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## **The effect of maxillary advancement on articulation of alveolar consonants in cleft patients**

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## Abstract

*Introduction:* Maxillary advancement may affect speech in cleft patients. The aim of this study was to evaluate the effect of maxillary advancement on Finnish alveolar consonants /s/, /l/, and /r/ in cleft patients.

*Materials and methods:* Fifty-nine Finnish-speaking nonsyndromic cleft patients, (~~35 females, 24 males~~) with CP (~~n=12~~), UCLP (~~n=31~~) and BCLP (~~n=16~~) who had undergone Le Fort I or bimaxillary osteotomies, were evaluated retrospectively. Production of the Finnish alveolar consonants /s/, /l/, and /r/ was assessed from pre- and postoperative standardized video recordings by two experienced speech pathologists. McNemar's test was used in the statistical analyses. Kappa statistics were calculated to assess reliability.

*Results:* The patients included 35 females and 24 males with CP ( $n = 12$ ), UCLP ( $n = 31$ ), and BCLP ( $n = 16$ ). There was a significant improvement in /s/ and /l/ sounds after maxillary advancement ( $p = 0.039$  and  $p = 0.002$ , respectively). The preoperative mean percentage of /s/ errors was 34%; postoperatively it was 20%. /L/ was misarticulated preoperatively by 34% of the patients and postoperatively by 19%. /R/ was misarticulated preoperatively by 47% of the patients and postoperatively by 42%. The level of mild articulation errors rose from 25% to 31%, while severe articulation errors decreased from 37% to 25%. The reliabilities were good.

*Conclusion:* When planning orthognathic surgery in cleft patients with maxillary retrusion and articulation errors, advancement of the maxilla might be a means for improving articulation of /s/ and /l/.

Key words: cleft palate, cleft lip and palate, maxillary advancement, articulation, osteotomy, speech

## Introduction

Patients with cleft lip and palate are at risk of developing maxillary hypoplasia and subsequent retrusion, which can affect not only appearance and occlusion but also speech and self-esteem. Speech sounds requiring bilabial, labiodental, linguodental, and alveolar production can be difficult to produce when the relationship of the jaws, teeth, and tongue is not optimal. Laitinen et al. (1999) studied alveolar consonant production in 260 Finnish non-syndromic cleft children aged 6–8 years. The patients represented all cleft types. The patients who misarticulated /s/, /l/, or /r/ sounds had crossbites significantly more often (73%) than subjects who correctly produced /s/, /l/, or /r/ (45%).

Cleft patients who develop maxillary retrusion often require maxillary advancement with Le Fort I osteotomy. According to previous studies the lowest need (13%) for Le Fort I osteotomy maxillary advancement is found in patients with CP (Antonarakis et al., 2015). In patients with UCLP or BCLP the need for maxillary advancement is significantly higher (40–76.5%) but this has varied greatly in recent literature (Daskalogiannakis and Mehta, 2009; Heliövaara and Rautio, 2011; Heliövaara et al., 2013).

Maxillary advancement may affect velopharyngeal function negatively and cause velopharyngeal insufficiency (Epker and Wolford, 1976; Witzel and Munro, 1977; Watzke et al., 1990; Haapanen et al., 1997; Maegawa et al., 1998; Trindade et al., 2003; Heliövaara et al., 2004; Janulewicz et al., 2004; Niemeyer et al., 2005; Pereira et al., 2013; Alaluusua et al., 2019). On

the other hand, maxillary osteotomy can improve articulation as occlusion and the relationships between the tongue and dental arches improve. In several studies, articulation has improved after maxillary advancement in cleft patients (Swarz and Gruner, 1976; Hagberg et al., 2019) and in noncleft patients (Witzel et al., 1980; Ruscello et al., 1986). However, in the study by Dalston and Vig (1984) on 40 noncleft patients articulation did not improve after maxillary advancement. In a review article by Pereira et al. (2013) the authors concluded that both perceptual and acoustic evidence on the impact of maxillary advancement on articulation are currently based on a noncleft clinical population. In the Finnish language most typical misarticulations are the alveolar sounds /s/, /l/, and /r/, making up 95% of all errors at the age of 7 years, and /l/ articulation error only 1% (Luotonen, 1995).

The aim of this study was to evaluate the effect of maxillary advancement on Finnish alveolar consonants /s/, /l/, and /r/ in cleft patients. The hypothesis was that patients who have maxillary retrusion and alveolar articulation errors would benefit from maxillary advancement and that the alveolar articulation errors would improve after maxillary advancement.

### **Materials and methods**

This retrospective study focused on 59 Caucasian patients with nonsyndromic cleft lip and palate or with isolated cleft palate who underwent a Le Fort I maxillary advancement to correct midfacial retrusion and crossbite at the Cleft Palate and Craniofacial Center (Cleft Center), Department of Plastic Surgery, Helsinki University Hospital between 2006 and 2016. Patients were operated on by two experienced, high-volume cleft surgeons (Board certified; over 10 years of experience; more than 30 operations per year). The osteotomies were grafted using bone from the iliac crest and were fixed with titanium plates. Five patients underwent an early osteotomy

during growth, and 18 patients underwent bimaxillary osteotomies. All patients had orthodontics with fixed appliances during the treatment.

~~The sample consisted of 35 female and 24 male patients. Patients' ages ranged from 11.5 to 45.3 years, with a mean of 17.9 years (SD 4.2). The cleft type distribution was as follows: 12 patients had palatal clefts (11 isolated cleft palate (CP) and 1 operated submucous palate (SMCP) and 47 had clefts of the lip and palate (31 UCLP and 16 BCLP).~~ The study excluded patients with the following characteristics: distraction was used to advance the maxilla; postoperative evaluation was missing; the cleft was part of a syndrome; the patient had severe dyspraxia; the patient was not able to read; the patient had a facial cleft. The patients included in the study all spoke Finnish as their mother tongue. The patient data according, to cleft type and gender, are given in Table 1.

The study protocol was approved by Helsinki University Hospital (§ 27/2015). The study did not fulfil characteristics of a medical study according to the Medical Research Act and did not need ethical permission. Principles outlined in the Declaration of Helsinki were followed.

### *Speech analysis*

Production of the sounds /s/, /l/, and /r/ was evaluated perceptually, from 59 pre- and postoperative video recordings, by two experienced speech pathologists from the cleft team. The preoperative video recordings had been done immediately before Le Fort I treatment and the postoperative ones 6–12 months (mean 8.7 months, SD 4.4) after the procedure. Video recordings were used instead of audio recordings so that the evaluators could not only perceive the errors audibly, but also see if the location of articulation was compromised.

Articulation of the sounds /s/, /l/, and /r/ was evaluated using 30 separate words, seven sentences, and a reading sample. The Finnish naming test originally designed for the Scandleft project (Lohmander et al., 2009) was used when evaluating separate words and sentences. The reading sample was a text (Vaahteranmäen Eemeli) routinely used to document connected speech at our center. This text contained 61 words and took approximately 30 seconds to read.

Articulation errors in the sounds /s/, /l/, and /r/ were evaluated and categorized as either *correct*, *distortion*, *substitution*, or *omission*. Errors in articulation were further classified as *mild* or *severe*. Substitutions and omissions fell automatically into the severe group. Distortions were mild if the location of articulation altered but the acoustical signal did not. /R/ was also classified as a mild articulation error in cases where the location of articulation did not change but the manner changed from trill to fricative or flap. Misarticulation was severe if location or manner of articulation changed and affected the perceived acoustic signal. Articulation errors are presented in more detail, with International Phonetic Alphabet (IPA) characters, in Appendix 1. The symbols used in the IPA character picker can be accessed at <http://schwa.dk/filer/ipacharpick/>. To increase reliability intra- and interrater agreements were calculated for the completed evaluations. The assessment was carried out separately in a quiet room, where the speech therapists listened to the samples through high-quality headphones (Creative Aurvana Live, Creative Technology Ltd, Singapore).

### *Cephalometric analysis*

Standardized lateral cephalometric radiographs were taken, with the head positioned in alignment with the Frankfort horizontal plane, and with molar teeth occluded and lips in repose. The radiographs were taken before the operation ( $x = 4.5$  months,  $SD = 4.1$ ) and 6–12 months ( $x$

= 7.2 months, SD = 2.7) after the operation. The cephalograms were traced by the same investigator using the Dolphin cephalometric program (Dolphin Imaging 11.95 Premium). To differentiate between horizontal and vertical changes, the cephalometric program uses an  $x$ - $y$  coordinate system. The subsequent cephalometric tracing was superimposed by a Sella-Nasion (SN) plane. Changes in maxillary position were calculated using changes in the value of cephalometric point A (the most concave point of the anterior maxilla). A positive value for horizontal advancement represented anterior movement of the maxilla; a negative value, posterior movement. In the vertical direction, a positive value represented movement of the maxilla to a more cranial position, while a negative value meant movement to a more caudal position.

#### *Statistical analyses*

Pre- and postoperative speech changes were tested statistically using McNemar's test. Kappa statistics were calculated to assess reliability of the speech assessment and the cephalometric digitalization. A statistician not involved in the study performed the analyses. The patients were blinded for both the speech evaluation and statistical analyses.

## **Results**

The sample consisted of 35 female and 24 male patients. Patients' ages ranged from 11.5 to 45.3 years, with a mean of 17.9 years (SD 4.2). The cleft type distribution was as follows: 12 patients had palatal clefts (11 isolated cleft palate (CP) and 1 operated submucous cleft palate (SMCP)) and 47 had clefts of the lip and palate (31 UCLP and 16 BCLP).



Using McNemar's test we found a statistically significant improvement in the production of alveolar sibilant /s/ and alveolar lateral approximant /l/ following maxillary advancement ( $p = 0.039$  and  $p = 0.002$ , respectively). The improvement in Finnish alveolar thrill was not statistically significant. Preoperatively, 63% (37/59) of the patients misarticulated 1–3 of the alveolar sounds /s/, /l/, or /r/.

/S/ was preoperatively misarticulated by 34% (20/59) of the patients. After maxillary advancement the percentage of misarticulated /s/ sounds was 20% (12/59). Two patients with BCLP had a normal /s/ articulation preoperatively and a mild articulation error in /s/ postoperatively. The /l/ sound was misarticulated preoperatively by 34% of patients and postoperatively by 19%. /R/ was misarticulated preoperatively by 47% (28/59) of patients and postoperatively by 42% (25/59) (Table 2).

Four of the patients' mild articulation errors for 1–3 alveolar consonants no longer existed after maxillary advancement. None of the severe articulation errors was rendered normal postoperatively. The level of mild articulation errors rose from 25% to 31%, while severe articulation errors decreased from 37% to 25% (Table 3). The sample was too small for statistical evaluation of differences between cleft types.

In total, 15 patients (five CP, six UCLP, four BCLP) had undergone corrective speech surgery before undergoing a Le Fort I osteotomy. Among these, 11 patients (three CP, six UCLP, two BCLP) had a velopharyngeal flap. Three patients (two CP, one BCLP) underwent Furlow's repalatoplasty and two patients with BCLP underwent two speech corrective surgeries — Furlow's repalatoplasty and a velopharyngeal flap.

The mean maxillary advancement of the cephalometric A point was 4.6 mm horizontally (range: -2.8 to 11.3) and -3.9 mm vertically (range -14.2 to 3.9). Reliability of the tracings was

determined by retracing and redigitalizing later radiographs of 20 randomly selected patients from our material. The values of the two tracings were compared using Kappa statistics.

Intrarater reliability was excellent (point A horizontally 0.934, point A vertically 0.830).

The perceptual re-evaluations completed by two speech pathologists were compared against each other, and the intra- and interrater reliabilities were calculated. These were found to vary between good and excellent agreement (inter 0.640–0.651; intra 0.766–0.792 and 0.641–0.709).

## **Discussion**

A positive impact on articulation was seen with the alveolar /s/ and /l/ sounds; improvements on both were statistically significant. The number of articulation errors was reduced and there was qualitative improvement towards a more normal articulation. The alveolar trill /r/ was not significantly improved.

### *Articulation errors*

Preoperatively 37/59 patients showed /s/, /l/, or /r/ articulation errors. Thirty-eight per cent (14/37) of these patients experienced improvement in their articulation. Previous studies have concluded that the sibilant sounds, including /s/, are particularly vulnerable to occlusional defects (Witzel et al., 1980; Ruscello et al., 1986; Kummer et al., 1989; Vallino et al., 1993; Lee et al., 2002; Hagberg et al., 2019). Estimates of the number of patients with occlusal problems who experience articulation impairment without a cleft vary between 50% and 60% (Witzel et al., 1980; Ruscello et al., 1986; Whitchill et al., 2001; Lee et al., 2002).

The Finnish language has only one /s/ phoneme. It is an unvoiced alveolar sibilant produced by the tip of the tongue forming a narrow constriction against the medioalveolar area. Forty percent (8/20) of the patients who preoperatively misarticulated /s/ were normalized after maxillary advancement. Two patients who had a normal /s/ preoperatively developed a significant VPI after maxillary advancement and an /s/ with nasal emissions. According to Laine et al. (1987), 16% of Finnish adults misarticulate /s/. That level of /s/ misarticulation is quite close to the status found after maxillary advancement (20%).

Hagberg et al. (2019) explored lay listeners' perception of articulatory change in Swedish cleft adults ( $n = 15$ ; seven UCLP, eight BCLP) post Le Fort I. In most cases, the lay listeners perceived no difference in accuracy between the /s/ sounds recorded preoperatively and postoperatively. On the other hand, two trained speech and language pathologists ascertained a significant positive change in the production of /s/ after maxillary advancement. This finding was supported by a statistically significant correlation with the results of acoustic analysis. The authors concluded that adult lay listeners do not notice differences in accuracy, or that the degree of distinctiveness in the /s/ sound in words is not crucial to understanding the word. In the study by Nyberg and Havstam (2016), 19 10-year-olds without a cleft listened to speech samples with different types of cleft speech characteristics assessed by speech and language pathologists and described what they heard. Interestingly, the 10-year-old children reacted to even minor articulatory difficulties in cleft peers and especially to distorted /s/ sounds.

The Finnish /l/ is a voiced alveolar lateral approximant similar to the English /l/. /L/ articulation errors in a normal adult population are somewhat nonexistent, with only 1% of the adult population misarticulating it (Laine et al., 1987). In our study 34% (20/59) misarticulated /l/ preoperatively and only 19% (11/59) postoperatively. Forty-five per cent (9/20) of /l/ articulation

errors were improved postoperatively. Kummer et al. (1989) also studied the effect of maxillary advancement on cleft patient articulation, but only one patient in their study of 16 patients misarticulated /l/ preoperatively. This one patient's /l/ was improved postoperatively.

The trilling /r/ is one of the most difficult sounds among the world's languages. Of the three sounds assessed in our study, /r/ was the only one not significantly improved after maxillary advancement. After maxillary advancement, 42% (25/59) of the cleft patients still misarticulated /r/. Three patients had a mildly distorted /r/ preoperatively — a fricative that was evaluated as normal postoperatively. No previous study has evaluated the change in the alveolar trill and for that reason comparison against previous studies is not possible. In a Finnish noncleft population of young adults, 3% had articulation errors in /r/ (Laine et al., 1987).

The degree of social burden of patients depends on whether errors are mild or severe. In four patients mild articulation errors were cured, while after maxillary advancement 18/59 (31%) patients exhibited mild articulation errors. Of the 22 patients who had severe articulation errors preoperatively, six were postoperatively evaluated as having mild articulation errors. However, the ability to compensate may vary individually.

### *Methodology*

The differences between results of previous studies may partly be explained by different evaluation methods and sample sizes (Chancareonsook et al. 2006, Pereira et al. 2013). Audio recordings, video recordings, and live evaluations have been used in the evaluation of articulation. In the study by Hagberg et al. (2019) the mean percentage of correct oral consonants in 15 cleft patients was 82% preoperatively. When the sounds were evaluated separately, the

mean correct articulation of /s/ was 34% preoperatively, while the postoperative value was 85%. Hagberg et al. (2019) used audio recordings and therefore did not evaluate visual misarticulations. We used video recordings to be able to evaluate visual, acoustic, and combined misarticulations. The downside to a video recording is the possibility of noticing whether maxillary advancement has already been performed. Our study was a perceptual study. Perceptual measurement is still considered an essential part of speech evaluation. Kim et al. (1999) found that perceptual rating of sounds correlated well with objective measurements of sound spectrometry.

Variable sample size might explain some differences in the results of previous studies. This issue was recognized by Chanchareonsook et al. (2006) and Pereira et al. (2013). Okazaki et al. (1993) evaluated the articulation of 10 cleft patients before and after maxillary advancement. In their study articulation after maxillary advancement did not improve. Kummer et al. (1989) evaluated the articulation of 16 patients: eight with a cleft (seven UCLP, one CP) and eight without. Eleven patients with preoperative articulation errors improved postoperatively.

A strength of our study is the fairly large sample size of cleft patients. The review articles have also emphasized the need for inter- and intrarater reliability tests, which in our study indicated good agreement and reliability. The timing of postoperative evaluation seems to be critical. Lee et al. (2002) evaluated acoustic and perceptual characteristics of the /s/ sound before and after orthognathic surgery in nine noncleft subjects. According to the study there appeared to be a positive change 3 months after surgery, but there was a 'relapse' towards the presurgical production of /s/ at 12 months after surgery. We evaluated speech 6–12 months after Le Fort I surgery. The amount of maxillary advancement was moderate, although the individual variation was large. A more specific analysis of the occlusion could have generated more information

about the effect of maxillary advancement on alveolar consonants in cleft patients.

Unfortunately, the sample sizes for different types of cleft were too small and we were not able to evaluate differences between the cleft groups.

### **Conclusion**

When planning orthognathic surgery in cleft patients with articulation errors, it should be considered that maxillary advancement may improve articulation of /s/ and /l/. If speech therapy is needed, it may be more effective after maxillary advancement.

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### **Conflicts of interest statement**

The authors have no conflicts of interest to disclose.

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**Table 1.** Patients by cleft type and gender ( $n = 59$ )

<b>Cleft type</b>	<b>Males</b>	<b>Females</b>	<b>Total</b>
<b>CP/SMCP</b>	2	10	12
<b>UCLP</b>	15	16	31
<b>BCLP</b>	7	9	16
<b>Total</b>	24	35	59

**Table 2.** Number of patients with pre- and postoperative articulation errors for /s/, /l/, /r/

	<b>CP (n = 12)</b>		<b>UCLP (n = 31)</b>		<b>BCLP (n = 16)</b>		<b>Total (n = 59)</b>		<b>Total (n = 59)</b>	
<b>Error</b>	Pre n	Post n	Pre n	Post n	Pre n	Post n	Pre n	Pre %	Post n	Post %
<i>/s/</i>	5	2	9	6	6	4	20	34	12	20
<i>/l/</i>	6	3	8	5	6	3	20	34	11	19
<i>/r/</i>	5	3	12	11	11	11	28	47	25	42

**Table 3.** Number of patients with normal, mild, and severe articulation errors pre- and postoperatively

	<b>CP (n = 12)</b>		<b>UCLP (n = 31)</b>		<b>BCLP (n = 16)</b>		<b>Total (n = 59)</b>		<b>Total (n = 59)</b>	
<b>Error</b>	Pre n	Post n	Pre n	Post n	Pre n	Post n	Pre n	Pre %	Post n	Post %
<b>normal</b>	3	6	14	16	5	4	22	37	26	44
<b>mild</b>	5	3	7	9	3	6	15	25	18	31
<b>severe</b>	3	3	9	6	10	6	22	37	15	25