Whistle deformity evaluation after Cronin labioplasty in unilateral cleft lip and palate cases from an anthropometry aspect

V M S Putra¹, I Tofani² and M S Hak³

¹Oral and Maxillofacial Surgery Residency Program, Faculty of Dentistry, Universitas Indonesia, Jakarta 10430, Indonesia
²Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Universitas Indonesia, Jakarta 10430, Indonesia
³Department of Oral and Maxillofacial Surgery, Harapan Kita Children and Maternal Hospital, Jakarta 10430, Indonesia

*E-mail: iwan.tofani@yahoo.com

Abstract: Whistle deformity of the lip usually occurs after labioplasty. We compared the height of Cupid’s bow on the normal side, midline, and cleft side after Cronin labioplasty in patients with unilateral cleft lip and palate (UCLP) in lip function and resting positions to determine the presence of whistle deformity. We evaluated whistle deformity according to an anthropometry scale from a profile photograph, including the height of Cupid’s bow on the normal side, midline, and cleft side in 24 patients with UCLP after Cronin labioplasty, while the lips in function and resting positions. Based on statistics, the results showed significant differences ($P < 0.05$) in the height of Cupid’s bow at midline in normal children and those with whistle deformity while the lips were at rest. On the other hand, significant differences also were found in the height of Cupid’s bow on the normal and cleft sides; on the normal side, cleft side, and midline in patients with whistle deformity in lip function and resting positions; on the right and left sides in normal children in lip function and resting positions; and in normal children and those with whistle deformity in the lip function position ($P > 0.05$). There was no significant difference between the height of Cupid’s bow on the normal and cleft sides; on the normal side, cleft side, and midline in patients with whistle deformity in lip function and resting positions; on the right and left sides in normal children in lip function and resting positions; and in normal children and those with whistle deformity in the lip function position. On the other hand, there was a significant difference between the height at midline in normal children and between normal children and those with whistle deformity while the lips were at rest.

1. Introduction

More than four million children around the world are born with a congenital defect every year. Cleft lip with and without cleft palate is the most frequent congenital craniofacial deformity with an estimation of more than 250,000 children born every year around the world. This deformity distorts...
the upper lip, nose, and palate. Approximately 65% of head and neck deformities are cleft lip and palate [1–4].

According to an epidemiology study on the distribution of clefts of the oral cavity, cleft lip and palate occurred in 46% of cases, cleft palate in 33%, and cleft lip in 21%. Among 60,038 babies born at Harapan Kita Children and Maternal Hospital between January 1995 and December 2013, 85 (0.14%, or 1:714) had cleft lip with or without cleft palate. Unilateral cleft lip occurred more frequently than bilateral cleft lip, more frequently on the left than the right sides, and more frequently in boys than girls. The prevalence of cleft palate was 1:2000 and occurred more often in girls [1,2].

The etiology of cleft lip with or without cleft palate was assumed to be a combination of genetic and environmental factors. Studies have shown that every parent had a 0.14% (1:700) chance of having children with cleft lip with or without cleft palate [1].

Optimal management of cleft lip and palate requires holistic treatment from multidisciplinary fields, such as oral and maxillofacial surgery, pediatrics, anesthesiology, otolaryngology (ENT), psychology, audiology, speech training, and orthodontics [1,4]. The goal of surgery is to obtain optimum aesthetics and function with as little scar tissue as possible. The objective of labioplasty is to create a balanced and symmetrical result with minimal scar tissue for the lip to function normally. According to Wilhelmsen and Musgrave in 1966, the conditions for performing labioplasty were the “rules of ten,” including weight more than 10 pounds or at least 4.5 kg, hemoglobin level more than 1 or equal to 10 g/dL, and leucocyte count less than 10,000 [1,5,6,7,8].

According to Millard (1965), labioplasty could be performed at any age, from newborn to adult; however, if the objective was the final result of surgery, it was better to perform the initial labioplasty after the baby was approximately three months old, so that the nose and lip components had a chance to grow, and the baby weighed approximately 10 to 12 pounds. Later on, since 1967, the rules for labioplasty (rules of ten) were determined to be age more than 10 weeks, weight more than 10 pounds or approximately 4.5 kg, hemoglobin level more than or equal to 10 g/dL, and leucocyte count less than 10,000 [7].

Patients with cleft lip tend to have various deformities according to the severity of the cleft lip itself, previous surgery techniques, the healing process, and growth pattern after primary labioplasty. Whistle deformity occurs most often after primary labioplasty [9–11].

Evaluation after labioplasty is performed according to functional and aesthetic aspects. Functional aspects are evaluated by speaking and vocalization. Otherwise, aesthetic aspects are evaluated according to the presence of a whistle deformity.

To date, to our knowledge, no anthropometry study has been done on whistle deformity after labioplasty via the Cronin technique. Therefore, this study was performed at the Harapan Kita Children and Maternal Hospital.

Whistle deformity after labioplasty was evaluated using an anthropometry technique, which is a method to obtain the most reliable body shape comparison using specific landmarks based on the most prominent anatomy. Using anthropometry, some landmarks can be determined as a reference for measurement, from which the size of the whistle deformity can be determined [1,7,12].

2. Methods

This observational analytic study was performed at Harapan Kita Children and Maternal Hospital from approximately June to July 2016 using a cross-section design on indirect anthropometry measurements. Subjects aged four to six years were diagnosed with unilateral cleft lip and palate (UCLP) and nonsyndromic cleft lip and palate. All subjects underwent labioplasty according to Harapan Kita Hospital protocols. Exclusion criteria included syndromic unilateral complete cleft lip and palate, noncooperative patients, and those who refused to follow the study. The control group included children aged four to six years with normal lips from Cut Nyak Dhien Islamic Kindergarten in Cimone who were healthy without congenital disease. Exclusion criteria from the control group included unhealthy children or those with a congenital disease, who were uncooperative, and who refused to follow the research.
Calculation of samples using G*power 3.0 with \( \alpha : 0.05, \beta : 0.80 \) resulted in a minimum of 24 samples per group. This study was accepted by the Dental Research Ethics Committee Faculty of Dentistry Universitas Indonesia and Harapan Kita Children and Maternal Hospital. Cronin labioplasty was the independent variable, and whistle deformity was the dependent variable. The controlled variables were the same operator (oral surgeon) for all labioplasty surgeries and the time of surgery.

Tools and materials used in this study were stationery, a Canon camera, Adobe Photoshop CS3, data processing sheets and data analysis, standard clinical photography tools, a computer, and statistical software (SPSS 22.0).

Data on patients who underwent labioplasty were obtained from patient ledger books at the Cleft Lip and Palate Unit of Harapan Kita Children and Maternal Hospital from 2010 to 2015. Based on the medical record, patients who matched the inclusion criteria were studied. Patients were recalled, and informed consent was obtained. Facial soft tissue (anthropometry) measurement was performed indirectly (photograph).

To obtain indirect anthropometry measurements, patients were seated and assumed a relaxed facial expression. Hair did not cover the lip parts. The patient assumed a straight forward view, chin up, and perpendicular to the floor. A photograph was taken using a camera at a 1 m distance from the patient. The measurement was obtained by two people, repeated two times, and the average values were noted in a table.

To measure the whistle deformity, we measured the vertical height of the tip of Cupid’s bow on the noncleft side, on the upper lip midline, and on the cleft side. Reliability was tested using an unpaired \( t \)-test (\( P < 0.05 \)).

This study used primary data. All results were put into the table and data were processed using SPSS software SPSS 22.0. The data were analyzed to determine whether there were similarities in vertical heights of the tip of Cupid’s bow on the noncleft side, the upper lip midline, and the cleft side after labioplasty.

### 3. Results

Various patients four to six years old underwent evaluation for whistle deformity after labioplasty using measurements on photographs. The landmarks used in this study were based on anthropometry introduced by Farkas and reported by others.

Parents and guardians were given explanation about the photographic procedure, and photographs were taken after they had provided informed written consent to participate. Demographic data of the 24 subjects studied are shown in Table 1.

| Diagnosis | Right UCLP | Left UCLP | Total |
|-----------|------------|-----------|-------|
| Sex       |            |           |       |
| Male      | 6          | 10        | 16    |
| Female    | 1          | 7         | 8     |
| Total     | 7          | 17        | 24    |

There were 16 male (66.67%) and 8 female (33.33%) patients. The UCLP was on the right side in 7 subjects (29.16%; 6 [37.5%] male; 1 [12.5%] female) and left side in 17 (70.83%; 10 [62.5%] male; 7 [87.5%] female). Three patients had whistle deformity.

The Shapiro–Wilk test for normality showed a \( P \) value of >0.05 for vertical height at the tip of Cupid’s bow on the noncleft and cleft sides, indicating a normal data distribution. A reliability test was done using an unpaired \( t \)-test with \( P < 0.05 \) indicating significance.
The average vertical height of the tip of Cupid’s bow was 7.17 ± 1.50 mm on the noncleft (normal) side and 7.16 ± 1.59 mm on the cleft side (Fig. 1). The average comparison coefficient was 0.99 ($P = 0.99$).

![Height of Cupid’s Bow](image1)

**Figure 1.** The average vertical height of the tip of Cupid’s bow on the noncleft (normal) and cleft sides

Based on SPSS analysis using an unpaired t-test, there was no statistical difference in vertical height of the tip of Cupid’s bow on the noncleft (normal) and cleft sides. This result matched the hypothesis stating that there is no difference in vertical height of the tip of Cupid’s bow on the noncleft side in patients with a complete cleft lip after labioplasty since all comparison coefficients were approximately 0.95 to 1.05.

In the patients with a whistle deformity, the average vertical height of the tip of Cupid’s bow was 8.27 ± 0.12 and 7.05 ± 1.78 mm with the lip in the resting and functional positions, respectively, on the cleft side (average comparison coefficient 0.85; $P = 0.38$); 8.35 ± 0.26 and 6.38 ± 1.41 mm, respectively, on the noncleft side (average comparison coefficient 0.76; $P = 0.17$); and 6.19 ± 0.17 and 5.80 ± 1.57 mm, respectively, in the midline (average comparison coefficient, 0.93; $P = 0.73$; Fig. 2).

![Patient with Whistle Deformity](image2)
Figure 2. The average vertical height on the tip of Cupid’s bow on the noncleft side (normal), cleft side, and midline during resting and functional lip positions in patients with whistle deformity

Based on SPSS analysis using an unpaired $t$-test, there were no statistical differences among the vertical heights of the tip of Cupid’s bow on the noncleft side (normal), cleft side, and midline during resting and functional lip positions in patients with whistle deformity. This result did not match the hypothesis stating that there are no differences among vertical heights of the tip of Cupid’s bow on the noncleft side (normal), cleft side, and midline during resting and functional lip position in patients with a whistle deformity after labioplasty, as the average comparison coefficients were out of the range of 0.95 to 1.05.

In normal lips, the average vertical height of the tip of Cupid bow was $5.90 \pm 1.45$ and $6.31 \pm 1.37$ mm during resting and functional lip positions, respectively, on the left side (average comparison coefficient, 1.06; $P = 0.45$); $6.05 \pm 1.59$ and $6.26 \pm 1.22$ mm, respectively, on the right side (average comparison coefficient, 1.03; $P = 0.70$); and $4.73 \pm 1.65$ and $5.73 \pm 0.98$ mm, respectively, in the midline (average comparison coefficient, 1.21; $P = 0.03$; Fig. 3).

Figure 3. The average vertical height of the tip of Cupid’s bow on the left side, right side, and midline during resting and functional lip positions in normal patients

Based on SPSS analysis with the unpaired $t$-test, there were no statistical differences in vertical height of the tip of Cupid’s bow on the right and left sides during resting and functional lip positions in patients with a normal lip. Otherwise, there was a statistical difference in midline height during resting and functional lip positions. This result did not match the hypothesis stating that there was no difference in vertical height of the tip of Cupid’s bow on the left side and midline during resting and functional lip positions with a normal lip, as the average comparison coefficient was outside of the range of 0.95 to 1.05. However, the results did match this hypothesis regarding the right side, as the entire average comparison coefficient was approximately 0.95 to 1.05.

The average vertical height of the tip of Cupid’s bow in patients with normal lips and those with a whistle deformity was $5.90 \pm 1.45$ and $8.27 \pm 0.12$ mm, respectively, during a resting lip position (average comparison coefficient, 1.40; $P = 0.01$) and $6.31 \pm 1.37$ and $7.05 \pm 1.78$ mm, respectively, during a functional lip position (average comparison coefficient, 1.12; $P = 0.45$; Fig. 4).
Figure 4. The average vertical height of the tip of Cupid’s bow in patients with a normal lip and whistle deformity during resting and functional lip positions

Based on SPSS analysis with an unpaired t-test, there was a statistically significant difference in vertical height of the tip of Cupid’s bow in patients with normal lips and those with whistle deformity during a resting lip position. However, no difference was found during functional lip positions. This result did not match the hypothesis stating that there was no vertical difference in the tip of Cupid’s bow between patients with normal lips and those with a whistle deformity during resting and functional lip positions, as the entire average comparison coefficient was outside the range of 0.95 to 1.05.

4. Discussion
We evaluated the whistle deformity in 24 cases of unilateral complete cleft lip after labioplasty at the Cleft Lip and Palate Unit Harapan Kita Children and Maternal Hospital, Jakarta, using anthropometric data from photographic measurements. Performance of labioplasty was determined by the “rules of ten” since 1967 [7].

The main objective of labioplasty was to create a balanced and symmetrical lip and nose with minimal scar tissue and a nicely functioning lip with a decreased effect of surgery on growth and development of the maxilla [5]. We used the Cronin labioplasty technique, which is expected to have a good result in cleft lip cases.

After primary labioplasty, cleft lip patients tend to have various deformities depending on the severity of the cleft lip, surgical technique used, healing process, and growth pattern. After labioplasty, the deformity that occurs most often in the abnormal vermilion is called the whistle deformity [9,13]. Whistle deformity is a concave defect in the middle of the upper lip vermilion, as the vermilion seems to be lifted upward due to complex causes. The basic cause of whistle deformity is intrinsic deficiency of soft tissue on the prolabium and absence of musculorbicularis oris continuity. The defect takes the shape of the lip when someone is whistling; however, it occurs during the resting position and becomes worse during lip function [9,11,14,15].

In cleft lip patients, the unmet orbicularis oris muscle or area unconnected with the opposite muscle fibers is well apparent, so the fibers will not pass through the maxilla midline, but rather position themselves parallel to the edge of the gap and the fiber leads to the base of the nose. The unity of the structure results in the side of the gap reaching into torn muscle that does not work. To achieve a good
labioplasty result and minimize the whistle deformity, the Cronin labioplasty requires management of the orbicularis oris muscle [7,16]. During the incision, the orbicularis oris muscle is being freed from mucosa and skin on the medial and lateral sides and then divided into three parts. On the pars peripheralis of the upper orbicularis oris, suturing is done with overlapping, and the pars peripheralis is cut diagonally according to the middle defect and suture interdigitations. On the pars marginalis of the orbicularis oris, suturing is done edge to edge. With this management, good muscle unity is expected [16].

In the case of whistle deformity, some surgical techniques can be performed to overcome this defect:

1. **Z-Plasty Technique** [17]: In cases of light deficiency of the vermilion, the upper lip itself can be used for reconstruction, just like in the Z-Pasty technique.

2. **Cross Muscle Flap** [8]: Before infiltration of local anesthetics, a muscle flap is designed on the lateral part of the lip with the whistle deformity, depending on severity and tissue deficiency volume. Then, 1% lidocaine and 1:100,000 epinephrine are infiltrated, and an incision is performed along the root line. On the lateral side of the vermilion, sharp dissection is performed between the orbicularis oris muscle and mucosa from the medial edge to 1.5 cm from the lateral aspect. Then, a muscle flap is created under one-half of the orbicularis oris muscle. On the tubercle media, dissection is performed between muscle and mucosa until the midline of Cupid’s bow on the middle of the vermilion in which volume deficiency occurs and a muscle flap is created under one-half of the orbicularis oris muscle. Then, both muscle flaps are flipped and crossed against each other. First, the muscle flap on the medial aspect is fixed 5 mm lateral to the orbicularis oris muscle. Then, they are sutured to each other with 4.0 chromic catgut. The lateral muscle also is sutured with 4-0 chromic on the medial edge point of the central tubercle. The rest of the mucosa is excised minimally.

3. **Bilobed Mucosal Flap Technique**: A bilobed flap is used commonly to repair a defect on the skin, and the use of this method to correct a secondary vermilion deformity in cleft lip has not been reported to our knowledge. Song et al. stated that this technique was suitable for patients with a whistle deformity, due to the simultaneous thickening of inadequate lateral vermilion and tubercle vermilion [18].

4. **Surgery technique**: Surgery begins with the use of local or general anesthetic. The first lobe (Lobe A) on the prolabium of the oral mucosa reaches the labiogingival groove and is less than 10 mm deep. The second lobe (Lobe B) is placed on the vermilion mucosa on the lateral side of the gap. The length and depth of the gap depend on the discrepancy between the vermilion at the gap edge and that of the normal side. A mucosal flap lifts the orbicularis oris muscle. Lobe A is used to correct the whistle deformity and reconstruct the tubercle vermilion. In patients with severe vermilion deficiency, Lobe A is longer in the design, so the distal part of the lobe can be demucosalized and folded inside for augmentation of the tubercle vermilion. Lobe B is rotated to repair the oral mucosa defects made by Lobe A. Harvesting lobe B simultaneously will thin the thick vermilion side and achieve correction of three deformities in one phase.

The advantages of bilobed mucosal flap are: (1) it is technically easy; (2) provides the largest subcutaneous and mucosal surfaces at the same time; (3) provides enough tissue to avoid upper lip constriction (the oral mucosa on the upper lip will provide adequate tissue for reconstruction); (4) because of good vascularization of the bilobed flap, unlike a free graft, it normally will heal without any risk of infection or absorption; and (4) little tissue is removed with this technique as the excess mucosa on the lateral vermilion effectively will be transferred to repair the defect in the oral mucosa. Disadvantages include a slightly dissimilar color between dry and wet mucosa. The bilobed mucosa flap simultaneously will repair the secondary deformity of the lip from cleft lip repair, including the whistle deformity, and thickening of inadequate lateral vermilion and tubercle vermilion. This technique will provide satisfying functional and esthetic results.
5. Conclusions
According to indirect anthropometry measurements (photograph), there was no difference in vertical height of the tip of Cupid’s bow on the noncleft side in patients with complete unilateral cleft lip after Cronin labioplasty. There were whistle deformities on the cleft sides after labioplasty in patients with unilateral cleft lip.

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