Speech in 3-year-old children with unilateral cleft lip and palate:
Impact of method for palatal repair and early intervention

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ABSTRACT

OBJECTIVE: The main objective of this study was to describe speech at 3 years of age in children born with UCLP and relate the findings to operation method and early intervention strategies.

METHODS AND PROCEDURE: 26 consecutive children born with unilateral cleft lip and palate operated with a one-stage palatal repair at 12 months (n=15) or a two-stage repair (n=11) with soft palate closure at 3-4 months and hard palate closure at 12 months. Two experienced speech and language pathologists independently transcribed single words from video recordings at age 3 years and intra- and inter-observer agreement was calculated. Mean percent correct consonants adjusted for age (PCC-A) and occurrence of active articulation errors were calculated from phonetic transcription and passive speech characteristics from scale ratings. Frequency and type of early intervention provided was extracted from patient records. Information about early speech production and estimated need for therapy at age 3 was obtained from a parallel project.

MAIN OUTCOME MEASURES AND RESULTS: PCC-A at 3 years was 68% (8 – 100%). Active articulation errors were noted in 46%. Concerning passive deviations moderately or severely deviant hypernasal resonance and perceived velopharyngeal dysfunction was present in 23%. No significant difference in speech results was found between the operation methods or could be related to level of early intervention received. The latter could not be related to frequency or type of the early consonant production. The estimated need for treatment at 3 years of age correlated well with the PCC-A value at 3 years.

CONCLUSIONS: There was no significant difference in speech results at age 3 years depending on if a one- or two-stage closure of the palate had been used. There was no indication that early intervention had had any impact on speech production at 3 years. No clear basis was found in early consonant production used for decisions about level of early intervention, whereas the SLP opinion of “need for treatment” at 3 years of age was well grounded in speech production.
BACKGROUND

The oronasal cavity differs anatomically in infants with a cleft lip and palate malformation from typically developing children. In order to normalize the development as much as possible different surgical methods are used to reconstruct the lip and palate in combination with orthodontic treatment and speech and language intervention.

It has been extensively discussed to what extent different operation techniques for palatal repair influence speech in children born with cleft palate (Peterson-Falzone, 1996, Peterson-Falzone et al, 2010, Lohmander, 2011a). The main issues have been to reach optimal maxillary growth by delaying the palatal repair versus the development of good speech thought to be enhanced by early palatal surgery. There is therefore no consensus regarding surgical protocol for primary palatal repair, which was demonstrated in a survey of clinical services for cleft palate in Europe showing that 201 clinical teams used 194 different protocols for primary surgical repair of unilateral cleft lip and palate (UCLP) (Shaw et al., 2000). The impact of type of primary palatal surgery on speech and dentofacial growth at the age of 5 years is currently being studied in the Scandcleft project, a randomized, international multicenter study (Semb, 2001). The project does not include earlier speech behavior, however. Since a restricted articulatory pattern has been shown already in the babbling stage of infants born with cleft lip and palate (CLP) compared to children without CLP it is interesting to investigate possible influence of surgical procedure and other intervention on speech production before age 5, which is the age commonly considered for speech to be sufficiently well developed (Grunwell, 1987). The findings in babbling of infants with CLP comprise a smaller consonant inventory and a lower occurrence of oral stops, a higher occurrence of nasal consonants and semi-vowels compared to in babbling of typically developing infants without clefts (e.g., Chapman et al., 2001; Jones et al., 2003, Scherer et al., 2008a). The concept of “true consonants” including all consonants except semi-vowels and glottals has been introduced, with findings that infants with CLP have lesser true consonants in their babbling than infants without CLP (e.g., Chapman et al., 2003). Also the place of articulation differs between infants with and without CLP but with not entirely consistent findings in different studies. Chapman et al. (2001) and O’Gara and Logemann (1988) reported more labial and glottal place of articulation than alveolar or velar, whereas more velar than alveolar place of articulation was found by Willadsen and Albrechtsen (2006) and Lohmander et al. (2011) in the infants with CLP compared to those without.

Even after palatal repair, oral stops or true consonants are often limited (Jones et al., 2003; Scherer et al., 2008a) and deviant articulatory patterns in 3-year-old children born with cleft lip and palate (CLP) have been reported in approximately 50% of the cases in several studies (Morris & Oxanne, 2003; Chapman et al., 2008; Lohmander & Persson, 2008; Willadsen, 2012; Klintö et al., 2013a). In these studies of young children different methods for primary palatal repair were practiced and thus similar problems were observed in the early speech regardless of staging, timing, and technique used for primary palatal repair.

Several studies have thus shown poorer speech at the age of 3 years in children born with cleft lip and palate compared to typically developing children. Most recently
Klintö et al (2013a) concluded that primary palatal surgery in one or two stages before or at around 1 year of age did not result in any significant differences in speech production at age 3 years. However, children with an un-operated hard palate at the time for speech assessment had significantly poorer speech production. Willadsen (2012) came to the same conclusion using a picture naming test with defined target sounds in single words, examining 3-year olds in two groups of children born with UCLP where one group had undergone closure of the soft palate at 4 months and the hard palate at 12 months and the other still had an un-operated hard palate. Both had a restricted phonological system and produced more cleft speech errors compared to a group of 35 typically developing children, but the late group to a considerably higher extent.

Cleft speech characteristics can be divided into passive and active errors (Harding & Grunwell, 1998). If there is a coupling between the oral and the nasal cavities caused by an unrepaired cleft, a fistula or inadequate velopharyngeal function, speech difficulties such as hypernasality, nasal emission of air, weak articulation of pressure sounds, and nasal realizations of consonants may occur. These typical cleft speech characteristics, directly caused by limited structure or function, are regarded as passive. In contrast, an active process is a compensatory strategy for the inability to produce sufficient intraoral pressure needed for high-pressure consonants, for instance consonants normally produced anteriorly to the undesired opening may be retracted to a place behind the oro-nasal opening. Another common and distinctive active deviation is the glottal stop substitution.

The concept of percent correct consonants (PCC) and adjusted for age, (PCC-A) has been found to be a stable measure of articulation skill in children with CLP (Lohmander & Persson, 2008; Scherer et al., 2008a; Klintö et al., 2011; Willadsen, 2012). It is calculated from transcriptions of single word productions and is an adaptation of the PCC measure developed by Shriberg et al (1997) who used transcriptions of connected speech. Appropriate PCC-A for typical 3-year-olds are scores at or above 80% (Lohmander & Persson, 2008; Klintö et al., 2011). Phonetic transcription for speech related to cleft palate is thoroughly described by Howard (2011).

Consonant use in vocalizations has been shown to be a consistent predictor of speech onset and of longer-term speech development in typically developing children (e.g. Vihman and Greenlee, 1987). Stable consonant production in the babbling period prepares the child for word production (McCune and Vihman, 2001). Considerable correspondence between typical sounds in babbling and in early words was noted by Stoel-Gammon (1989). Measures of consonant production at 18 months of age, such as number of consonant types, frequency of occurrence of oral stops and of anterior stops have also been associated with speech production at 3 years of age in children with CLP (Lohmander and Persson, 2008; Scherer et al., 2008a; Klintö et al., 2013a).

Based on this continuity principle, stimulation of vocabulary and of sound production has been proposed to reduce compensatory articulation in later speech (Scherer & Kaiser, 2007; Scherer et al 2008b; Hardin-Jones & Chapman 2008). It has been hypothesized that early intervention could prevent active deviances from being established (Dixon-Wood, 1997) and even been claimed that intensive early
intervention (EI) programs are necessary in order to change already established behavior (Henningsson, 1982). The literature is sparse, however, and there is no or little evidence for improved speech results after EI. Studies are generally based on few participants and the methodology for intervention differs (Scherer, 2008b; Dynesius et al, 1993). In a systematic review of articles on EI Meinusch & Romonath (2011) further emphasized that there is limited empirical support for the effectiveness of early language intervention for children with CLP. Also, that the methodological quality is heterogeneous and in general, not convincing.

According to Scherer et al (2007, 2008b) parents can be trained to deliver sufficient home training. The authors in those studies investigated parent implemented EI to 10 children with cleft lip and palate and deviant articulatory development compared to 10 typically developing children, 14 – 36 months of age. While the intervention resulted in speech gains for the children with clefts, speech measures did not exceed those made by the children without clefts. Dynesius et al (1993) found no significant difference in speech between two groups of children, at the age of 5 years with isolated cleft palate: four receiving early speech stimulation and four children with no intervention.

In Stockholm, an intensive EI program to children, 12 months to 3 years of age born with CLP, who in their sound production lacked anterior, oral articulation, sometimes in combination with glottal articulation, was used from the end of 1970s to the end of 1990s. This intervention program consisted of intensive therapy sessions by the SLP two to three times a week for several months and specific advice to parents for home training. The method included non-speech oral-motor activities as well as sound play with toys and items containing anterior, oral sounds such as bilabial and dental high pressure consonants. It included close eye-contact, many repetitions, and encouragement to turn-take. The SLP initially acted as a role model with the child when new elements were introduced, with the parent gradually taking over. The method was inspired by a behavioristic approach and has been clinically described by Henningsson (1982), but has not been evaluated.

In the late 1990s and throughout the first decade in 2000, the protocol gradually changed towards a program with different levels of intervention. In addition to the intensive EI described above, a more consultative approach was also used. It contained standardized advice to parents for home training during the time between a post operative review session after palatal surgery with a SLP at about 15-18 months and 3 years of age. A slightly closer intervention model including contact with an SLP, information about speech-language development, individually tailored advice about specific home training, combined with SLP sessions was also used. In a recent chart review of 79 consecutive infants born with different types of CLP in Stockholm 2005-2007 type of articulation patterns and type and frequency of EI to the children were investigated (Raud Westberg & Lohmander, 2011; 2012). The survey revealed that most children had received intervention according to the routines described above. However, according to the assessment of early speech production before EI only 42% were considered to have a deviant speech production. It was therefore concluded that the basis for decision about EI was unclear and no obvious reason was found for providing almost all children (89%) with EI.
Thus, there is rather good knowledge about the early development of speech production and about the relationship to later speech but little about the possible influence of method for surgical treatment and even lesser of EI.

**AIM**

The main objective of this study was therefore to investigate speech production at 3 years in children born with UCLP treated according to two different methods for primary palatal repair and different EI strategies.

**RESEARCH QUESTIONS**

- How is speech at 3 years of age in children with UCLP?
- Is there a difference in speech related to whether the children are treated according to a one- or two-stage palatal repair?
- Is there a relationship between speech in children at 3 years and frequency and type of EI?

We were also interested in the basis for decision on intervention, that is, the relationship between speech at 18 months of age and received early intervention and also between speech at 3 years of age and the need of treatment as estimated by a speech-language pathologist at 3 years of age.

**METHOD**

**Subjects**

A consecutive series of 36 children (27 boys and 9 girls) born with UCLP participate in the Scandcleft project and were possible participants in the present study. All children were born between 1998-2006 and were patients at Karolinska University Hospital, Stockholm, Sweden. They were native Swedish speaking and had no known additional malformations or syndromes. Since they were treated at the Stockholm center and participants in the Scandcleft project, the children were randomized to palatal closure either in one session (Op 1) with the so called Minimal Incision Technique, MIT (Mendoza, 1994) at 12 months of age (n = 19) or in two stages (Op 2) with lip and soft palate closure at 3 to 4 months and hard palate closure at 12 months (n = 17) (Rautio et al., 2013). Two surgeons had performed all operations. Twenty of the children were treated with ventilation tubes before the age of 3 years, mostly simultaneously with palatal surgery (Tengroth et al., 2013). Data on hearing status at 3 years was not available, since the clinical routines at that time only included formal testing at 5 years and older.

Two children had to be excluded due to poor cooperation and refusal to participate in the video recording situation, and another two for producing less than 50% of the target words. Two video recordings proved to be without sound, and another one had poor picture quality. Three remaining children were excluded due to missing video recordings. The final study group thereby consisted of 26 children (72%), 19 boys and 7 girls.
Table 1. Total number of participants, distributed for gender and operation method with exclusions.

<table>
<thead>
<tr>
<th>Participants</th>
<th>Op 1</th>
<th>Op 2</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Boys</td>
<td>Girls</td>
<td>Boys</td>
</tr>
<tr>
<td>Total</td>
<td>15</td>
<td>4</td>
<td>12</td>
</tr>
<tr>
<td>Excluded</td>
<td>4</td>
<td>1</td>
<td>3</td>
</tr>
<tr>
<td>Included</td>
<td>12</td>
<td>3</td>
<td>7</td>
</tr>
</tbody>
</table>

Procedure

Video recordings were obtained at the age of 3 years. The mean age was 36.8 months, with a range of 35-42 months. The recordings took place in a quiet room at Karolinska University Hospital, during interaction with a SLP and with a parent being present in the room. They lasted between 30 and 45 minutes. A video camera (Sony DCR-TRV30E) on a tripod, with an external microphone (AKE ECM-MS957) was used. The microphone was placed in the middle of the table in front of the seated child, with the SLP straight across the table.

A picture naming test with 30 single words developed in the Scandcleft project to assess cleft palate speech was used (Lohmander et al., 2009). Each word had one defined target consonant in initial or final position. Each target consonant occurred at least three times in the test which minimized the risk for random production of a target consonant. The recordings were performed by three experienced SLPs, not including the author or the raters. The SLP held up a picture card at the time in front of the child. The elicitation of words was obtained either by spontaneous production from the child, or by the child repeating the production of the SLP. Not all children responded to all thirty pictures. The median number of words elicited was 27 (range: 15 – 30 words). If less than 50% of the words were produced the child was excluded.

The video recordings were edited and each test word production was saved in a separate file in mpeg format. The editing was performed by an independent person using the Correl Video Studio Pro X4. In most cases each file included, as a reference, the sequence where the word with the target sound was either used by the SLP for elicitation or repeated by the SLP as reinforcement and as a reference (Lohmander et al., 2009). A computer with Windows Media Player and headphones Denon AH D1001 or Sennheiser HD 218 were used for the assessments, which were performed by two observers who are SLP’s, with 18 respective 9 years of experience of cleft palate speech. Every target sequence could be watched and listened to repeatedly. Narrow phonetic transcription of each single word was performed, blinded and independently. The International Phonetic Alphabet IPA (2005) and ExtIPA for disordered speech (2002) were applied and the target consonants were categorized regarding place and manner of articulation as well as presence or absence of voicing (table 2).
Table 2. Categories for the transcriptions of the target consonants.

<table>
<thead>
<tr>
<th>Place of articulation</th>
<th>Bilabial, labiodental</th>
<th>Interdental, dental, alveolar, postalveolar, retroflex (common variation of dentals in Swedish)</th>
<th>Palatal, velar, uvular (common variations of velars in co-articulation in Swedish)</th>
<th>Glottal</th>
</tr>
</thead>
</table>

<table>
<thead>
<tr>
<th>Manner of articulation</th>
<th>Stop</th>
<th>Fricative</th>
<th>Nasal</th>
<th>Realizations of /r/</th>
<th>Lateral /l/</th>
<th>Approximant</th>
</tr>
</thead>
</table>

<table>
<thead>
<tr>
<th>Voicing</th>
<th>Voiced</th>
<th>Unvoiced</th>
</tr>
</thead>
</table>

A 4-step rating scale was applied to the variables for the passive speech deviations hypernasality, hyponasality, audible nasal emission of air, and perceived velopharyngeal function (table 3).

For calibration and recalibration the two observers earlier the same year had undertaken six hours of transcribing together, rating in consensus, and discussing the use of different symbols and diacritics from video recordings of the same picture test on 3-year-old children born with cleft palate but not included in the study. The calibration sessions also included practicing the use of a 4-step rating scale for passive deviations.

Table 3. Variables and rating scales for passive speech deviations.

<table>
<thead>
<tr>
<th>Scale Step</th>
<th>Hypernasality</th>
<th>Hyponasality</th>
<th>Audible Nasal Emission of Air</th>
<th>Perceived Velopharyngeal Function</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Normal</td>
<td>Normal</td>
<td>No presence</td>
<td>Competent</td>
</tr>
<tr>
<td>2</td>
<td>Mild</td>
<td>Mild</td>
<td>Occurs sometimes</td>
<td>Mildly reduced</td>
</tr>
<tr>
<td>3</td>
<td>Moderate</td>
<td>Moderate</td>
<td>Occurs often</td>
<td>Moderately reduced</td>
</tr>
<tr>
<td>4</td>
<td>Severe</td>
<td>Severe</td>
<td>Occurs all the time</td>
<td>Severely reduced</td>
</tr>
</tbody>
</table>

The level of intensity, type, and frequency of EI after palatal surgery, during the time span between 18 months and 3 years of age, was collected from patient records, and categorized into three levels (table 4).
Table 4. Level of intensity, type, and frequency of EI between 18 months and 3 years.

<table>
<thead>
<tr>
<th>Level</th>
<th>Description</th>
<th>Frequency</th>
</tr>
</thead>
<tbody>
<tr>
<td>Level 1</td>
<td>Reviews with general advice/counseling to parents about stimulation of oral anterior articulation</td>
<td>1-3 times</td>
</tr>
<tr>
<td>Level 2</td>
<td>Reviews and intervention, including consultations with specific, individually customized advice to parents about stimulation of oral anterior articulation and oral motor activities, combined with SLP therapy sessions</td>
<td>4-9 times</td>
</tr>
<tr>
<td>Level 3</td>
<td>Early intensive intervention in SLP therapy sessions with specific, individually customized stimulation of articulation and oral motor activities, combined with parental advice</td>
<td>More than 10 times</td>
</tr>
</tbody>
</table>

Information about the type of early consonant production at 18 months of age (i.e., number of consonant types and number of true consonants) as well as the need for intervention at 3 years of age, as estimated by a SLP was collected from a previous study, which included the same children (Höglund Santamarta & Karlsson, 2011). Ethical approval had been obtained by the Regional Ethical Review Board, number 97-372, within the Scandcleft project, and all parents had given their written informed consent for participation.

Analysis
Percent correct consonants adjusted for age, PCC-A (Shriberg et al., 1997, Klintö et al., 2013a) was calculated from the transcriptions as a measure of articulation skill. Varying types of deviant /s/-articulation, such as inter-dental, retroflex, lateral, and palatal production of /s/, were accepted as age adequate and scored as correct, as well as deviant or missing production of /r/. The not accepted productions were glottal articulation, retracted oral articulation when other than /s/, and omission of target sound. A PCC-A of 80% and higher was considered as typical for the age group in this study (Klintö et al., 2013a). For the ratings of passive deviations percent occurrence of each deviance was calculated.

Reliability
Intra- and inter-observer agreement of transcriptions of target sounds in the word naming test for all children at 3 years of age was calculated by means of percent agreement point by point. The consonants compared had to be identically transcribed for place, manner and voicing, to be considered as agreed. Differences in transcriptions of diacritic symbols were not taken into account. The ratings were compared within and between the two observers. Thirty percent of the material was re-assessed by both observers four weeks later for intra-observer agreement. The inter-observer agreement was calculated from the assessments of all material for both observers.
For transcriptions the exact mean intra-observer agreement for both observers was 92% (Table 5). The exact mean intra-observer agreement for observer 1 on the 4-point scale for ratings of passive deviations was 86% and for observer 2 89%. The observer with the highest intra-observer reliability was used as the reference in the following analysis of the children's production.

Table 5. Mean intra-observer agreement and range for the two observers (O1 and O2).

<table>
<thead>
<tr>
<th>Exact intra-observer agreement</th>
<th>Mean (%)</th>
<th>Range (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Transcriptions</td>
<td>92</td>
<td>57-100</td>
</tr>
<tr>
<td>Ratings</td>
<td>86</td>
<td>78-100</td>
</tr>
</tbody>
</table>

The exact mean inter-observer agreement was 90% (table 6). The exact mean inter-observer agreement on the 4-point scale, was 84%, with 80% for hypernasality, 88% for hyponasality, 88% for audible nasal air leakage, and 80% for perceived velopharyngeal function. With an acceptance of one scale step difference on the 4-point scale as agreement both intra- and inter-observer agreement was 100% for all variables.

Table 6. Mean inter-observer agreement and range between two observers.

<table>
<thead>
<tr>
<th>Exact inter-observer agreement</th>
<th>Mean (%)</th>
<th>Range (%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Transcriptions</td>
<td>90</td>
<td>50-100</td>
</tr>
<tr>
<td>Ratings</td>
<td>84</td>
<td>80-88</td>
</tr>
</tbody>
</table>

Statistical analysis
Descriptive statistics with mean values and ranges were calculated. The Statistical Package for Social Sciences, IBM SPSS, version 20 was used. Difference in the PCC-A measure between groups was analyzed with independent t-test. Kolmogorov-Smirnovs test was not significant (Z=.64, p=.81) and thus revealed normal distribution. Crosstabs and Cramer’s V were used for nominal data (active speech deviances), and Mann-Whitney U-test for ordinal data (passive speech deviances). Spearman rank correlation test or Kendall’s tau_b was carried out between speech outcome variables at 3 years and the dependent variables level of early intervention, early speech production and estimated need for treatment. A p-value of < .05 was considered significant.

RESULTS
Articulation skill
For the whole group the overall mean PCC-A in transcriptions was 68% (range 8 – 100). Of the 26 studied children 9 (35%) had a higher PCC-A than 80% (range 85-100), whereas 17 (65%) proved to have a PCC-A lower than 80% (range 8-79%). Moreover, four of these exhibited a very low PCC-A (8-36%).
There was no significant difference in PCC-A between the two groups with different operation methods ($t = 1.514$, $df=24$, $p = .143$)

Active speech deviations
Two types of deviant articulation were noted in 12 children (46%): glottal articulation in one child (8.3%), retracted oral articulation in 10 children (83.3%), and in one child who used both glottal and retracted oral articulation (8.3%). (Table 7). There was no significant correlation found between retracted oral articulation (Cramer’s $V = .012$ $p = .951$) and the operation method. Also when glottal and retracted oral articulation were regarded as one category of compensatory articulation there was no significant correlation.

Table 7. Compensatory articulation in relation to operation method.

<table>
<thead>
<tr>
<th></th>
<th>Compensatory articulation</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>No</td>
<td>Yes</td>
</tr>
<tr>
<td>Op 1</td>
<td>8</td>
<td>7</td>
</tr>
<tr>
<td>Op 2</td>
<td>6</td>
<td>5</td>
</tr>
<tr>
<td>Total</td>
<td>14</td>
<td>12</td>
</tr>
</tbody>
</table>

Passive speech deviations
Figure 1 a-c, shows the perceptual assessment of hypernasality, audible nasal emission of air, and perceived velopharyngeal function at 3 years of age distributed by operation method. Twenty (77%) of the children had a normal or mildly hypernasal resonance, and six (23%) moderately or severely hypernasal resonance. In 22 (85%) of the children there was no audible nasal emission, or occurring only sometimes, whereas it occurred often in four children (15%). The velopharyngeal function was perceptually assessed as competent or mildly reduced in 20 (77%) of the children and moderately reduced in six (23%). Hyponasality was rated as normal in 23 children (88%) and mild in three children (12%). No statistically significant difference in the passive speech deviations in relation to operation technique was found (hypernasality: $Z = -.139$, $p = .889$; nasal emission of air: $Z = -.905$, $p = .366$; velopharyngeal function: $Z = -.452$, $p = .651$).
Figure 1. a) Prevalence of presence of perceived hypernasality, b) audible nasal emission of air, and c) velopharyngeal function in 26 children born with UCLP in relation to operation method 1 or to operation method 2, rated on a 4-step rating scale.

Speech at 3 years and level of early intervention
Half (n=13) of the children had received EI on level 1 which meant a few reviews with general advice and counseling to parents, about stimulation of oral anterior articulation. Reviews and counseling to parents on level 2 with more specific and individually tailored advice about stimulation of oral anterior articulation and oral motor activities, combined with some SLP therapy sessions had been provided to 11 children. Intensive EI on level 3 with SLP therapy sessions, combined with specific parental counseling, more than 10 times had been provided to two children (table 7).

Table 7. Level of intervention between 18 months and 3 years to 26 children born with UCLP.

<table>
<thead>
<tr>
<th>Level of intervention</th>
<th>Number of patients</th>
</tr>
</thead>
<tbody>
<tr>
<td>Level 1</td>
<td>13 (50%)</td>
</tr>
<tr>
<td>Level 2</td>
<td>11 (42%)</td>
</tr>
<tr>
<td>Level 3</td>
<td>2 (8%)</td>
</tr>
</tbody>
</table>

Of the studied group of children nine (35%) had a PCC-A of 80% or higher. EI on level 1 was provided for six of them and on level 2 for three of them. Seventeen children (65%) had a PCC-A lower than 80%. Intervention on level 1 was provided for seven of them and on level 2 and 3 for ten. No significant correlation was found between level of EI between 18 months and 3 years and PCC-A at 3 years of age (rho = -.210, p = .197).

Nor was there any significant correlation between the active errors glottal articulation (rho = -.02, p = .914) or retracted oral articulation (rho = .287, p = .139) at age 3 years and the level of EI.

Basis for decision on intervention
The median number of consonant types at 18 months was 5 (range: 2-10) and the median number of true consonants was 4.5 (range: 2-10). There were no significant correlations between the variables of consonant production at 18 months and the level of EI (rho = .287, p = .155; rho = .257, p = .205).

Data from the parallel project revealed that sixteen of the children were at 3 years of age estimated to need treatment, which was significantly correlated with PCC-A at the same age (Kendall’s tau_b = .663, p< .000).

Background characteristics for individuals with low performance
Four children exhibited very low PCC-A values (8 – 36%). When scrutinizing the records for possible common traits it was found that all four children had been judged as “in need for treatment” at the age of 3 years. Three of them had received EI at level 2 or 3, whereas one had only had a couple reviews at level 1 in the time span between 18 months and 3 years of age. Three of the children had deviant articulation, either retracted oral or glottal or, as in one case, both. The child with the lowest PCC-A (8%) had four consonant types at the age of 18 months and only two
true consonants. Three out of the four children had ventilation tubes in the eardrums at the age of 3 years.

DISCUSSION
The aim of this study was to investigate speech at age 3 years in a group of children born with UCLP, operated with either a one-stage or a two-stage palatal surgery and the possible influence of EI between 18 months and 3 years of age. As little is known about the applied procedures, as well as the effect of EI, it was considered important, in order to provide a basis for future evaluation, to gain knowledge of the type and extent of EI being performed. The study showed that 65% of the 3-year olds had a PCC-A lower than 80%, which has been used as cut-off score for 3-year-olds (Klintö et al., 2013b). Furthermore, there was a presence of moderate to severe hypernasal resonance and perceptually assessed VPI in 23% of the children with CLP indicating that this group of children is more burdened altogether with limitations in their speech ability than typically developing children without CLP.

Deviant articulatory patterns in 3-year-old children born with cleft lip and palate have been reported in approximately 50% in several previous studies (Morris & Ozanne, 2003; Chapman et al., 2008; Lohmander & Persson, 2008; Willadsen, 2012; Klintö et al., 2013a). This corresponds quite well with the findings in the present study, as 46% of the children had deviant articulation. The deviant articulation pattern for the children in this study was either glottal, retracted oral or both. Other researchers have also found that the place of articulation differs between infants with and without CLP but with not entirely consistent findings in the different studies. The results showed no significant difference in the speech results when comparing the two operation methods, which is in agreement with Klintö et al. (2013a). Similar problems and poorer production compared to typically developing children at 3 years of age have also been observed and reported previously regardless of surgical method used for primary palatal repair (Morris & Ozanne, 2003; Chapman et al., 2008; Lohmander & Persson, 2008; Willadsen, 2012). However, children with an unrepaired cleft in the hard palate at age 3 years have been demonstrated to have poorer speech at age 3 compared to children with palatal repair in one or two stages before or at around 12 months of age in these studies.

Somewhat unexpected, no relationship was found in the speech production in the children at 3 years of age and the received frequency and type of EI in the time span between 18 months and 3 years. Half of the children and their parents had received EI in the form of a few reviews with general advice and counseling, about stimulation of oral anterior articulation. Even though it was difficult to determine from the records how and what type of exercises the children actually had undergone during the time span between 18 months and 3 years, it was clear that both children with normal and deviant articulation patterns had received EI. A belief among SLP’s that EI can prevent undesired speech deviations to develop, as hypothesized by Dixon-Wood (1997), could be a possible reason for the EI given to children with normal articulation as well. It has also been claimed that behaviourally based intensive EI programs are necessary in order to change already established behavior (Henningsson, 1982), which has led to established clinical SLP routines for all
children at our center. Parental worry and insecurity of what to expect in typical child speech and language development might also have motivated the given EI. The routines with EI have thus been well intentioned but unfortunately not evidence based. The same finding was recently reported from a survey of 79 children treated by the Stockholm Craniofacial team, i.e., both children with and without typical articulation patterns had received EI (Raud Westberg & Lohmander, 2011; 2012). In that study it was concluded that the basis for decision about early intervention was unclear, which holds true for the present as well.

Regarding effectiveness of EI, certain models for intervention might be relevant. Scherer & Kaiser (2007), Scherer et al. (2008b), and Hardin-Jones & Chapman (2008) have shown that stimulation of vocabulary and of sound production gives a reduction of compensatory articulation. Furthermore, parents and maybe also preschool teachers are a resource to take into account for delivering training at home or in kindergarten. Scherer et al. (2007; 2008b) has, for instance shown that mothers can be trained to deliver intervention reliably. The so called naturalistic methods using natural routine and play situations in the child’s everyday life, are already practiced in speech and language stimulation programs at our center. Greenberg & Weitzman (2002) in the so called Hanen method, also discuss the importance of a small child’s closest family environment for positive learning success. More studies will show if these are successful in order to prevent established speech deviances at a later age in children with CLP and in addition, the basis for decision needs to be clarified.

In contrast, the SLP opinion about need for treatment at the age of 3 years seemed well grounded in speech production at 3 years of age for this group. There thus seems to be good agreement among SLP’s on what in a child’s speech needs to be taken into account and needs to be attended to. Whether effective speech therapy approaches are carried out is an issue for further research (Bessel et al, 2013).

LIMITATIONS AND STRENGTHS
Unfortunately, there were some serious limitations in this study but also important strengths. Large study groups are always to prefer, but not always possible to obtain in this patient group at a specific center during a limited time span. Ten children in this study had to be excluded, which presumably influences the power of the results. Four children were excluded due to poor co-operation or too small speech samples, a fact that very seldom is possible to foresee or influence. The remaining six exclusions though, were due to technical reasons and could possibly have been prevented with better documentation routines and equipment handling skills. It has been shown in several studies that hearing in children born with cleft palate can be affected. The incidence of otitis media with effusion (OME) and related mild to moderate hearing loss is high among children born with cleft palate (Flynn et al., 2009). For this group of children it was not possible to extract hearing status for comparison since the clinical routines were not consistent at that time and data could therefore not be obtained before the age of 5 years.

Data about the articulation and speech and language development was incomplete in some records, as well as information about treatment contents. A calibration among SLP’s of what data is compulsory in patient records is called for.
Both intra- and inter-observer reliability was good, which might be related to the preparation of the assessment. Before this study was carried out thorough calibration exercises were performed between the two observers.

There was a presence of moderate to severe hypernasal resonance and perceptually assessed VPI in 23% of the children. Three years, though, is a quite low age with difficulty to make secure statements about velopharyngeal function, i.e., whether due to a continued structural deficit or to the developmental level of the child. Follow up studies of speech in the same group, are therefore of great interest.

CONCLUSION AND CLINICAL IMPLICATIONS
Based on the results from these assessments it can be concluded that the speech results at the age of 3 years of children with UCLP correspond with other studies, and that the children performed poorer compared to what has been reported on typically developing peers. Interestingly, there was no significant difference in speech results at age 3 years depending on if the cleft palate had been closed in one or two stages up to 12 months of age.

There was no indication that EI had had any impact on speech production when assessed at 3 years of age and no clear basis in the early consonant production was found used for decisions about level of EI. However, the SLP opinion of “need for treatment” at 3 years of age was well grounded in speech production and can be relied on.

General reconsideration of routines in order to obtain more efficient working models is needed. Measures to address issues such as standardization of data collection and documentation have already been initiated. Changes in EI strategies have also been introduced. Information and advice as well as speech therapy delivered in small groups of children with their parents observing and doing follow-ups in the home have been tried with preliminary promising clinical results. Parent group sessions (Pepper & Weitzman, 2004) and pre-school teacher sessions (Greenberg & Weitzman, 2002; Greenberg, 2006) using the Hanen method for intervention is offered. Future evaluation will show the effectiveness of these new routines.
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