



DOKTORITÖÖ TUTVUSTUS: The first study of voice and resonance related treatment outcomes of Estonian cleft palate children

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LIST OF ORIGINAL PUBLICATIONS

Lehes, L., Horn, R., Lippus, P., Padrik, M., Kasenõmm, P., Jagomägi, T. (2018). Normative nasalance scores for Estonian children. *Clinical Linguistics & Phonetics*, 32 (11), 1054–1066.

Lehes, L., Sõber, L., Padrik, M., Kasenõmm, P., Numa, J., Jagomägi, T. (2020). The effect of velopharyngeal insufficiency on voice quality in Estonian Children with Cleft Palate. *Clinical Linguistics & Phonetics*.

Lehes, L.; Aria, C.; Padrik, M.; Kasenõmm, P.; Jagomägi, T. (2023). Pilot Study: Correlation between nasalance scores and cephalometric parameters in Estonian cleft palate children. *Stomatologija. Baltic Dental and Maxillofacial Journal*, 2023, Vol.25, No.1

ORAL CLEFTS (OC)

- common birth defect (1:700)
- approximately 70% - non-syndromic

Prevalence varies according to

- ethnicity;
- gender;
- socioeconomic factors.

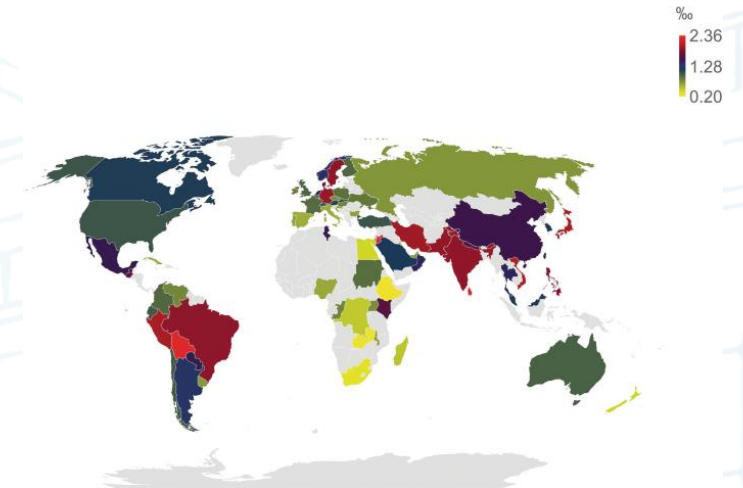
OCs are divided based on the extent of the cleft:

- cleft lip (CL) with or without primary palate;
- CL with or without cleft palate (CP);
- isolated cleft palate (CP);
- submucous cleft palate (SMCP).

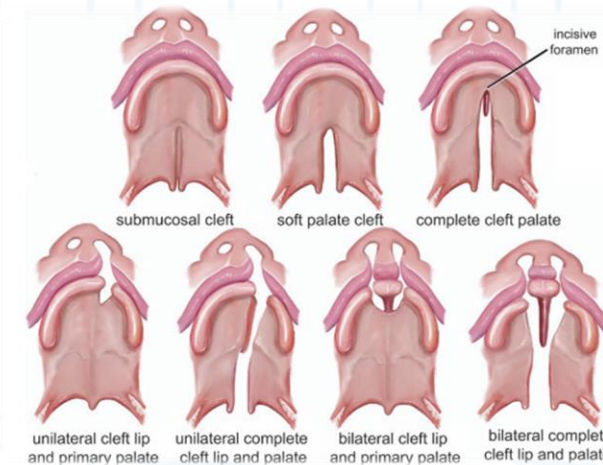


<https://www.cdc.gov/ncbddd/birthdefects/surveillancemanual/chapters/chapter4-6.html>

Estimated birth prevalence of orofacial clefts by country



Denadai, R., Lo, L.J. (2022). Epidemiological Features in Cleft Lip and Palate. In: Denadai, R., Lo, L.J. (eds) Current Concept in Cleft Surgery. Springer, Singapore



Swanson, J. W. (Ed.). (2021). Global Cleft Care in Low-Resource Settings.

CHALLENGES IN LIFE

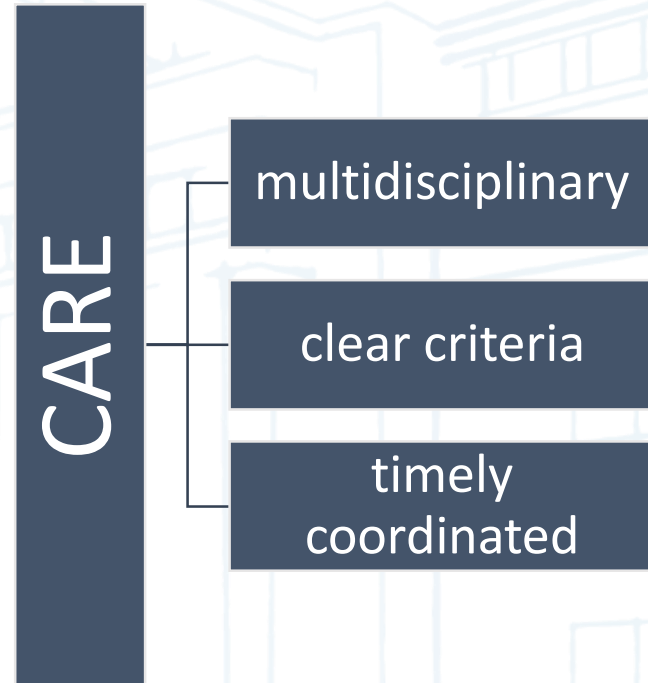
Multiple problems:

- early swallowing and feeding difficulties;
- abnormal articulation, **resonance, voice disorders**;
- **craniofacial growth deviance** and orthodontic abnormalities;
- hearing loss and poor language acquisition skills;
- psychosocial issues and learning difficulties at school.



Appearance and speech seem to be mostly associated with increased emotional and social difficulties

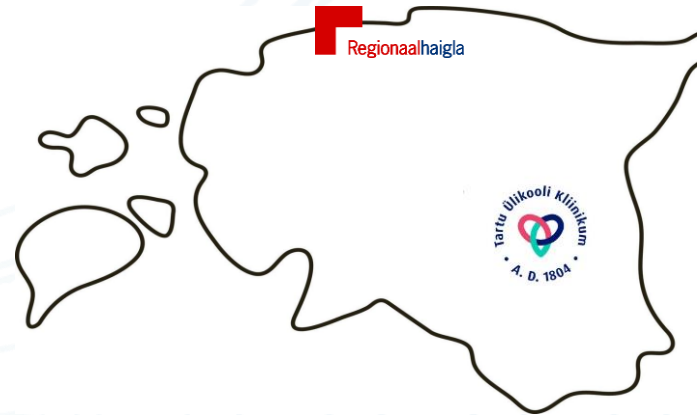
One of the primary treatment goals in CP±L therapy is to **achieve normal speech**.



CLINICAL MANAGEMENT OF CP±L CHILDREN IN ESTONIA

Minimal standardas for CLEFT TEAM:

- orthodontist;
- surgeon;
- otolaryngologist;
- speech and language therapist.



EUROCLEFT PROJECT

Table F.1 — Complete Cleft Lip, Alveolus and Palate (UCLAP and BCLAP)

Timing	Models	Lateral skull radiograph	Photographs	Speech	Audiometry/tympanometry
Primary surgery	√		√		
3 years				√ ^a	√ ^a
5/6 years	√		√	√	√
10 years	√	√	√	√	√
18+ years	√	√	√	√	

^a If hard palate is closed.

Tartu University
Hospital

North Estonia
Medical Centre

Primary surgery:
1-stage palatal
repair

Primary surgery:
2-stage palatal
repair

standardized care
procedures based
on EU guidelines

VELOPHARYNGEAL DYSFUNCTION

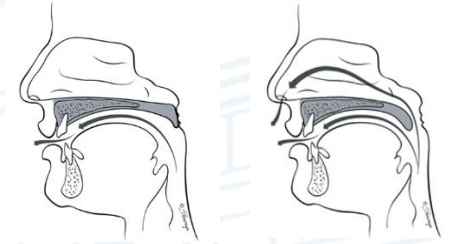
... soft palate and pharyngeal walls are **unable to form an effective closure**.

Affects:

- resonance, speech sound errors, voice;
- eating, drinking (nasal regurgitation).

VPD in CLEFT PALATE population:

- shortened soft palate or its irregularity of the movements;
- severity (scale 0-4): pin-hole opening (1) to opening of entire VP port (4);
- **30-50%** of individuals after surgical intervention



<https://www.nationwidechildrens.org/conditions/velopharyngeal-dysfunction>



CAREFUL SPEECH ASSESSMENT

speech therapy (70%)
OR
secondary surgical intervention

normal speech
AND/OR
continuous speech therapy

RATIONALE FOR THE INCLUDED STUDIES

1. Understanding the mechanisms of abnormal nasalance and voice disorders would considerably facilitate the work of multidisciplinary teams (surgeons, SLPs, otolaryngologists, orthodontists).
2. We need **evidence-based approach**, and objective **thresholds**.
3. Resonance and voice disorders in speakers of **Estonian** have not been described.
4. **Nasalance score** is an objective measurement of treatment outcome.
5. Velopharyngeal dysfunction may cause **laryngeal voice disorders**.
6. **Craniofacial growth** may impact speech quality.
7. **Speech quality** and **appearance** are major factors to be considered when describing the effectiveness of the treatment received by CP±L children.

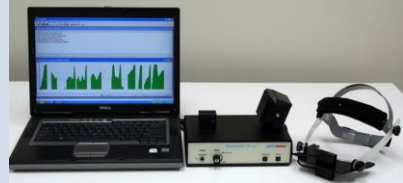
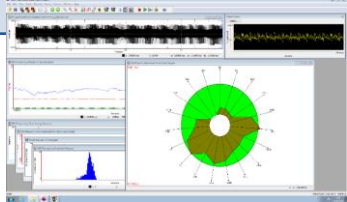

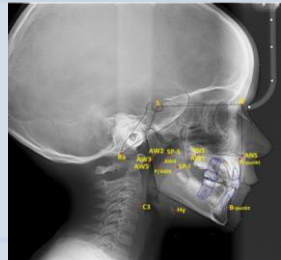
GENERAL AIM

The general aim of the study was to develop specific assessment protocols tailored to the Estonian speech and language and to describe the voice and resonance related treatment outcomes in Estonian cleft palate children.

SPECIFIC OBJECTIVES:

- (1) to develop Estonian-specific optimised speech stimuli for Nasometer II and establish normative nasalance scores for Estonian (Paper I);
- (2) to develop Estonian specific optimised speech stimuli for videonasoscopy and analyse the correlation between the severity of VPD and the severity of voice disorders (Paper II);
- (3) to assess the voice quality and its impact on quality of life in Estonian children born with CP±L (Paper II);
- (4) to determine the cephalometric parameters that distinguish the healthy group from the cleft-palate one as well as the cephalometric parameters that predominantly relate to resonance disorders (Paper III).

Approval (no. 263/T2) for the study was obtained from the Ethics Review Committee on Human Research of the University of Tartu prior to beginning the research.

	Enrolment	No. of subjects	Methods and materials
Study I	March 2015 – February 2017	14 CP±L 92 healthy	Estonian stimuli for Nasometer II pilot study modification of speech stimuli normative nasalance scores 
Study II	October 2016 – February 2017	18 CP±L 79 healthy	Estonian stimuli for VNE acoustic analysis of voice (MDVP) auditory-perceptual analysis of voice (GRBAS) validation of pVHI questionnaire stimuli for videonasoendoscopy stimuli for videolaryngostroboscopy  
Study III	September 2019 – March 2020	11 CP±L 17 healthy	lateral cephalograms nasalance scores cephalometric parameters 

NORMATIVE NASALANCE SCORES FOR ESTONIAN (PAPER I)

DEVELOPMENT OF ESTONIAN SPEECH STIMULI

24 utterances divided into three groups:

- I. oronasal stimuli (ONS) (10% of nasal phonemes);
- II. oral stimuli (OS) – include only oral phonemes;
- III. nasal stimuli (NS) – loaded with nasal phonemes (30% of nasal phonemes).

Examples:

Isal on pikk habe. (ONS) (*Daddy has a long beard.*)

Kaja pugib kooki. (OS) (*Kaja munches on a cake.*)

Mamma pani akna kinni. (NS) (*Granny closed the window.*)

*Each stimulus group consists of eight utterances of at least six syllables each.

*Estonian is a highly phonetic language, i.e. each letter has one sound and is generally sounded consistently.



NORMATIVE NASALANCE SCORES FOR ESTONIAN (PAPER I)

Speech stimuli	N	M	SD	M +/- 2 SD cut-off scores	Max	Min	CI (95%)
ONS	HG92 CP14	30,5 47.1	5,8	18,9 – 42,1	56,3	19,5	29,2–31,6
NS	HG92 CP14	57,8 60.9	5,8	46,2 – 69,4	72,0	41,7	56,6–59,0
OS	HG92 CP14	15,9 37.0	6,0	3,9 – 27,9	45,9	6,8	14,6–17,1

Language	Authors	Age group	N	OS	ONS	NS
Korean	Park et al., 2014	7-11 18-29	216	11.69	34.0 4	31
Greek	Okalidou et al., 2011	18-32	80	12.4	25.5	8.6
Dutch	Van der Heijden et al., 2011	4-6	55	11/13	27	11.6
Swedish	Brunnegård & van Doorn, 2009	4-11	220	12.7	29.5	12.9
Hungarian	Hirschberg et al., 2006	4-9	30	11	31.7	25
English (Ireland)	Sweeney et al., 2004	5-7	70	14.16	26	11
Flemish	Van Lierde et al., 2003	7-13	58	11.3	31.9	11.7
English (Australia)	van Doorn & Purcell, 1998	4-13	245	13.1	-	-
Finnish	Haapanen, 1991	3-54	42	13.6	-	-
Vietnamese	Nguyen et al., 2017	7-9	106	13.0	30.7	56.9
Brazilian Portuguese	Sampaio-Teixeira et al. 2019	6-35	62	18/18/21	-	-
Chilean	Inostroza-Allende et al. 2022	18-35	76	14.15	25.3 8	52.13
English (American)	Alfwaress et al. 2021	100	10-18 19-25	11.75/8.13	-	64/5 5
Canadian French	Rochet et al., 1998	9-85	153	8.8-14.5	24- 30	33.4- 35.6

- 1) phonological quantity contrast does not affect nasalance scores on the sentence level;
- 2) nasalance scores were higher for the girls compared to the boys, the differences were not significant;
- 3) determination of cut-off values is essential for applying nasalance scores in clinical practice.

EFFECT OF VELOPHARYNGEAL DYSFUNCTION ON VOICE QUALITY (PAPER II)

VOICE-RELATED QUALITY OF LIFE (pVHI)

- parents filled the questionnaire (score min 0 max 92)
- CP±L children scored higher in all subscales and in total scores;
- voice-related quality of life is significantly more affected in the CP±L group;
- results may be affected by resonance and articulation disorders.

	Group	N	Mean	SD	p
Functional	CP±L	18	6.56	3.714	< .05
	Healthy	79	2.99	2.844	
Physical	CP±L	18	5.72	4.443	< .05
	Healthy	79	1.95	3.071	
Emotional	CP±L	18	5.06	4.820	< .05
	Healthy	79	1.48	2.297	
Total score	CP±L	18	17.33	11.209	< .05
	Healthy	79	6.42	6.725	

EFFECT OF VELOPHARYNGEAL DYSFUNCTION ON VOICE QUALITY (PAPER II)

ACOUSTIC ANALYSIS OF VOICE (MDVP)

Acc. Par.	Group	Mean	p
F ₀	CP±L	285.09	0.314
	healthy	273.25	
Jitter	CP±L	1.37	0.000
	healthy	0.74	
Shimmer	CP±L	0.422	0.016
	healthy	0.345	
NHR	CP±L	0.13	0.009
	healthy	0.11	
VTI	CP±L	0.044	0.089
	healthy	0.035	
SPI	CP±L	10.8	0.201
	healthy	13.8	

- based on literature 6 acoustic parameters were compared;
- increased values of all parameters in CP±L group, decreased *SPI*;
- previous studies have demonstrated that jitter (frequency variation) is higher in children with VPD;
- jitter may be related to the presence of vocal nodules/oedema - confirmed by our findings from VLS;
- shimmer (amplitude variation) did not exceed the cut-off limits;
- increased values of the NHR was an expected outcome;
- CP±L children's voice appears pressed, hoarse and strained;



EFFECT OF VELOPHARYNGEAL DYSFUNCTION ON VOICE QUALITY (PAPER II)

AUDITORY-PERCEPTUAL ANALYSIS OF VOICE (GRBAS-scale)

	G	R	B	A	S
F (n=5)	1	.80	.60	.00	.40
M (n=13)	.85	.85	.77	.23	.38
TOTAL	.89	.83	.72	.17	.39

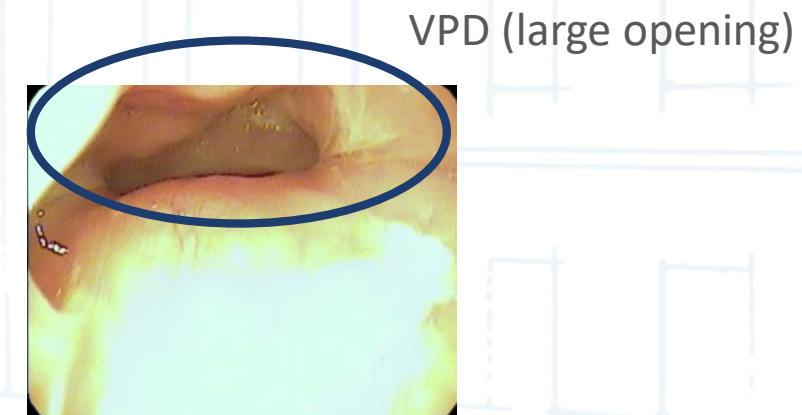
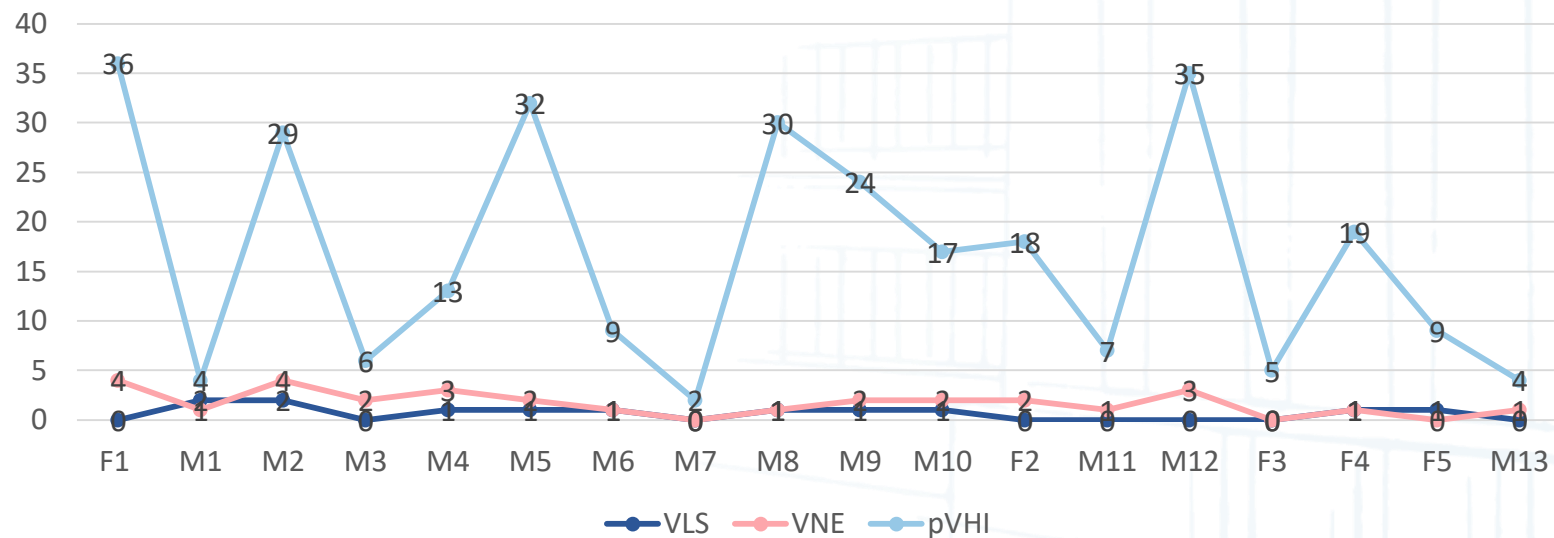
- 18 speech samples graded by 2 independent listeners (scale 0-3);
- inter-rater reliability $\kappa=0.89$;

COMPONENT		DESCRIPTION
Grade	G	Overall grade of hoarseness
Roughness	R	Impression of irregularity of the vibration of the vocal folds
Breathiness	B	Degree to which air escapes from between the vocal folds
Asthenia	A	Degree of weakness heard in the voice
Strain	S	Extent to which strain or hyperfunctional use of voice is heard

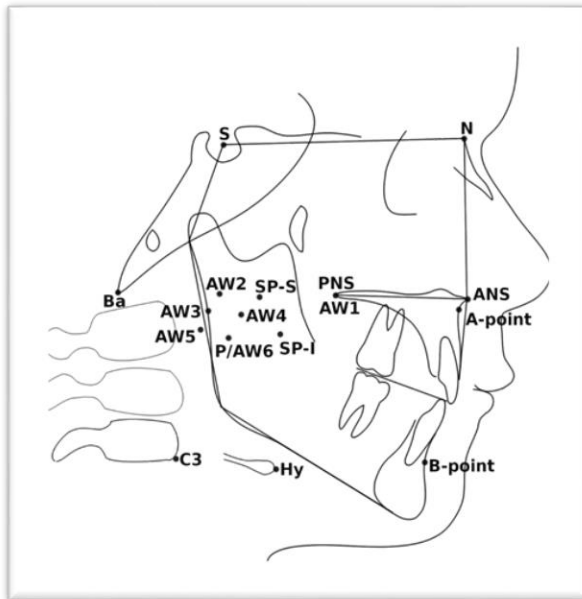
EFFECT OF VELOPHARYNGEAL DYSFUNCTION ON VOICE QUALITY (PAPER II)

INSTRUMENTAL FINDINGS OF THE VELOPHARYNGEAL FUNCTION AND VOCAL FOLD FUNCTION

1. Videonasoscopy (VNE) (scores 0-4)
2. Videolaryngostroboscopy (VLS) (scores 0-2)
3. Pediatric voice handicap index (pVHI)



CEPHALOMETRIC PARAMETERS AND THEIR RELATION TO NASALANCE SCORES (PAPER III)



Lehes, L.; Aria, C.; Padrik, M.; Kasenõmm, P.; Jagomägi, T. (2023). Pilot Study: Correlation between nasalance scores and cephalometric parameters in Estonian cleft palate children. Stomatologija. Baltic Dental and Maxillofacial Journal, 2023, Vol.25, No.1

- cephs of 28 children were graded by 2 independent rater;
- inter- and intra-rater reliability was sufficient.

Ceph. parameter	Participants	Dif. in par. ($p < .05$)	Cor. btw ceph.par. and ONS (CP±L)		Cor. btw ceph.par. And-NS (CP±L)		Cor. btw ceph.par. and OS (CP±L)	
			<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>	<i>r</i>	<i>p</i>
PNS-A (mm)	CP±L (n=11)	0.03*	-0.52	0.05*	-0.45	0.08	-0.58	0.03*
	Healthy (n=17)							
Hy-C3 (mm)	CP±L (n=11)	0.75	-0.64	0.02*	-0.45	0.08	-0.59	0.03*
	Healthy (n=17)							
ANB (°)	CP±L (n=11)	0.31	-0.50	0.06	-0.64	0.02*	-0.54	0.04*
	Healthy (n=17)							
Ba-S-N	CP±L (n=11)	0.55	-0.36	0.14	-0.57	0.03*	-0.27	0.21
	Healthy (n=17)							
PNS-P	CP±L (n=11)	0.01*	-0.38	0.13	-0.39	0.12	-0.27	0.22
	Healthy (n=17)							
AW5-AW6 (mm)	CP±L (n=11)	0.04*	-0.04	0.45	0.06	0.43	-0.08	0.401
	Healthy (n=17)							

CONCLUSIONS AND CLINICAL IMPLICATIONS

1. Most Estonian CP±L children **exhibit both resonance and voice disorders quite expressively**:
 - (1) nasalization is significantly higher compared to what is shown by healthy children,
 - (2) laryngeal voice disorders and morphological changes of vocal folds are common,
 - (3) craniofacial morphology differed compared to the norms.
2. We developed **Estonian-specific optimized speech stimuli for Nasometer II** and nasoendoscopic evaluation. Determination of cut-off scores for nasalance is essential for applying nasalance scores into clinical practice.
3. **Velopharyngeal dysfunction** impacts the prevalence of voice disorders. **Laryngeal dysphonia is frequent** among CP±L children. Therefore, SLPs should include voice therapy in their everyday practice.
4. Changes in **craniofacial morphology may have an impact on speech quality**. These results are of practical use to multidisciplinary CP±L teams, especially orthodontists and surgeons.
5. **Treatment of CP±L children is a teamwork**. In older children, accurate speech assessment identifies children who require secondary surgical intervention or who benefit from speech therapy for optimizing speech outcome.

RESEARCH LIMITATIONS AND FUTURE

1. The principal limitation was the sample size. In Estonia, about 20 CP±L children are born every year.
2. Since the sample size was limited, the mean scores may have been artificially inflated or deflated.
3. In addition to the small sample size, the cleft palate group was very heterogeneous.
4. We should have instructed parents more thoroughly how to fill in the pVHI questionnaire.

We are in progress of creating a **National Registry** for CP±L children.

We are planning a follow-up study.



THANK YOU

ACKNOWLEDGEMENTS

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