Application for doctoral studies

1. Curriculum: Educational Science 80338

2. Title in Estonian: Standardiseeritud tegevusjuhise loomine eesti huule-ja/või suulaelõhededega laste arengu toetamiseks multidistsiplinaarses meeskonnas

3. Title in English: Development of Standardized Protocol for Supporting Estonian Cleft Palate Children: Evaluation and Management in Multidisciplinary Team

4. CERCS speciality.
Logopedics S271 Special didactics
Clinical Medicine B610 Otorhinolaryngology, audiology, auditive system and speech

5. Candidate for PhD studies:

<table>
<thead>
<tr>
<th>First name</th>
<th>Surname</th>
<th>Current Activity</th>
<th>E-mail:</th>
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6. Supervisors:

<table>
<thead>
<tr>
<th>First name</th>
<th>Surname</th>
<th>Affiliation</th>
<th>Position</th>
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<tbody>
<tr>
<td>Evelyn</td>
<td>Kiive</td>
<td>University of Tartu, Institute of Education, Department of Special Education</td>
<td>Professor of Special Education, PhD (supervisor)</td>
</tr>
<tr>
<td>Marika</td>
<td>Padrik</td>
<td>University of Tartu, Institute of Education Department of Special Education</td>
<td>Lecturer in Logopedics and Theory of Learning Difficulties, PhD (co-supervisor)</td>
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</table>

7. Research group where the doctoral thesis is being done:

<table>
<thead>
<tr>
<th>First name</th>
<th>Surname</th>
<th>Affiliation</th>
<th>Position</th>
<th>PhD Student</th>
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<tbody>
<tr>
<td>Evelyn</td>
<td>Kiive</td>
<td>SHHI</td>
<td>Professor of Special Education, PhD</td>
<td>No</td>
</tr>
<tr>
<td>Marika</td>
<td>Padrik</td>
<td>SHHI</td>
<td>Lecturer in Logopedics and Theory of Learning Difficulties, PhD</td>
<td>No</td>
</tr>
<tr>
<td>Triin</td>
<td>Jagomägi</td>
<td>University of Tartu,</td>
<td>Associate Professor,</td>
<td>No</td>
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SHHI
8. Description of necessary available resources (equipment, finances):

The group does not have separate funding. The research group has all necessary equipment for conducting the research. The research is conducted using a digital video camera, KayPentax Nasomeeter II (Model 6450) hardware and software; KayPentax voice analyzer (Multi-Dimensional Voice Program) with high-quality calibrated microphone, KayPentax endoscopy and stroboscopy system (Model 7245D) with digital video and audio for recording and playback.

9. Doctoral thesis’ innovativeness and importance for development of this scientific direction:

It is an interdisciplinary research project that focuses on cleft palate children (CLP) as children with special needs. The project enables us to monitor and better understand these children’s complexity of development and fully realize what individuals with CLP need from their treatment, and how best to address these needs and improve their quality of life. In addition, these findings give us the base to design their unique, individually-tailored developmental environment and educational programs. As a result of this project the quality of life will increase for Estonian children with a history of cleft. The results of this study will be also interesting in international comparison in two main aspects: (1) CLP specific speech disorders in Estonian compared to the findings in other languages; (2) standard and quality of helping Estonian CLP children as children with special educational need in comparison with other countries.

Introduction

Cleft lip (CL), cleft lip with or without cleft palate and isolated cleft palate (CP), collectively termed oral clefts (OC), are the second most common birth defects among newborn. These defects arise in about 1 in 700 liveborn babies, with ethnic and geographic variation [1].
National statistics regarding orofacial clefts is nonexistent in Estonia. The only prevalence findings known to us were those of Lõvi-Kalnin [2], conducted during 1970–1980. On the basis of the data from the study, the current rate of occurrence of clefts in Estonia would be 1 case per 777 live births. The only way to estimate the number of children affected by clefts is to use the pre-existing information from previous visits to maxillofacial surgeons which are carried out in two different hospitals in Estonia: The Tartu University Hospital and The North Estonia Medical Centre. It would be easier to follow CLP children’s quality of care, general development, and quality of life if we’d have an overall database of statistics and that can be used during the rehabilitation process by all the specialists who work with the CLP child. One of the outcomes of this work would be a database development that includes all needed aspects, and would be accessible to all the specialist.

**Developmental Aspects: Speech, Language, and Cognition**

Although nonsyndromic CLP is usually not a life-threatening condition, these children often demonstrate multiple complex issues. These issues may include feeding and nutritional problems, digestion, developmental delay or learning disabilities, speech, language, resonance and voice disorders, middle-ear ventilation and hearing, obstructive sleep apnea, facial and dental development may be disturbed because of the structures involved. All these problems, in turn, may cause emotional, psychosocial and educational difficulties.

The literature on the developmental status of CLP children is contradictory. This is partly because CLP group is very heterogeneous population. CLP children may differ not only in the type and severity of the cleft but also with respect to other conditions such as chronic middle ear effusion and hearing loss; the number of hospitalizations, the type and effectiveness of surgical repairs and parental attitudes and involvement. For children and teenagers with a nonsyndromic CLP, intelligence seems to be in the average range [3,4], however several authors have reported that these children show some early deficits in language skills and cognitive development [5,6,7,8]. Nevertheless, these delays seem to disappear with time [9, 10,11]. By contrast, some authors have noted, that many children with CLP do need extra attention because of their speech and hearing difficulties. Children with oral clefts have been shown to be at elevated risk for learning problems compared with unaffected children, particularly in reading and related tasks [12, 13, 14, 15, 15]. In considering speech sound development, even with early surgical repair, a majority of preschoolers demonstrate delays in speech sound development and have typical cleft palate speech [17]. Children who have speech problems often lack self-confidence in reading aloud, which may influence the teachers’ evaluation of their abilities [18]. Therefore, speech and resonance, in addition to language and cognitive development, should be carefully monitored throughout preschool and school years.

**Societal Issues: Appearance, Speech Quality and Hearing Difficulties**

Humans are naturally social beings who require human interaction, communication and acceptance by others. Individuals with clefts are often hindered in their communication by speech and hearing difficulties. In addition, they can be viewed more negatively by others because of their appearance [18]; even teachers have more favorable expectations of attractive children than unattractive children [19]. Children with a history of cleft are probably more teased than their unaffected peers [20]. Teasing as well seems to be influenced by the child’s physical appearance and speech differences, because children tend to report less teasing after surgeries that address those problems. It has been found that experience of being teased and/or bullied affected children more than having the cleft per se [21]. Bullying is associated with
depression, anxiety, and fear of negative evaluation [22]. Self-perception plays a pivotal role in influencing an individual's self-esteem and psychological adjustment affected. Children with a history of cleft have consistently been found to have a more negative self-concept when compared to their unaffected peers [23, 24, 25, 26].

In individuals with CLP, errors in speech production are noticed due to the abnormalities in oronasal structure/function, orofacial structure and growth, learned neuromotor patterns during early infancy, and/or disturbed psychosocial development [27]. Broadly, cleft type errors of speech sound production are classified into two types: obligatory and compensatory [28]. Obligatory errors include errors in production due to interference of structural abnormalities. These errors cannot be corrected through speech therapy unless the underlying structural deformity is corrected. Compensatory errors include errors that occur due to maladaptive articulatory placements. These errors can be corrected only through speech therapy [29]. It is important to identify compensatory and obligatory errors in articulation, in order to choose the right treatment method. CLP children may have hypernasal resonance. Speech resonance is the result of the transfer and modification of the acoustic signal produced in the larynx through the vocal tract [30]. Hypernasal resonance, or excessive nasal resonance in speech [31], is a significant perceptual feature of velopharyngeal dysfunction, most noticeable on vowels and approximants [30]. Severity of speech problems depend on child's general development and health, surgical intervention and orthodontic treatment, timing and quality of speech therapy. In conclusion, studies on cleft have shown relationships between (a) facial appearance and teacher perception, (b) behavioral inhibition and lower school achievement, and (c) speech defectiveness and self-esteem [26].

Many children with a history of cleft have some degree of hearing impairment. The primary cause is Eustachian tube dysfunction due to impaired function of levator veli palatine muscle. It can lead to either otitis media with effusion (OME), retraction pockets, partial or complete adhesion of the tympanic membrane and in some patients to the development of cholesteatoma [32, 33]. Among them, the most common condition is OME, which in children with CLP appear in younger age and have higher prevalence compared with children without cleft [34]. All those conditions are associated with a mild-to-moderate conductive hearing loss with levels fluctuating between 0 to 55 dB across the speech frequencies [35]. Having a hearing impairment can add to the social judgments that people make [18]. In addition, hearing loss in childhood, even mild, brings learning difficulties. Children with mild hearing loss may present problems in language development, reading disabilities and behavioral disorders [36].

CLP speech in Estonian has been studied by few students in University of Tartu, Institute of Education Department of Special Education: Terras [37], Palo [38] ja Ird, Suvi [39]. However, there have been done no studies that describe the quality of life, including social withdrawal and psychosocial issues, satisfaction with physical appearance, self-perception, and hearing impairment. Furthermore, there is no data available about CLP children’s academic skills, school performance and availability of special assistance, e.g. speech and language therapy, psychological support.

To accomplish coordinated and integrated care, the team approach to management is required for these children. With the team approach, the child is more likely to receive quality of services, continuity of care, and long-term follow-up in order to achieve the best outcome. The entire rehabilitation process may last form infancy into adulthood. Long term care may affect the child’s and his/her parents’ well-being, including psychological and social aspects.
Minimal standards of record taking have been established by Americleft [40] and Euroclef [41] projects:

<table>
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<tr>
<th>Timing</th>
<th>Models</th>
<th>Lateral Cephs</th>
<th>Photos</th>
<th>Speech</th>
<th>Audiometry</th>
<th>Patient/Parent Satisfaction</th>
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<td>Primary Surgery</td>
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<td>5/6 Years</td>
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These standards include medical intervention and speech evaluation for cleft palate teams. In Estonia, we have two cleft teams that are based in Tartu and in Tallinn. The goal of the cleft or craniofacial team is to ensure that care is provided in a coordinated way and consistent manner with the proper sequence of evaluations and treatments. This should be done with consideration for the child’s overall developmental, medical and psychological needs [41].

Researches stress that follow-up studies for evaluation of all affected aspects are necessity. In order to meet the requirements in Estonia, there is a need for (1) adopt the minimal record taking standards so the outcomes could be compared on a global scale (2) validate Estonian Speech Test for CLP children; validate pediatric voice handicap index (pVHI) and develop normal nasality scores for Estonian language, (3) make a survey of CLP children’s educational options, special needs and support, (4) find out the opportunities for the families to get support and information.

Scientific relevance of the research is to describe factors that affect CLP children’s quality of life, special educational needs, including speech and voice disorders, hearing impairment, self-esteem, and quality and availability of speech therapy and medical intervention. Practical relevance of the research is development and standardization of protocol for supporting and treating cleft palate children.

The goal of the dissertation is the development of guidelines for supporting Estonian CLP children regarding to their special needs in multidisciplinary team. Based on the objective, we need to study speech and voice quality, psychosocial problems, quality and availability of support and quality of treatment. In addition, based on the findings we describe to their special needs related to their quality of speech, voice and velopharyngeal dysfunction, finding association between speech therapy, surgical, orthodontic intervention and overall speech outcome, and its effect on quality of life, including academic achievement.

10. Doctoral thesis project (up to 4000 characters with spaces):
1) developing the minimal record taking standards for speech and language and psychological evaluation; validating Estonian Speech Test for CLP children, including voice evaluation, and developing normal nasality scores for Estonian language – Sept 2016 – June 2017 (1st article);

2) collecting data and describing speech and voice outcomes and their correlation in Estonian CLP children – Sept 2017 – June 2018 (2nd article);

3) collecting data about CLP children’s quality of life and their family’s well-being, rehabilitation process, educational needs and quality of support from different specialists – Sept 2018 – June 2019 (3rd article);

4) writing doctoral thesis - Sept 2019 – June 2020

11. Summary in Estonian

Pealkiri: Standardiseeritud tegevusjuhise loomine eesti huule- ja/või suualõhenedega laste arengu toetamiseks multidistsiplinaarses meeskonnas

Juhendajad: Evelyn Kiive, PhD, juhendaja; Marika Padrik, PhD, kaasjuhendaja

Kokkuvõtte tekst


12. Summary in Estonian

Title: Development of Standardized Protocol for Treating Estonian Speaking Cleft Palate Children: Evaluation and Management in Multidisciplinary Team

Supervisor(s): Evelyn Kiive, PhD, juhendaja; Marika Padrik, PhD, kaasjuhendaja
It is an interdisciplinary research project that focuses on cleft palate and/or lip children as children with special needs. Cleft lip, cleft lip with or without cleft palate and isolated cleft palate, collectively termed oral clefts, are the second most common birth defects among newborn. These defects arise in about 1 in 700 liveborn babies, with ethnic and geographic variation. The project enables us to monitor and better understand these children’s complexity of development and fully realize what individuals with cleft palate and/or lip need from their treatment, and how best to address these needs and improve their quality of life. In addition, these findings give us the base to design their unique, individually-tailored developmental environment and educational programs. As a result of this project the quality of life will increase for Estonian children with a history of cleft. Although nonsyndromic CLP is usually not a life-threatening condition, these children often demonstrate multiple complex issues. These issues may include feeding and nutritional problems, digestion, developmental delay or learning disabilities, speech, language, resonance and voice disorders, middle-ear ventilation and hearing, facial and dental development may be disturbed because of the structures involved. All these problems, in turn, may cause emotional, psychosocial and educational difficulties. Rehabilitation is successful with integrated care, including different specialists and children’s families. The goal of the dissertation is the development of guidelines for supporting Estonian CLP children regarding to their special needs in multidisciplinary team. Based on the objective, we need to study speech and voice quality, psychosocial problems, quality and availability of support and quality of treatment. In addition, based on the findings we describe to their special needs related to their quality of speech, voice and velopharyngeal dysfunction, finding association between speech therapy, surgical, orthodontic intervention and overall speech outcome, and its effect on quality of life, including academic achievement. Using standardized protocols enables us to collect data during long period and compare our results with the results of different cleft palate teams.