Stereophotogrammetric Evaluation of Labial Symmetry After Surgical Treatment of a Lymphatic Malformation

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Abstract: Lymphatic malformations (LMs) are rare, nonmalignant masses, frequently involving the head and neck, potentially causing impairment to the surrounding anatomical structures. Major LMs frequently cause facial disfigurement with obvious consequences on self-esteem and social functioning. The attempt to restore symmetry is thus one of the main goals of treatment. In this study, the authors present a not-invasive method to objectively quantify the symmetry of the labial area before and after surgical treatment of a LM, affecting a 16-year-old woman. This was done with sequential three-dimensional stereophotogrammetric imaging and morphometric measurements. The method showed a high reproducibility and supplied quantitative indicators of the local degree of symmetry, helping clinicians in its objective assessment, and facilitating treatment planning and evaluation. A quantitative appraisal of the results can additionally improve patient adherence to a usually multistage therapy.

Key Words: Asymmetry, lips, lymphatic malformation, stereophotogrammetry

Lymphatic malformations (LMs) are rare and nonmalignant masses, made of lymph containing vessels and chambers, which frequently involve the head and neck. Depending on their localization, they can compress or obstruct surrounding structures, thus causing different problems that can compromise facial appearance and aesthetics.[1] Treatment for macrocystic LMs is generally based on sclerotherapy, while microcystic LMs require a surgical approach based on gross debulking and/or camouflage procedures.[2,3]

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Diagnosis and follow-up evaluations of craniofacial diseases can take advantage of computed tomography (CT) and magnetic resonance imaging. Unfortunately, these techniques present some disadvantages such as high costs, radiation exposure (CT), and quite long acquisition time, which make them not suitable for daily use and for repeated follow-up examinations.[4,5] While the visualization of inner hard tissues can be obtained only using these volumetric techniques, the advancement of noninvasive surface technologies has permitted new solutions for the morphological analysis of the external soft tissues only.[6]

In particular, optical systems, like stereophotogrammetry, can obtain three-dimensional (3D) reconstructions of the facial soft tissues in a safe and rapid way, thus allowing repeated assessments, with high levels of accuracy and reproducibility.[6,7]

In this study, we present a series of follow-up stereophotogrammetric evaluations of the labial symmetry of a Caucasoid 16-year-old woman affected by a microcystic LM, who was surgically treated. The evaluations were performed to objectively monitor the treatment progression and final outcome.

The treatment of microcystic LM has always been quite challenging. Indeed, the risk of recurrence is high because complete removal of the malformation is impossible or not advisable, being microcystic LMs generally multifocal.[8,9] Due to the difficulties in the surgical treatment and the recurrence of the disease, patients can lose motivation to adhere to a multistage (more frequently than not) treatment. The method described here allows for easy and fast assessment of the on-going achieved results, providing objective and easy-to-understand indicators, both for the surgeon and, especially, for the patients, who can be encouraged to carry on the treatment phases.

METHODS

The patient involved in this study came at our observation when she was 16 years old. She had a microcystic LM of her right hemiface that had been treated with several partial removals in another hospital since she was 12 years old. During one of these surgical sessions, the facial nerve was injured. The long-standing unilateral facial paralysis was treated with a free gracilis muscle transfer, innervated by homolateral masseteric nerve.[10] The residual deformity involved the right facial soft tissues in the labial area, parasymphysis, and mandibular body.

During the first surgical phase, by careful inspection of the muscle and vermilion, a new commissure was made by removing 2 myomucosal wedges at the angle of the mouth, to symmetrize the mediolateral position of the commissure.[11,12] At the same time, a skin flap was made through an incision into the nasolabial fold to improve the vertical position of the commissure. Then, 6 months after the first surgical phase, an osteotomy of the mandibular body with genioplasty was performed. Seven months after mandibular remodeling, a suspension of the right cheek with fascia lata was made.[13,14]

Three stereophotogrammetric evaluations were performed. The first one was made before the reconstruction of the oral commissure; the second one 6 months after the reconstruction of the oral commissure and before mandibular osteotomy and genioplasty; the last one 6 months after suspension with fascia lata. Figure 1 shows a 3D reconstruction of the patient facial skeleton, obtained from CT data, before and after the surgical procedure. Figure 2 shows the stereophotogrammetric reconstructions of the face taken at the same phases.

All stereophotogrammetric acquisitions were performed using the VECTRA M3 system (Canfield Scientific Inc, Fairfield, NJ). Stereophotogrammetric systems are safe and noninvasive instruments that allow repeated assessment, without any risk for the patient.[5] Furthermore, the system is fast (it needs 3.5 ms to capture

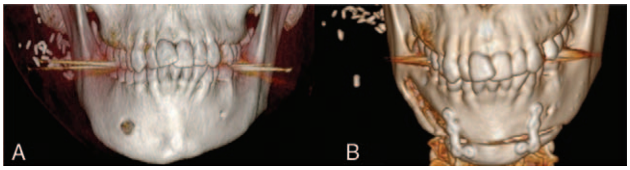

FIGURE 1. Three-dimensional computed tomography reconstruction of the patient facial skeleton. (A) Before surgical treatment. (B) After surgical treatment.

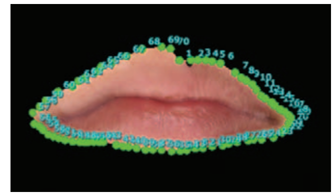

FIGURE 3. Example of labial surface segmentation.

facial images) and allows to obtain 3D reconstructions of the face with a geometry resolution of 1.2 mm.[15]

In detail, the procedure consisted of a preliminary phase, where a set of anatomical landmarks was identified by palpation or visual inspection, and marked on the patient face, using black, biocompatible, liquid eyeliner. This operation was performed by an expert and well-trained operator and executed following a protocol, validated and widely used in our laboratory, which ensures a good accuracy and reproducibility in the measurement of facial soft tissues.[16] In the second phase, the patient was asked to seat in front of the stereophotogrammetric system, in a relaxed way, keeping the face in a neutral expression and teeth in loose contact, and 3D facial images were taken.

After the 3D reconstructions of the face, an off-line working protocol was applied. In this step, the anatomical landmarks were digitally marked on the facial reconstruction, following the previously mentioned protocol.[16] Then, a subset of them was used to automatically select a portion of the facial surface that allows the automatic detection of the midline plane of facial symmetry, excluding confounding areas such as hair and neck. The facial area (FA) and symmetry plane identification processes, performed through the Mirror imaging software (Canfield Scientific Inc, Fairfield, NJ), are described and validated in the literature.[17]

Once the area of interest was defined, the labial surface (LS) was manually segmented from it. Intraoperator repeatability of LS selection was evaluated in a sample of 10 FAs of reference healthy subjects of the same sex, age, and ethnic group of the patient, selected form our dababase. Repeatability analysis was performed only on LS selection, since it was the only manual step of the procedures that had not been validated in previous studies.[17] Linear regression and Bland and Altman[18] analysis were used to test the repeatability of LS selection; an example of LS segmentation is depicted in Figure 3.

During lip segmentation, the operator delimitated the LS using an arbitrarily defined number of 70 points, starting from the landmark "labiale superius" (ls), which is the midline point, located in the upper vermillion line; continuing, clockwise direction, to the same landmark (Fig. 3). Then, using these points, the machine software allowed the automatic selection of the LS, deleting the surrounding FA. The midline plane that had been automatically selected in the previous image processing steps was used to copy and reflect the LS. Subsequently, the root mean square deviation (RMSD) between the original and the reflected LS was calculated. An example of copied and reflected LS is shown in Figure 4.

This off-line protocol was applied to all stereophotogrammetric acquisitions, to quantify the symmetry variations of the LS during the different phases of the surgical treatment. Root mean square deviation evaluation had already proved to be good instrument to evaluate the level of asymmetry.[17,19]

Taking the landmarks "subnasale" (sn, the point located in the midline in the lowest part of the columella), "cheilion" (ch, the point located at the oral commissure), and "stomion" (sto, the point located in the midline between the lips) into account, a series of linear measurements were also performed during the different treatment phases (Fig. 5). In particular, mouth width ($ch_r - ch_l$) and the ch_r-sn, ch_l-sn, ch_r-sto, and ch_l-sto linear distances were calculated (r and l indicate left and right sides of the mouth). The obtained measurements were compared with values coming from the previously selected group of 10 reference women, and *z*-scores values were computed (*z*-scores are obtained subtracting the mean value of the reference group from the patient value and dividing by the standard deviation of the reference group). *Z*-scores were also calculated for RMSD, using reference values coming from the same group.

RESULTS

Linear regression analysis for LS repeated measurements showed an R^2 value of 0.99, indicating a very high correlation between these measurements. Figure 6 shows the Bland–Altman plot for the same measurements. Bias value was very low ($-0.02\,cm^2$), indicating that LS was measured with almost identical values in both the repetitions, and reproducibility was very high (95.9%).

Root mean square deviation evaluated during the subsequent phases of the surgical treatment was respectively 5.8, 2.5, and 1.7 mm. Their progressive reduction, alongside with the surgical

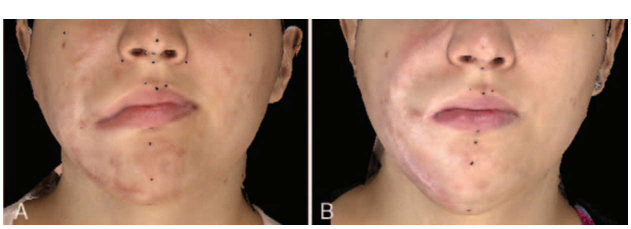

FIGURE 2. Three-dimensional stereophotogrammetric reconstruction of the patient face. (A) Before surgical treatment. (B) After surgical treatment.

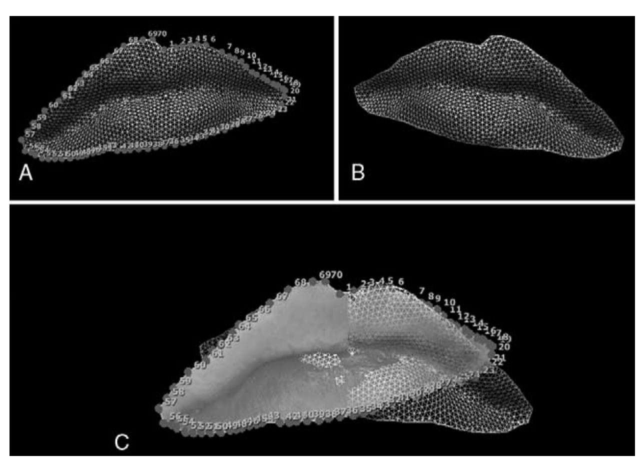

FIGURE 4. (A) Example of segmented labial surface. (B) Labial surface copied and reflected around the Y axis. (C) Superimposition of the original and copied labial surfaces. Segmented lips correspond to the presurgical treatment stage.

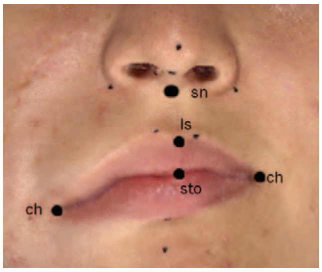

FIGURE 5. Landmarks used to calculate linear distances.

treatment, corresponds to a consequent increase in terms of labial symmetry. Values closer to 0 indicate high symmetry level, while negative or positive values correspond to negative or positive deviations from the perfect symmetry condition.

Z-score values for both the linear measurements and the RMSD are shown in Table 1. During the treatment, the *z*-scores progressively reduced, becoming very similar to those of the reference group (that are equal to 0), and showing the successful result of the surgical interventions.

DISCUSSION

The objective assessment of labial symmetry is a very important element for maxillo-facial and plastic surgeons. Labial asymmetry can be associated with different pathological conditions, which include not only lymphatic but also different malformations, such as cleft lip and palate, macrostomia, Parry Romberg syndrome, and others.[20,21]

Nonetheless, labial asymmetry, as facial asymmetry in general, can affect patients' quality of life, both from functional, aesthetic, and psychological points of view, so a quantitative assessment of the degree of imbalance is more and more required.[17,22]

The definition of morphological parameters to quantitatively assess labial asymmetry may be very useful in clinical practice. It can help surgeons and clinicians to define the best therapeutic options, which drive to satisfying results and permit to motivate the patient to better accept surgery and to be more compliant to the therapy.[20]

Many techniques have been proposed for the assessment of labial asymmetry, ranging from direct, two-dimensional, or 3D assessments; among them, 3D methods have been suggested as the more appropriate.[23] Despite McKearney defined these last, though reliable and repeatable, expensive and time-consuming, optical systems are very fast.[5,20] Furthermore, they permit to acquire 3D

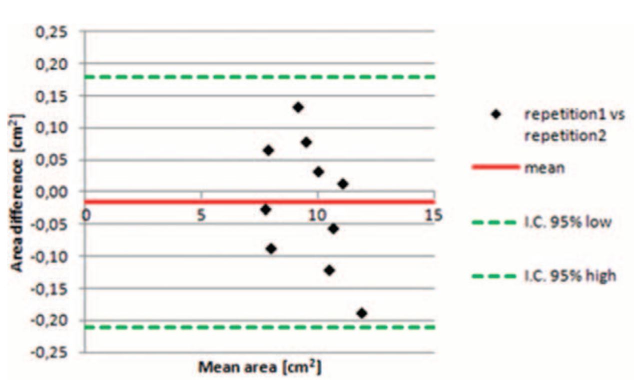

FIGURE 6. Bland and Altman plot for the repeated measurements of the labial area. Continuous line indicates the bias, dashed lines the intervals of confidence.

TABLE 1. Summary of the *Z*-Scores Values Calculated During the Different Treatment Phases for Both Linear Measurements and Root Mean Square Deviation

Z Score	Surgery #1	Surgery #2	Surgery #3
ch-ch	2.5	1.3	0.3
ch_r-sn	5.6	2.8	0.1
ch_l-sn	1.7	1.1	0
ch_r-sto	3.5	0.6	−0.8
ch_l-sto	1.2	1.5	0.5
RMSD	10.6	3.1	1.3

r and l indicate, respectively, the right and left facial sides. *Z* scores of the unaffected side are also reported.

ch, the point located at the oral commissure; RMDS, root mean square deviation; sn, the point located in the midline in the lowest part of the columella; sto, the point located in the midline between the lips.

facial, and consequently labial, morphology at virtually no cost, except for the initial price of the instrument and its software. Additionally, being noninvasive, they permit executing multiple acquisitions without any danger for patients and operators.

Furthermore, a 3D approach allows rotating the image around all the axes, thus facilitating, for example, the segmentation of the region of interest, as the LS. This procedure, in fact, becomes more precise once an arbitrary number of points are defined, which can be placed around the labial profile and visualized from multiple points of view. The aforementioned procedure improves, indeed, the intraoperator repeatability of LS selection, compared with that achieved by freehand drawing, as performed by Russell et al in two-dimensional labial images.[24,25]

Indeed, in the measurement protocol used in the current study, FA selection and labial segmentation remain the only manual steps. Facial area selection has proved to be repeatable and reliable.[17] For what concerns labial segmentation, Bland and Altman[18] analysis confirmed the fine intraoperator repeatability. The bias of −0.16% indicates a negligible systematic error of under-estimation, compared with the measurement dimension, as depicted in Figure 6.[18]

Furthermore, RMSD, which was used to objectively assess labial asymmetry, proved to be a good asymmetry indicator, confirming literature findings.[17,19,26] Indeed, the patient analyzed in the present study underwent a remarkable reduction of the RMSD (and corresponding *z*-score value), after the first surgical step (Table 1). This means a marked decrease of labial asymmetry and justifies the surgical reconstruction of the oral commissure as first-line treatment: this choice was done to meet the patient's compelling requirements, since the unsatisfying results achieved by the previous operations. The increase of symmetry was maintained during the successive surgical treatments, as demonstrated by the additional reduction of the RMSD *z*-scores.

The same reduction was observed for the *z*-scores of the calculated linear distances. For example, at the beginning of the treatment the patient's mouth width was 2.5 SD bigger than the average mouth width of the reference women. At the end of the treatment it was 0.3, practically identical to the average value of the reference group.

It is not possible to achieve a perfect symmetry in biological systems, and perfect symmetrical facial features are not always considered attractive; however, in case of severe asymmetry, social life and psychology of the affected people can be seriously influenced.[27,28] Since many conditions cause the asymmetry of labial area, our study provides a reliable, fast, easy, and safe method to help clinicians in the initial evaluation of the involved area, and its follow-up. Furthermore, the possibility to easily quantify the surgical results motivates the patients who, having an objective parameter for treatment assessment, can be more compliant to the therapy and more confident of clinicians' and surgeons' therapeutic decisions. This holds particularly true when dealing with facial microcystic LMs where the anatomy is usually severely subverted and where clear guidelines to symmetrize the face are still lacking.

ETHICS CONSENT AND PERMISSION

Before the acquisition of the face through a stereophotogrammetric system, all the procedures were explained to the patient and her parents. They read and voluntarily signed an informed consent, whose written copy is available for the Editor in Chief of this Journal. Procedures were also previously approved by the local Ethics Committee [Università degli Studi di Milano, June 27, 2014, no. 266 230 92/2014]. All clinical procedures were explained to the patient and her parents, and informed consent was obtained according to the local hospital guidelines. The patient was free to leave the treatment and morphometric evaluations in any moment.

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