufficient for our analysis. See Table 1 for patient characteristics. The majority of these patients (87%) had a known history of cleft palate. Primary Indications for tonsillectomy included obstructive sleep apnea in 11 (48%), staged tonsillectomy prior to pharyngoplasty in 10 (43%), recurrent strep tonsillitis and VPI in 1 (4.3%), and obstructive dysphagia in 1 (4.3%). Average tonsillectomy size (as graded on a 0–4 + scale) was 3.6 with a median value of 4.0, indicating a strong preponderance of tonsillar hypertrophy in this group.

Overall there was no statistically significant difference between pre- and postoperative speech intelligibility (with trend towards improvement, $P = 0.13$) or velopharyngeal competency (no trend $P = 0.83$) in this population using the Wilcoxan rank-sum. With respect to speech intelligibility, one patient demonstrated significant worsening, and 2 patients demonstrated significant improvement as defined by a change in 2 points on the scale. With respect to velopharyngeal insufficiency, only one person had significant worsening of their insufficiency. No significant trends were noted when comparing patient outcomes with respect to cleft type, age of patient, gender, indication for surgery, or tonsil size.

Ten of the patients underwent both pre- and posttonsillectomy nasal endoscopy, and their data is presented in Table 2. Palatal closure remained the same in 5 (50%), improved in 3 (30%), and worsened in 2 (20%). Lateral wall movement remained the same in 3 (30%), improved in 5 (50%), and worsened in 2 (20%).

4. Discussion

The role of tonsils on velum position and function is poorly characterized. The velum position during speech depends on

Table 2: Nasal endoscopy scores.

Pt.	Indication for tons.	CP type	Initial cleft repair	Protons. endoscopy	Posttons. endoscopy	Speech intelligibility			VPI Severity		
						Preop.	postop.	Diff.	Preop.	Postop.	Diff.
4	VPI	SM	Unk.	Palate 9 lat 4	Palate 9 lat 4	2	2	0	2	2	0
5	VPI	None	None	Palate 7 lat 2	Palate 9 lat 4	2	2	0	3	2	−1
6	VPI	UC	2 flap	Palate 0 lat 0	Palate 0 lat 1	3	2	−1	2	2	0
7	VPI	UC	2 flap	Palate 9 lat 2	Palate 9 lat 1	3	2	−1	1	2	1
9	VPI	UC	2 flap	Palate 1 lat 1	Palate 2 lat 1	3	4	1	4	4	0
12	VPI	UC	2 flap	Palate 5 lat 1	Palate 5 lat 3	4	4	0	4	4	0
15	Recurrent strep + VPI	I	2 flap	Palate 8 lat 4	Palate 9 lat 4	2	1	−1	1	2	1
16	VPI	None	None	Palate 9 lat 4	Palate 7/8, lat 1	3	3	0	3	3	0
17	OSA	UC	2 flap	Palate 8 lat 0	Palate 8 lat 1	2	2	0	2	2	0
22	VPI	SM	furlow	Palate 4 lat 0	Palate 3.5 lat 1	4	4	0	3	3	0

Patient characteristics. Tons: tonsillectomy. Cleft type: UC: unilateral complete, BC: bilateral complete, SM: submucous, I: incomplete. Unk.: unknown. Endoscopy scores: palate: palatal movement, lat: lateral wall movement. Scores according to the Golding-Kushner Scale, adjusted to 0–10 scale.

Table 3: Ranking of speech intelligibility.

Speech intelligibility		
0	Normal	100% intelligible
1	Minimal	95–99% intelligible
2	Mild	80–94% intelligible
3	Mod	50–79% intelligible
4	Severe	<50% intelligible

Table 4: Ranking of velopharyngeal insufficiency.

Velopharyngeal insufficiency	
0	Normal
1	Minimal
2	Mild
3	Mod
4	Severe

the complex balance of vector forces created by palatal elevators, depressors, and constrictors [8]. Elevation is primarily achieved by the levator veli palatini, and transverse closure is mediated primarily by the superior constrictor. The palatopharyngeal and palatoglossal muscles, between which the palatine tonsils reside, serve to depress the palate. The position of these arches can vary based on the size and shape of the tonsils, and, theoretically, the vector forces exerted on the palate may be affected. While there are many theories and practices, the effect of tonsillectomy in patients with VPI is largely unknown, yet there remains widespread hesitancy to perform tonsillectomy in these patients.

Few studies have characterized the influence of tonsils on speech. A 1994 study by Finkelstein et al. examined tonsil size and position in relation to speech function. They concluded that in most cases markedly enlarged tonsils do not appear to affect velopharyngeal closure as they are typically positioned below the level of velum closure [9]. However, several case reports have described VPI associated with patients with superior pole hypertrophy in which the pole extends above the level of the velum. Endoscopic exam in these cases revealed prominent superior poles which appear to contribute to VPI by extending into the lateral ports, thus, displacing the palatopharyngeus muscle anteriorly [9–11]. It is argued that tonsillectomy may play a role in improving VPI in such cases.

On the opposite side of the spectrum, certain cases of VPI may be expected to worsen with tonsillectomy. In some, the tonsils are thought to act as lateral obturators, particularly in patients who have had pharyngeal flaps that are narrow or low placed. In these cases tonsillectomy would not be recommended or may necessitate simultaneous or staged flap augmentation or revision [12]. These cases should be identified with careful endoscopy. Thus, the role of preoperative endoscopy is emphasized.

In our study, patients with VPI who underwent tonsillectomy had very little overall change in speech parameters. Our findings are consistent with several studies. D'Antonio et al. in 1996 demonstrated improved or unchanged speech parameters in 15 patients at risk for VPI after tonsillectomy [13].

Similar results have been demonstrated in other groups. In a recent Taiwanese study by Hu et al. comparing management of VPI in the presence of tonsillar hypertrophy, a subset of patients who underwent an isolated tonsillectomy either alone or for staged pharyngoplasty had similar speech outcomes to our study. In their study, 19 of the patients underwent tonsillectomy without a simultaneous velopharyngeal procedure. Of these, 14 patients had no change in function, three patients improved, and two patients worsened after tonsillectomy [14].

Potential biases of this study include observer bias and selection bias. The evaluators were not blinded to the pre-versus postoperative status in these cases. The selection of patients for surgery was not randomized and was based on

clinical judgment. An argument against this bias in this study, however, is the inclusion of the subset who had severe VPI and underwent tonsillectomy as a first stage prior to definitive pharyngoplasty. In these patients it was felt that a procedure to improve speech, which invariably narrows the pharynx, would likely result in sleep-disordered breathing postoperatively. These patients arguably may have been the most "at risk" for worsening speech parameters given the preoperative decision that the patient would need VPI surgery. However, this group did not have significantly different outcomes.

It should be noted that the average tonsil size in this study was large (3−4 + in the majority of patients). Thus, the results must be interpreted with respect to this. Studies of tonsil size and speech characteristics are sparse. In one study of healthy male adults, Mora et al. demonstrated that tonsil size was directly related to the degree of audible speech changes after tonsillectomy, notably, the degree in change of improvement of hyponasality [15]. However, data on velopharyngeal competency with respect to tonsil size is sparse.

Furthermore, we must address the fact that only half of the patients in our VPI database who underwent tonsillectomy had adequate pre- and postoperative speech evaluation with standardized perceptual speech analysis. We excluded those patients with speech assessments that did not quantify the two parameters of interest, namely, speech intelligibility and velopharyngeal competency. Several patients also were lost to followup or had a delay in postoperative evaluation, such that a direct comparison of pre- and postoperative speech parameters would not be useful. Ideally, a study of this design should have standardized time intervals for speech evaluation.

5. Conclusion

In this study, tonsillectomy without adenoidectomy in patients with VPI and tonsillar hypertrophy did not significantly alter speech intelligibility or velopharyngeal competence. This must be interpreted with respect for adequate clinical judgment. More research is needed to further elucidate the impact of tonsillectomy on patients with or at risk for velopharyngeal insufficiency, particularly given the high prevalence of OSA in this population.

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