Running title: Identifying oral-nasal balance disorders: clinical approach

Clinical application of a new approach to identify oral-nasal balance disorders based on nasalance scores

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Abstract

Objective. A new approach to classify oral-nasal balance disorders based on instrumental measurements was developed based on linear discriminant analysis (LDA) of nasalance scores of simulated oral-nasal balance disorders by de Boer and Bressmann (2015). The current study aimed to apply the newly developed functions to clinical data to investigate the applicability of this new approach.

Design. Retrospective diagnostic accuracy study.

Setting. Tertiary university hospital.

Participants. Fifty-five Dutch-speaking Flemish children (age 4-12y) with normal (n=20), hypernasal (n=18), hyponasal (n=12) or mixed nasality (n=5).

Interventions. Nasalance scores of an oral and a nasal text were used to calculate three sets of LDA function scores. Predicted classification was consecutively based on the function values of the group centroids originally determined by de Boer and Bressmann (2015) and adapted LDA functions and group centroids based on clinical data.

Main Outcome Measures. Discriminatory power of the linear discriminant formulas.

Results. Based on the original LDA functions, 56% of the speech samples matched the perceptual classification. Applying a correction factor for age and language differences resulted in a 67% correct classification, although 83% of the hyponasal samples were ranked as ‘normal resonance’. Re-derivation of the LDA functions based on current clinical data resulted in an 80% correct classification.

Conclusions. The new approach of classifying oral-nasal balance disorders based on a combination of nasalance scores was promising. However, further clinical research is needed to refine the LDA functions and group centroids before clinical application is possible.

Key words: *nasalance, oral-nasal balance, nasometry, hypernasality, hyponasality, mixed nasality*

Introduction

To determine the presence and severity of an oral-nasal balance disorder, most clinicians rely on a combination of perceptual and instrumental assessment techniques. Perceptual judgments by a trained speech-language pathologist are considered the gold standard because no instrumental assessment can yet surpass the capabilities of a trained ear (Kuehn & Moller, 2000). Nevertheless, perceptual judgments can be subject to several influences which may limit their reliability and validity in clinical practice. For example, a patient's physical appearance or knowledge of the patient's medical history can influence perceptual judgments. Additionally, the listener's familiarity with a specific speaker can influence the perception of the severity of a speech problem because listeners may adapt to atypical patterns of speech production. This can introduce bias during speech assessments (Kent, 1996). Other confounding variables may be related to the rating task itself, such as clarity, or lack thereof, of definitions of the perceptual symptoms to be assessed and the corresponding rating scale, as well as the listener's familiarity with the rating scale (Kreiman, Gerratt, Kempster, Erman, & Berke, 1993).

One of the most widely used instruments to complement and corroborate the perceptual assessments of oral-nasal balance is the Nasometer^{TM}. This device, originally developed by Fletcher and Bishop (1973), measures the amount of nasal contribution to the speech signal. Oral and nasal acoustic signals are captured with an oral and a nasal microphone attached to a sound separation plate which is placed under the nose of the patient. After bandpass filtering, the nasal sound pressure is divided by the sum of the nasal and oral sound pressure. The resulting quotient is multiplied by 100 to obtain the nasalance score as a percentage. This nasalance score represents an indirect measure of nasality. To detect hypernasality, mean nasalance scores of an oral text passage, devoid of nasal consonants, are usually compared with normative values which are available for different languages (see D'haeseleer, Bettens, De Mets, De Moor, and Van Lierde (2015) for a recent overview). Hypernasal speakers will demonstrate higher nasalance scores than established norms for oral text passages. A nasal text passage, including a high number of nasal consonants, can be applied to detect hyponasality. Hyponasal speakers will demonstrate lower nasalance scores than established norms for speech stimuli loaded with nasal sounds. However, mean nasalance scores cannot always discriminate between the presence and absence of hypernasality (Van Lierde, Wuyts, Bonte, & Van Cauwenberge, 2007; Watterson, McFarlane, & Wright, 1993), especially when patients with borderline or mild hypernasality are assessed (Bressmann, Klaiman, & Fischbach, 2006; Dalston, Neiman, & Gonzalezlanda, 1993). Possible influence of gender, age, language and dialect have to be taken into account when interpreting or comparing nasometric scores (see Bettens, Wuyts, De Graef, Verhegge, and Van Lierde (2013) and D'haeseleer et al. (2015) for an overview). Finally, variable results have been reported for the correlation between the perceived degree of nasality and nasalance scores, resulting in correlation coefficients between 0.29 and 0.78 (Brancamp, Lewis, & Watterson, 2010; Brunnegard, Lohmander, & van Doorn, 2012; Dalston et al., 1993; Keuning, Wieneke, & Dejonckere, 2004; Lewis, Watterson, & Houghton, 2003; Sweeney & Sell, 2008; Watterson et al., 1993). The variability of the reported correlations may result in part from methodological differences between studies. For example, the use of different or similar speech stimuli for perceptual assessments and Nasometer™ recordings has been reported as a factor that can influence the correlation between perceptual judgments and nasalance scores (Sweeney & Sell, 2008). Furthermore, experience may influence listener agreement (Brunnegard et al., 2012; Lewis et al., 2003). Brunnegard et al. (2012) reported variable listener agreement depending on speech stimulus and experience of the listeners. The highest levels of agreement, measured with correlations, were reported for the perceptual judgment of hypernasality based on spontaneous speech by speech-language pathologists and nasalance scores based on an oral stimulus. Another source of variability between studies is the inclusion of different patient populations. For example, the inclusion of patients who underwent pharyngeal flap surgery may decrease the correlation between nasalance scores and the perceptual ratings of nasality (Hardin, Demark, Morris, & Payne, 1992; Nellis, Neiman, & Lehman, 1992).

In order to overcome the frequent discrepancies between acoustic and perceptual assessments of hypernasality, several authors developed diagnostic algorithms based on combinations of different instrumental measurements. For example, the Nasality Severity Index (NSI) was first developed by Van Lierde et al. (2007) and revised as the NSI 2.0 by Bettens, Van Lierde, Corthals, Luyten, and Wuyts (2015). In its current version, the NSI 2.0 is based on the combination of the nasometric scores of the vowel /u/ and an oral text and an acoustic analysis of the vowel /i/. Bressmann et al. (2000) proposed two measures based on the combination of the nasalance scores of an oral and a nasal stimulus, namely the nasalance distance and nasalance ratio (Bressmann et al., 2000). However, these new diagnostic algorithms were only used to assess hypernasality. Hypernasality affects speech intelligibility and acceptability more than hyponasality and therefore is clinically more relevant (Shprintzen et al., 1979). However, some authors have highlighted the need to also measure hyponasality and to approach the diagnosis of oral-nasal balance disorders as a simultaneous assessment of hyper- and hyponasality (Bressmann et al., 2000; de Boer & Bressmann, 2015). This results from the notion that oral-nasal balance disorders are often complex due to the frequent co-occurrence of hypernasality and a reduced nasal patency in patients with cleft lip and palate, resulting in mixed nasality. This nasal obstruction can be due to septum deviation, narrow vestibule, maxillary retrusion or as a result of secondary velopharyngeal surgery such as a pharyngeal flap (Fukushiro & Trindade, 2011; Kummer, 2011; Nellis et al., 1992; Shprintzen et al., 1979). Following this reasoning, de Boer and Bressmann (2015) also used nasalance scores from an oral and a nasal stimulus but attempted to discriminate between normal resonance, hypernasality, hyponasality and mixed nasality on the basis of the two nasalance scores. Their aim was to develop a tentative statistical procedure for nasometric data with a simultaneous reflection of hyper- and hyponasality to arrive at a comprehensive quantitative classification for the speaker's oral-nasal balance, which could then supplement the clinician's perceptual assessment. In order to provide a reliable ground truth, de Boer and Bressmann's (2015) experiment was based on simulations. Eleven normal female speakers of Canadian English who were students of speech-language pathology were recorded with their normal speaking voice and simulating hypernasality, hyponasality and mixed nasality based on the authors' instructions. A linear discriminant analysis (LDA) with the nasalance values of the oral and nasal stimulus as predictors was applied to discriminate the four simulated conditions. This resulted in the derivation of two discriminant functions. To predict which oral-nasal balance condition a specific speech sample belonged to, the minimal Euclidean distance was determined between the pair of function values of that specific speech sample and that of the oral-nasal balance group centroids. This resulted in the correct classification of 88.6% of the simulations.

Although the statistical analyses of the simulations were successful, clinical data of patients with real oral-nasal balance disorders are necessary to confirm the discriminatory power of the proposed algorithms. The simulations of hypernasality tended to reflect only severe hypernasality which facilitated a clear discrimination. Therefore, the purpose of the present study was to clinically test the discriminatory power of the linear discriminant formulas developed by de Boer and Bressmann (2015) by using nasalance scores obtained from children with a perceived range of hypernasality, hyponasality or mixed nasality as well as children without oral-nasal balance disorders. The research was explorative and not driven by specific hypotheses. Two different modifications of the linear discriminant analysis that tailored the discriminant formulas more specifically to the current study group were also tested. It was expected that all different variations of the classification algorithms would discriminate the four oral-nasal balance groups with a higher probability than chance alone (25%).

Method

This study was conducted according to the World Medical Association Declaration of Helsinki and approved by the institutional review and ethical board of the Ghent University Hospital (B670201213214). Written informed consent was obtained from the parents.

Participants

Data were collected retrospectively from a database of 73 children with oral-nasal balance disorders who attended the Ghent University Hospital Craniofacial Centre between 2013 and 2016, as well as a second database of normal speakers. Only children were selected because the number of available speech samples from adults was insufficient. All participants' speech had been analyzed with a Nasometer II model 6450™ (Kay Pentax, Lincoln Park NJ). For the current study purpose, only the nasalance scores of the oral and nasal text passages for Dutch were analyzed. The oral text passage contains no nasal sounds and is used to detect hypernasality, comparable to the Zoo passage in English. The nasal text passage is heavily loaded with nasal phonemes and is used to detect hyponasality, comparable to the nasal sentences in English (Van de Weijer & Slis, 1991).

The participants were selected based on the following criteria as reported in the database: age between 4 and 12 years old, native language Dutch, perceived hypernasality, hyponasality or mixed nasality due to hypernasality in combination with nasal obstruction, and no additional cognitive or neuro-motor disorders. Complete nasalance records had to be available for participants to be included. Additionally, recorded speech samples had to be available to enable the verification of previous clinical perceptual judgments mentioned in the database.

In total, the data of 19 children with perceived hypernasality (7 boys, 12 girls, mean age 7.8y, SD 2.71) and 7 children with perceived mixed nasality (3 boy, 2 girls, mean age 7.3y, SD 3.15) fulfilled the criteria. No patients with only hyponasality were identified from the clinical database. Nine patients were born with cleft palate only, eight were born with unilateral cleft lip and palate, six with bilateral cleft lip and palate and three patients underwent adenotomy resulting in hypernasality. In children born with cleft palate, the palate was closed at a mean age of 11 months (SD 2.06, range 8-15 months).

A second database including speech data of children without craniofacial disorders was used to obtain data of normal resonance as well as hyponasality. The database contained randomly selected children between 4 and 12 years old from several (pre-school) kindergartens and elementary schools. Data for the present study were selected based on the following criteria: age between 4 and 12 years old, native language Dutch, perceived normal resonance or suspected hyponasality due to a cold, no hearing problems, neurological or velopharyngeal problems, developmental delay, general disability, orthodontic treatment or oral surgical interventions reported. Additionally, complete nasalance records had to be available. Seventeen children with suspected hyponasality (9 boys, 8 girls, mean age 7.7y, SD 2.50) fulfilled the criteria. Data of 17 children without oral-nasal balance disorders (9 boys, 8 girls, mean age 8.6y, SD 2.37) were randomly selected from a pool of 114 children with normal resonance.

Perceptual assessments

To verify the previous clinical diagnoses of the oral-nasal balance conditions documented in the databases, spontaneous speech samples of all children were judged by two speech-language pathologists with experience in oral-nasal balance disorders. One of them (rater 1) was blind to the study goals. A comprehensive re-evaluation of the samples was conducted because no sufficiently detailed information was available about the clinical ratings (e.g., experience of the listener, listener's familiarity with a specific speaker, content of the speech samples used for the ratings). Due to the retrospective character of this study, only samples of spontaneous speech were available for all participants. Spontaneous speech was elicited by asking questions about school, leisure activities and their favorite TV show. This resulted in speech samples of approximately 3 minutes of spontaneous speech. The samples had been video-recorded using a Sony HDR-CX280 camera in a quiet room at the clinical department of the Ghent University Hospital or in a quiet classroom at the child's school. To limit possible listener bias based on children's physical appearance, all samples were converted to audio samples (.wav files) using audio converter software (Freemake audio converter, version 1.1.0.66) at a sampling frequency of 48kHz. The samples were blinded and randomized for presentation. To rate the oral-nasal balance and audible nasal airflow, the audio samples were presented through a pair of over-ear headphones (Sennheiser EH150). The rating scales and definitions of the Cleft Audit Protocol for Speech – Augmented (John, Sell, Sweeney, Harding-Bell, & Williams, 2006) formed the basis for the perceptual judgments. The degree of hypernasality was judged on a 5-point scale (0 = absent, 1= borderline, 2 = mild, 3 = moderate, 4 = severe) and hyponasality was quantified using a 3-point scale (0 = absent, 1 = mild, 2 = marked). A child was classified as having a mixed oral-nasal balance when the degree of both, hyper- and hyponasality was scored above 0. Due to a possible influence of audible nasal airflow and/or articulatory distortions on the ratings of oral-nasal balance, the frequency of audible nasal airflow (i.e. audible nasal emission and turbulence) was rated on a 3-point scale (0 = absent, 1 = occasionally heard, 2 = frequently heard). Furthermore, cleft type speech characteristics (CTCs) (i.e. anterior oral CTCs, posterior oral CTCs, non-oral CTCs and passive CTCs) were rated based on the randomized video samples according to the definitions of the Cleft Audit Protocol for Speech-Augmented (John et al., 2006; Sell, 2005). To assess intra-rater reliability, 10% of the samples were repeated. Each sample could be listened to as often as needed. However, once the listener moved on to the next sample, it was not possible to return to a previous rating task.

Statistical analysis

IBM SPSS Statistics software version 25.0 (IBM Corp., Armonk, NY) was used for the statistical analysis of the data. Inter- and intra-rater reliability were determined using quadratic weighted kappa (Fleiss & Cohen, 1973) due to the ordinal character of the variables.

Differences between the nasalance scores of the four oral-nasal balance groups were analyzed using Kruskal-Wallis tests with post-hoc pairwise comparisons based on Mann-Whitney U tests because the assumption of homogeneity of variances was not fulfilled. A probability level of $p<0.05$ was considered significant. Effect sizes were reported using Pearson *r* coefficients (Field, 2012).

The nasalance scores of the four oral-nasal balance groups were analyzed with three sets of LDA functions. For the *first analysis*, the two original LDA functions developed by de Boer and Bressmann (2015) were applied. The formulas were D1 = [0.099*nasalance oral stimulus (%) – 0.050*nasalance nasal stimulus (%) + 0.068] and D2 = [-0.016*nasalance oral stimulus (%) + 0.094*nasalance nasal stimulus (%) – 4.594]. For the *second analysis*, the original algorithm was linearly transformed to better match the nasalance scores from the Dutch-speaking Flemish children. The original LDA algorithms were based on data from female speakers of Canadian English. As mentioned above, possible influence of gender, age, language and dialect have to be taken into account when interpreting or comparing nasometric values. Comparing the mean nasalance values reported by de Boer and Bressmann (2015) in the normal speaking condition (i.e., mean oral text (SD): 10.2% (3.2); mean nasal text (SD):

62.0% (4.2)) to the normative values of Dutch-speaking Flemish children (i.e., mean oral text (SD): 10% (3.2); mean nasal text (SD): 55% (5.1%); Bettens, Wuyts, Jonckheere, Platbrood, and Van Lierde (2017)) resulted in a difference of 7% for the nasal text passage. This may be explained by the difference in language, because higher normative scores for the nasal text passage are reported for English-speaking Canadian children (girls mean (SD): 59.5% (5.7), boys: 62.0%(5.2); Rochet, Rochet, Sovis, and Mielke (1998)). Additionally, some authors have reported significantly lower nasalance values in children compared to adults, especially in stimuli including nasal consonants (Hirschberg et al., 2006; Prathanee, Thanaviratananich, Pongjunyakul, & Rengpatanakij, 2003; Rochet et al., 1998; Van Lierde, Wuyts, Bodt, & Van Cauwenberge, 2003). To adjust the formula for this difference, a correction of 7% was applied to the nasal nasalance value in both formulas. This resulted in two adapted LDA formulas: D1 = [0.099*nasalance oral stimulus (%) – 0.050*(nasalance nasal stimulus (%) + 7) + 0.068] and D2 = [-0.016*nasalance oral stimulus (%) + 0.094*(nasalance nasal stimulus (%) + 7) – 4.594]. For the *third analysis*, the LDA functions were re-calculated based on the nasalance scores from the Dutch-speaking Flemish children to account for the influence of age and language using the procedures described by de Boer and Bressmann (2015).

The minimal Euclidean distances between each sample's pair of function values and those of the group centroids for each of the LDAs were calculated. Based on these distances, the efficacy of all three LDA functions to classify speech samples according to their oral-nasal balance group was calculated and compared to the 25% chance level. Finally, multinomial logistic regression analyses were performed to specify the amount of variance in the calculated LDA function scores that could be explained by the difference among the four oral-nasal balance groups.

Results

Perceptual assessments

Reliability. Based on the guidelines by Altman (1991), good to very good *intra-rater reliability* was found in both raters for the perceptual judgments of hypernasality (rater 1: κ=0.94; rater 2: κ=1.00), hyponasality (rater 1: κ=0.70; rater 2: κ=1.00), audible nasal airflow (rater 1: κ=1.00; rater 2: κ=0.91) and all cleft type speech characteristics (rater 1: κ=1.00; rater 2: κ=1.00). Furthermore, good to very good *inter-rater reliability* was found for the judgments of hypernasality (κ=0.87), audible nasal airflow (κ=0.92) and the non-oral (κ=0.73) and passive CTCs (κ=0.84). A moderate reliability was found for hyponasality (κ=0.59), and the anterior (κ=0.53) and posterior CTCs (κ=0.41).

Subsequent analysis regarding the classification of samples into the four oral-nasal balance categories based on perceptual judgments revealed that 77% (46/60) of the samples were classified in the same category by both raters (i.e., whether the sample belonged to the hypernasal, hyponasal, mixed or normal group). For the other 23% (14/60) of the samples, the raters only disagreed about the presence or absence of hyponasality. Consequently, these 14 samples were judged in consensus (i.e., both raters listened together to each sample and discussed the presence or absence of hyponasality until a consensus could be reached). As a result of this consensus analysis, three of these samples were classified with hyponasality and 11 without hyponasality.

Perceptual judgments. Two patients with the label 'hypernasality' and one patient with the label 'mixed nasality' in the clinical database were judged as having normal oral-nasal balance, although audible nasal airflow was judged to be present by the two raters. Additionally, two children from the group with normal resonance from the normative database were judged as having atypical resonance (one hypernasal, one cul-de-sac resonance). These five samples were excluded from the study in order to create clear groups (e.g., no inclusion of samples with audible nasal airflow in the group of normal oral-nasal balance). Furthermore, 5/17 children with suspected hyponasality were judged as having normal oral-nasal balance. They were therefore included in the group with 'normal oral-nasal balance'. One patient with the initial label 'mixed nasality' was judged as having only hypernasality, resulting in the placement of this patient into the hypernasal group. As a result, 55 children were included in the statistical analysis: 18 children with perceived hypernasality (mean age 7.6y, SD 2.68, 8 boys, 10 girls), 5 children with perceived mixed nasality (mean age 6.6y, SD 2.88, 3 boys, 2 girls), 12 children with perceived hyponasality (mean age 7.0y, SD 2.41, 8 boys, 4 girls) and 20 children with normal oral-nasal balance (mean age 8.6y, SD 2.12, 8 boys, 12 girls). A Kruskal-Wallis test revealed no statistically significant difference in age between the four groups ($H(3)=6.256$; $p=0.100$). Regarding the cleft type speech characteristics (CTCs), 52% (12/23) of the patients had anterior oral CTCs, including dentalization, interdentalization, lateralization and/or palatalization. Fifty-nine percent (19/32) of the control children, however, also presented with dentalization and/or interdentalization. Furthermore, 30% (7/23) of the patients showed posterior oral CTCs, including backing of alveolar consonants to a velar or uvular articulation place, 35% (8/23) had non-oral CTCs, including pharyngeal and/or glottal articulation, and 57% (13/23) had passive CTCs, including weak and/or nasalized consonants and nasal realization of plosives and/or suspected passive nasal fricative. The results of the perceptual judgments with representation of the different oral-nasal balance categories are displayed in Table 1.

Table 1. Results of the perceptual judgments.

	Oral-nasal balance condition			
	Normal	**Hypernasal**	**Hyponasal**	**Mixed**
Degree of hypernasality				
- within normal limits	20/20	0	12/12	0
- borderline	0	1/18	0	3/5
- mild	0	7/18	0	1/5
- moderate	0	6/18	0	1/5
- severe	0	4/18	0	0
	Oral-nasal balance condition			
	Normal	**Hypernasal**	**Hyponasal**	**Mixed**
Degree of hyponasality				
- within normal limits	20/20	18/18	0	0
- mild	0	0	9/12	1/5
- marked	0	0	3/12	4/5
	Oral-nasal balance condition			
	Normal	**Hypernasal**	**Hyponasal**	**Mixed**
Degree of audible nasal airflow				
- absent	20/20	2/18	12/12	1/5
- occasionally heard	0	6/18	0	2/5
- frequently heard	0	10/18	0	2/5
	Oral-nasal balance condition			
	Normal	**Hypernasal**	**Hyponasal**	**Mixed**
Cleft type characteristics (CTCs)				
- absent	6/20	2/18	7/12	0/5
- anterior oral CTCs	14/20	11/18	5/12	1/5
- posterior oral CTCs	0	4/18	0	3/5
- non-oral CTCs	0	6/18	0	2/5
- passive CTCs	0	12/18	0	1/5

Instrumental assessments

Kruskal-Wallis tests. Descriptive statistics of the nasalance values are provided in Table 2. Kruskal-Wallis tests revealed overall statistically significant differences between the four oral-nasal balance groups for the nasalance scores of the oral and nasal text passages ($p<0.001$). Post-hoc pairwise comparisons with adjusted significance levels based on a Bonferroni correction showed a statistically significant difference for the nasalance scores of the oral text passage between the normal and hypernasal group ($p<0.001$), between the hypernasal and hyponasal group ($p<0.001$) and between the hyponasal and mixed group ($p=0.016$). Regarding the nasalance values of the nasal text passage, statistically significant differences were found between the normal and hyponasal ($p=0.036$), the hypernasal and hyponasal group ($p<0.001$), and the hypernasal and mixed group ($p=0.018$). Test statistics, p-values and effect sizes of all pairwise comparisons are provided in Table 3.

Table 2. Descriptive values and overall results of the Kruskal-Wallis tests for the instrumental measurements of the four oral-nasal balance groups.

Oral-nasal balance group	N	Nasalance oral text (%)		Nasalance nasal text (%)	
		Median	Q1-Q3	Median	Q1-Q3
Normal	20	13	8;14	54	51;58
Hypernasal	18	39	31;49	59	54;64
Hyponasal	12	9	8;11	47	44;48
Mixed	5	23	17;45	43	31;58
H statistic		36.213		21.421	
p value		<0.001*		<0.001*	

*statistically significant difference, $p<0.05$

Table 3. Results of the post-hoc pairwise comparisons between the four oral-nasal balance categories for the variables 'nasalance score of an oral text (%)' and 'nasalance score of a nasal text (%)'.

Nasalance oral text			
Category	**Hypernasality**	**Hyponasality**	**Mixed**
Normal	Z = 4.574 p < 0.001* r = 0.743	Z = 1.359 p = 1.000 r = 0.208	Z = 2.208 p = 0.164 r = 0.446
Hypernasality		Z = 5.319 p < 0.001* r = 0.938	Z = 0.756 p = 1.000 r = 0.196
Hyponasality			Z = 3.006 p = 0.016* r = 0.730
Nasalance nasal text			
Category	**Hypernasality**	**Hyponasality**	**Mixed**
Normal	Z = 1.740 p = 0.404 r = 0.421	Z = 2.744 p = 0.036* r = 0.485	Z = 1.876 p = 0.475 r = 0.351
Hypernasality		Z = 4.206 p < 0.001* r = 0.768	Z = 2.974 p = 0.018* r = 0.620
Hyponasality			Z = 0.120 p = 1.000 r = 0.017

***Post-hoc pairwise comparisons with adjusted significance levels (Bonferroni correction), p≤0.05; Z = standardized test statistic, p = significance value, r = effect size*

First classification based on original LDA functions and oral-nasal balance group centroids determined by de Boer & Bressman (2015). Based on the minimal Euclidean distances between each sample's pair of function values and those of the group centroids determined by de Boer and Bressmann (2015) (Table 4), 56% of the speech samples were classified correctly. A graphical representation of this result is provided in Figure 1. Seven of the 20 speech samples with perceived normal oral-nasal balance were classified as hyponasal. Of 18 samples judged to be hypernasal, two were classified as normal, three as hyponasal and six as mixed. Additionally, four of the 12 perceived hyponasal samples were classified as normal. Of the five mixed samples, one was classified as hyponasal and one as hypernasal.

Table 4. Function values of the group centroids for four oral-nasal balance conditions as reported by de Boer & Bressman, 2015 (table 5, p 179) and based on the data of Dutch-speaking Flemish children.

Oral-nasal balance condition	*Function 1 - original*	*Function 2 - original*	*Function 1 - Dutch*	*Function 2 - Dutch*
Normal	-2.020	1.041	-1.356	0.431
Hyponasal	-1.305	-1.189	-1.387	-0.609
Hypernasal	1.802	1.162	2.201	0.358
Mixed	1.522	-1.014	1.478	-1.551

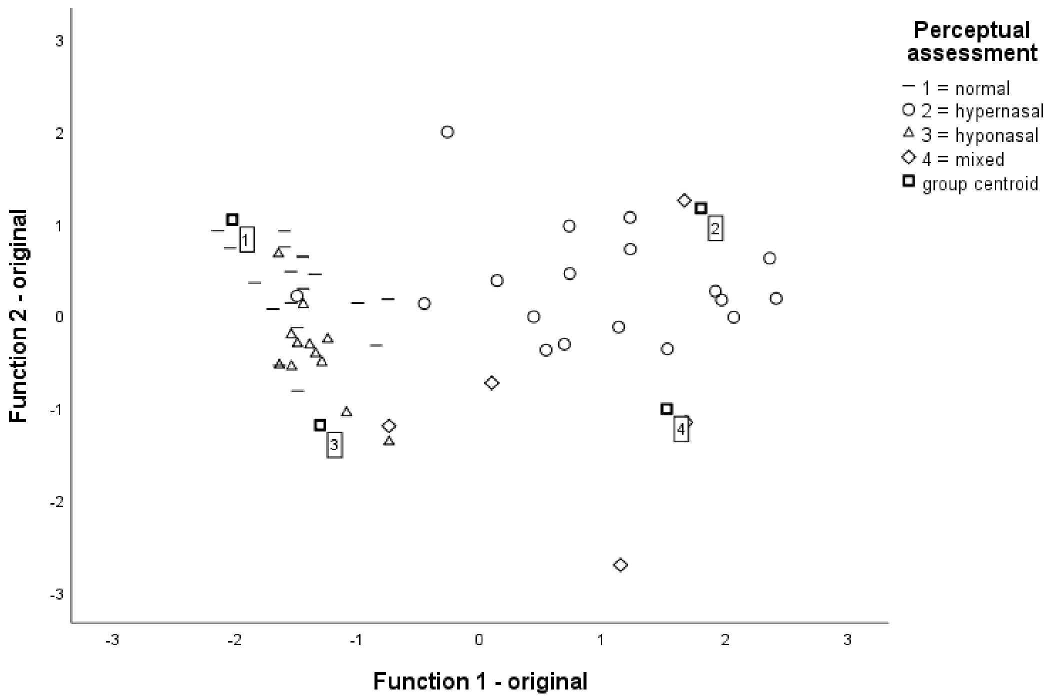

Figure 1. Scatterplot of the original linear discriminant analysis (LDA) function values with group centroids for four oral-nasal balance conditions as reported by de Boer & Bressmann, 2015 (table 5, p 179).

Second classification based on adapted LDA functions and oral-nasal balance group centroids determined by de Boer & Bressman (2015). After adapting both LDA functions with a correction factor of 7% for the nasalance value of the nasal text, the classification analysis was run a second time. As a result of this adaptation, the amount of correctly classified samples increased to 67%. A graphical representation of this result is provided in Figure 2. One of 20 speech samples with perceived normal oral-nasal balance was classified as hyponasal and four of 18 speech samples with perceived hypernasality were classified as normal. However, ten of the 12 hyponasal samples were classified as normal. Of the five mixed samples, two were classified as hyponasal and one as hypernasal.

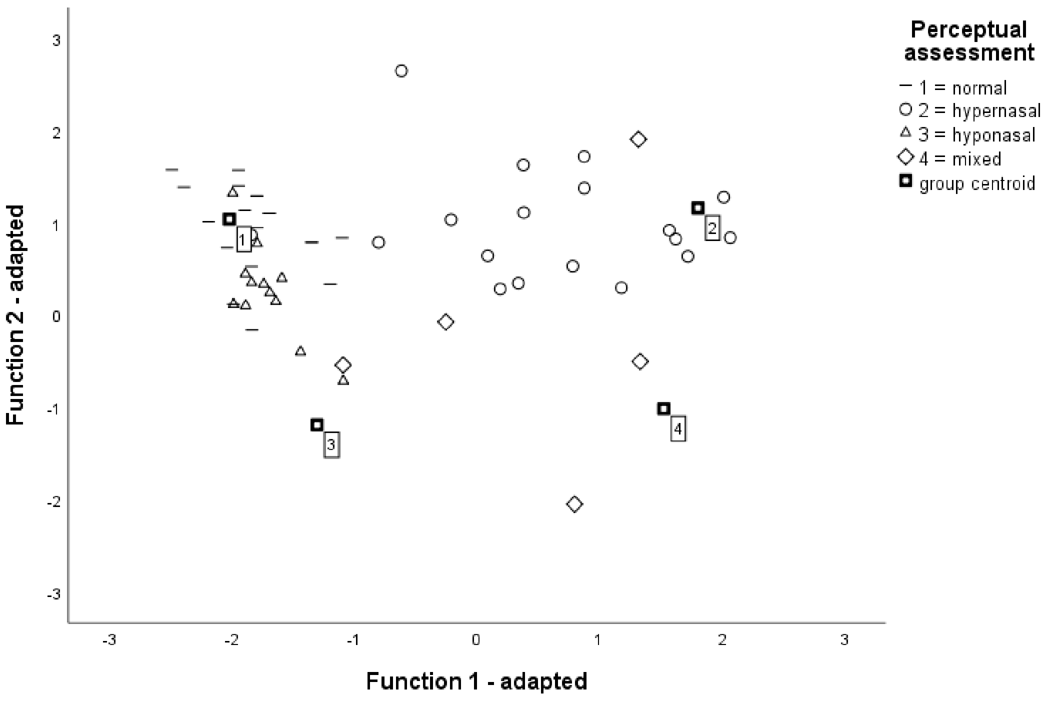

Figure 2. Scatterplot of linear discriminant analysis (LDA) function values adapted with a correction factor of 7% and group centroids for four oral-nasal balance conditions as reported by de Boer & Bressmann, 2015 (table 5, p 179).

Third classification based on newly calculated LDA functions and oral-nasal balance group centroids specific to the Dutch-speaking study group. Descriptive and linear discriminant analysis were applied on the data of the Dutch-speaking Flemish children with the nasalance values of an oral and nasal text as predictors and the four oral-nasal balance groups as classification variables. Because equal variances could not be assumed (Box's M $p<0.001$), a separate groups covariance matrix was used. Significant values were found for the Wilks' Lambda of both functions (combined $\Lambda = 0.181$, $\chi^2[6] = 87.120$, $p<0.001$; residual $\Lambda = 0.694$, $\chi^2[2] = 18.639$, $p<0.001$), indicating that the nasalance values differentiated significantly among the oral-nasal balance groups. The differences among the four groups accounted for 86.0% of the variances in values of the two discriminant functions and 55.3% by the second discriminant function. Based on the obtained canonical discriminant function coefficients, the two new Dutch adapted discrimination function formulas are:

$$D1_{Dutch} = (0.135 * \text{nasalance oral text}) - (0.047 * \text{nasalance nasal text}) - 0.411$$

$$D2_{Dutch} = -(0.032 * \text{nasalance oral text}) + (0.149 * \text{nasalance nasal text}) - 7.212$$

For each oral-nasal balance group, centroids were determined (Table 4). Based on these newly developed group centroids, the classification of speech samples was repeated a third time. Eighty percent of the samples were correctly classified. Five of 20 samples with perceived normal oral-nasal balance were classified as hyponasal. Of 18 samples with perceived hypernasality, one was classified as normal, another as mixed. Two of 12 hyponasal samples were classified as normal. For the five mixed samples, one was classified as hypernasal and one as hyponasal. A graphical representation of this result is provided in Figure 3.

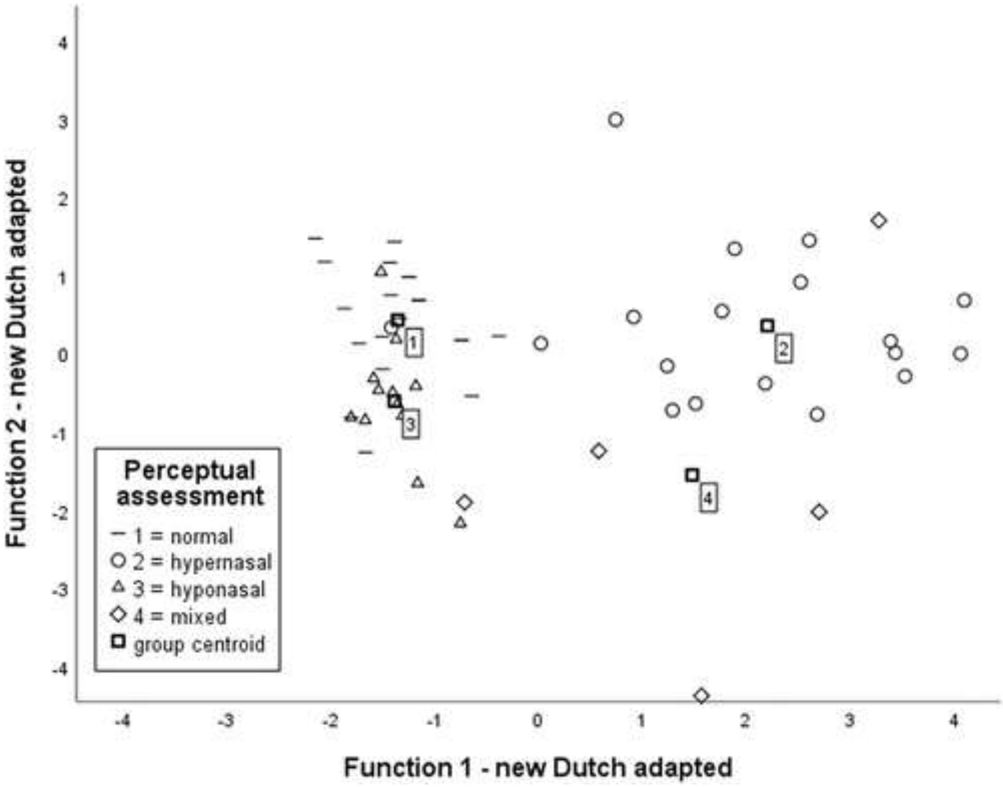

Figure 3. Scatterplot of newly calculated linear discriminant analysis (LDA) function values and group centroids for 4 oral–nasal balance conditions based on data of Dutch-speaking Flemish children.

Multinomial regression analysis. A multinomial regression analysis, with the two original LDA functions entered as covariates, revealed that both functions had a statistically significant contribution to the regression model (D1: $\chi^2(3)=58.863$, $p<0.001$; D2: $\chi^2(3)=18.945$, $p<0.001$). Furthermore, 82.5% of the variance (Nagelkerke's R^2_N) in the calculated LDA function scores could be explained by the difference among the four oral-nasal balance groups. A regression analysis with the two adapted LDA functions entered as covariates provided the same results because of the linear transformation. Entering the two new Dutch adapted LDA functions as covariates revealed comparable results. Both functions contributed to the regression model in a statistically significant way ($D1_{Dutch}$: $\chi^2(3)=59.535$, $p<0.001$; $D2_{Dutch}$: $\chi^2(3)=19.177$, $p<0.001$). Again, 82.5% of the variance (Nagelkerke's R^2_N) in the calculated Dutch adapted LDA function scores could be explained by the difference among the four oral-nasal balance groups.

Discussion

Current diagnosis of oral-nasal balance disorders is based on perceptual as well as instrumental assessments. However, instrumental results do not always reflect the perceptual judgments of a clinician. A possible explanation for this inconsistency was proposed by de Boer and Bressmann (2015) who suggested that some of the disagreement between perceptual judgments and nasalance values may be attributed to the presence of hypernasality in combination with hyponasality. Based on this reasoning, two discriminant formulas, including an oral and nasal stimulus, were developed resulting in a two-dimensional representation of oral-nasal balance disorders instead of a continuum of hypernasality alone. The purpose of the current study was to apply the LDA functions to clinical data from children with perceived normal oral-nasal balance, hypernasality, hyponasality and mixed nasality.

The blinded and randomized perceptual assessment showed that, despite the limited number of participants per group, the samples represented a suitable range of oral-nasal balance disorders. Based on the inter-rater reliability analyses, however, only a moderate agreement was found for the perceptual ratings of hyponasality. Most of the contentious samples (11/14) were finally classified as ‘perceptually normal oral-nasal balance’ after consensus rating. A possible difficulty may have been the task of using only spontaneous speech samples to judge the amount of hyponasality. In contrast, a speech sample loaded with nasal consonants may have facilitated the detection of hyponasality and might have resulted in better agreement between raters. However, a previous study found only fair to moderate inter-rater agreement for the perceptual ratings of hyponasality, despite the use of nasal speech samples (Chapman et al., 2016). Furthermore, the use of a three-point scale could also have had an impact on the reliability. The use of a binary scale could probably have induced a more conservative judgment in which samples with a minimal degree of hyponasality were rated as normal in a more consistent manner. This underlines the need for additional instrumental measurements to supplement the clinicians' diagnostic assessment of hyponasality.

Because audible nasal airflow and articulation errors are commonly associated with oral-nasal balance disorders and may influence perceptual and instrumental assessments of oral-nasal balance, these aspects were additionally rated. A high percentage of patients in the hypernasal and mixed groups presented with audible nasal airflow and articulation errors. Dattilo (2016) reported that speech-language pathology students rated the degree of hypernasality as more severe when more articulation errors were present in children with repaired cleft palate. However, to the best of our knowledge, no studies are yet available regarding the influence of articulation errors on the judgments of hypernasality by experienced raters. Furthermore, a Nasometer cannot discriminate between the acoustic energy from nasal resonance and energy from aerodynamic phenomena (such as audible nasal emission and turbulence) (Karnell, 1995; Sweeney & Sell, 2008; Watterson, Lewis, & Deutsch, 1998). As a result, nasalance scores may have been more severely increased in those patients with perceived audible nasal airflow. Moreover, the presence of a pharyngeal fricative may increase nasalance scores (Garcia et al., 2014). These influencing variables have to be taken into account when interpreting the results of this study and when applying this assessment in clinical practice. Remarkably, 59% of the children without craniofacial anomalies showed (inter)dentalization of alveolar consonants. However, previous studies also reported high prevalence of (inter)dentalization in Dutch-speaking Flemish adults (D'haeseleer et al., 2016; Van Borsel, Van Rentergem, & Verhaeghe, 2007). Another explanation for this high percentage is that many of the included children were in the mixed dental stage, including tooth eruptions of the incisors, which may had an influence on the production of the alveolar consonants.

The nasalance scores for the normal speakers were comparable to scores found in previous studies of Dutch-speaking Flemish children (mean oral text (SD): 12% (3.9) vs. 10% (3.2); mean nasal text (SD): 54% (5.2) vs. 55% (5.1%); Bettens et al. (2017)). As mentioned in the methods section, nasalance scores for the nasal stimulus were lower compared to the results reported by de Boer & Bressmann (2015) due to the possible influence of language and age.

The nasalance scores of the speakers with oral-nasal balance disorders differed numerically from the results for the simulations reported by de Boer and Bressmann (2015). The lower nasalance values of the hypernasal group in the current study (mean oral text (SD): 39% (11.5) vs. 53.2% (20.2) and mean nasal text (SD): 59% (6.8) vs. 70.5% (9.9); de Boer and Bressmann (2015)) may be explained by the inclusion of samples with more variety in the severity of hypernasality while de Boer and Bressmann (2015) reported that most of their simulations represented severe hypernasality. Additionally, higher nasalance values were found in the hyponasal group (mean oral text (SD): 9% (2.0) vs. 5.0% (1.3) and mean nasal text (SD): 46% (5.6) vs. 37.2% (12.3); de Boer and Bressmann (2015)). This may be explained by the inclusion of mainly samples with only a mild degree of hyponasality in the current study. For the mixed nasality group, lower nasalance values were observed in the current study for the oral text passage (mean oral text (SD): 29% (15.8) vs. 37.2% (14.4) whereas they were identical for the mean nasal text (SD): 44% (17.1) vs. 44.6% (17.1), de Boer and Bressmann (2015)). The lack of samples with severe hypernasality in this group may explain these differences. Since the group compositions and the premises of the two data collections were sufficiently different, no further inferential testing of these differences was undertaken.

Based on the nasalance scores of the oral text passage, significant differences and high effect sizes were found between the normal and hypernasal group, between the hypernasal and hyponasal group, and between the hyponasal and mixed group, which was expected because the oral text passage is used to determine hypernasality. Furthermore, significant differences and high effect sizes were found between the normal and hyponasal group, the hyper- and hyponasal group and the hypernasal and mixed group based on the nasalance scores of the nasal text. This was expected because the aim of the nasal text passage is to discriminate hyponasality.

The differences among the four oral-nasal balance groups accounted for 82.5% of the variance in all three sets of calculated LDA function scores. Based on the original LDA functions and group centroids, 56% of the samples were correctly classified into the expected diagnostic groups. Exploration of the data points revealed that many samples were assigned to the hyponasal group, although they were perceptually judged as normal or hypernasal. This can be explained by the difference in language and age of the speakers in the current study compared to those in the study by de Boer and Bressmann (2015). The original LDA algorithm was based on data from female speakers of Canadian English (de Boer and Bressmann (2015)). To account for the lower scores in the typical speakers of Flemish Dutch, a correction factor was applied to the nasal stimuli, resulting in a second set of LDA functions. This adaptation induced an increase of the percentage of correctly classified samples to 67%.Although this classification was still based on the group centroids derived from simulations of oral-nasal balance disorders by adult female speakers of Canadian English, the results were considerably better than chance (i.e. 25%) and confirmed that both LDA functions showed reasonable diagnostic accuracy. Most of the samples judged as normal resonance or hypernasal were correctly classified. However, only 17% (2/12) of the hyponasal samples were correctly classified, indicating that the correction factor based on the normative values may have been too coarse. Adding a 7% correction factor to the nasalance score of the nasal stimulus resulted in a shift of the data points, especially in function 2 which assigns the highest weight to the nasal stimulus. Consequently, the distance between the group centroids and data points of the hyponasal and mixed group increased, resulting in a misclassification of especially these samples. Moreover, with 83% (10/12) of the samples identified as hyponasal by both raters, this agreement suggests that there was perceptible hyponasality and that the classification by the adapted LDA functions was incorrect. The median and Q1-Q3 interval of the nasal nasalance values in this group also confirm decreased nasal resonance (Table 2). Another possible explanation for this misclassification can be that most of the samples were judged with only mild hyponasality. In the study by de Boer and Bressmann (2015), hyponasality was simulated by blocking the more patent nostril of the participant. This simulation may have resulted in a more than mild hyponasality and may not have reflected actual clinical presentation of hyponasality. The adapted LDA algorithms also only classified 40% of the mixed samples correctly. The actual clinical cases may have presented with more variety than the more homogenous simulated cases by de Boer and Bressmann (2015).

These findings led to the exploratory development of a third classification algorithm based on the clinical data of the included Dutch-speaking Flemish children, resulting in the derivation of new LDA functions and group centroids adapted for language and age. Based on these newly derived functions and centroids 80% of the samples matched the perceptual classification. More hyponasal samples were now correctly classified (10/12), although more normal samples were now classified as hyponasal (5/20). As can be seen from Figure 3, the LDA function data points of perceptually normal and hyponasal samples are close to each other which prevents a clear discrimination between both groups. As mentioned above, this can be due to the inclusion of samples with mainly mild hyponasality. Moreover, these functions were only based on small sample sizes per oral-nasal balance group and need to be validated with larger clinical data sets in future research.

As can be inferred from Figures 1 to 3 and the results of the multinomial regression analyses, the combination of both LDA functions showed good potential to arrive at a clinically reasonable classification of a speaker's oral-nasal balance. However, prospective research with more speakers will be necessary to further adapt the LDA functions and to develop new group centroids based on naturalistic clinical groups, taking into account the possible influence of gender, age and language.

Although the results were overall encouraging, the current study had a number of limitations. First, small sample sizes in the different speaker groups limit the ability to generalize the research findings to other patient populations. This was due to the retrospective character of the study resulting in a lot of incomplete data for several children in the database. Second, speech samples for perceptual assessment (i.e., spontaneous speech) and calculation of nasalance values (i.e., oral and nasal text) differed. This discrepancy may have influenced the perceptual assessment. Due to the retrospective character of this study, however, it was not possible to use the same samples for the perceptual analyses that had been used for the nasometry. Furthermore, only spontaneous speech samples were available for all included participants due to unstandardized clinical protocols. Although high correlations were reported between perceptual judgments of hypernasality based on spontaneous speech and nasalance scores of oral stimuli (Brunnegard et al., 2012), the use of only spontaneous speech may also have limitations due to the lack of control regarding the phonetic content of the sample (Sell, 2005). Only moderate inter-rater reliability was found for the variables 'hyponasality', 'anterior oral CTCs' and 'posterior oral CTCs' in this study. A more comprehensive speech sample, including spontaneous speech, automatic speech and sentence repetition may facilitate ratings and may increase reliability (Sell, 2005). These shortcomings should be taken into account in future research. Fortunately, a new Dutch assessment protocol for the perceptual evaluation of speech in patients with cleft palate was recently developed and introduced in our clinical and research practice (Bruneel et al., 2018). This will result in the availability of a more standardized and comprehensive data collection for future research. Third, the purpose of the LDA is to provide an alternative to listener ratings because of the known limitations of perceptual evaluations (Kreiman et al., 1993). Therefore, it could appear as somewhat circular reasoning to then test the algorithm against listener ratings. However, in order for the LDA-based classification to be acceptable for a speech-language pathologist undertaking a perceptual evaluation, it will be necessary to first demonstrate that the LDA can accurately reflect clinical opinion. Finally, some interspeaker variability in nasalance scores exists, even in persons without oral-nasal balance disorders (1). This may detract from the robustness of the LDA classifications. More complex diagnostic measures based on different instrumental measurements, such as the NSI 2.0, could potentially provide more stable scores. This will have to be investigated in future research.

Conclusion

This study describes a first application of recently developed LDA functions to clinical data to classify four oral-nasalance balance conditions (i.e. normal resonance, hypernasality, hyponasality and mixed nasality) in a two-dimensional diagnostic continuum based on the nasalance scores of an oral and a nasal stimulus. Although the overall amount of correctly classified samples was high (67%), most hyponasal speech samples were classified as normal. A first exploratory derivation of new Dutch LDA functions based on clinical data resulted in a better classification (80%). However, future research should focus on prospective clinical data collection to further adapt the LDA functions and to explore the possibilities to include more complex acoustic measures. Nevertheless, this new approach of classifying oral-nasal balance disorders based on a combination of nasalance scores appears promising and may in the future assist clinicians in their perceptual assessments.

Declaration of Conflicting Interests

The authors declare that there is no conflict of interest

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