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The use of SymNose for quantitative assessment of lip symmetry following repair of complete bilateral cleft lip and palate

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Introduction: The SymNose computer program has been proposed as an objective method for the quantitative assessment of lip symmetry following unilateral cleft lip repair. This study aims to demonstrate the use of SymNose in patients with complete bilateral cleft lip and palate (BCLP), a group previously excluded from computer-based analysis.
Methods: A retrospective cohort study compared several parameters of lip symmetry between BCLP cases and non-cleft controls. 15 BCLP cases aged 10 (±1 year) who had undergone primary repair were recruited from the patient database at the South West Cleft Unit, Frenchay Hospital. Frontal facial photographs were selected for measurement. 15 age-matched controls were recruited from a local school. Lip symmetry was expressed as: percentage mismatch of left vermillion border and upper lip area over the right, horizontal lip tilt and lateral deviation of the lip.
Results: A significant increase in lip asymmetry was found in the BCLP group expressed as upper vermillion border mismatch across computer-defined and user-defined midlines (mean difference was 16.4% ($p < 0.01$) and 17.5% ($p < 0.01$) respectively).
Conclusions: The results suggest that a significant degree of lip asymmetry remains in BCLP patients even after primary repair. This challenges previous assumptions that those with bilateral defects would be relatively symmetrical.

1. Introduction

Children with cleft lip and palate are at risk of developing psychological and social sequelae through lack of personal adjustment to their facial deformity. One of the main aims of cleft surgery is to reduce this visible deformity to enhance patient-related quality of life. However, given the highly subjective nature of facial appearance and attractiveness, the reliable assessment of treatment outcome remains a challenge. As of yet, no single method of quantifying treatment success has been agreed upon, making it difficult for cleft units to audit their results and compare surgical techniques.

A method of objective facial assessment was developed by Pigott and Pigott (2010) in the form of the SymNose computer program. The SymNose program uses two-dimensional (2-D) digital images of faces and requires the user to mark important anatomical landmarks including the medial canthi, nasal bridge, nose tip and middle upper lip. SymNose uses these landmarks to calculate both user- and computer-defined midlines. Tracings made by the user are reflected across these midlines in order to calculate the degree of facial asymmetry. Degree of asymmetry, or lack thereof, is used as a proxy for treatment success.

The importance of symmetry in areas close to the midline has been emphasised previously (Springer et al., 2007). In this study, panel assessment concluded that slight but visible lateral orbital asymmetries did not diminish attractiveness. Moreover, lateral asymmetries were found to be significantly more attractive than those close to the midline. These findings were applied to the field of cleft surgery by Meyer-Marcotty et al. (2011), who analysed facial symmetry and perceived attractiveness in patients with unilateral cleft lip (UCL) compared to those with severe Class III malocclusion. It was found that although there were no quantitative differences in facial symmetry between patient groups, UCL patients were rated as significantly less attractive than malocclusion patients by panel assessment. It was concluded that not only the extent, but the location of asymmetry has an influence on attractiveness. Through the objective assessment of lip symmetry in areas close to the midline, SymNose focuses on the area of the face considered most important towards affecting perceived attractiveness.

SymNose has been proposed as a time-efficient and straightforward tool for the objective quantitative assessment of lip symmetry

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following cleft lip repair, and has been used to compare patients with UCL to non-cleft controls (McKearney et al., 2012). Results showed a significant increase in lip asymmetry in the cleft group, suggesting that a degree of facial deformity remains in these patients even after primary surgical repair. The authors suggested that the use of SymNose is limited to the measurement of unilateral defects, due to its reliance on data based on midline symmetry. For this reason, individuals with bilateral cleft lip and palate (BCLP) have thus far been excluded from computer-based assessment. Given that children with BCLP require an equal standard of audit as those with unilateral defects, the efficacy of SymNose for use in this patient group remains an important research question. This study therefore aims to assess the effectiveness of SymNose for measurement of lip symmetry following primary surgical repair of complete BCLP.

A retrospective cohort design is used to compare 15 complete BCLP cases with 15 age-matched non-cleft controls using the five parameters of lip symmetry measured by SymNose. A statistically significant difference in one or more of these parameters will be used as evidence against the null hypothesis that there is no difference in lip symmetry between BCLP cases and controls.

2. Material and methods

2.1. Patients and participants

Cases were identified from a computer database of all cleft lip repairs carried out in the UK at the South West Cleft Unit, situated in Frenchay Hospital, Bristol All repairs were performed between 1991 and 2002 by the same consultant cleft surgeon. Eligibility criteria included age 10 years ($\pm$1 year) at the age of follow-up as well as the completeness of the defect as documented in the patient notes using the LAHSAL code (Kriens, 1989). Only patients coded as having complete bilateral cleft lip as well as complete cleft palate were included. Frontal photographs were taken in clinic prior to alveolar bone grafting, as per the unit's audit protocol. Patients with Taunton and Gloucester postcodes (medium sized towns within the catchment area of Frenchay Hospital) were excluded from the study due to difficulties obtaining their digital photographs. One further patient was excluded due to lack of written consent. The digital photographs of a series of 15 children were thus obtained from the medical illustrations department to be evaluated using SymNose.

A series of 25 photographs of age-matched, non-cleft controls from a local primary school was made available to us. These were frontal facial views taken using the same image protocol as the case series. Eight controls were excluded due to the subject either smiling or having their mouth open. From the remainder, 15 were randomly selected to be included.

2.2. Materials

Lip symmetry was assessed using the SymNose program installed on an Apple Macintosh OSX 10.4. Photographs were obtained from the medical illustrations department and scanned onto the hard drive using a Canon CanoScan 5600F Film Scanner at 4800×9600 dpi resolution. Images were cropped vertically using Adobe Photoshop CS6 to leave just the canthi, nose, upper lip border and labial commissures visible. Tracing was carried out by enlarging the image onto a Wacom Intuous 3 digitising pad and using a compatible digital stylus to trace directly onto the screen.

2.3. Protocol

Frontal facial photographs were selected from images supplied by the medical illustrations department. Photographs were taken using a Nikon D3 + 105 mm lens with studio lighting at a distance of 1.8 m to minimise parallax. The patient was facing the camera directly in a head-neutral posture with their mouth closed and facial muscles relaxed in order to minimise any asymmetry relating to facial expression. Images were scanned and subsequently saved into a single folder. Cropping was carried out by opening the image document in Adobe Photoshop CS6 and using the 'Crop' tool to preserve just the canthi, nose, upper lip border and labial commissures. Surrounding facial features were excluded so as not to influence any subsequent measurements (Asher-McDade et al., 1991). Cropped images were saved into a new folder and opened using SymNose. Images were projected onto the digitising pad and magnified up using the scale bar until the inter-alar distance reached 150 mm. This magnification was chosen as it was the point at which the image could be accurately traced around without becoming overly pixelated. Cross-wire bull's-eyes (or 'roundels') were dragged by the user onto the right and left canthi, nasal bridge, centre nose tip and middle upper lip (Fig. 1, A). The roundel placed on the middle upper lip creates a user-defined midline, whilst the roundels placed on the canthi allow SymNose to draw a horizontal line between them and a vertical line bisecting the midline (computer-defined midline). Tracing was carried out by the user using a digital stylus directly onto the pad (Fig. 1, B). The lower nasal border was traced starting from the superior aspect of the right alar groove, beneath the nostrils and columella to the opposite alar groove. Any mistakes were corrected at the time of tracing. A second line was traced along the upper lip border from right labial commissure along the upper vermillion border to the opposite commissure. The 'Calculate' function was then selected using the 'Actions' tab. At this point, SymNose reflects the left vermillion border and left lower nose border over the right to produce an area of non-overlap (Fig. 1, C). This area is presented as a percentage mismatch between left and right, and is a proxy for asymmetry. In order to account for intra- and inter-observer variability, each image was traced twice by two different assessors, with each set of tracings carried out on separate days.

2.4. Statistical analysis

The five parameters of lip symmetry of the BCLP group were compared against controls using a two-way Mann–Whitney *U* Test. This is the non-parametric equivalent of a two-sample *t*-test, and is used to provide evidence of a statistically significant difference between the samples. The resulting *p* value is used to provide evidence for or against the null hypothesis that there is no difference in lip symmetry between BCLP patients and non-cleft controls. A *p* value of $<$0.01 was accepted as statistically significant after Bonferroni correction of the 0.05 level for $n = 5$ comparisons. Bonferroni correction reduces the likelihood of incorrectly rejecting the null hypothesis (Type I error) by maintaining the familywise error rate across all five comparisons. The data was then normalised by presenting the BCLP group as a percentage of the control value normalised at 100%.

Intra- and inter-observer reliability were quantified using Spearman's rank correlation coefficient. Two sets of tracings from the same assessor measuring upper vermillion border mismatch across a user-defined midline (UALip) were compared. The resulting correlation coefficient was used as a measure of intra-observer reliability. The mean value for this parameter was compared against the corresponding mean from the second assessor as a measure of inter-observer reliability.

2.5. Ethics and consent

This study was approved by a National Health Service ethics committee, and full informed consent was obtained from the legal

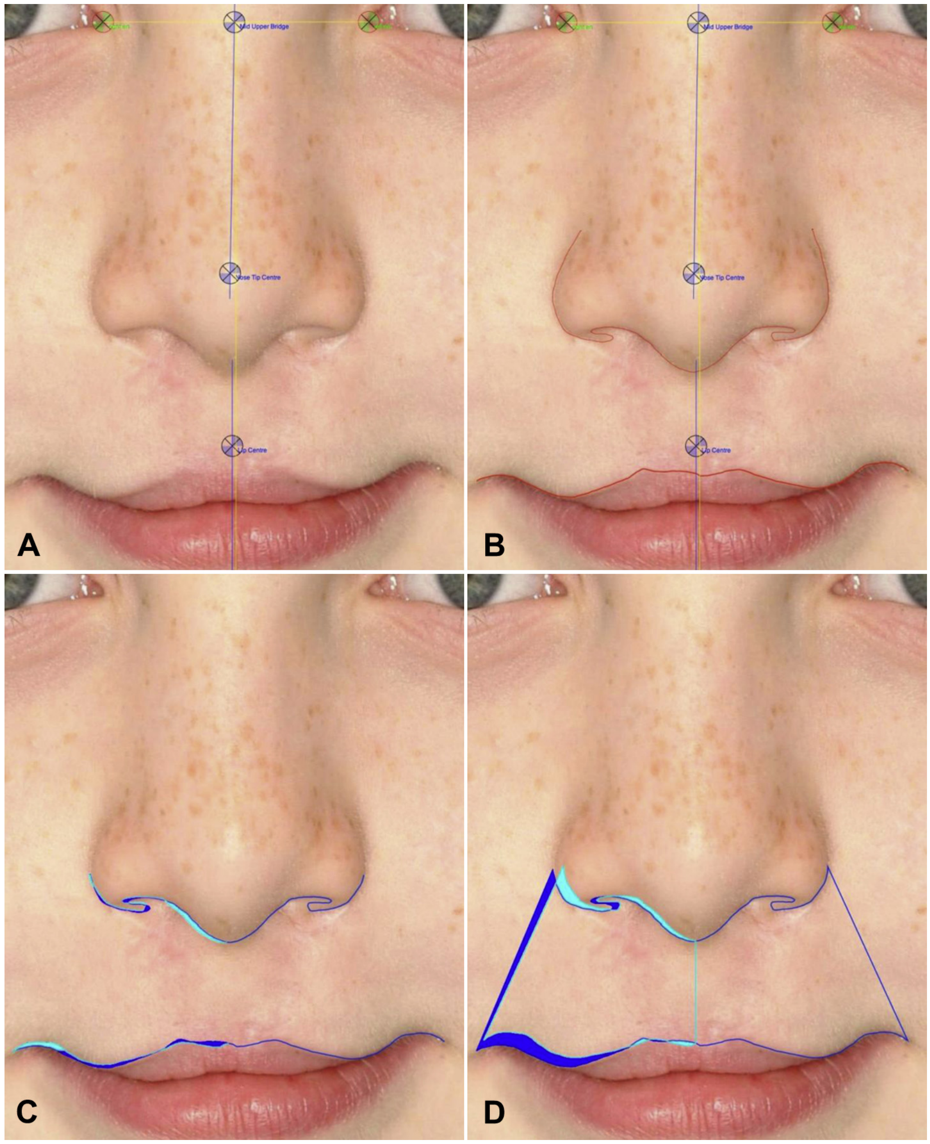

Fig. 1. A: Roundels are placed by the user on the canthi, allowing SymNose to draw a horizontal line between them with a vertical line bisecting the midline (computer-defined midline). Roundels are then placed on the upper nasal bridge, nose tip, and mid-upper lip. B The lower nasal border is traced starting from the superior aspect of the right alar groove, beneath the nostrils and columella to the opposite alar groove. A second line is traced along the upper vermillion border from the right labial commissure 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). D: The upper lip borders are enclosed laterally by lines created by SymNose from the labial commissures to the widest parts of the alar bases. This area is used to calculate the Upper Lip Difference.

guardians of all participants prior to obtaining images for use in this study.

3. Results

In total, 15 BCLP cases and 15 age-matched non-cleft controls were included in the study. Frontal facial photographs were traced using SymNose, and results compared in terms of values given for lip symmetry. Five parameters of lip symmetry were calculated by SymNose, which included Computer Axis Lip (CALip), User Axis Lip (UALip), Upper Lip Difference (UPLipDiff), Lip Obliquity (LipObliq) and Endocanthus to Cheilion Ratio (En-Ch). The BCLP group demonstrated statistically significant increases in both CALip and UALip ($p < 0.01$) (Table 1). UpLipDiff, LipObliq and En-Ch did not differ significantly between groups ($p > 0.01$), however values were normalised to show the data in graph form (Fig. 2).

3.1. Computer Axis Lip (CALip)

SymNose reflects the users' tracing of the left upper lip over that of the right side across a computer-defined midline. Areas of non-overlap are presented as a percentage mismatch, referred to as CALip. The larger the percentage mismatch, the higher the degree

Table 1
Comparison of lip symmetry between BCLP and control groups.

	CALip (%)	UALip (%)	UPLipDiff (%)	LipObliq (°)	En-Ch (%)
BCLP ($n = 15$), mean	23.8	25.8	8.4	1.2	1.4
Control ($n = 15$), mean	7.4	8.3	6.7	1.1	1.2
Difference between means	16.4*	17.5*	1.7	0.1	0.2
95% confidence interval	5.6–27.2	5.9–29.1	−2.4–5.8	−0.5–0.7	−0.4–0.8

*$p < 0.01$ by two-way Mann–Whitney U test.

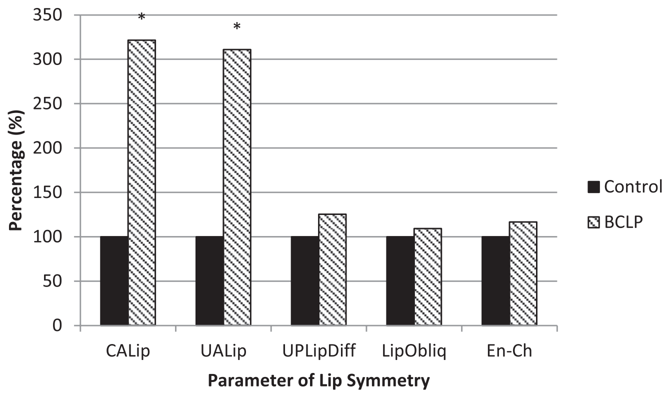

Fig. 2. Bar chart showing BCLP group as a percentage of normalised controls for five parameters of lip symmetry. $^{*}p < 0.01$ by two-way Mann–Whitney U test.

of asymmetry. It was found that the cleft group had a mean percentage mismatch of 23.8% compared with 7.4% in the control group (difference between the means = 16.4%; $p = 0.0028$).

3.2. User Axis Lip (UALip)

UALip is similar to CALip, however the midline is calculated according to where the middle upper lip roundel is placed by the user. The cleft group had a mean percentage mismatch of 25.8% versus 8.3% in the control group (difference between the means = 17.5%; $p = 0.0042$).

3.3. Upper Lip Difference (UpLipDiff)

UpLipDiff refers to the percentage mismatch between right and left upper lip areas. The upper lip area is defined as that between the upper lip border and the lower border of the nose. The lateral edges of the upper lip area are formed by lines created by SymNose between the labial commisures to the widest part of the alar bases (Fig. 1, D). The midline is calculated according to where the middle upper lip roundel is placed by the user, and right and left sides of the upper lip area are subsequently folded over this midline. The cleft group had a mean percentage mismatch of 8.4% versus 6.7% in the control group (difference between the means = 1.7%; $p = 0.6818$).

3.4. Lip obliquity (LipObliq)

LipObliq describes the angle at which the lips are deviated from the horizontal plane. SymNose presents this according to the direction of tilt (clockwise: positive, anticlockwise: negative). In this study, directionality was removed in order to quantify mean lip tilt as an absolute value. The mean value in the cleft group was 1.2° whereas the control group had a mean value of 1.1° (difference between the means = 0.1°, $p = 0.8026$).

3.5. Endocanthus to Cheilion Ratio (En-Ch)

En-Ch measures the distance between the medial canthus (endocanthus) and labial commissure (chelion). This distance is used as an indicator of how far the lips deviate from the vertical midline. Values for both left and right sides are measured and are presented as a ratio of (right − left/larger of right or left). This value is multiplied by 100 to give a percentage. Mean value in the cleft group was 1.4% compared with 1.2% in the control group (difference between the means = 0.2%; $p = 0.8026$).

3.5.1. Accuracy

Intra-observer reliability was assessed by calculating Spearman's rank correlation coefficient between first and second sets of tracings made by the same assessor for UALip (Fig. 3). The analysis showed a high level of intra-observer correlation for both assessors (Spearman $r = 0.94$ ($p < 0.00001$) and 0.92 ($p < 0.00001$) for Assessor 1 and Assessor 2 respectively).

Inter-observer reliability was assessed by calculating Spearman's rank correlation coefficient between both assessors for the measurement of UALip (Fig. 4). The analysis showed a high level of inter-observer correlation (Spearman $r = 0.94$, $p < 0.00001$).

4. Discussion

A number of methods have been used to assess facial appearance following cleft lip and palate surgery. A systematic review carried out by Al-Omari et al. (2005) found 40 publications relating to the measurement of cleft-related deformity, categorising them into direct clinical, 2-D and three-dimensional (3-D) assessment.

Direct clinical evaluation has been carried out using both qualitative and quantitative means. Farkas et al. (1993) used anthropometric measurements of the cleft lip and nasal complex taken from live subjects. These were compared with normal children of similar age. It was concluded that quantitative determination of facial asymmetry provides valuable information for surgical correction of cleft deformities, however direct clinical evaluation is

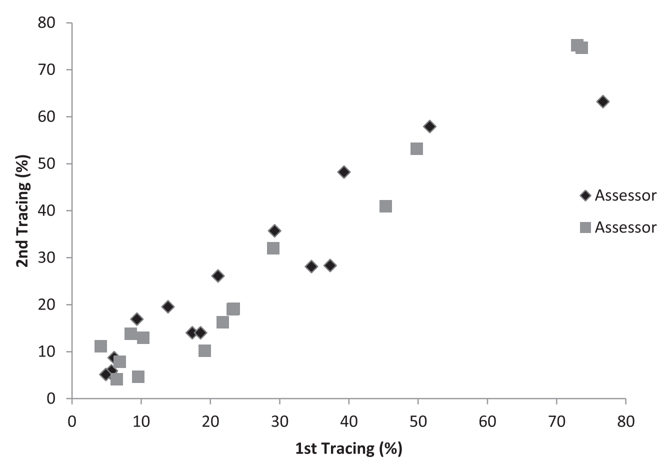

Fig. 3. Intra-observer reliability. Scatter plot showing the correlation between 15 repeated tracings performed by Assessor 1 ($r = 0.94$, $p < 0.00001$) and Assessor 2 ($r = 0.92$, $p < 0.00001$) for UALip.

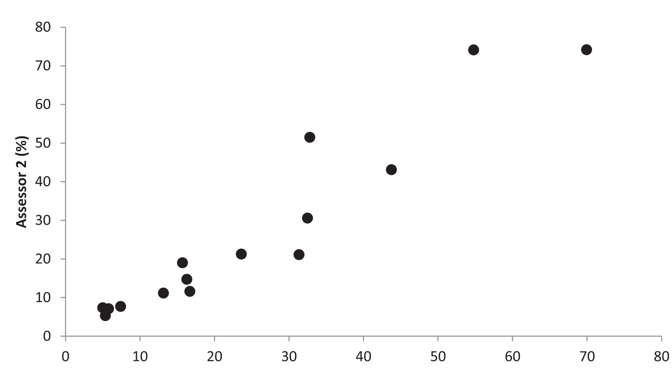

Fig. 4. Inter-observer reliability. Scatter plot showing the correlation between 15 tracings made by Assessor 1 and the corresponding tracings made by Assessor 2 ($r = 0.94$, $p < 0.00001$) for UALip.

a time-consuming process which must be carried out with the patient present.

Assessment using 2-D media addresses this problem by using photographs of patients that can be obtained in clinic in minimal amounts of time. Both qualitative and quantitative approaches have been used to assess facial appearance using photographs. The former approach does not require the assessor - or more frequently a panel of assessors - to make any measurements, however it is time-consuming and highly subjective. Pitak-Arnnop et al. (2011) noted that using the traditional gold standard of panel assessment, the composition of the panel may influence the outcome of assessment, with laypersons judging patients more empathetically and thus are likely to raise their ratings of facial appearance. A mixed panel of cleft specialists and laypersons may give a more unbiased assessment, whilst reflecting the impact cleft deformity has on everyday social interactions. This, however, may be difficult to achieve in clinical practice. Quantitative methods utilise a variety of photographic views including frontal and lateral facial views (Al-Omari et al., 2003; Eliason et al., 1991; Johnson and Sandy, 2003) as well as inferior or "worm's eye" views (Cussons et al., 1993). Increased levels of reliability and reproducibility have been reported by cropping the frontal image so that only the nasolabial area is exposed, thus reducing the influence of surrounding facial features on assessment of facial deformity (Asher-McDade et al., 1991). Computer-based assessment of lip symmetry following surgical repair is not a recent development in the field of cleft lip and palate surgery. Coghlan et al. (1987) first reported this method as an objective measure of nasal symmetry in a series of unilateral complete cleft patients. Digital images were traced and analysed using a microcomputer. The distance from regularly spaced points on the outline of one side to the nearest point of the reflection on the opposite side was used as a proxy for nasal symmetry. High levels of intra- and inter-observer reliability were demonstrated, and the results based on this method agreed well with subjective rankings given by a lay panel.

Eight studies were found which used 3-D media for the assessment of facial deformity relating to cleft lip. CT scanning has been used (Dado et al., 1997; Vannier et al., 1993) but its use is limited to the study of hard tissues. This supplies little information relating to the appearance of the facial surface, and so the technique is unsuitable for the assessment of cleft-related deformity. A computer-aided diagnostic system was developed by Yamada et al. (1999; 2002a; 2002b) in which facial symmetry was measured using an optical scanner and automatically extracted 3-D coordinates. The reliability of the technique was reported to be approximately 1 mm when the volunteer was measured in seven different postures. This technique has thus far only been applied to nasal anatomy. Although demonstrated to be useful, 3-D techniques require considerable operator skill as well as equipment that is expensive and of limited availability. This makes the routine use of 3-D assessment a relative impossibility in current clinical practice.

The SymNose computer program designed by Pigott and Pigott (2010) utilises 2-D images to evaluate symmetry of both nose and lip. Until recently, computer-aided methods of measuring facial symmetry had focused mainly on the nose and nostrils (Coghlan et al., 1987; Yamada et al., 1999) without reference to the lip. This gap in knowledge was partially addressed by McKearney et al. (2012) who evaluated the use of SymNose for the assessment of lip symmetry in a group of UCL patients 10 years post-primary surgical repair. A significantly higher amount of asymmetry was found in the cleft group for three out of the five parameters of lip symmetry measured. This supported the authors' assumption that unilateral defects would create a degree of asymmetry due to their location on one side of the midline.

The current study differs from that of McKearney et al. (2012) in the inclusion of BCLP patients. This group has been overlooked in all previous publications relating to the computer-based assessment of lip symmetry (Bilwatsch et al., 2006; Proff et al., 2006; Yamada et al., 2002a,b). However, due to the potential of bilateral defects to cause more severe deficits of form and function, as well as being more challenging to manage clinically, it is important that this group are audited to an equally high standard. This study revealed that 10 year-old children following primary repair of bilateral cleft lip had less symmetrical lips than age-matched non-cleft controls. This suggests that a significant degree of lip asymmetry remains in BCLP patients even after primary surgical management. This challenges previous opinion that those with bilateral defects would be relatively symmetrical. Nose symmetry was found to differ little between both groups, although a basal view as used by Pigott and Pigott (2010) may provide more information in this area. One of the main concerns regarding use of SymNose is the inherent element of subjectivity involved. The placement of roundels and subsequent tracing is dependent on the performance of the user as well as the quality of the image, which makes it, at best, a "semi-objective" process (Pigott and Pigott, 2010). To account for these factors, a precise protocol was followed to standardise the images, and tracings were carried out twice by each assessor on separate days. The accuracy of SymNose has been documented previously (McKearney et al., 2012; Pigott and Pigott, 2010) with the latter authors producing high levels of intra-observer ($r = 0.76$) and inter-observer ($r = 0.67$) reliability. Even higher values were found in this study, leading to the conclusion that one set of tracings would be adequate to achieve a high level of accuracy.

SymNose is well-suited for use in the clinical setting due to its simplicity and relatively low equipment cost. Tracings can be carried out quickly without the need for the patient to be present. Moreover, photographs are taken regularly as part of the standard audit protocol of many centres, and are thus readily available. These advantages make SymNose preferable to lengthy 3-D analyses which have thus far been confined to the research environment. Work has already been carried out comparing values measured by SymNose to the traditional gold standard of panel assessment (Gujral et al., 2011). Preliminary results show a strong agreement between the two methods ($r = 0.795$), thus further supporting the more widespread use of SymNose.

This work also highlighted some the limitations of SymNose, which include the potential to miss features such as scars within the lip which may be scored poorly by a panel. Features to account for this could be factored in to future versions of the program. Another limitation of SymNose is the difficulty faced in achieving standardisation of digital images. Whilst a precise photographic protocol was set up in this study to minimise any asymmetry relating to head position or facial expression, this may be harder to achieve in the clinical setting. Slight movement of the subject's head may lead to a degree of measurement error. A possible solution to this problem would be to use an external device to stabilise the subject's head during image capture. Facial expression is indeed harder to standardise, although the neutral expression has been adopted successfully in previous studies without reported measurement errors (Pigott and Pigott, 2010; McKearney et al., 2012).

This study has identified the need for a standardised, objective and reliable method of assessing post-operative outcomes following cleft lip and palate surgery. One key area this would facilitate would be in comparisons between novel techniques of cleft lip repair, with two different views of the face (frontal and basal) enabling the assessor to analyse both lip and nasal symmetry separately. The frontal view, as used in this study, is best suited to the analysis of vermillion border asymmetry. Vermillion notching or 'whistle deformity' is a common complication of cleft lip repair, occurring after primary cheiloplasty in patients with UCL. Lee et al. (2011) recently described a revision technique involving the sufficient lengthening of the medial oral lining, and taking the white roll with the greatest vermilion fullness as the standard point of the height of cupid's bow on the cleft side. Good aesthetic outcomes were expressed as those deemed satisfactory by the patient, and were achieved in all cases. A more objective follow-up, as could be achieved using SymNose, would facilitate a comparison between the results of this technique and those used by other surgeons, or indeed between results of the same technique practised at different centres. The basal view, as used by Pigott and Pigott (2010), is best suited to the analysis of nose and nostril symmetry. Nasal deformities commonly associated with BCLP include a shortened columella, depressed nasal tip, eversion of the alar bases, and bilateral dislocation of the alar cartilage.

A number of novel techniques for correcting the nasal deformities associated with BCLP have recently been described (Nakamura et al., 2011). The assessment of aesthetic outcomes in this study was achieved using 3-D laser scanning to measure the facial forms of the patients followed by image reconstruction using specialised software. Outcome assessment could be improved further using SymNose to quantify symmetry of the nostrils and nasal tip. Furthermore, adverse outcomes including scarring or deformity of the upper lip or columella base could be assessed using digital images taken in the frontal view and analysed using SymNose.

5. Conclusion

The results of this study support the use of SymNose as an objective method for assessing lip symmetry following primary repair of complete BCLP. Contrary to previous assumption (McKearney et al., 2012), a significant degree of lip asymmetry was found in patients with bilateral defects. Interestingly, the same degree of asymmetry was not found in the nose, a result which may contradict panel assessment. Further research could focus on the basal view of the face in order to explore nose and nostril symmetry as carried out previously in a group of UCL patients (Pigott and Pigott, 2010). Overall, the authors of this study found SymNose simple to use, time-efficient and highly reliable. The widespread introduction of SymNose into clinical practice could obviate the requirement for more lengthy or costly methods of facial assessment such as 3-D analysis.

Conflict of interest statement

None.

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