ORIGINAL ARTICLE

Quantitative Computer-Based Assessment of Lip Symmetry Following Cleft Lip Repair

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Objective: To demonstrate an objective method of measuring lip symmetry after cleft lip repair by comparing patients with unilateral cleft lip (UCL) to non-cleft lip controls using the SymNose computer program.

Design: Retrospective cohort study comparing several parameters of lip symmetry between UCL cases and controls. Participants were represented by digital photographs that were traced using SymNose.

Setting: This study was performed at the South West Cleft Unit, Bristol, U.K.

Participants: Forty-four participants aged 10 years (± 1 year) were recruited into and completed the study. A consecutive case series of 22 patients with UCL \pm palate had previously undergone primary repair. Twenty-two age-matched controls were recruited from a local primary school.

Main Outcome Measures: Lip symmetry, expressed as the percentage mismatch of an overlaying of the left upper lip area and upper vermillion border area over the right, horizontal lip tilt, and lateral deviation of the lips.

Results: A significant increase in labial asymmetry was seen in the UCL group in the following parameters: upper lip mismatch and upper vermillion border mismatch. No significant differences were observed in horizontal lip tilt or lateral lip deviation.

Conclusion: This study supports the use of SymNose for the objective quantitative assessment of lip symmetry as an outcome measure of surgery following cleft lip repair. It allows comparison of surgical techniques and can be used to perform audits. It is a time-efficient process, relatively inexpensive, and straightforward to use.

KEY WORDS: computer-based, long-term follow-up, photographs, symmetry, SymNose, unilateral cleft lip and palate

The expression "beauty is in the eye of the beholder" characterizes the difficulties faced in attempting to objectively measure facial appearance as an outcome of cleft lip repair. In order to improve patient care through audit, current practice must be quantified against a measurable standard. This is particularly difficult for surgical procedures in which improved appearance is a primary outcome. Effective assessment is required to determine treatment success that, among other things, is affected by the surgical technique used and the time of intervention. Surgery is efficacious in improving the aesthetic outcome of cleft lip and has been shown to reduce the psychological effects associated

with it, improving self-esteem and confidence (Lefebvre and Munro, 1978). Despite advances in surgical techniques, facial appearance is not always completely restored to how it would be had the child not been affected by cleft lip.

Panel assessment of facial symmetry following cleft lip repair has been used to measure surgical outcome, but it is a time-consuming process that is not quantitative. The need for objective assessment has led to the development of various craniofacial anthropometric techniques, which perform a more objective two-dimensional (2D) or three-dimensional (3D) assessment. Although 3D assessment has been shown to be useful and accurate, it is limited by the current unavailability of equipment for routine clinical use, the training required, and the cost of the equipment.

A possible solution to the difficulties encountered in objective facial assessment was devised by Pigott and Pigott (2010) with the creation of the SymNose computer program. It assesses 2D digital images of faces and evaluates symmetry as a proxy for cosmetic surgical outcome. A systematic search performed on Medline (1950 to July 2011) of 17 key search terms identified only four papers using a computer-based assessment of lip

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symmetry after cleft lip repair (Yamada et al., 2002a, 2002b; Bilwatsch et al., 2006; Proff et al., 2006). All four used 3D technology, which is currently not routinely available. SymNose is able to assess symmetry from the frontal facial view for the nose and lips, and sub-mental ("worm's-eye") view of the base of the nose. Objective computer-based methods of assessing surgical outcome have typically focused on nasal anatomy (Coghlan et al., 1987; Yamada et al., 1999). For this reason this study made use of the frontal view to assess lip symmetry in order to evaluate SymNose as a tool for assessing surgical outcome. It measures the degree of residual lip asymmetry in children with UCL compared to children without cleft lip of the same age. The aim of this study is to provide new information on a novel 2D technique for measuring lip and nose symmetry following cleft lip repair that could be standardized for routine clinical use.

This retrospective cohort study compares 22 patients with repaired unilateral cleft lip (UCL) ± cleft palate to 22 controls to determine if there is a quantifiable difference in lip symmetry from 2D photographs as determined by SymNose.

METHODS

Patients and Participants

The photographs of a consecutive series of 22 children with a history of unilateral cleft lip and palate treated at the South West Cleft Unit were selected to be evaluated using SymNose. The children had undergone primary lip repair by the same cleft surgeon shortly after birth. The photographs were taken at follow-up clinics at age 10 years (±1 year) prior to alveolar bone grafting, in keeping with the standard audit protocol. Children with bilateral cleft lip and palate were excluded. This is because SymNose is limited to producing data based on midline symmetry and these cases are therefore likely to be relatively symmetrical. Additionally, unilateral cleft lip and palate cases form the basis of international audit and research.

The noncleft control cases were recruited from a local primary school. A series of 22 high-quality digital images were made available to us. They were frontal facial views, comprising of 19 boys and three girls aged 10 years (±1 year). The participants were matched for age, sex, and geographical region of England.

In a pilot run, 22 participants in each group were traced by one assessor, and the mean difference in lip symmetry (measured in percentage mismatch of left upper vermillion border reflected upon the right) was found to be 12.1% (control group, 10.4%: UCL group, 22.5%). Based on this preliminary finding, a power calculation demonstrated that 21 participants in each group were required in order for a Mann-Whitney U test to have a 90% chance of detecting a difference in mean lip symmetry of 10% with the level of significance set at P < .05.

Materials

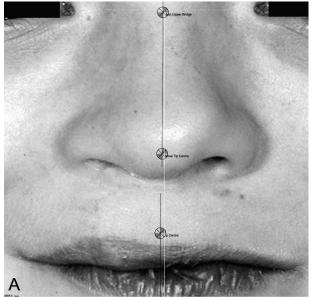
Assessment of lip symmetry was performed using SymNose on an Apple Macintosh OSX 10.4. This was used in conjunction with a digitizing pad (Wacom Intuous 3 PTZ930) and dedicated stylus to allow the operator to accurately trace around the lips of a 2D enlarged photographic image. Photographs were taken using a Sony Cybershot camera. A full description of the methods and materials is available from Pigott and Pigott (2010) who first described the use of this method of assessing facial symmetry.

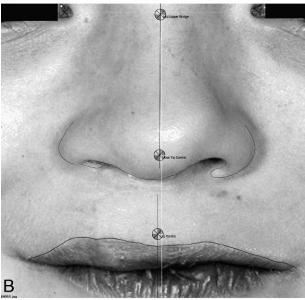
Protocol

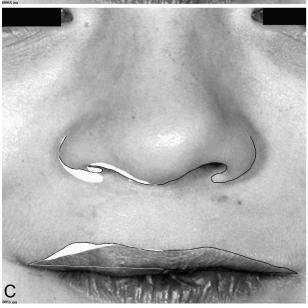
Frontal photographs were taken at age 10 years (± 1 year) using studio lighting with the patient facing the camera directly and their facial muscles relaxed. Photographs were taken at a distance of 1.8 m to minimize parallax, using a 105-mm lens. Photos were cropped vertically, leaving just the canthi and labial commissures visible to maintain patient anonymity. Using SymNose, target roundels (bullseyes) were placed on the canthi, upper nose bridge, center nose tip, and middle upper lip (Fig. 1A). These anatomical landmarks have been recognized and used in previous studies of facial symmetry (Amaratunga, 1988; Farkas et al., 1993). The user then traced from the superior aspect of the right alar groove, beneath the nostrils and columella to the contralateral alar groove. A second line was traced along the upper vermillion border from the right to left commissure (Fig. 1B). SymNose then reflected the tracing across either a user- or computer-defined midline to calculate the percentage mismatch (Fig. 1C). To reduce intra- and interobserver variability, each image was assessed four times; twice by a separate assessor, each on separate days. A mean value for these four was taken for each of the five parameters of symmetry that SymNose calculates.

Statistical Analysis

The five parameters of lip symmetry of the repaired UCL group were compared to the control group using a two-way Mann-Whitney U test. The data were then normalized by presenting the repaired UCL group data as a percentage of the control value normalized at 100%. We accepted a P value of <.01 after Bonferroni correction of the .05 level for n=5 comparisons, to disprove the null hypothesis that there is no difference in lip symmetry between the UCL group and the control group for each parameter of lip symmetry. Bonferroni correction was used because multiple (five) comparisons were made between groups. In order to maintain the familywise error rate (i.e., a probability of .05 of rejecting a true null hypothesis) each individual hypothesis was tested against a fraction of the statistical significance level desired.







To measure the amount of intra- and interobserver reproducibility, Spearman's rank correlation coefficient was calculated, comparing the tracings used to calculate the percentage of upper vermillion border mismatch for the UCL group. This parameter and group were chosen because we anticipated that this group would be harder to reproducibly trace and this parameter relies on both user roundel placement and tracing. This would therefore demonstrate the level of reproducibility for images that are the most challenging to trace and for the most userdependent parameter. Two sets of measurements from one user were used to calculate the correlation coefficient as a measure of intra-observer reproducibility. Two sets of measurements, one from two separate users were used to calculate the correlation coefficient as a measure of interobserver reproducibility.

Ethics and Consent

This study was approved by a National Health Service ethics committee, and informed consent was obtained from the legal guardians of all patients prior to obtaining the images for their use in this study.

RESULTS

In total, 44 white children aged 10 years old (± 1 year) participated in the study. These children formed two groups: a repaired UCL \pm cleft palate group (n = 22; 19 boys, three girls) and a noncleft control group (n = 22), matched for age and sex.

The UCL group demonstrated a significant increase in lip asymmetry for three out of the five parameters measured. A statistically significant (P < .01) difference in means between the groups was found for the following parameters (Table 1):

- vermillion border mismatch across a user-defined axis (UALip)
- vermillion border mismatch across a computer-defined axis (CALip)
- upper lip mismatch (UpLipDiff).

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FIGURE 1 A: Frontal view photograph of upper lip and nose. Cross-wire roundels placed by the user on the canthi allow SymNose to draw a horizontal line between them with a vertical line bisecting the midline. Roundels are then placed on the upper nasal bridge, nose tip, and mid-upper lip. B: The user then traces the lower nasal border from the right alar groove, beneath the nostrils and columella to the left alar groove. A second line is traced from the right labial commissure and along the upper vermillion border to the left commissure. C: SymNose draws a vertical midline as defined by the user-positioned roundels and reflects the left vermillion border and nose tracings onto the right to calculate the area of non-overlap (shaded white).

UPLipDiff (%) CALin (%. UALin (%) Lip Oblique (degrees) En-Ch (%) UCL (n = 22): Mean (SD) 19.1 (± 12.1) $20.2 (\pm 11.7)$ $11.8 (\pm 6.1)$ $2.1 (\pm 1.6)$ $2.9 (\pm 1.8)$ Control (n = 22): Mean (SD) $8.5 (\pm 4.6)$ $9.9 (\pm 4.9)$ $6.5 (\pm 2.8)$ $1.5 (\pm 0.7)$ $1.9 (\pm 1.0)$ 10.6** 10.3** 5.3* Difference between means 0.6 1.0 95% Confidence interval 5.1 to 16.1 4.9 to 15.7 2.5 to 8.1 0 to 1.3 0.1 to 1.9

TABLE 1 Comparison of Lip Symmetry Between UCL and Control Groups^{†‡}

The obliquity of the lip (Lip Oblique), defined as the angle that the lips are deviated from the horizontal plane, and the lateral lip deviation (En-Ch) showed no statistical difference between groups. The data were normalized to allow the proportional increase in lip asymmetry in the UCL group to be numerically and graphically compared to the control group (Fig. 2).

Center Axis Lip (CALip)

The CALip is the percentage mismatch of the left vermillion border compared to the right side, across a computer-defined midline. The percentage mismatch is a proxy for asymmetry. The greater the mismatch, the less symmetry there is. The control group had a mean mismatch of 8.5% whereas the UCL group had a mismatch of 19.1% (difference between means = 10.6%; P = .0002).

User Axis Lip (UALip)

UALip is similar to CALip, except that the left vermillion border is reflected upon the right side across a user-defined midline, according to where the upper lip center roundel is positioned. The percentage mismatch of the left vermillion border upon the right is then calculated. Controls had a mean mismatch of 9.9% versus 20.2% in the UCL group (difference between means = 10.3%; P = .0002). Normalizing the data (Fig. 2) shows that the UALip parameter detects less mismatch than the CALip parameter.

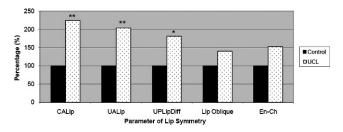


FIGURE 2 Bar chart showing the UCL group as a percentage of the normalized control group. *P < .01, **P < .001 by two-way Mann-Whitney U test.

Upper Lip Difference (UpLipDiff)

UpLipDiff is the percentage mismatch of the left upper lip compared to the right upper lip area. The upper lip borders are enclosed laterally by lines created by SymNose from the labial commissures to the widest parts of the alar bases. The inferior edge is formed by the vermillion border. The superior border is formed by the observer-drawn tracing of the alar base and columella. The midline is defined by the user from the roundel placed over the philtrum. When the right and left sides are folded across this midline, the percentage area mismatch is calculated by SymNose. Controls had 6.5% mismatch, whereas the UCL group had 11.8% (difference between means =5.3%; P=.001).

Lip Oblique

Lip Oblique looks at the degree of lip tilt in the horizontal plane, based on the intercanthal line. The directionality of lip tilt (clockwise: positive, anticlockwise; negative) was removed to allow the mean lip tilt to be quantified as an absolute value. The control group had a mean horizontal lip tilt of 1.5° whereas the UCL group had a mean tilt of 2.1° . However, this difference was not statistically significant (difference between means = 0.6° ; P = .403).

Endocanthus to Cheilion Ratio (En-Ch)

En-Ch is the lateral deviation of the lips from the vertical midline. It looks at the distance between the medial canthus (endocanthus) and labial commissure (cheilion) on the left and right side and compares them in the form of a ratio of (right - left/larger of right or left) and multiplies this by 100 to create a percentage. The control group had and En-Ch ratio of 1.9% and the UCL group ratio was 2.9% (difference between means = 1.0%; P = .0325).

Accuracy

One observer retraced 22 different images to produce a value for UALip. The analysis showed a high level of intraobserver correlation (Spearman r = .76, P < .0001) (Fig. 3). A high level of interobserver correlation between

[†] Table showing the differences between the UCL and control groups for each parameter of lip symmetry reported by SymNose.

[‡] CALip – Percentage mismatch of left vermillion border compared to right across a computer-defined axis; UALip – percentage mismatch of left vermillion border compared to right across a user-defined axis; UpLipDiff – percentage mismatch of left upper lip compared to right; Lip Oblique – horizontal lip tilt; En-Ch – lateral deviation of the lips from the vertical midline.

* P < .01, ** P < .001 by two-way Mann-Whitney U test.

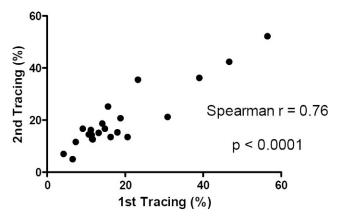


FIGURE 3 Intra-observer reproducibility. Scatter plot showing the correlation between 22 repeated tracings analyzed for lip asymmetry (UA Lip) performed by the same observer.

two users who traced the same 22 images was also observed (Spearman r = .67, P < .0006) (Fig. 4).

DISCUSSION

Al-Omari et al. (2005) systematically reviewed the literature relating to the objective assessment of cleft-related facial appearance and found a variety of studies using vastly different techniques for the same purpose. Although anthropometric measurements and 3D techniques provide quantifiable and reproducible measurements of facial form, they are time-consuming, costly, and often based in academic research centers. There is therefore a need for a standardized, objective, and reliable method for assessing the outcome of cleft lip repair in order to assess differences between surgeons and for use in research and audit. This would facilitate the comparison of surgical outcomes between clinics and promote multicenter collaboration, with the ultimate goal of improving patient care.

SymNose offers a method to address these needs, and this study evaluated its suitability by investigating how it assesses labial symmetry. Whilst SymNose is capable of

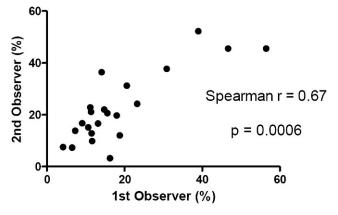


FIGURE 4 Interobserver reproducibility. Scatter plot showing the correlation between 22 tracings analyzed for lip asymmetry (UA Lip) performed by two observers.

assessing nasal symmetry, the study focused solely on the lips because there is an extensive literature base on the nasal form. This study revealed that children at the age of 10 years with repaired UCL had less symmetrical lips in comparison to a matched cohort of children without cleft lip. It should be noted that in the control group, a degree of facial asymmetry was normal across all of the parameters. The difference in symmetry between groups was significant for three out of the five parameters measured by SymNose. The parameters of Lip Oblique and En-Ch did not show any significant difference between the groups. This latter result is in keeping the findings of Kyrkanides et al. (1996), who studied the symmetry of the upper lip at its midpoint.

The accuracy of SymNose has previously been demonstrated by Pigott and Pigott (2010), who produced a standard deviation of 0.25 based on a mean lip mismatch (UA Lip) of 22.53%. A high level of intra-observer (r = .76) and interobserver reproducibility (r = .67) was found in this study. Repeat tracings are therefore not necessarily required; however, reproducibility may be confirmed by tracing each image twice.

A potential limitation of SymNose is that there is an inherent minor element of subjectivity involved because it requires user input in the form of image tracing and anatomical landmark positioning. This is dependent on the performance of the user and the quality of the images being used. It is therefore recommended that some time is taken to become familiar with the program and the procedure for tracing images, prior to data collection. Repeating measurements on separate days and times, between different users, will help to improve the reliability of the results.

SymNose reports data relating to lip symmetry across five different parameters, which means that there is no single global score for labial symmetry. A future version of SymNose could achieve this by combining the parameters in a weighted equation to produce an overall outcome score for symmetry.

It takes less than 3 minutes for an assessor to trace an image and for SymNose to calculate the values for all of the parameters. Allowing for repeat measurements to improve intra- and interobserver reliability, a case load of around 30 new cleft cases per year could therefore be audited in around 3 hours once familiar with the program. This processing rate is comparable to cephalogram (lateral skull radiograph) tracings performed by orthodontists for audit purposes.

The evaluation of outcomes after cleft lip surgery is challenging. Although the use of SymNose for the objective quantification of facial appearance is valuable, it represents a simplified method of addressing a highly complex problem and should not be relied upon as a stand-alone measure. It should ideally be coupled with other important outcomes such as scar quality and lip function and be used alongside overall clinical judgment, taking the patients' and their carers' views into consideration. Nevertheless, this study supports the use of SymNose for assessing labial symmetry in patients with UCL. This method is easy to use,

quick, reliable, reproducible, and relatively inexpensive to set up. In contrast to other techniques that are based in large research institutions, SymNose can be readily used in a clinic setting for auditing purposes, allowing surgeons to compare techniques and outcomes. Future work leading on from this study could include a SymNose-based measure of symmetry in patients with UCL before and after surgery compared to a panel-based assessment, similar in format to a previous study by Laitung et al. (1993).

CONCLUSION

This study found that SymNose is a useful tool for assessing labial symmetry in UCL patients, with significant differences in three out of the five parameters reported. The program is free and easy to use, requiring minimal training. SymNose has allowed us to produce standard data from a group of noncleft images, which could be used for future auditing. This study represents the first time that SymNose has been evaluated for the assessment of labial symmetry in UCL patients. Our findings support its use for this purpose, and we recommend SymNose as an effective and reliable method of examining lip symmetry for the purposes of audit and research. While the program has a few limitations, it offers several distinct advantages over other assessment methods and allows objective evaluation of the results of different techniques and surgeons.

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