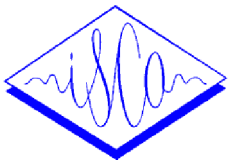

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Higher Prevalence of Voice Disorders in Patients with Cleft Lip and Palate? Results from Two Studies.

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This paper addresses two questions about voice disorders in patients with cleft lip and palate (CLP): 1. What is the actual prevalence of voice disorders in CLP and is this prevalence higher than in the non-cleft population? 2. Is there an interaction between hypernasality and phonatory dysfunction which can be assessed in terms of signal perturbation quotients? In the first study, 154 CLP patients were examined perceptually and with the MODIAS software. Severe voice disorders were found in 6.5% of the CLP group which is numerically only slightly higher than in the normal population. None of the pertubation quotients differentiated between modal and disordered voices. In the second study, 22 hypernasal and 20 non-hypernasal CLP patients were compared to 14 tumor patients using the EZVoice software. The perturbation measures differentiated dysphonic patients from patients with normal voices. An effect of hypernasality on the signal perturbation could not be verified.

Introduction

It has been discussed whether hypernasality has a detrimental effect on voice quality, especially in patients with congenital cleft lip and palate (CLP). The complex craniofacial disorder of CLP affects the oro-naso-pharyngeal system but does not extend to the larynx. Although the phonatory system is not directly affected by a CLP, a number of cleft patients present with voice disorders. According to Warren [1], the nasal loss of air pressure in patients with velopharyngeal insufficiency triggers compensatory regulatory mechanisms. In order to compensate for the decrease in intraoral air pressure, the subglottic air pressure increases. The heightened air pressure on the phonatory level also implies a modification of the laryngeal gestures in speech. This explains why subjects with cleft palate may be more prone to develop voice disorders.

McWilliams et al. [2] conclude from their review of the literature: "... phonation deficits are considerably more common in subjects with clefts than in subjects without clefts." However, the empirical evidence is not at all unequivocal. Epidemiological studies report prevalence rates of voice disorders in cleft palate as high as 10% [3], 0.6% [4], 34% [5], and 41% [6]. While Bzoch [7] reports 15% hoarseness and 31.3% breathiness in 1000 CLP patients, Bzoch et al. [8] report 0% hoarseness and 10% breathiness in a follow-up examination of 50 patients using exactly the same methodology.

It also remains unclear if the hypernasal resonance disorder *per se* has an effect on vocal quality that can be captured in terms of signal perturbation quotients. To our best knowledge, this issue has only been addressed in two studies so far. Zajac & Linville [9] report a strong correlation between jitter and hypernasality in a study

using electroglottography with ten children with hypernasality and hoarseness and five children with normal nasal resonance. Shimmer was correlated to the degree of perceived hoarseness. However, since their patients also presented with mild to moderate degrees of hoarseness it is questionable if the increased jitter quotients should really be attributed to the hypernasality or if they might have been induced by the hoarse voices. Lewis et al. [10] used a combination of aerodynamic and acoustic recording procedures when they examined voice quality in 27 CLP patients. They could not find a significant relationship between hypernasality ratings and the perturbation quotients, but found that the perturbation quotients were numerically higher. These findings will have to be interpreted with caution since the age range in this group was relatively wide (6-14 years) in a rather small sample (N=27). Factors of growth and mutation may have been a source of considerable variation.

We conducted two studies in order to answer two questions:
1. What is the prevalence of voice disorders in CLP patients?
2. Is there an interaction between hypernasality and voice quality which can be assessed by standard voice quality analysis measures such as jitter, shimmer or harmonics to noise-ratio (HNR)?

Study 1

Patients:

154 patients (102 males and 52 females) with CLP participated in this study. The median of age in these patients was 17 years with a standard deviation of 7.23 years and a range from 10 to 66 years.

Method:

Voice quality was assessed perceptually according to the RBH-system (roughness, breathiness, hoarseness [11]). Suprasegmentals and nasal resonance were assessed as well. Assessments on the four-point scales were made by the first author online during a standardized interview procedure and offline, using the DAT recordings made from the interviews.

The four sustained German vowels /a:/, /i:/, /u:/, and /y:/ were recorded with the Modular Diagnostic System (MODIAS) for MATLAB, developed by Ziegler and Merk [12, 13]. The Voice Quality module of the MODIAS program package was used to analyse these vowel samples. Measures for pitch, intensity and pertubation (jitter and shimmer) were obtained [14].

Results:

The intra-rater agreement was 91.9%, with a Kendall's tau-b of 0.866. Table 1 gives the results of the perceptual analysis for the RHB-scales and the hypernasality ratings. In order to summarize the data, we grouped patients according to their highest mark in any of the three RBH-scales. A mean value of 2.5 of the two listener ratings in any of the three scales was defined as the cutoff for a voice disorder that is severe enough to require therapy (Table 2). According to this definition, the prevalence of severe voice disorders in CLP patients was 6.5 % which is lower than in most other studies reported. The Spearman correlation coefficient between the highest RBH-rating and the degree of hypernasality was in the low range ($r=0.2537$, $p<0.01$). The incidence of voice disorders in our CLP patients was numerically only slightly higher than in the normal population when compared to the base-line data of Brindle & Morris [15] and Laguaite [16]. A more detailed account of these results has been given elsewhere [14].

The acoustic measurements from MODIAS confirmed the results of the perceptual analysis. As the prevalence of severe voice disorders was low, none of the pertubation quotients differentiated between modal and disordered voices. A weak correlation was found between jitter and the ratings for hoarseness.

Table 1: Results of the two RBH- and hypernasality-ratings

Mean of two RBH-ratings	Roughness: N	%	cum. %	Breathiness: N	%	cum %	Hoarseness: N	%	cum %
0	72	46.8	46.8	51	33.1	33.1	71	46.1	46.1
0.5	10	6.5	53.2	15	9.7	42.9	15	9.7	55.8
1	49	31.8	85.1	54	35.1	77.9	50	32.5	88.3
1.5	7	4.5	89.6	9	5.8	83.8	2	1.3	89.6
2	13	8.4	98.1	18	11.7	95.5	13	8.4	98.1
2.5	0	0	98.1	2	1.3	96.8	0	0	98.1
3	3	1.9	100	5	3.2	100	3	1.9	100

Mean of two hypernasality-ratings	Hypernasality N	%	cum. %
0	40	26.0	26.0
0.5	7	4.5	30.5
1	44	28.6	59.1
1.5	11	7.1	66.2
2	24	15.6	81.8
2.5	5	3.2	85.1
3	23	14.9	100

Table 2: Re-grouping of the patients with respect to the highest RBH-value scored individually. A value of 2.5 was defined as the cutoff for a severe voice disorder.

highest RBH-value (mean from two ratings)	N	%	cum. %
0	1	0.6	0.6
0.5	4	2.6	3.2
1	86	55.8	59.1
1.5	14	9.1	68.2
2	39	25.3	93.5
2.5	2	1.3	94.8
3	8	5.2	100.0

Conclusions from Study 1:
While it is appropriate to assume a higher potential risk for vocal dysfunction in CLP patients the actual prevalence in the patients examined is numerically only slightly higher than in normals. For patients with voice disorders, apparative diagnostics of voice quality is a useful and practical adjunct to perceptual analysis which helps profiling individual patients. Due to the relatively low prevalence of moderate and severe voice disorders in the patients examined we were not surprised that we did not obtain statistical significance in most of our acoustic data.

Study 2
Patients:
A total of 56 adult subjects were examined. 20 CLP subjects had perceptually normal nasal resonance and vocal quality, 22 CLP patients had marked hypernasality but no overt voice problem, and 14 patients with tumors of the oral cavity had marked hoarseness but no hypernasality.

Procedure:
The four sustained German vowels /a/, /i/, /u/ and /y/ were recorded for each patient and analysed with the EZVoicePlus (see Figure 1 [17]) signal analysis software. Jitter, Shimmer, Harmonics to Noise-Ratio (HNR) and Discrete Fourier Transformation Ratio (DFTR, the ratio of low frequency to high frequency energy in the voice spectrum, derived by discrete Fourier transformation) were computed

and compared to the perceptual assessment of hoarseness and hypernasality.

Results:
Table 3 shows the results from the non-parametric statistical analysis with the Kruskal-Wallis test and the subsequent Mann-Whitney U-test. The results indicate that the perturbation measures, especially jitter and HNR, are useful to differentiate dysphonic patients from patients with normal voices. An effect of hypernasality on the signal perturbation could not be verified. The spectral measure DFTR of /a/ differentiated the hypernasal patients from the other two patient groups.

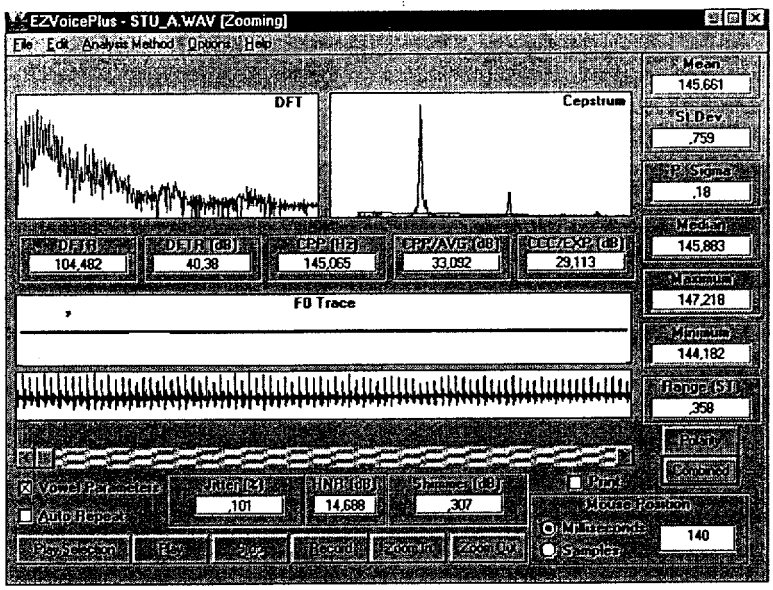

Figure 1: Screenshot from the EZVoicePlus program [18]

Conclusions from Study 2:
On the background of the results reported, we conclude that we cannot find a systematic relationship between signal perturbation and hypernasality. Hypernasality may better be assessed by a spectral measure such as the DFTR or by comparison of nasal and oral levels of sound pressure as measured with instrumentation such as NasalView [18, 19].

Main Conclusions
The relationship between a hypernasal resonance disorder and a vocal dysfunction is not as straightforward as Warren's regulation/ control theory suggests [1]. While hypernasality was present in a number of patients in this group, the correlation between the degree of the resonance disorder and the RHB-ratings was in the low range. The overall prevalence of severe voice disorders was low in the patients examined. Because of this, there was no significant interaction between the apparative measurement with MODIAS and the perceptual ratings either. In th second study, we re-evaluated the relationship between signal perturbation and hypernasality. Again, the systematic relationship that was postulated by other authors ([9, 10] could not be replicated. While this does not mean that the regulation/ control theory is all wrong, our findings implicate that this theory is too simplistic regarding the complex relationship of voice and resonance disorders.

Table 3: Overview of the results of the Mann-Whitney U-tests. Significant differences between the groups are marked with * ($p<0.05$). Tendencies towards significance are marked with (*) ($p<0.1$). If there were no statistically significant differences between the groups this is marked with -. If no U-test was calculated, this is indicated by /.

	controls vs. hypernasal patients	controls vs. dysphonic patients	hypernasal vs. dysphonic patients
Jitter (males)	-	*	*
Jitter (females)	-	(*)	*
Jitter (males and females)	-	*	(*)
Shimmer (males)	(*)	-	*
Shimmer (females)	/	/	/
Shimmer (males and females)	-	*	*
HNR (males)	-	*	*
HNR (females)	-	*	*
HNR (males and females)	-	*	*
DFTR of /a/ (males)	*	-	(*)
DFTR of /a/ (females)	/	/	/
DFTR of /a/ (males and females)	*	-	*

References:

[1] Warren, D. Compensatory speech behaviours in cleft palate: a regulation/ control phenomenon. Cleft Palate 1986; 25: 251-60.

[2] McWilliams, B, Morris, H, Shelton R. Cleft Palate Speech, 2nd ed.Philadelphia: Decker, 1990.

[3] Brooks, A, Shelton R. Voice disorders other than nasality in cleft palate children. Cleft Palate Bulletin 1963, 13: 63-71.

[4] Takagi, Y, Mcglone, R, Millard, R. A survey of the speech disorders of individuals with cleft. Cleft Palate 1965; 2: 28-31.

[5] Marks, C, Barker, K, Tardy M. Prevalence of percieved acoustic deviations related to laryngeal function among subjects with palatal anomalies. Cleft Palate 1971; 8: 201-11.

[6] D'Antonio, L, Muntz, H, Providence, M, Marsh J. Laryngeal/ Voice findings in patients with velopharyngeal dysfunction. Laryngoscope 1988; 98: 432-38.

[7] Bzoch, K. Measurement and assessment of categorical aspects of cleft palate speech. In: K. Bzoch (ed): Communicative disorders related to cleft lip and palate, 2nd ed. Boston: Little, Brown. 1979: 161-91.

[8] Bzoch, K, Kemker, F, Dixon-Wood V. The prevention of communicative disorders in cleft palate infants. In: N. Lass (ed): Speech and Language: Advances in Basic Research and Practice, Volume 10. New York: Academic Press, 1984: 59-110.

[9] Zajac, D, Linville, R. Voice perturbations of children with percieved nasality and hoarseness. Cleft Palate 1989; 26: 226-31.

[10] Lewis, J, Andreassen, M, Leeper, H, Macrae, D, Thomas, J. Vocal characteristics of children with cleft lip/ palate and associated velopharyngeal incompetence. Journal of Otolaryngology 1993; 22: 113-7.

[11] Wendler, J. Stimmstörungen: Schwerpunkte der Diagnostik und Therapie. Laryngo-Rhino-Otologie 1997; 76: 327-31.

[12] Merk, M, Ziegler, W. MODIAS - A PC-based system for routine acoustic analysis of neurogenic speech disorders. In: Maassen, B, Groenen, P (eds). Pathologies of Speech and Language. London: Whurr. 1999: 315-21.

[13] Bressmann, T, Merk, M, Sader, R, Ziegler, W, Horch, HH. Computergestütze akustische Sprechanalyse bei Patienten mit Lippen-Kiefer-Gaumenspalten. Biomedizinische Technik, 1997, 42 (Supplement 2): 93-94.

[14] Bressmann, T, Sader, R, Merk, M, Ziegler, W, Busch, R, Horch, HH. Perzeptive und apparative

Untersuchung der Stimmqualität bei Patienten mit Lippen-Kiefer-Gaumenspalten. Laryngo-Rhino-Otologie, 1998, 77: 700-708.
[15] Brindle, B, Morris, H. Prevalence of voice quality deviations in the normal adult population. Journal of Communication Disorders, 1979, 12: 439-45.
[16] Laguaite, J. Adult voice screening. Journal of Speech and Hearing Disorders 1972; 37: 147-51.
[17] Awan, S. EZVoicePlus signal processing software. VoiceTek Enterprises, RR#2, Box 2490, Nescopeck, PA 18635, USA.
[18] Bressmann, T, Sader, R,, Awan, S, Busch, R, Zeilhofer, HF, Brockmeier, J, Horch HH. Nasalanzmessung mit dem NasalView bei der Therapiekontrolle von Patienten mit Lippen-Kiefer-Gaumenspalten. Sprache - Stimme - Gehör, 1998, 22: 98-106
[19] Awan, S, Bressmann, T, Sader, R, Horch, HH. Measures of RMS Nasalance Using NasalView in Cleft Palate Patients. In: Maassen, B, Groenen, P (eds): Pathologies of Speech and Language: Advances in Clinical Phonetics and Linguistics. London: Whurr,1997, 333-341