Lipofilling in patients with a cleft lip (and palate) – a pilot study assessing functional outcomes and patients' satisfaction with appearance

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Abstract

Introduction: Lipofilling of the upper lip as secondary treatment in patients with a cleft lip (and palate) (CL ± P) has been proposed to improve projection and volume especially in profile view. The purpose of the present study was to document differences in functional (i.e. logopaedic) and self-reported aesthetic outcomes by comparing pre- and postoperative results after lipofilling of the upper lip in patients with CL ± P.

Methods: Eight Dutch-speaking youngsters and young adults (three women, five men) with CL ± P were included. The median age was 19 years (range: 14–24 years). Logopaedic outcomes (i.e. assessment of orofacial myofunctional behavior, articulation and lip strength) and self-reported aesthetic outcome (i.e. patients' satisfaction using the Cleft Evaluation Profile) were determined.

Results: Neither for lip strength and orofacial myofunctional behavior nor for articulation statistically significant differences were found when comparing measurements before and after lipofilling. Regarding patients' satisfaction, a statistically significant increased self-evaluation of appearance in profile was found after lipofilling.

Conclusion: Regarding functionality, the present study did not find any differences when comparing outcomes before and after lipofilling. Nevertheless, patients were more satisfied with appearance in profile after performance of this technique. As this is a small sample study, further research and long-term follow-up studies are necessary.

Keywords

Cleft lip (and palate)
Lipofilling
Functional outcomes
Patients' satisfaction

1. Introduction

A cleft lip with or without a cleft palate (CL ± P) is one of the most common craniofacial birth defects in children [1]. Repair of the lip is performed when the child is around 3 months old [2]. Palatal repair is mostly performed between 6 and 14 months of age [2] although the timing is still subject for discussion [3]. The main goals of surgical reconstruction of the lip are to achieve normal facial appearance and functional reconstruction of the orbicularis muscle necessary for oral competence. Nevertheless, surgical reconstruction may lead to unfavorable results such as scars, which are not always easy to treat [4]. Thereby, reconstructions may influence patients' psychosocial functioning [5] and satisfaction with facial appearance [6]. Turner, Thomas et al. [6] investigated satisfaction with facial appearance among 111 patients with CL ± P with an age of 10, 15 or 20 years old using a subjective ordinal scale (for assessing satisfaction with facial appearance) and the Childhood Experience Questionnaire (for assessing psychosocial adjustment). Results showed that the 20-year-old participants were, on average, significantly more satisfied with their appearance than the 10- ($p = 0.009$) and 15-year-olds ($p = 0.012$). Furthermore, it was found that patients with visible stigmata of cleft lip surgery were significantly more dissatisfied than those without. Hunt, Burden et al. [7] determined self-reported psychosocial effects among children and young adults with CL ± P compared to a control group consisting of children without CL ± P. The study included 160 children with and 113 children without CL ± P between 8 and 21 years old (mean age: 13.6 years). Children with CL ± P reported significantly more behavioral problems and symptoms of depression when compared to children without CL ± P. It was found that children with CL ± P were bullied more often and were less happy with their facial appearance and speech. No statistically significant differences were found in terms of anxiety or self-esteem between both groups. However, having a history of being bullied was a significant predictor of problems with psychosocial functioning. Furthermore, the Eurocleft study [8] showed that 74% of the patients with CL ± P have been teased because of their appearance. A more recent study of Rivera et al. [9] investigated

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parents' satisfaction with repair of cleft lip and palate in Honduras. Pre- and post-operative interviews were taken from parents of 45 patients (mean age: 4 years). Pre-operatively, parents indicated that they hoped for improvement in speech, appearance and/or feeding. Post-operative interviews (i.e. after lip closure or palatal surgery) showed that only 14 of 22 parents had all their pre-operative expectations fulfilled. However, despite unmet expectations, parents of children who received surgery expressed satisfaction with the surgical outcomes.

To further optimize patients' satisfaction and psychosocial functioning, additional surgical procedures can be performed during adolescence or after facial skeletal maturity has been reached. When an inadequate primary lip closure has been performed resulting in puckering, whistle deformity or a too short lip, secondary lip repair (re-repair) has to be performed [10–12]. Additionally, other studies proposed lipofilling (i.e. fat injection/grafting) of the upper lip as an alternative technique for secondary lip correction [13–16]. Lipofilling is an important component in aesthetic surgery. It consists of the injection of an amount of autologous fat in different body parts [17]. One of the main applications of this technique is the rejuvenation of the aging face by restoring or creating fullness [18–20]. Since full lips are associated with an attractive and youthful appearance, many individuals desire lip augmentation [21]. In this case, lipofilling is used to reverse the subcutaneous loss of volume and vertical tissue descent due to aging [17]. Another application of lipofilling is reconstruction of acquired (e.g. post-traumatic) or congenital (e.g. CL ± P) soft tissue defects [22]. Rieger and Pierer [23] investigated the effects of combined autologous dermis graft and lipofilling in the case of an acquired anomaly (i.e. correction of post-traumatic upper lip notching). Two patients (a 40-year-old female and a 29-year-old male), both with upper lip notching after a bike accident with post-traumatic upper lip volume loss, were treated with this technique. The authors argued that the combination of dermal grafts and lipofilling was a safe and easy procedure to correct volume deficiencies of the upper lip with excellent aesthetic outcome. Duskova and Kristen [13] used lipofilling to resolve low projection of the upper lip in patients with CL ± P. As these patients often present with an abnormal harmonic profile between the lower and upper jaw, it is hard to achieve a good nasal profile using conventional methods. Therefore, lipofilling might be a solution for these patients [24].

Two studies [15,16] investigated fat grafting in primary cleft lip repair. Postoperative photographs were assessed revealing minimal residual cleft stigmata [15] and significant improvement in scar appearance and contour of the fat-grafted area when compared to a group of patients who underwent primary cleft lip repair without fat grafting [16]. More recently, Talmant, Talmant and Lumineau [11] and Bae et al. [25] investigated the secondary treatment of cleft lip and palate using lipofilling. Results showed the usefulness of this technique in the case of interlabial disharmony [11]. Furthermore, a statistically significant volume increase was found [25]. Regarding aesthetic outcomes after lipofilling, patients' satisfaction should also be taken into account. However, literature regarding satisfaction after lipofilling in patients with CL ± P is scarce (Table 1). Duskova and Kristen [13] explored patients' satisfaction after augmentation by autologous fat grafting in cleft lip and nose. All patients were pleased and satisfied with the approach as well as with the results. Unfortunately, the desired effect only lasted an average of 7 months (range: 5–9 months) due to resorption of the injected fat. More recently, Jones et al. [12] found that patients were significantly happier with their appearance after lipofilling ($p < 0.001$). When patients were asked about the ease of the surgery and rate of recovery, they answered uniformly positive. Koonce et al. [26] also assessed autologous fat grafting in the treatment of cleft lip volume asymmetry and discussed that all patients and caregivers were subjectively pleased with the results and reported they would choose to have the procedure again.

As lips have a principal aesthetical and functional role in the face [27], it may be questioned whether or not this technique has functional consequences as well. Haneke [28] investigated complications after injection of different types of fillers. Interestingly, loss of function was mentioned as a late complication appearing after about 2–6 weeks. Lipofilling in the atrophied lip in facial palsy patients was investigated by Biglioli et al. [29]. These patients often show an atrophy of the musculus orbicularis oris resulting in oral incompetence, speech difficulties and nutritional problems [29]. Lipofilling was performed in the lower lip of the included patients. The results showed an increased lip volume with a great satisfaction among the patients (aesthetic part) together with an ameliorated lip competence (functional part). To the best of our knowledge, this was the only study investigating functional outcome after lipofilling. Unfortunately, the assessment procedure for functional outcome (i.e. lip competence) was not applicable.

In summary, literature regarding functional outcomes and patients' self-reported satisfaction with appearance after lipofilling is scarce. Therefore, the aim of present pilot study was to document differences in functional (i.e. logopaedic outcome: orofacial myofunctional behavior, articulation and lip strength) and aesthetic (i.e. patients' self-reported satisfaction) outcomes by comparing pre- and postoperative results after lipofilling of the upper lip in patients with a cleft lip (and palate). It was hypothesized that orofacial myofunctional outcomes and lip strength would be lower after performance of lipofilling. Since bilabial consonants require an active involvement of the upper lip, an increased percentage of distortions of the bilabial consonants was hypothesized after performance of this technique. Full lips are associated with attractiveness and youthfulness, thus an increased self-reported satisfaction with appearance after performing lipofilling was hypothesized.

2. Methods and materials

This study was approved by the Ethics Committee of the Ghent University Hospital (B670201215561) and the described research adhered to the tenets of the Declaration of Helsinki. All patients participated voluntarily and signed an informed consent.

2.1. Participants

Patients from the Ghent University Hospital who underwent lipofilling between 2014 and 2016, performed by the same plastic surgeon (N.R.), were invited to participate in this study. Lipofilling was performed when patients were dissatisfied with appearance in profile. This was the case in patients with a disharmonic profile between the lower and upper lip, combined with decreased volume of the upper lip. Moreover, decreased flexibility of the scar tissue resulting in problems with upper lip mobility was another indication for performance of lipofilling.

Eight Dutch-speaking youngsters and young adults (three women, five men) with CL ± P agreed to participate in the present study (Table 2).

Median age was 19 years (range: 14–24 years). Three patients had a left sided unilateral cleft lip and palate (CLP), three patients had a right-sided unilateral CLP, one patient had a bilateral cleft lip (CL) (with an incomplete right sided cleft lip) and one patient had a left-sided unilateral CL. Lip repair was performed at a median age of 4.5 months (range: 3–7 months). Palatal repair was performed at a median age of 17.50 months (range: 13–28 months). At a median age of 9.65 years (range: 8.11–10.3 years), bone grafting was performed. All individuals underwent lipofilling at a median age of 18 years (range: 14–24 years). Median time between lipofilling and post-operative assessment was 8.0 weeks (range: 6–15 weeks).

2.2. Surgical procedure

All patients were operated in day care under general anesthesia. Autologous fat was harvested from the inner knees with an 11 Gauge 3 mm rasp cannula after infiltration with standard Klein's solution. After centrifugation for 3 min at 3000 rpm, the fat was transferred to

Table 1
Summary of recent studies investigating the effect of lipofilling on aesthetic outcome.

Authors	Participants	Methods	Results
Talmant, Talmant and Lumineau (2016) [11]	Patients with CLP (more information was not applicable)	- Secondary treatment of CLP using velar re-repair and lipofilling	Results showed: - That lipofilling was useful when there was interlabial disharmony with a slightly flat upperlip
Jones et al. (2017) [12]	Patients with CL (n = 18) Mean age: 16 years (range: 6–43 years)	- Aesthetic outcome and symmetry was evaluated using the Asher-McDade scale	Results showed: - That symmetry and aesthetics were improved based on the nasal form and vermillion border - That patients were significantly happier with their appearance after fat grafting - That patients were uniformly positive when questioned about the ease of surgery and rate of recovery
Duskova and Kristen (2004) [13]	Patients with CL (n = 5) (4 females, 1 male) Age range: 26–38 years	- Outcome measures NA	Results showed: - That all included patients were pleased and satisfied with the approach as well as with the results
Balkin et al. (2014) [15]	Patients with CL - Fat grafting n = 20 (12 females, 8 males) - No fat grafting n = 40 (22 females, 18 males) Mean age: 3.5 months (range: 1.5–6.4 months)	- Postoperative photographic assessment by three blinded judges using a 5-point ordinal scale *(type of judges: NA)*	Results showed: - Minimal residual cleft stigmata after gat grafting - That the mean ordinal scale values were improved for the fat grafted subjects compared to those non-fat grafted
Zellner et al. (2015) [16]	Patients with CL - Fat grafting n = 19 (13 females, 6 males) - No fat grafting n = 16 (9 females, 7 males) Mean age: 4.9 months (range: 2–20 months)	- Postoperative photographic assessment by three blinded judges using a 5-point ordinal scale (type of judges: *NA)*	Results showed: - A statistically significant improvement in scar appearance and contour of the fat-grafted area
Bae et al. (2016) [25]	Patients with CL - Fat grafting n = 19 (6 females, 13 males) Mean age: 25 years (*SD*: 9.41)	- Aesthetic outcome was evaluated using a questionnaire	Results showed: - A statistically significant volume increase in upper lip - That the volume increase was satisfactory for the patients
Koonce et al. (2018) [26]	Patients with CL (n = 52)	- Pre- and postoperative photographs were randomly assessed by six blinded reviewers using the Asher-McDade scale (type of judges: *NA)*	Results showed: - That postoperative assessment revealed improved volume symmetry in all patients - That all patients or families were pleased with the results

CL: cleft lip CLP: cleft lip and palate *SD* = standard deviation NA = not applicable.

1 cc syringes and injected through a stab incision at both lip commissures with a 14 Gauge 2 mm cannula in the (preoperatively marked) full aesthetic unit of the upper lip, starting from the nasolabial folds and additionally in the mucosa of the upper lip. Injected fat volume was 20–40 cc. After surgery, the patients were instructed to use fatty ointment on the mucosa of the upper lip.

2.3. Assessment using the Iowa Oral Performance Instrument

The Iowa Oral Performance Instrument (IOPI) (model 2.1; IOPI medical LLC, Carnation, WA) was used to measure differences in lip strength before and after lipofilling. The IOPI is a portable device that uses an air-filled plastic bulb. This bulb is connected via a clear plastic tube to measure pressure in kilopascals (kPa) exerted on the bulb. In the present study, lip strength before and after lipofilling was measured on the affected side, non-affected side, central side without tongue blades and central side with tongue blades using the IOPI. The protocol described by Clark and Solomon [30] and Van Lierde, Bettens [31] was used. The subjects were instructed to protrude the lips and to push and squeeze the lips around the bulb (placed between wooden tongue blades) with maximum effort. The lip strength was measured on the basis of three test trials per side with a brief resting period of 30 s between each trial. The highest pressure was used for the statistical analysis.

2.4. Cleft evaluation profile

To measure self-perceived satisfaction before and after lipofilling, the Dutch version of the Cleft Evaluation Profile (CEP) was used [32]. This questionnaire consists of an eight-item list: (1) speech, (2) hearing, (3) lip, (4) nose, (5) teeth, (6) breathing, (7) appearance of bite and (8) appearance of profile. Patients were asked to rate their satisfaction on a seven-point Likert scale ranging from 1 to 7, respectively very satisfactory and very unsatisfactory.

2.5. Articulatory assessment

For the assessment of articulation, a picture naming test was used [33]. Participants were shown a series of 135 black and white drawings of common objects and actions. They were asked to name these pictures. This test contains all Dutch single speech sounds and most clusters in all positions [33]. The assessment took place in a sound-protected room at the Ghent University Hospital and was videotaped using a Sony Handycam HDR-CQ280E with a built-in high quality microphone. Percentage distortions of the bilabial (/b/, /p/, /w/, /m/) and the labiodental (/v/, /f/) consonants were compared before and after lipofilling. The video-recorded speech samples were anonymized and randomized. The assessment of articulation was performed by two judges (two speech-language pathologists: K.B. and C.A.), blinded to pre- and postoperative measurements, with respectively 6 and 2 years

Table 2
Demographic, cleft, and surgical details for the participating patients.

Patient	Gender	Cleft type	Age at lip closure (months) + surgical procedure	Age at palatal closure (years, months) + surgical procedure	Age at bone graft	Age at lipofilling (years) + additional surgery	Additional surgeries (years)
1	M	UCLP left	3 m – type NA[1]	2y4m – soft palate – type NA 7y1m – hard palate – type NA	8y11 m	20y - Revision lip correction + lipofilling left upper lip	18y - osteotomy
2	F	UCLP left	4 m – Tennison	1y2m – Van Langenbeck	10y3m	15y – revision lip correction + lipofilling left upper lip	
3	M	BCL (incomplete right)	5 m – Tennison 6y0m – Z-plasty left 9y8m – revision Mulliken	NA	10y2m	16y – rhinoplasty + lipofilling upper lip	
4	M	UCLP right	Age NA – Tennison	NA *(no information, procedure was performed in another hospital)*	NA	24y – revision lip correction, Fisher + lipofilling right upper lip	
5	M	UCL left	3 m – Tennison	NA	10y10 m	21y – rhinoplaty + septoplasty + lipofilling left upper lip	
6	F	UCLP right	7 m – Tennison	1y3m – Furlow	9y6m	15y – rhinoplasty + lipofilling right upper lip	
7	M	UCLP right	5 m – Tennison	1y1m – Van Langenbeck	9y6m	14y – nose correction + revision lip correction + lipofilling right upper lip	
8	F	UCLP left	NA	NA	NA	19y – rhinoplasty + septoplasty + revision lip correction + lipofilling left upper lip	Pharyngoplasty 18y – osteotomy Le Fort I

NA: not applicable UCLP: unilateral cleft lip and palate UCL: unilateral cleft lip BCL: bilateral cleft lip.

experience in assessing articulation disorders in children with CL ± P. Inter-rater reliability was calculated by the average measures intraclass correlation coefficient (ICC) with a 2-way, mixed-effects model.

2.6. Assessment of orofacial myofunctional behavior

Six oral motor functions were compared before and after lipofilling as proposed in the protocol of Lembrechts et al. [34]. These exercises included lip closure, lip protrusion, lip broadening, lip trill without phonation, lip stretching and lip plopping. The motor functions were assessed on a three-point rating scale (0 = normal function, 1 = decreased function, 2 = function impossible).

2.7. Statistical analysis

Statistical analysis was performed using SPSS version 24.0 (SPSS Inc., Chicago, Illinois). Significance level was set at $\alpha < 0.05$. All continuous variables were non-parametrically distributed, resulting in the use of a Wilcoxon signed-rank test. For the analysis of categorical data, the McNemar test was used.

3. Results

3.1. General postoperative findings

All patients were discharged at the same day of surgery reporting minimal discomfort and morbidity. Stitches at the fat harvest donor sites were removed after 7 days. No pressure garments were prescribed, bruising was minimal and no complications occurred.

3.2. Assessment using the IOPI

Table 3 provides results of the IOPI measurement of strength before and after lipofilling. No statistically significant differences in lip/tongue strength for the different IOPI measurement conditions were found before and after lipofilling ($p > 0.05$).

3.3. Cleft evaluation profile

A statistically significant difference was found in self-evaluation of appearance in profile before and after lipofilling ($Z = -2.271$, $p = 0.023$). For the other parameters of the CEP, no statistically significant differences were found before and after lipofilling (Table 4).

3.4. Articulatory assessment

Results regarding the inter-rater reliability on the articulatory assessment are provided in Table 5. Based on the average ICC measures, very good inter-rater reliability was found for all assessments of the different consonants.

In Table 6, results of the comparison of distortions of the bilabial and labiodental consonants before and after lipofilling are provided. For none of the bilabials, a statistically significant difference was found in distortions before and after lipofilling: /p/ ($Z = -1.540$, $p = 0.123$), /b/ ($Z = -0.922$, $p = 0.357$), /w/ ($Z = -1.014$, $p = 0.310$) and /m/ ($Z = -0.447$, $p = 0.665$). However, a decrease of distortions was seen for the production of the /p/ (8.3% distortions before lipofilling, 2.8% after lipofilling), the /b/ (4.7% distortions before lipofilling, 3.1% after lipofilling) and the /w/ (25% before lipofilling, 10.2% after). For none of the labiodental consonants, statistically significant differences in distortions were found before and after lipofilling: /v/ ($Z = 0.000$, $p = 1.000$), /f/ ($Z = 0.000$, $p = 1.000$).

3.5. Assessment of orofacial myofunctional behavior

In Table 7, results of the comparisons of oral motor functions before

Table 3
Comparison of measurements using the IOPI (kPa) before and after lipofilling. Statistical analysis based on Wilcoxon signed rank test with significance level set on $p < 0.05$.

IOPI measurement condition	*n*	Before lipofilling		After lipofilling		Z	Significance level
		Median (kPa)	Q1-Q3 (kPa)	Median (kPa)	Q1-Q3 (kPa)		
Affected side	6	27	20–34	27	21–30	−0.135	0.893
Non-affected side	6	30	22–36	32	27–35	−0.405	0.686
Central without tongue blades	6	13	10–20	11	8–17	−0.271	0.786
Central with tongue blades	7	15	13–25	17.5	16.25–26.75	−0.931	0.352

Table 4
Comparison of self-assessment based on the Cleft Evaluation Profile [32] before and after lipofilling. Statistical analysis based on Wilcoxon signed rank test with significance level set on $p < 0.05$.

Parameter	*n*	Before lipofilling		After lipofilling		Z	Significance level
		Median	Q1-Q3	Median	Q1-Q3		
Speech	7	2	2–2.75	2	1–2	−1.732	0.083
Hearing	7	1.5	1–2.75	2	1–2	< 0.001	1.000
Appearance of teeth	7	2	2–3	2	2–2	−0.816	0.414
Appearance of lip	6	3	3–3.75	2	1.75–3.25	−1.857	0.063
Appearance of nose	7	4	4–4.75	3	2–5	−1.633	0.102
Breathing through nose	7	4	3.25–4.75	3	2–5	−0.687	0.492
Appearance of profile	7	4	3.25–5	3	2–4	−2.271	0.023*
Appearance of bite	7	2	1–3	2	1–2	−1.342	0.180

*Statistical significant difference, $p < 0.05$.

Table 5
Inter-rater reliability by means of an average two-way mixed ICC (consistency) of the two listeners for the articulatory assessment.

	Average ICC consistency		95% CI average ICC consistency		Interpretation of the average ICC[a]	
	Before lipofilling	After lipofilling	Before lipofilling	After lipofilling	Before lipofilling	After lipofilling
Distortions /p/	0.99	1.00	0.99–1.00	1.00–1.00	Very good	Very good
Distortions /b/	0.99	1.00	0.96–1.00	1.00–1.00	Very good	Very good
Distortions /w/	1.00	0.98	0.99–1.00	0.99–0.99	Very good	Very good
Distortions /m/	1.00	1.00	0.99–1.00	1.00–1.00	Very good	Very good
Distortions /v/	1.00	1.00	0.99–1.00	1.00–1.00	Very good	Very good
Distortions /f/	1.00	1.00	0.99–1.00	1.00–1.00	Very good	Very good

[a] Based on Altman (1990) [37]: ICC < 0.20: poor, 0.21–0.40: fair, 0.41–0.60: moderate, 0.61–0.80: good, 0.81–1.00: very good.

Table 6
Comparison of distortions of the bilabial consonants before and after lipofilling, expressed in %. Statistical analysis based on Wilcoxon signed rank test with significance level set on $p < 0.05$.

Consonant	*n*	Before lipofilling		After lipofilling		Z	Significance level
		Median	Q1-Q3	Median	Q1-Q3		
Distortions /p/ (%)	8	8.3	6.0–20.8	2.8	0.7–9.4	−1.540	0.123
Distortions /b/ (%)	8	4.7	3.1–8.6	3.1	0.1–6.3	−0.922	0.357
Distortions /w/ (%)	8	25.0	6.0–44.2	10.2	1.1–29.9	−1.014	0.310
Distortions /m/ (%)	8	0.0	0.0–2.6	0.0	0.0–2.0	−0.447	0.665
Distortions /v/ (%)	8	0.0	0.0–0.0	0.0	0.0–0.0	0.000	1.000
Distortions /f/ (%)	8	0.0	0.0–0.0	0.0	0.0–0.0	0.000	1.000

and after lipofilling are provided. No statistically significant differences ($p > 0.05$) in oral motor exercises were found when comparing measurements before and after lipofilling.

4. Discussion

The aim of this pilot study was to assess functional (i.e. logopaedic outcome: orofacial myofunctional behavior, articulation and lip strength) and self-reported aesthetic (i.e. patients' satisfaction) outcomes in patients with cleft lip (and palate) by comparing pre- and postoperative results after lipofilling of the upper lip.

In general, patients were statistically significant more satisfied with appearance in profile after lipofilling (Table 4). Similar findings were described by Jones et al. [12]. Nonetheless, a methodological difference between latter and present study should be mentioned. The present study investigated different aspects of appearance (e.g. appearance of the nose and lip and appearance in profile) whereas Jones et al. [12] investigated overall appearance without making further divisions. Given that patients with CL ± P often present with a disharmonic profile between lower and upper jaw, satisfaction with appearance in profile is important. To the best of our knowledge, this was the first study investigating this specific aspect. Regarding appearance of the lip,

Table 7
Comparison of oral motor functions before and after lipofilling. Statistical analysis based on McNemar test with significance level set on $p < 0.05$.

Oral motor exercise	Before lipofilling		After lipofilling		Significance level
	Normal	Affected	Normal	Affected	
Lip closure	63% (5/8)	37% (3/8)	75% (6/8)	25% (2/8)	1.000
Lip protrusion	57% (4/7)	43% (3/7)	71% (5/7)	29% (2/7)	1.000
Lip broadening	100% (7/7)	0% (0/7)	100% (7/7)	0% (0/7)	NA
Lip trill	33% (2/6)	67% (4/6)	67% (4/6)	33% (2/6)	0.500
Lip stretching	67% (4/6)	33% (2/6)	17% (1/6)	83% (5/6)	0.250
Lip plopping	83% (5/6)	17% (1/6)	67% (4/6)	33% (2/6)	1.000

NA: Not applicabled.

patients reported more satisfaction with appearance of the lip after lipofilling, although not statistically significant (Table 4). This finding is in line with the hypothesis and is most likely associated with the increased volume of the upper lip.

No statistically significant differences in distortions were found when comparing measurements before and after lipofilling (Table 6). However, assessment of articulation of bilabial consonants showed that the interquartile range (*IQR:* Q1-Q3) of distortions of the /p/ decreased from 6.0 to 20.8 before lipofilling to 0.7–9.4 after lipofilling. For the sound /b/, the IQR decreased from 3.1 to 8.6 before lipofilling to 0.1–6.3 and for the /w/, the IQR decreased from 6.0 to 44.2 to 1.1–29.9. This is an interesting finding suggesting that patients were able to use their upper lip more actively in the production of these consonants after performance of lipofilling.

When comparing distortions of the labiodental consonants /v/ and /f/ before and after lipofilling, again no statistically significant differences were found. In addition, results showed a low percentage of distortions of these consonants both before and after performance of this procedure (Table 5). Given that in phonetics, the labiodentals are articulated with the lower lip and upper teeth, these findings seem reasonable. Furthermore, no lip rounding nor lip protrusion is needed for articulation of labiodental consonants because the air stream is obstructed between the central part of the incisors and the upper lip. Therefore, no contribution of the upper lip is needed for articulation of the labiodentals. A CL ± P is a defect of the upper lip so that a normal contact between the upper teeth and lower lip is possible, which may explain this finding for the labiodental consonants.

Neither for lip strength (Table 3) nor for oral motor functions (Table 6), statistically significant differences were found when comparing measurements before and after lipofilling. A possible explanation is the hypothesis that the sudden increase of lip volume results in perioral stiffness and hence, affects lip strength and lip mobility Haneke [28]. Moreover, postoperative swelling of the injection place [26] may also affect lip movements. In the light of these ideas, a decreased normal performance of lip stretching was observed after lipofilling, although not statistically significant. More specifically, 83% (5/6) of the patients had difficulties with this function after lipofilling, in which they were unable to stretch the upper lip beyond their teeth. Because postoperative measurements were performed relatively short after fat injection (on average 1.8 months), it is expected that this region will regain flexibility when fat resorption occurs [25]. However, no consensus is reached regarding long-term outcomes of lipofilling. Some authors found a temporary effect of lipofilling, which necessitated repeated application every 7 months [13]. Other authors argued that subsequent autologous fat transplantations should be conducted at an interval of at least 6 months [35]. Additionally, the resorption rate is unpredictable, with resorptions up to 70% of the volume [35]. Therefore, future studies including longitudinal follow-up after lipofilling are necessary.

The main limitation of this pilot study was the small sample size ($n = 8$), which may negatively affect validity. Furthermore, including more patients might probably result in some statistically significant differences. Another limitation of this pilot study was the effect of observer bias which may possibly explain the increase of normal performance of the oral motor functions lip closure, lip protrusion and lip trill without phonation (Table 6). Observer bias possibly occurs when outcome measures are influenced by the raters' (un)conscious predispositions (e.g. expectations influencing the outcomes of the experimental intervention) [36]. Hence, it is possible that the two raters (K.B. and C.A.) had a preconceived idea of the effects after lipofilling. Additionally, it is possible that the visual perception of a normal lip volume resulted in a more positive assessment of these functions. Moreover, in 62.5% of the patients (5/8), lipofilling was combined with rhinoplasty. Latter procedure may also have influenced not only raters' perception but also self-assessment. A possible solution is to include assessors blinded to the purpose of the study.. In the light of this idea, it might be interesting to include a control group in order to compare outcomes in patients who have and have not undergone this procedure. In summary, assessment before and after lipofilling was largely subjective (i.e. CEP, assessment of articulation and orofacial myofunctional behavior). Nevertheless, this perceptual analysis was supplemented by an objective measure in terms of the Iowa Oral Performance Instrument.

Despite these limitations, the present pilot study was the first to assess functional outcomes after performance of lipofilling providing important information for (plastic) surgeons and speech-language pathologists treating patients with CL ± P. Given the findings of the present study, one may wonder whether or not this technique should be included in the standard multidisciplinary plan of care for patients with CL ± P. To the authors' opinion, lipofilling could be useful in some patients but an individual approach is necessary. In addition, further research and long-term follow-up studies are required.

5. Conclusion

Lipofilling of the upper lip in patients with a cleft lip (and palate) is a new technique for secondary lip correction. Findings of the present study showed that there were no statistically significant differences in functional outcomes before and after this procedure. However, patients' satisfaction with appearance in profile was increased after performing lipofilling. Therefore, further research including larger sample sizes and long-term follow-up is necessary.

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Conflicts of interest

There is no conflict of interest.

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