

# Nostril Morphometry Evaluation before and after Cleft Lip Surgical Correction: Clinical Evidence

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Abstract Introduction The purpose to this work is to review systematically the morphological changes of the nostrils of patients undergoing surgery for correction of cleft lip and identify in the literature the issues involved in the evaluation of surgical results in this population. Review of Literature A review was conducted, searching for clinical evidence from MEDLINE. The search occurred in January 2012. Selection criteria included original articles and research articles on individual subjects with cleft lip or cleft palate with unilateral nostril anthropometric measurements before and after surgical correction of cleft lip and measurements of soft tissues. There were 1,343 articles from the search descriptors and free terms. Of these, five articles were selected. **Discussion** Most studies in this review evaluated children in Eastern countries, using **Keywords** different measurement techniques but with the aid of computers, and showed ► cleft lip improved nostril asymmetry postoperatively compared with preoperatively. anthropometry **Conclusion** There is a reduction of the total nasal width postoperatively compared with preoperative measurements in patients with cleft lip. nose

# Introduction

Nasal deformities associated with unilateral cleft lip are characterized by asymmetry, which gradually progresses with severity of the cleft, and nostril structures with morphological changes.<sup>1–4</sup> The main supporting structure of the nasal wing, the nasal lower lateral cartilage, in patients with cleft lip is concave with a depressed nasal tip, and separates from the opposite side, not the cleft, resulting in depression and collapsed nasal tip asymmetry,<sup>5</sup> which is very common, even after surgical treatment of cleft lip.<sup>6</sup>

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DOI http://dx.doi.org/ 10.1055/s-0033-1352506. ISSN 1809-9777. The creation of a symmetrical nose is a big challenge, and it is also difficult to evaluate an outcome after surgery. The appearance of the deformity is the result of primary relationship of different factors. Subjective evaluation by expert surgeons in cleft lip surgery is the standard; however, a simple objective measure would be important if it could faithfully reflect this pattern.<sup>3</sup> Use of anthropometric techniques for preoperative quantitative evaluations of morphological changes in soft and stiff tissues of the cleft face is essential to objectively determine the facial anatomy and surgical treatment result.<sup>7</sup>

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Understanding how the nostril is modified after the surgical treatment of cleft lip may contribute to the clinical practice of health professionals who work on the nose. This knowledge may influence the surgical program focused on the specific need of each patient and the rehabilitation process of lip mobility and facial expression, which are closely linked to the muscles of the superior lip and nose base.

Considering that few studies have investigated this theme, the present study aims to review the literature on the condition of the nostril morphometry in patients with cleft lip before and after cleft surgery, as well as identify the issues involved in assessing these changes in this population.

### **Review of Literature**

A literature review was performed from the databases of the Medical Literature Analysis and Retrieval System Online (MEDLINE), and the data search occurred in January 2012. A specific strategy was developed for crossing the descriptors (MeSH)—keywords for retrieving subjects from literature— and scientific terms not found in MeSH terms but of relevance to the research.

In MEDLINE, using the PubMed search engine we performed a search strategy using the syntax: "Cleft lip" (MeSH) AND "Anthropometry" (MeSH); "Cleft lip" (MeSH) AND nostril; "Cleft lip" (MeSH) AND "Morphometry"; "Anthropometry" (MeSH) AND "nostril"; "nostril" AND "Morphometry."

Inclusion criteria were original articles (excluding editorials and case reports) reporting on individuals with unilateral cleft lip or cleft lip and palate who underwent anthropometric measurements of the nose or nostril before and after cleft lip surgical correction. Exclusion criteria were measurements performed on bony structures or other facial structures that did not include the nostrils or the nose; studies that compared surgical techniques; articles written in Oriental languages (Mandarin, Japanese); and articles not found by switching bibliographic system (COMUT).

Article selection was conducted in three stages. In the first step, we performed a reading of the titles of the studies. We excluded those that clearly did not fit any of the criteria for inclusion in this study. Then, we read remaining abstracts and excluded those that clearly did not fit any of the inclusion criteria previously established. In the third stage, all the studies that were not excluded in these first two steps were read in entirety for the selection of articles to be included in this review (**-Fig. 1**).

In MEDLINE via PubMed, crossing the free terms "nostril" and "Morphometry" found three articles, of which all were excluded by the title. Crossing the keyword "Cleft lip" and the free term "Morphometry" found seven articles that were excluded by the title. Crossing the keyword "Anthropometry" and the free term "nostril" found 71 articles, of which 61 were excluded by the title, 8 were excluded after reading the abstract, and 2 were selected for full reading. Crossing the keyword "Cleft lip" and the term free "nostril" found 177 articles, of which 131 were excluded by the title, 41 were excluded after reading the abstract, and 5 articles were selected for full reading. Crossing the keywords "Cleft lip" and "Anthropometry" found 1,085 articles, of which 923 were excluded by the title and 147 were excluded after reading the abstract, leaving 15 articles selected for full reading. Of the 22 articles selected for full reading, we excluded 5 papers that were repeated, 1 written in Mandarin, and another not found by switching bibliographic system (COMUT).

Considering inclusion and exclusion criteria, of the 15 fully read articles, only 5 articles were chosen to include in this systematic review. Ten articles were excluded for different reasons: 3 because they did not perform morphometric comparison between pre- and postoperative nostril, 2 because they were not performed in patients with clefts, 2 because they were performed without analyzing the nose or nostrils, 1 because a morphometric study was not performed, 1 because it used cephalometric radiographs for analysis, 1 because it conducted analysis and validation of measurement method without evaluating and interpreting results of the evaluations.

There were no sufficient data to conduct a meta-analysis because the articles' heterogeneity did not allow grouping for statistical analysis (**-Table 1**). Thus, the results of this study are in the form of systematic review without meta-analysis. According to Cochrane in situations where meta-analysis is not practical, the researcher should feel encouraged by the line of research on building a field for randomized clinical trials.

For a better presentation of the results, the following variables of the selected articles were considered: author and year, country, number of patients, measurement method, patient age, follow-up, associated procedures, cleft type, and measurements and differences after surgery (**-Table 2**).

#### Discussion

The studies are from the end of the 1990s (one article) and the 2000s (four articles). Although study of cleft lip deformities began at the late 19th century,<sup>8</sup> it is clear that the association with anthropometric studies of the face only occurred beginning in the 1960s; however, measurements were performed with very simple criteria.<sup>9,10</sup> We believe that the absence of studies that compared pre- and postoperative anthropometric measures in that period arises from difficulty in standardizing measures in the early days of anthropometry, as well as the difficulty of techniques using direct measurements, which are difficult to obtain in children.<sup>11</sup> Studies using standardization emerged in the 1980s,<sup>6,12,13</sup> noting the concern of choosing anthropometric points.<sup>14</sup> The manner in which anthropometric data are collected should be considered when comparing the values obtained and the parameters reported in the literature, due to the small variation between the values acquired directly versus those acquired indirectly.<sup>15</sup>

Recent research shows concern about the use of modern techniques for measurement. A example of this are two articles found in this study reporting the use of photogrammetry<sup>5,16</sup> and three studies using three-dimensional measurements.<sup>17–19</sup>

Digital photogrammetry is a noninvasive, inexpensive, and common method to investigate pre- and postoperative

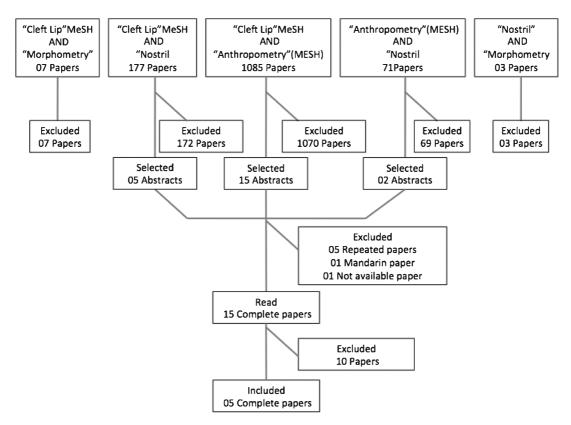


Fig. 1 Revew of Literature Stages.

changes and provides a permanent record of patients. Additionally, data can be stored and managed in a digital format that takes measurements using software.<sup>20,21</sup> Advancement in technology has allowed three-dimensional images,<sup>22</sup> such as computed tomography, that require expensive equipment and has limited ability to determine the characteristics of soft tissues.<sup>23</sup> Moreover, ethical considerations limit the use of radiation for studies, especially in children.

For these reasons, techniques for three-dimensional surface imaging such as laser scanning and stereophotogrammetry have been developed to capture the soft tissue and facial structures. Studies based on three-dimensional images appear to be the best alternative for assessing children with clefts both pre- and postoperatively; these methods provide more information than two-dimensional methods. According to Harris and Smith, most deductions that occur in studies are derived from statistical analysis and, in addition to concerns about accounting adequately for known sources of variation within the research project, a major source of variability is measurement error.<sup>24</sup> With the increasing access to data collection methods, computers have improved the ease of incorporating repeated measures on statistical models, with increased chance of finding biologically true differences when they exist.

All articles selected for this review are from research conducted in Eastern origin populations. One study was in a Japanese population,<sup>17</sup> three in a Taiwanese population,<sup>5,16,18</sup> and one study occurred in Cambodia.<sup>19</sup> According to Dixon et al,<sup>25</sup> cleft lip and palate affects  $\sim$ 1/700 live births, and in general, Asian and Amerindian populations have the

	Yamada et al. <sup>19</sup>	Liou et al <sup>5</sup>	Pai et al <sup>18</sup>	Seidenstricker-Kink et al <sup>20</sup>	Schwenzer-Zimmerer et al <sup>21</sup>
1. Inclusion criteria specified	Yes	Yes	Yes	Yes	Yes
2. Control group	Yes	No	No	Yes	Yes
3. Random allocation	No	No	No	No	No
4. Blind allocation	No	No	No	No	No
5. Blind subjects	No	No	No	No	No
6. Blind therapists	No	No	No	No	No
7. Statistical analysis	Yes	Yes	Yes	Yes	Yes
8. Statistical comparison between groups	Yes	No	No	Yes	Yes

Table 1 Methodological classification of selected papers

Authors and year	Country	Number of patients	Measurement method	Patients age	Follow-up	Associated procedures	Cleft type	Differences after surgery	Measurements
Yamada et al. <sup>19</sup>	Japan	1	3-D reconstruction with computerized analysis of models	4 mo	2 то	Not used	Unilateral complete	Nasal width reduced with improved sym- metry; reduction of columellar angle	Nasal width, height and length; axial, frontal and side angles; no nostril measurements
Liou et al <sup>5</sup>	Taiwan	25	Photogrammetry	Newborns (operated at 3 mo of age)	Up to 3 y (annual revaluations)	Preoperative nasoalveolar molding	Unilateral complete	Significant improve- ment of nostril sym- metry after surgery; vertical nostril grow- ing and nasal domus significantly lower at cleft side	Nostril width and height: nasal domus and columellar height
Pai et al <sup>18</sup>	Taiwan	57	Photogrammetry	3 mo	Up to 1 y	Preoperative nasoalveolar molding	Unilateral complete (n = 34); unilateral incomplete (n = 23)	Improved symmetry after surgery: differ- ences in nostril width (10%), height (20%), and columellar angle (4.7%) between pre- and postoperative	Nostril width, height and colu- mellar angle
Seidenstricker-Kink et al <sup>20</sup>	Taiwan	26 (7 excluded because loosing data); overall 19	3-D computerized tomography	3 mo (025 y + 0.06)	10 mo	Not used	Unilateral complete	Preoperative nasal and cleft nostril wider than postoperative; improved nostril symmetry after surgery	Columellar length and nostril width
Schwenzer- Zimmerer et al <sup>21</sup>	Cambodia	11	3-D surface laser scanner	From 12 to 41 y (13.8 average)	6 mo after surgery	Preoperative nasoalveolar molding (9 patients); lip taping (all)	Unilateral complete	Improvement of nos- tril symmetry after surgery (43%)	Nostril width, cleft width, and colu- mellar length

highest prevalence, often as high as 1/500. European populations have intermediate prevalence rates of  $\sim$ 1/1,000, and Africans have the lowest prevalence rates of  $\sim$ 1/2,500.

These data justify the predominance of studies in Eastern countries, but we emphasize that population-based studies to estimate the true incidence of cleft lip and palate are scarce. Sporadic reports suggest that there has been some decrease in the incidence of cleft deformities; the reasons for this decline are multifactorial.<sup>26</sup>

These studies have small numbers of patients, from 1 to 57.<sup>16,17</sup> According to Harris and Smith, researchers commonly infer characteristics about populations from relatively small samples of study that can lead to errors in the evaluation of the results.<sup>24</sup> The amount of variability in the numbers of individuals evaluated reflects the lack of homogeneity in the samples. Other factors that influence the definition of clinical evidence is heterogeneity in the age of attainment of the first surgery and clinical procedures associated with the surgical procedure, such as the use of presurgical orthopedics or labial adhesion.<sup>27</sup>

There was variation in periods of postoperative follow-up. Some studies cited shorter periods of around 2 months, and others cited up to 6 months, and others even longer periods, up to 3 years.<sup>5,16–19</sup> The medical literature suggests that monitoring children with cleft lip and palate should continue during their growth, including adolescence,<sup>27</sup> both for evaluation of results and to monitor facial growth. Shorter termlimited follow-up or search for other surgical procedures to be performed on children can sometimes be justified, such as palatoplasty at 9 months of life. We believe following patients longer term allows better characterization morphological evolution of the nostril, which usually has increased asymmetry over the months. Thus, control of the growth is accepted as a useful tool in evaluating the natural state of health of an individual.<sup>28</sup>

Numerous methods have been described for repair of cleft lip deformity. Repair of unilateral deformity is usually approached by rotation and advancement technique as described by Millard<sup>29</sup>, under the concept of advancement of a flap on the lateral side of the upper lip combined with the rotation of the medial segment. This technique preserves both Cupid's bow and the philtrum.<sup>30</sup>

Procedures associated with surgery, such as nasoalveolar molding or lip adhesion, were performed in three studies.<sup>5,16,19</sup> Some authors aligned the alveolar segments to create the foundation for the primary lip surgery to obtain good results and, to achieve this, early assessment and initiation of preoperative orthopedics should happen in the first days of life.<sup>27</sup> Other authors, who disagree with the preoperative procedures, claimed that the secondary deformities on the nose are caused by many factors, but the greatest determinant of nasal appearance after treatment is the primary deformity,<sup>3</sup> and cited that associated procedures generate higher costs and are very dependent on the cooperation patient and family for a good result.<sup>2</sup> Among the results, we noticed that patients who used a nostril mold had improvement of asymmetry even before surgery.

He et al performed a retrospective study to correlate the width of the cleft with the severity of unilateral nasal

deformity in patients with cleft lip and palate before repair primary lip and found that the width of the nose, nasal length, and width of the lip were larger in patients with complete clefts.<sup>31</sup> The preoperative facial asymmetry in patients with unilateral complete cleft is obvious and is widely established, and deficit of transverse tissue is generally more severe in complete clefts.<sup>6</sup> At preoperative evaluation of unilateral complete cleft, the width of the cleft is a reliable guide to the severity of the other parameters.<sup>4</sup> These data show that unilateral complete cleft represents a greater deformity compared with an incomplete fissure and indicate the need for uniformity of the population when comparing studies; only four articles described patients with unilateral complete cleft, and in only one work were these patients the majority of the group.<sup>5,17–19</sup>

Some authors only measured width, height, and length of the nose without including measurements of the nostrils.<sup>17</sup> The majority of authors surveyed agreed that these measures should be included, but only allowed the observation, already established, that the cleft nose was wider preoperatively than postoperatively.<sup>3,6,32</sup> Experts are certainly capable of subjectively grading patients according to degree of nasal deformity, and two anthropometric measures that are easily obtained objectively-nostril width and columellar angle-can correlate with the expert's ranking.<sup>3</sup> Four studies performed more elaborate measurements,<sup>5,16,18,19</sup> based on anthropometric points described in the literature, allowing them to apply a more detailed analysis of the entire nasal morphometry to compare the possibility of a greater number of variables and use different ratios in assessment of results.<sup>6</sup> Visual judgment is influenced by the most impressive disproportions and cannot determine the factors causing the disproportions; the consensus among researchers is that the quality of facial morphology results can be estimated only by the proportions shown by quantitative data.<sup>7</sup> A standard morphometric assessment outside the nose could be a reliable parameter for comparing rhinoplasty results.<sup>33</sup> We believe that measurements should contain the maximum information possible, therefore covering all nostril points, and anthropometric measurements should include the angle of the columella, allowing a richer analysis when comparing the periods evaluated.

Among the residual deformities after surgery for complete unilateral cleft, asymmetry of the width of the floor of the nostrils was the most common finding, followed by columellar length asymmetry, low nasal bridge, broad nose, flat nasal tip, and low and shorter columella on the cleft side.<sup>5,6</sup> We believe the application of this knowledge will allow an individualized approach to the patient—for example, if after measurements nostril asymmetry is observed, with a greater breadth of cleft nostril, treatment is aimed at correcting that deformity during surgical procedure (cleft lip surgery).

## Conclusion

Although all studies have established and described inclusion criteria, none of them observed all the criteria for randomization, which encourages future researchers to try to complete new work with greater methodological rigor in the future.

The articles concluded that there is an important improvement in nostril asymmetry when comparing preoperative and postoperative measurements. The main changes that occurred after surgery were reduction of columellar angle, reduction of the width of the cleft nostril, and increase in the height of the cleft nostril.

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