

ORIGINAL RESEARCH ARTICLE

## Long-term speech outcome after anterior distraction osteogenesis of the maxilla in patients with cleft lip and palate

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### ABSTRACT

Advancement of the maxilla may increase the distance between the soft palate and the posterior pharyngeal wall in patients with cleft lip and palate, implying a risk of velopharyngeal dysfunction. The aim was to evaluate long-term speech outcome in a consecutive series of patients treated with distraction osteogenesis (DO). Fourteen out of the 16 patients agreed to participate. A long-term speech follow-up was performed 1.5 to 13.5 years after DO. For two participants, audio recordings before DO were missing, and for another one, it was incomplete. The percentage of consonants correct (PCC) based on phonetic transcription and perceived velopharyngeal competence rated on a three-point scale were assessed before and after DO by three independent judges, based on audio recordings of reading of standardised sentences. Also, the participants were asked how they perceived their speech after DO. Changes in PCC were insignificant. Four participants perceived deteriorated speech related to DO. In two cases, the subjective deterioration did not correlate to results from perceptual assessment. In two others, the subjective deterioration correlated with the perceptual assessment, and the velopharyngeal function was judged as being incompetent after DO. After secondary velopharyngeal surgery, velopharyngeal function improved to competent in one case and marginally incompetent in the other. The results need to be interpreted with caution due to methodological limitations but indicate that some patients develop deteriorated velopharyngeal function after DO. The impact on articulation needs to be further explored. It is important that patients are informed before treatment of the risk of velopharyngeal dysfunction after DO.

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### Introduction

In Sweden, about 175 children are diagnosed with some type of cleft lip and palate every year [1]. They are treated at one of the six Swedish cleft lip and palate centres, from birth up to adulthood. The cleft lip and palate centre at Skåne University Hospital handles approximately 30 new patients with some type of cleft lip and palate each year, and this corresponds to a catchment area of 1.7 million inhabitants. The basic principle for decades has been that lip plasty is performed at 3 to 9 months of age, and primary palatal surgery in one stage at 12 to 18 months. The residual cleft in the alveolar ridge is closed by a cancellous bone graft in the mixed dentition, at seven to 11 years of age. In cases of persisting velopharyngeal dysfunction (i.e., inability to achieve sufficient closure between the oral and nasal cavity during the production of oral speech sounds), secondary speech-improving velopharyngeal surgery is performed when needed. In patients with maxillary hypoplasia, conventional orthognathic surgery or distraction osteogenesis (DO) in the more severe cases is used for the advancement of the maxilla when needed.

To allow fully developed bone growth and to achieve a long-lasting result, surgical maxillary advancement is usually performed in the late teens [2]. Conventional orthognathic surgery and DO are based on Le Fort I osteotomy, to make the maxilla freely movable in the movement forward. In the conventional orthognathic surgery, the maxilla is advanced in one step with rigid fixation, sometimes in

combination with bone transplantation [3]. The disadvantage with conventional orthognathic surgery is that only limited advancement is possible, about 5 mm, and there is a risk of relapse [4]. DO has been used in orthognathic surgery in humans since the 90s [4]. It implies a mechanical bone lengthening procedure that generates new bone with gradual distraction through Le Fort I osteotomy, supplemented with a rigid external distractor [5,6]. The whole procedure takes about 3 months [3,4,7].

Previous research has highlighted that advancement of the maxilla may increase the distance between the soft palate and the posterior pharyngeal wall in patients with cleft palate with or without cleft lip [4]. This implies a risk of velopharyngeal dysfunction that may affect speech, with a postoperative need of secondary velopharyngeal surgery [4]. The risk for deteriorated velopharyngeal function has been shown to be higher in patients who have signs of velopharyngeal dysfunction already before maxillary advancement [8]. However, there seems to be no correlation between the amount of maxillary advancement and postoperative speech outcome [9–12]. In a Swedish study on speech and velopharyngeal function in patients with cleft lip and palate treated with conventional maxillary advancement, it had no significant impact on velopharyngeal function on group level, but three out of 13 patients had speech symptoms of deteriorated velopharyngeal function to some degree postoperatively [12]. On the other hand, since maxillary retrognathia

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may impact lip and tongue positions negatively, advancement may in some cases improve the articulation of anterior consonants [13–15]. In a Swedish study, on impact of conventional maxillary advancement on consonant proficiency in patients with cleft lip and palate, 11 out of 15 patients had improved articulation postoperatively, especially on the /s/-sound [15]. In studies comparing speech outcome in groups treated with conventional orthognathic surgery and DO, no group differences have been seen [9,10].

DO has been carried out at Skåne University Hospital since 2004. The initiative for the present project was taken in 2017, when the basis for preoperative patient information was found to be insufficient. The first consecutive series of patients treated with DO of the maxilla was identified. In five parallel studies, long-term results after DO concerning the patient-reported perspective, speech, skeletal relations, occlusion, and the exterior were assessed. Long-term results after DO concerning the patient-reported perspective have been published previously [16]. The aim of this study was to evaluate long-term speech outcome in a consecutive series of patients treated with DO.

## Material and methods

### Participants

Patients treated with DO since the start of 2004 were invited by ordinary mail to participate in the long-term follow-up, which, in addition to speech recordings, included dental casts, lateral cephalograms, photographs and answering a questionnaire on the patient's perspective of the treatment. Out of the 16 possible individuals, 14 accepted the invitation. Two individuals declined participation because they resided too far away.

Of the 14 participants, eight were females and six males (Table 1). Three were born with bilateral cleft lip and palate, 10 with unilateral cleft lip and palate and one with cleft soft and hard palate only. Two had an additional diagnosis which may affect speech (#1 and #10). The median age of DO was 20.1 (range = 15.7–25.3). Five participants were treated with secondary velopharyngeal surgery before DO (#1, #8, #11, #12 and #13), and one of them twice (#1). This participant was treated twice with secondary velopharyngeal surgery before DO and was treated with velopharyngeal surgery one more time after the long-term speech follow-up. Another two participants were treated

with secondary velopharyngeal surgery after the DO and before the long-term speech follow-up (#7 and #9) (Table 1). In all cases of secondary velopharyngeal surgery, pharyngeal flap surgery was performed.

### Speech documentation

The audio recordings before DO were performed in connection to routine follow-ups at our cleft lip and palate centre. For 10 participants, the preoperative audio recordings were performed about 1 to 5 months before the DO, for two participants, more than 2 years before DO, and for two participants, the preoperative audio recording was missing (#5 and #6) (Table 1). The preoperative audio recordings were performed by speech-language pathologists (SLPs) specialised in cleft palate speech, using an audio recorder (TASCAM HD-P2, California) or a PC with Soundswell software (Saven Hitech, Sweden), and a condenser microphone (Sony ECM-MS957, Japan, or Pearl CC3, Sweden, or Sennheiser MKE 2 P-C, Germany). The speech material in the recordings varied somewhat. However, in all recordings except one (#13), the sentences in the Swedish test for assessment of nasality and articulation (SVANTE) [17] were included. In this study, the participants read the sentences.

In connection to the long-term speech follow-up after DO, speech assessment and documentation was performed in a studio. The time for long-term speech follow-up in relation to time for DO varied from about 1.5 to 13.5 years (Table 1). One SLP performed 13 assessments and another SLP performed one, and both were specialised in cleft palate speech. A PC with Soundswell software (Saven Hitech, Sweden) and a condenser microphone (Pearl CC3, Sweden) was used for documentation. The participants read the words and sentences of SVANTE [17,18], and read and retold a short text. In addition, the participants were asked if they felt that the DO had affected their speech, and if so in what way. Two participants (#7 and #9) were treated with secondary velopharyngeal surgery after the DO. For these two participants, speech was documented before velopharyngeal surgery as well (Table 1), as described earlier.

Since all recordings before and after DO, except one, contained reading of sentences, it was decided to base the speech analysis on the sentences. For one participant, the recording before the DO only included reading of a short text. This recording was used in the perceptual analysis of perceived velopharyngeal competence (VPC),

**Table 1.** Sex, diagnosis, age at distraction osteogenesis surgery (DO), maxillary advancement measured as difference in millimetres (mm) overjet on dental casts before DO and at long-term follow-up, age at secondary velopharyngeal surgery (VS), time at speech assessment (speech ass.) before DO, time at speech ass. after DO in cases where the need of VS was indicated and the time of long-term speech follow-up in relation to DO.

ID	Sex	Diagnosis	Age at DO (years)	Maxillary advancement (mm)	Age at VS (months)	Time of preoperative speech ass. in relation to DO (years)	Time of postoperative speech ass. after DO if the need of VS was indicated (months)	Time of long-term speech follow-up in relation to DO (months)
1	F	BCLP+	24.3	8.5	7.9; 13.7; 27.3	6	-	21
2	F	UCLP	16.3	6.2	-	3	-	138
3	M	UCLP	23.1	9.5	-	46	-	98
4	M	UCLP	21	9.6	-	1	-	96
5	M	BCLP	20.1	10.5	-	N/A	-	153
6	F	UCLP	15.7	9.1	-	N/A	-	145
7	M	UCLP	19.5	8.3	20.3	5	7	32
8	M	UCLP	19.4	10.0	7.6	5	-	85
9	M	UCLP	19.8	17.0	22.9	1	12	61
10	F	BCLP+	21.3	13.0	-	1	-	163
11	F	SHP	25.3	5.6	13.8	3	-	44
12	F	UCLP	20	7.4	7.8	1	-	54
13	F	UCLP	21.4	7.0	6.7	26	-	149
14	F	UCLP	19.5	8.4	-	2	-	30

F: female; M: male; BCLP: bilateral cleft lip and palate; BCLP+: bilateral cleft lip and palate with additional diagnosis that may affect speech; UCLP: unilateral cleft lip and palate; SHP: cleft soft and hard palate; N/A: not available.

but not in the analysis of percentage of consonants correct (PCC). All recordings were saved in .wav format and de-identified and edited using Audacity (Free Software, General Public Licence GPL). Ten randomly chosen recordings were duplicated to allow for assessment of intra-judge agreement.

### Perceptual speech assessment and analysis

Three SLPs, the one who performed 13 long-term assessments of speech and two from two other Swedish cleft lip and palate centres, performed independent perceptual speech analysis of all included speech material. The SLPs' experience of cleft palate speech varied from 6 to 18 years. They used headphones (Creative Aurvana Live, Singapore, or Yamaha HPH-MT7, Germany, or Denon AH-D100, Japan) when listening. The recordings were randomly presented, and the SLPs were blinded to the time of recording. The SLPs transcribed the consonants in the sentences (maximum 32 consonants) with 'semi-narrow' transcription according to the International Phonetic Alphabet [19], using supplement diacritics common in cleft palate speech.

Long-term speech outcome was evaluated with the measures PCC and VPC. The SLPs judged the consonants as correct or not based on the phonetic transcriptions and calculated PCC by dividing the number of correct consonants by the total number of elicited consonants [20]. Active articulatory and phonological errors were scored as incorrect if the phonetic symbol differed from the target phonetic symbol in the sentences of SVANTE [17]. In addition, distorted s-sounds were scored as errors. Passive cleft speech characteristics such as audible nasal air leakage and reduced pressure on consonants were not scored as errors in the PCC analysis [18]. The SLPs also performed an overall rating of VPC, based on the impression of hypernasality, audible nasal air leakage and reduced pressure on consonants, on a three-point scale with the scale values *competent*, *marginally incompetent* and *incompetent* [18].

### Reliability

The results on agreement were interpreted according to Cichetti [21]. Absolute agreement for PCC was calculated with the single measures intra-class coefficient (ICC) using a two-way mixed effects model. Intra-judge agreement ICC values for PCC were excellent and varied between 0.994 and 1, with a 95% confidence interval (CI) between 0.975 and 1. Inter-judge agreement was also excellent (0.967) with a CI between 0.938 and 0.984. In the presentation of the results, the mean PCC score of the three judges for each participant was used.

Intra-judge agreement for VPC as calculated with quadratic Kappa was excellent (1; CI: 1–1) for all three SLPs. Inter-judge agreement for VPC when pairwise comparisons were performed was poor (0.319; CI: 0.037–0.602), moderate (0.552; CI: 0.220–0.884) and good (0.771; CI: 0.338–1). Exact percentage inter-judge agreement when pairwise comparisons were performed was good in one case (85.7%) and poor in two cases (53.6 and 57%). When a difference of one scale value was allowed, it was excellent (100, 96.4 and 100%). In the presentation of the results, the median VPC value of each participant was used. Thus, the values of the SLPs with the closest ratings were used.

### Ethics

This study was approved by the Regional Ethical Review Board in Lund, Sweden (Dnr: 2017/690). All participants gave their written informed consent for participation.

### Results

Eight of 11 participants with available PCC data before and after DO had a PCC score of 100 at both assessments (Table 2). For one participant (#7), the PCC score was 97 before DO, 94 after DO and before secondary velopharyngeal surgery, and 99 at the long-term speech follow-up. For the three participants with missing PCC data before DO (#5, #6 and #13), two had a mean PCC score of 100 after DO, and one had a score of 91. For the two participants with additional diagnoses that may have affected speech (#1 and #10), the PCC scores before DO were 26 and 53, respectively, and at the long-term speech follow-up, they were 25 and 56.

In two cases (#7 and #9), VPC changed from marginally incompetent before DO to incompetent after DO. For these two participants, at the long-term speech follow-up after velopharyngeal surgery, VPC changed to competent for one of them (#9) and to marginally incompetent for the other (#7). In two other cases (#3 and #13), VPC changed from competent before DO to marginally incompetent at the long-term speech follow-up. In one case (#10), VPC changed from marginally incompetent before DO to competent after DO. In the remaining seven cases, there was no change in VPC before DO and at the long-term speech follow-up.

Out of the 14 participants, 11 commented on how they perceived their speech after DO. Four perceived that their speech did not change after DO (#3, #6, #8 and #13), and two (#4 and #11) perceived that their speech improved. Five participants (#2, #7, #9, #10 and #14) perceived that their speech deteriorated, and in two cases (#7 and #9), this corresponded to the VPC scores. In these two cases, secondary

**Table 2.** Speech outcome before distraction osteogenesis (DO), after DO before secondary velopharyngeal surgery (VS) and at long-term follow-up.

ID	Speech before DO		Speech after DO before VS		Speech at long-term follow-up		
	PCC	VPC	PCC	VPC	PCC	VPC	Subjective comment on speech after DO
1	26	1	-	-	25	1	None
2	100	1	-	-	100	1	Deteriorated
3	100	0	-	-	100	1	Unchanged
4	100	1	-	-	100	1	Improved
5	N/A	N/A	-	-	100	1	None
6	N/A	N/A	-	-	100	1	Unchanged
7	97	1	94	2	99	1	Deteriorated after DO; improved after VP surgery
8	100	1	-	-	100	1	Unchanged
9	100	1	100	2	100	0	Deteriorated after DO; improved after VP surgery
10	53	1	-	-	56	0	Deteriorated for other reason than DO
11	100	0	-	-	100	0	Improved articulation of /s/
12	100	0	-	-	100	0	None
13	N/A	0	-	-	91	1	Unchanged
14	100	1	-	-	100	1	Deteriorated

PCC: percent consonants correct; VPC: perceived velopharyngeal competence (0 = competent, 1 = marginally incompetent, 2 = incompetent); N/A: not available.

velopharyngeal surgery was performed, which the participants perceived improved their speech. One of the participants with deteriorated speech (#10) did not associate the deterioration with the DO surgery (Table 2).

Thus, four (33%) of the 12 participants with available speech recordings before and after DO had deteriorated VPC after DO, and in three of the cases (25%), the impairment was stated to be related to the DO.

## Discussion

In this study, long-term speech outcome after DO was followed-up. Between 2004 and 2017, 16 patients had been treated with DO, and 14 agreed to participate in this study. They were born over a period of 17 years from the beginning of the 1980s. In some cases, the assessment and documentation of speech before DO were not standardised, and in some cases, data were missing. Despite these limitations, we consider the results important. Speech and velopharyngeal function in Swedish-speaking patients with cleft lip and palate after conventional maxillary advancement has been evaluated previously [12,15]. However, this is the first study on DO and speech in Swedish-speaking patients. In addition, we have only found one previous published study with a stringent methodology for speech assessment, including assessment of inter- and intra-judge agreement, which has evaluated the speech after DO after 1 year or more [9].

Two of the 14 participants (#3 and #13) were judged as having marginally incompetent velopharyngeal function, and two (#7 and #9) were judged as having incompetent velopharyngeal function after DO. This is in accordance with findings in earlier studies, where some patients had deteriorated speech after DO [9,10], and also with findings in a Swedish study on speech related to conventional maxillary advancement [12]. The risk of deteriorated velopharyngeal function to incompetent has been stated to be higher in patients who already have signs of velopharyngeal dysfunction before conventional maxillary advancement [8]. This is in line with the results of the present study, where both participants with incompetent velopharyngeal function after DO had marginally signs of velopharyngeal dysfunction before DO. No relationship was seen between the amount of maxillary advancement, measured from dental casts, and postoperative speech outcome, which also is in line with the results from previous studies [9–12].

The PCC-measure did not detect any differences in results before and after DO. Looking at specific consonants, one participant (#11) perceived improved articulation of /s/. This was not verified in the PCC results or when reviewing the phonetic transcriptions of this participant. The production of /s/ was judged as correct both before and after DO. In the sentences, only 16% of the consonants consisted of /s/. With a higher occurrence of /s/ in the speech material, for example, by the use of conversational speech for perceptual assessment and documentation with audio–video recordings which may give extra visual cues [22], a difference before and after DO might have been detected. The review of the transcriptions also revealed that one patient (#10) produced /s/ with velar placement, both before and after DO. The rest of the patients produced /s/ with correct placement pre- and postoperatively.

Studies using audio–video recordings for perceptual speech assessment have found articulation of /s/ [13,14], /l/ [13] and /f/ [14] to be improved after maxillary advancement. Furthermore, Hagberg et al. [15] found a significantly improved articulation of /s/ when perceptually assessed from audio recordings. That no articulatory improvement was seen in the present study was likely because few of the patients displayed any articulatory abnormalities in the recorded speech material.

Of the five patients (#2, #7, #9, #10 and #14) who perceived deteriorated speech after DO, one patient (#10) had an additional diagnosis, which was judged to be the cause of the deterioration. The subjective deterioration only correlated with results from perceptual speech assessment in two patients (#7 and #9). How SLPs judge the speech of individuals with cleft lip and palate does not have to correlate with how satisfied the individuals are with their speech [23]. However, the results in the study by Hagberg et al. [15] indicated a relationship between results from perceptual speech assessments and the patients' perceptions of their speech.

Hypernasality is a distinctive symptom in cleft palate speech, but it may be difficult to achieve sufficient agreement in ratings of hypernasality [10], which has been obvious in earlier Swedish studies [24,25]. In the present study, we therefore assessed VPC instead, and the intra-judge agreement was excellent. However, the inter-judge agreement for VPC was poorer than in previous studies [26]. The small group of participants might have contributed to this. We handled this by using the median values, which means that the values of the SLPs with the closest ratings were used. Other limitations of this study were small number of participants, missing data and the varying time-points of follow-up in relation to DO. Thus, the results need to be interpreted with caution.

## Conclusion

Due to methodological limitations, the results of this study need to be interpreted with caution. However, the results are very similar to those after maxillary advancement with conventional Le Fort I osteotomy and indicate that some patients develop deteriorated velopharyngeal function after DO. After treatment, they may need secondary velopharyngeal surgery. Thus, it is important that patients are informed of the risk before treatment. The impact on articulation needs to be further explored.

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## Disclosure statement

The authors have no conflicts of interest to report.

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