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Objective evaluation of vertical Z-plasty with double transposition vermillion flaps for secondary whistling deformity correction: A method for uni- and bilateral correction

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ABSTRACT

Objectives: This study describes a modified method for secondary correction of whistling deformities in patients with unilateral and bilateral cleft lip/palate (CL/P), using a horizontal double transposition vermilion flap, including parts of the orbicularis oris muscle. The pre- and postoperative results were objectively evaluated.

Study design: 34 patients with a whistling deformity who underwent secondary reconstruction between 07/2013 and 11/2018 were included in this study (mean age 20.2 ± 11.6 years). 24 patients were male and 10 female. 30 patients presented with cleft lip and palate (CLP) — 15 bilateral, nine on the left side and six on the right. Four patients had only a left-side cleft lip (CL).

The whistling deformity reconstruction was carried out using two triangular transposition vermilion flaps with muscle parts, for a vertical Z-plasty. The surgical procedure is normally performed under local anesthesia in all patients older than 10 years. For statistical evaluation, the size of the whistling defect in the vermilion was determined on photographs before and 6–9 months after surgery. The individual defect score (DS) was evaluated pre- and postoperatively. In all patients, no additional surgical procedures, such as rhinoplasty or scar correction in the upper lip, were carried out simultaneously.

Results: Minor (DS < 400), moderate (DS 400–1400), and severe (DS > 1400) whistling defects were surgically corrected. The whistling defect score was significantly reduced in all patient groups ($p < 0.001$). In six patients the result of surgery was rated as 'acceptable' (DS > 30), in five patients as 'good' (DS 10–30), and in 23 patients as 'very good' (DS 0–10).

Conclusions: This study describes a modified method for whistling deformity reconstruction in uni- and bilateral clefts. The aesthetic results are based on a reconstruction of the subcutaneous muscle layers and the creation of a symmetrical lip contour and prolabium using transposition flaps from the lateral side of the cleft. The great advantage is the uncomplicated performance under local anesthesia, even for all children over 10 years, and the short operation time. Postoperative complications did not occur.

1. Introduction

Patients with a cleft lip (CL) and/or cleft lip and palate (CLP) show a variety of secondary deformities after primary cheiloplasty. Henkel et al. (1998) describe an incidence of secondary surgery for cleft lip patients of about 35%. One of these secondary deformities — the whistle deformity — is common and usually requires surgical correction. It is defined as a central vermilion notching due to complex causes (Choi et al., 2012). The typical clinical findings for the whistle deformity are an upper lip notching, bulging on the lateral lip segment, and volume deficiency at the upper vermilion (Reddy et al., 2009). According to Bhandari (2014) the

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noncontinuity of the muscle at the vermilion border is responsible for whistle deformity. This noncontinuity can also be due to a deficiency of the mucosa or of the submucosal or subcutaneous tissue at the free vermilion border.

These clinical manifestations are caused by insufficiently reconstructed anatomical structures at the time of first lip reconstruction. In bilateral cleft patients the tissue deficiency, especially of orbicularis muscle fibers in the prolabium, the incorrectly and insufficiently reconstructed orbicularis muscle, and muscle fibers from the orbicularis muscle still entering the nostrils are the main causes of whistling deformity (Duffy, 1971). Aside from this, vertical shortage of lip in the midline, especially when the prolabium is small, can also lead to whistle deformity after bilateral cleft lip repair (Bhandari, 2014).

Various surgical techniques have been described to address these problems, including fat grafting (Patel and Hall, 2004; Baum et al., 2017) or intraoral grafts such as free palatal mucosa (Vecchione, 1982) or free tongue graft (Cohen and Kawamoto, 1991). In free grafts the resorption rate is unpredictable (Baum et al., 2017) and so local flaps in combination with autologous grafts are used. Local triangular mucosal flaps in combination with artificial dermis transplants have been used to prevent postoperative scar contracture and shrinking (Wakami et al., 2010). Bilayer local flaps, such as the bilobed mucosal flap (Song et al., 2011) and the cross-muscle flap (Choi et al., 2012) are used for whistling deformity reconstruction. The Kapetansky and 'Kapetansky-Juri' advancement flap techniques (Kapetansky, 1971; Juri et al., 1976; Kiran et al., 2014) put additional tissue in the central region of the upper lip to decrease the deformity, using two double pendulum flaps.

The musculocutaneous transposition labial flap, originally described by Abbé in 1898 for double cleft lip reconstruction (Sabattini, 1838; Abbé, 1898) has been modified by various surgeons (Takato et al., 1996; Cohen and Kawamoto, 1991). The soft tissue deficit and underdeveloped orbicularis muscle ring in the prolabium can be built up using this technique. The transposition of full-thickness parts of the lower lip is still a common procedure for building up the missing muscle ring in the prolabium in bilateral clefts, but views on indications for an Abbé plasty as standard surgery vary in the literature, especially with regards to children (Momma et al., 1974). A modification of the Abbé flap, in which a musculocutaneous flap from the central portion of the lower lip vermilion and a tiny portion of skin are transposed, can be used for the correction of mild tightness of the vermilion tubercle and Cupid's bow in cleft lip patients (Takato et al., 1996). Nevertheless, the vertical scar and donor side morbidity remain obvious disadvantages. Moreover, postoperative care and food supplementation associated with this method must always be considered, especially in children.

There is a great variety of surgical methods used for reconstructing whistling deformity. The surgical method used depends on the preoperative situation and on the degree of deformity.

Our prospective study evaluates the measurable aesthetic results of whistling deformity reconstruction in patients with uni- and bilateral cleft lip/palate (CL/P). The whistling deformity correction was carried out using a vertical Z-plasty with double vermilion transposition flaps (dry mucosa).

2. Materials and methods

2.1. Patients

Of all patients with secondary cleft lip reconstruction who visited the hospital from June 2013 to November 2018, 34 patients were included in the study (Fig. 1). Informed consent was given by all patients or their legal representatives.

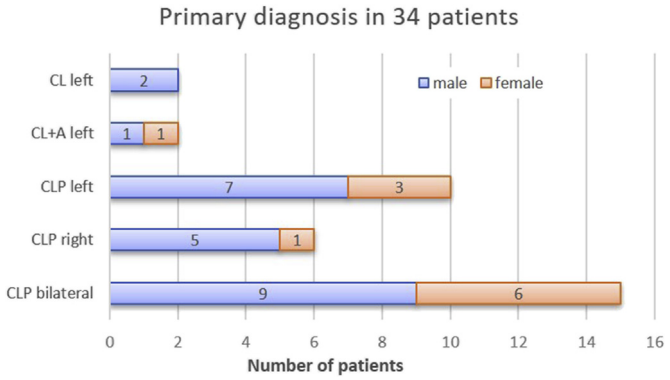

Fig. 1. Gender-related cleft manifestation in patients with whistling deformity ($n = 34$). The patient collective comprised 10 female and 24 male patients. The main diagnosis was a bilateral complete CLP.

The mean age of all patients was 20.1 ± 13.1, with a range of 5.6–54.9 years. At the time of treatment most patients were adolescents/young adults (10–20 years).

The study included 24 men and 10 women (ratio 1:0.42), so more male patients than female patients presented for whistling deformity reconstruction.

2.2. Cleft appearance

15 of 34 patients showed a complete bilateral CLP (44.1%), nine patients a complete CLP on the left side (26.5%), and six on the right (17.6%). Two of the remaining four patients had a CL on the left side with alveolus (CL + A, 5.9%) and two without (CL, 5.9%).

2.3. Inclusion and exclusion criteria

Inclusion criteria for this study were: lip closure according to Pfeifer's technique between the 4th and 7th month of life; presence or prosthetic replacement of the front teeth; no combination with other plastic or reconstructive procedures, such as rhinoplasty, nasal columella reconstruction, or complete upper lip reconstruction procedures; and informed consent of each patient (or parents, when patient was under 18 years). Patients who had undergone previous attempts to correct whistling deformity or who had simultaneous correction of the upper lip length were excluded from the study. The mean follow-up period was 20.5 ± 10.9 months and ranged from 6 to 31 months.

2.4. Measurement and defect score

In accordance with to Nkenke et al. (2013) and Wakami et al. (2010) the horizontal defect ratio ((b 'maximum defect width'/a 'intercommisural distance') × 100) and the vertical defect ratio ((d 'maximum defect height'/c 'maximum height of the upper lip') × 100) were calculated on photographs taken before and 6–12 months after surgery (Fig. 2). The product of both ratios was defined as the defect score (DS). Minor (DS < 400), moderate (DS 400–1400) and severe (DS > 1400) whistling defects were corrected surgically. In accordance with Nkenke et al., 2013 the outcome was rated 'very good' when there was almost no sagittal vermilion deficiency, a well filled vermilion tubercle, and a postoperative defect score of 0–10. It was rated 'good' when the sagittal vermilion deficiency was resolved in a way such that the anterior teeth were hidden but the vermilion tubercle was unclear or flat (DS 11–30). An 'acceptable' or almost poor outcome showed

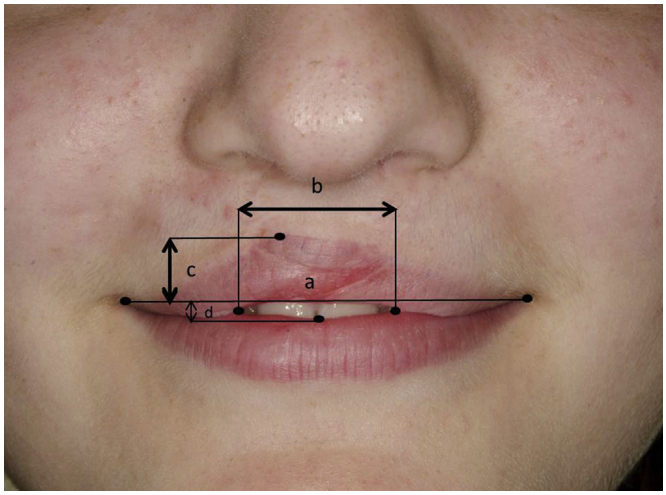

Fig. 2. Method of calculating the defect score with the width of the upper lip (a), the width of the whistling deformity (b), the height of the upper lip (c), and the height of the whistling deformity (d). The product of the defect ratio of the labial width (horizontal defect score: (b/a) × 100%) and the defect ratio of the labial height (vertical defect score: (d/c) × 100%) is the defect score (Nkenke et al., 2013) or defect point (Wakami et al., 2010), essential for further analysis.

persisting sagittal vermilion deficiency, as well as vermilion tubercle deficiency (DS > 30).

2.5. Surgical procedure

Surgical procedure (operation time: 37.4 ± 9.2 min) in all patients over 10 years ($n = 27$) was carried out under local anesthesia; in children aged of 5–9 years ($n = 7$) in sevoflurane inhalation narcosis and oral intubation were applied (operation time: 30.5 ± 8.8 min). No other orofacial reconstruction procedures, such as nasal revision, rhinoplasty, or iliac bone grafts for secondary osteoplasty, were performed at the same time. All operations were performed by the same surgeon (K.S.).

After marking the double Z-plasty in the vermilion (dry mucosa) for vertical extension (Fig. 3a,b), local infiltration anesthesia was carried out using Ultracain® D-S (Sanofi, Germany) (40 mg articaine hydrochloride, 0.006 mg epinephrine hydrochloride). The initial incision was made from the white skin roll (Millard, 1964) to the center of the whistling deformity. Two triangular lobules were formed in the dry mucosa above the red line (Fig. 2). The incision included the mucosa and the superficial part of the underlying orbicularis oris muscle. Subsequently, instead of the horizontal V-incision on the opposite aspect of the defect, as used in a VY-plasty, lateral and medial triangular flaps were created and extended to the white line (a, b). These myomucosal triangular flaps were transposed to the opposite side of the deformity (Fig. 2). The middle part of upper vermilion was thus reconstructed using a vertical Z-plasty (Fig. 3). It is important to take care with the red line — the division between the dry vermilion and the mucous membrane of the oral cavity (Milard, 1964; Nordhoff, 1984). In this respect, the 'dirty lip trick' according to Jones and Verchere (2008) can be useful.

A small triangular skin excision was applied to the scar of the vermilion border if there was an unattractive peaking of the vermilion at the Cupid's bow (Fig. 2a,b). The cutaneous incisions were closed using resorbable sutures (Monocryl 6-0, Ethicon, Germany). The vermilion triangular flaps were transposed to the end of the incision on the contralateral side and the incisions were closed with resorbable sutures (Monocryl 5-0, Ethicon, Germany). No perioperative antibiotics were given. Pain therapy was carried out with NSAIDs, with dose adapted to weight.

The wound dressing consisted of steristrips at the white skin roll. Wound care involved regular mucosal disinfection with Octinisept® (Schülke & Mayr GmbH, Norderstedt, Germany) and the application of oxytetracycline eye ointment for 5 days (Jenapharm GmbH & Co. KG, Jena, Germany).

2.6. Operation-specific side effects

There was minimal scar formation in the vermilion for all the patients, and it was possible to create a peaked tubercle. Postoperative aesthetic outcome was favorable, with no inflammation, loss of tissue, or wound dehiscence.

A specific side effect of this method is shown in the preoperative (Fig. 4a) and postoperative (Fig. 4d) images of the patient. A small part of dry mucosa can be seen in the middle of the whistling deformity preoperatively and more cranially located in the reconstructed upper lip postoperatively (Fig. 4d). This part has been transposed by moving the triangular vermilion flaps. This is one disadvantage of this method.

2.7. Statistical evaluation

All calculations were made using SPSS version 14.0 for Windows (SPSS, Chicago, USA). Mean values are given with standard deviations. The Wilcoxon test was used for comparison of continuous variables in paired samples, and p-values ≤ 0.05 were considered significant.

2.8. Photography

The photographic standards required for plastic surgery, according to DiBernardo et al., 1998, have been respected. A Nikon

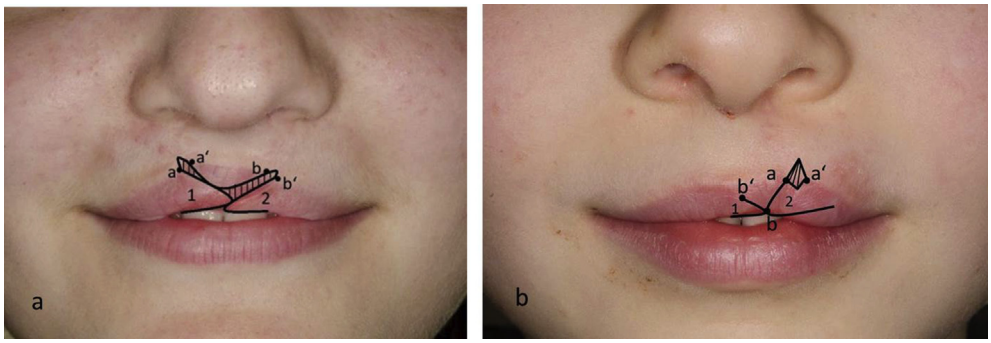

Fig. 3. Technique for whistling deformity reconstruction via a vertical Z-plasty, using a bilateral triangular vermilion transposition flap for vertical lengthening of the vermilion in (a) bi- and (b) unilateral cleft patients.

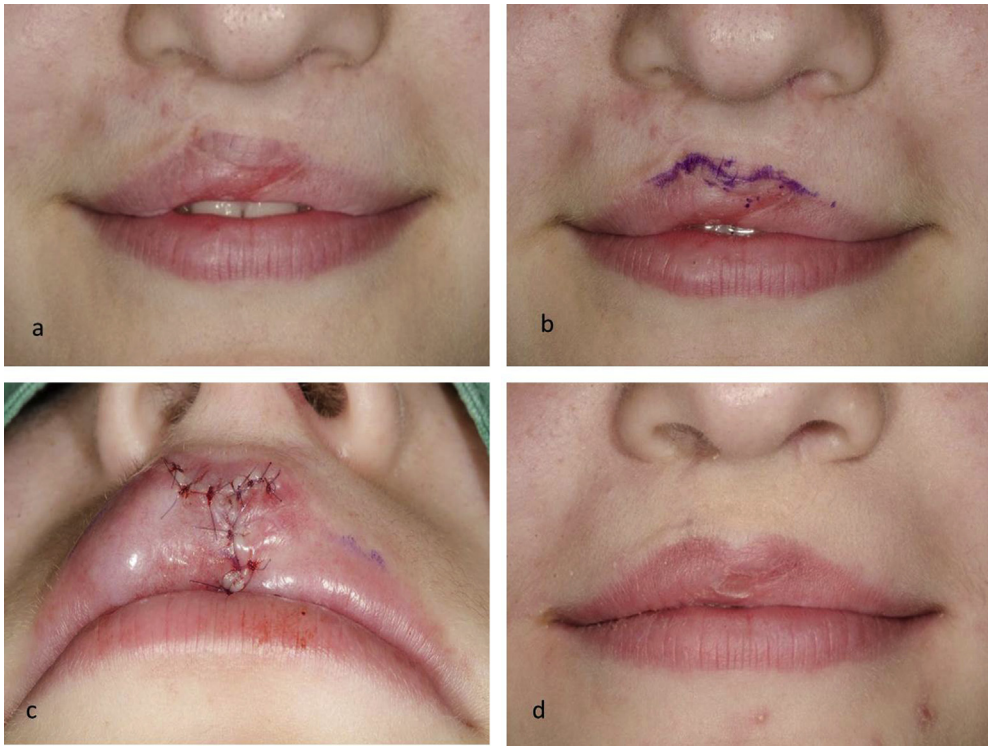

Fig. 4. Vertical Z-plasty with double transposition of myocutaneous vermilion flaps for secondary whistling deformity correction under local anesthesia. Preoperative situs (a) and marking of the desired harmonic Cubid's bow (b). Situs after transposition of two vermilion transposition flaps (c) and postoperative control after 6 weeks (d).

D750 camera (Nikon, Japan) with a 24.3 megapixel sensor (24 × 36 mm) was used with a Sigma 24–105 mm F4.0 DG OS HSM lens (82 mm filter thread) and a Sigma EM-140DG macro ring flash with dual flash tubes (Sigma GmbH, Germany) and high-speed synchro flash (1/120 s). The patient was required to take up certain standardized positions. We were using the PSA and FHL (Frankfort horizontal plane) lines (Ettorre et al., 2006), asking the patient to relax all facial muscles, close their mouth naturally, and look straight ahead.

3. Results

3.1. Defect score

The defect score (DS) for all patients ($n = 34$) was calculated preoperatively (DS: 812.8 ± 901.4) and postoperatively (DS: 28.4 ± 71.2). In 14 patients the defect score was preoperatively classified as a minor defect (DS: 63.5–1372.5), in 14 patients as a moderate defect (DS: 439.6–1372.5), and in six patients as a severe defect (DS: 1672.2–4794.0).

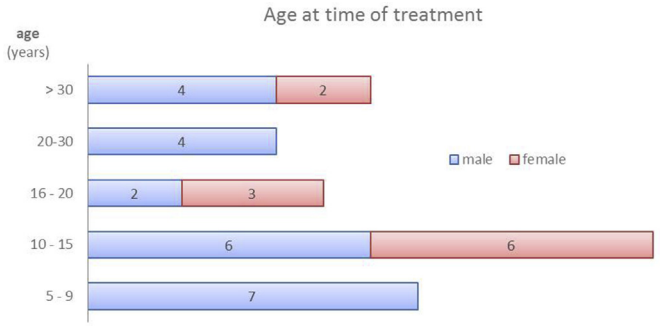

Fig. 5. Age of all patients with whistling deformity reconstruction ($n = 34$) at time of operation.

3.2. Postoperative outcome

After whistling deformity reconstruction (Fig. 5), the defect score was reduced significantly in all three groups: in the minor group (DS < 400) the postoperative scores ranged from 0 to 24.0 ($p_{minor} < 0.001$), in the moderate group (DS: 400–1400) the scores ranged from 0 to 392.2 ($p_{moderate} < 0.001$), and in the severe group (DS > 1400) 0–150.4 ($p_{severe} < 0.001$) (Fig. 6).

In patients with a preoperative minor defect ($n = 14$) the postoperative outcome was 'very good' in 11 (78.6%) and 'good' in three patients (21.4%). Patients with a moderate defect ($n = 14$) showed a 'very good' postoperative result in 10 cases (71.4%), a 'good' result in one (7.2%), and an 'acceptable' result in three (21.4%). Patients with a severe defect ($n = 6$) had a 'very good' postoperative result in two cases (33.3%), a 'good' result in one (16.7%), and an 'acceptable' result in three (50.0%).

The clearest improvement of 97.7% ± 2.8%) was found in patients with a preoperatively severe defect (DS > 1400). In the moderate group the improvement was 95.6% ± 8% and in the minor group 96.7 ± 3.4%. Overall, 'very good' aesthetic and functional results were found in 67.6% across all groups of patients. 14.7% of all patients showed a 'good' result and 17.6% an 'acceptable' one (Fig. 7).

3.3. Limitation of the defect score

The postoperative evaluation of patients with a severe whistling defect (Fig. 8a) clearly shows a much better aesthetic result than the preoperative finding. This is especially evident in the percentage improvement. The criteria used in this evaluation described this

defect score postoperatively as only showing an 'acceptable' result (DS: 366.3) (Fig. 8b).

The defect score used in this study has its limitations in terms of a clear description of the complex causes of a whistling deformity, and describes only the structural deficit in the vermilion (Bhandari, 2014; Choi et al., 2012; Reddy et al., 2009).

4. Discussion

Whistling deformity defects are common after primary lip reconstruction (Reddy et al., 2008). Regardless of the method used for lip repair, these deficits are often observed in the vermilion (Henkel et al., 1998). Many different methods have been described for whistling deformity reconstruction, representing different surgical approaches. Local flap plastics (Choi et al., 2012; Kapetansky, 1971; Nkenke et al., 2013; Song et al., 2011; Wakami et al., 2010) are suggested, as well as the transplantation of different autologous grafts (Baum et al., 2017; Cohen and Kawamoto, 1991; Patel and Hall, 2004; Vecchione, 1982) in order to replace the missing tissue volume and to prevent scar-related shrinkage.

The method for whistling deformity correction described in this study can be performed in patients with uni- or bilateral clefts. The main difference from other methods is the direction of the first cut. A modified Z-plasty in the vermilion is used for vertical extension of the shortened vermilion part of the whistling deformity. The orbicularis oris muscle bundles on both sides of the deformity are not dissected from the subcutaneous tissue or freed up to the nasolabial line. The covering vermilion mucosa with the superficial parts of the underlying muscle bundles is transposed and used to give more volume to the vermilion deficit. A very important point in the reconstruction of the whistling deformity is the reconstruction of the orbicularis muscle (Duffy, 1971) which, through its function and volume, contributes to a harmonious upper lip.

The main disadvantage in this method is that parts of the dispersed wet mucosa can be displaced to the dry vermilion. Prominent deformities can remain because of the exposed oral mucosa in the vermilion region. Patient complaints include

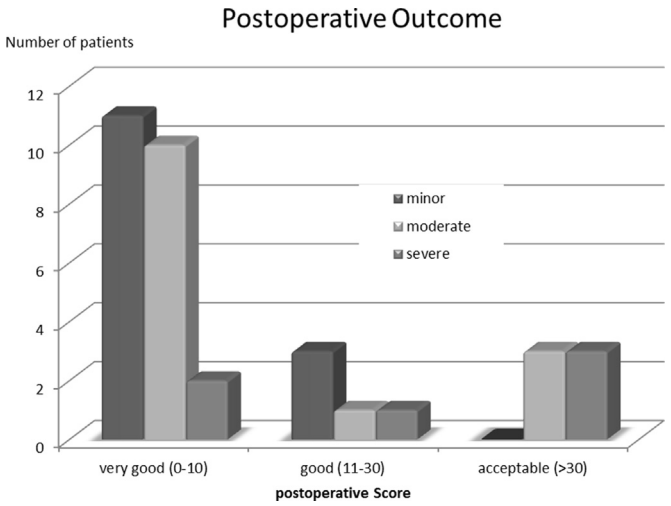

Fig. 7. Postoperative outcome in all patients in relation to preoperative defect score (minor, moderate, severe).

constant scab formation (Mitsukawa and Hosaka, 2008) when the wet lip is placed in a dry environment. This exposure is a result of the difference in the widths of the dry lip and wet lip. Knowledge of the anatomical structure of the normal lip is thus very important. Whistle deformity reconstruction must be performed with regard to the natural muco-vermilion border, also called the 'red line', to avoid this complication (Millard, 1964; Noordhoff, 1984).

The method of evaluation and classification of whistling defects used in this study (Nkenke et al., 2013; Wakami et al., 2010) is shown to be objective with regards the deficit in the vermilion. It cannot describe the whole shape of the deformity and says little about aesthetics and function in the patient. The standardization of the photographs is another weakness (DiBernardo, 1998; Ettorre et al., 2006; Rhee, 2017), but the conditions of a photographic shoot are never identical and sometimes even the complexion of the patient can change. Since the evaluation is based on length

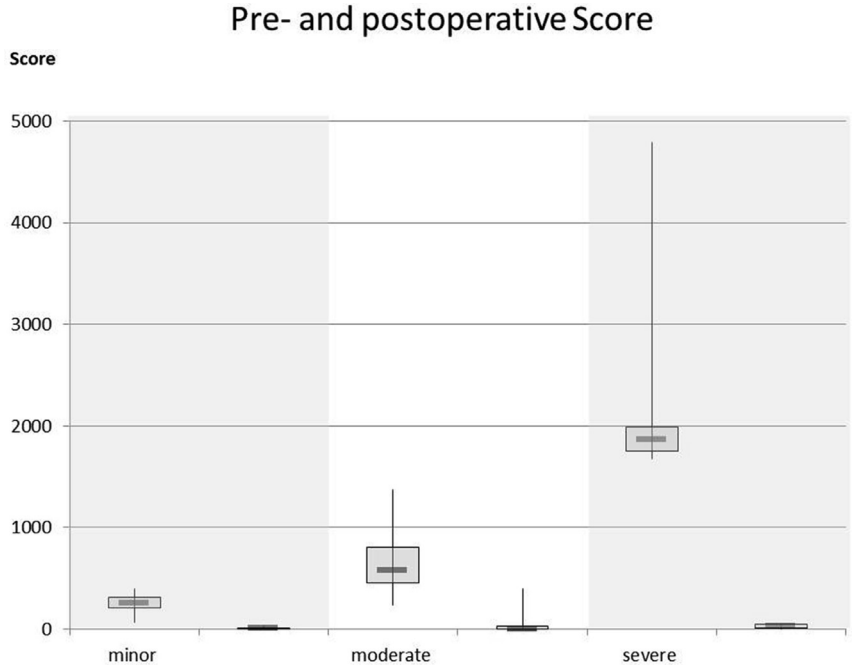

Fig. 6. Pre- and postoperative defect score for all patients ($n = 34$) in relation to primary defect score. A significant postoperative improvement was seen in all groups ($p < 0.001$).

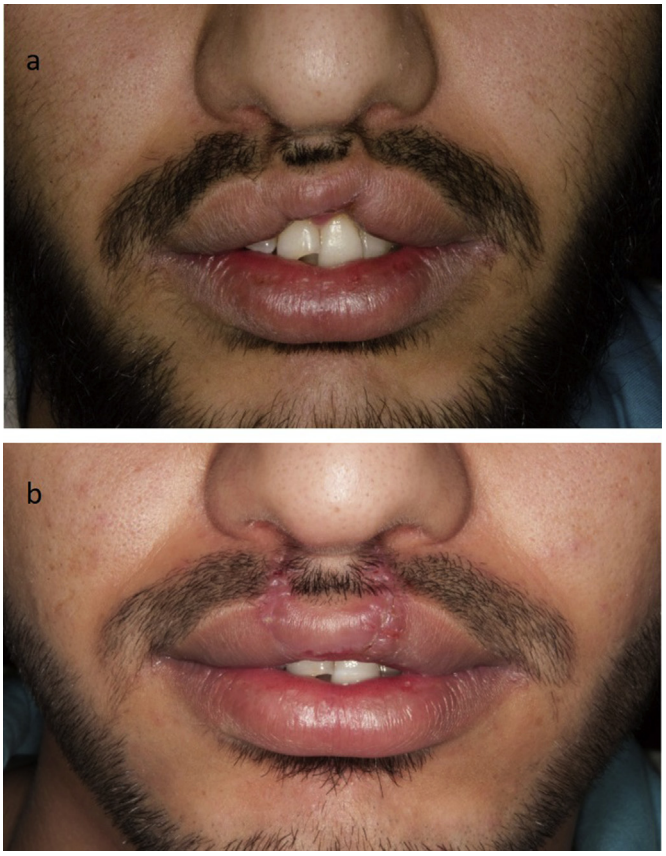

Fig. 8. 21-year-old patient No. 31 (cleft surgery, alio loco) with severe whistling deformity before (a) and after reconstruction (b). The patient was asked to keep his lips slightly open (b). The postoperative result in this case was rated 'acceptable' or 'poor' (postoperative score: 366).

relationships and not absolute lengths, the values obtained can be compared with each other.

5. Conclusion

The advantage of this method is the short operation time and the relatively good aesthetic and functional results. This method is practicable under local anesthesia. The special anatomical conditions of the vermilion and mucosa must be considered in order for the transposition of the musculo-mucosal flaps of the vermilion to lead to a good, long-lasting result.

Conflicts of interest

The authors disclose no conflicts of interest in this study.

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