

CORRECTION OF WHISTLE DEFORMITY BY BILOBED MUCOSAL FLAP TECHNIQUE IN UNILATERAL CLEFT LIP

Ari Raheem Qader *, Hawree A Hassan **

and Dana Abdulmajid ***



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ABSTRACT

Background

The whistle deformity is one of the common sequelae of secondary cleft lip deformities. It is often accompanied by asymmetric upper lip thickness and insufficient vermilion tubercle. The bilobed mucosal flap was described by Song Tao. To correct these deformities in a single stage, we have used the same technique in our study and combined with other procedure to correct wider array of secondary cleft lip deformities.

Objectives

To evaluate the reliability and versatility of bilobed mucosal flap in reconstruction of whistle deformity, insufficient tubercle and lateral vermilion redundant.

Methods

Nine patients with whistling deformities were included in our study. Their ages ranged from 5-34 year. All cases were corrected by bilobed mucosal flap, which consist of two lobes The first lobe located at the oral mucosa of the lateral affected lip side, used to correct whistle deformity and augmenting the tubercle. The second lobe elevated from the lateral vermilion mucosa on the affected side and used to repair the oral mucosal defect, at the same time it decreases the excess on the lateral side.

Results

Minimum follows up was 6 month. All patients were satisfied with the aesthetic outcomes. In all of the patients, whistle deformity were corrected, tubercle augmented and more symmetric vermilion appearance were achieved. Major complications (necrosis of mucosal flap, infection and recurrence of whistling deformity) did not occur in any of the patient.

Conclusion

A bilobed mucosal flap is a safe and simple procedure, effectively performed to correct combined secondary deformities after primary cheileoplasty in unilateral cleft lip patient. It can be combined with other procedures to revise upper lip scar, repair muscular diastases, and correction of severe whistle deformity

Keywords: *Whistle deformity, cleft lip, Scar .*

* Department of Surgery, School of Medicine, Faculty of Medical Sciences, University of Sulaimani.

Correspondence: ari.gader@univsul.edu.iq

** Sulaimani Burn and Plastic Surgery Hospital.

*** Department of Community Health, Technical College of Health, Sulaimani Polytechnic University

INTRODUCTION

The whistling deformity was defined as insufficient tissue in the lower border of a repaired cleft lip, giving the appearance of whistling while in repose and worsening on activation ⁽¹⁾. It is characterized by inadequate fullness of the central upper lip with abnormal exposure of the central incisors ⁽²⁾. This deformity is usually accompanied by excessive redundancy of mucosa and the vermilion on the affected side ⁽³⁾, (Fig 1). It is not only unsightly, but it may interfere with speech because of inability to produce a lip seal—so necessary to form the plosive consonant sound ⁽⁴⁾. This deformity is more common in bilateral cleft lip than in unilateral and frequently more severe ⁽⁵⁾.

The cause could be failure of alignment of orbicularis oris and orbicularis marginalis at the time of primary lip surgery, scar contracture of the primary lip repair or dehiscence of orbicularis oris or marginalis. Apart from that, vermilion volume deficiency of the upper lip may occur due to normal patient growth, infection of suture line and dehiscence of the repaired site. These factors often lead to combination of notching over the free border of the lip, inadequate tubercle size, or deficiency in volume of the free border of the lip ^(6, 7).

Many techniques have been described for correction of whistle deformity. When the deformity is mild, the upper lip itself can be used for reconstruction, such as V-Y mucosal advancement and Z-plasty ⁽⁸⁾, by placing the central limb of the Z on the existing scar ⁽³⁾. Matsuo et al. ⁽⁹⁾ developed bilateral lateral vermilion border transposition flaps, which they are marked inside the vermilion-mucosal junction. Kapetansky ⁽¹⁰⁾ reported double pendulum flaps for augmentation of the central defect. Juri et al. ⁽¹¹⁾ developed the bilateral mucomuscular flaps to fill up the insufficient central tubercle in the Kapetansky technique. Robinson used double V-Y procedure, by V shape incision done in lower border of vermilion which is closed in Y manner ⁽⁴⁾.

For the moderate or severe cases, the adjacent tissue, including the tongue and lower lip, can be used. Guerrero-Santos ⁽¹²⁾ used two types of tongue flap for the reconstruction of the upper lip. Jeffrey D. Wagner ⁽¹³⁾ used bipedicle axial cross-lip flap for major deformities. Patel and Hall ⁽¹⁴⁾ used dermal fat grafting to correct a whistle deformity, although it has been conventionally used to correct a facial contour or nose deformity. James Belyea et al. ⁽¹⁵⁾ used autologous fat graft to augment the free border of the lip and correct whistling deformity.



Figure 1. Showing combination of whistle lip, inadequate tubercle and lateral lip excess.

Niechajev ⁽¹⁶⁾ also reported lip enhancement using various alternatives including implants, autologous fat graft, and derma fat graft. Dermal fat grafting also used by Craig Staebel ⁽¹⁷⁾ putting in a manner that the dermis side inward facing the muscle. Cohen et al and K. Kawamoto ⁽¹⁸⁾ they used free tongue graft by making incision along free vermilion border, tongue graft taken below the papillary line, undermining in the direction of sagittal deficiency and graft placement. Millard DR. ⁽¹⁹⁾ described the use of dermal fat graft in combination of V-Y advancement in sever cases. KN Lee and ks Koh ⁽²⁰⁾ developed new technique to overcome sever cases of absolute shortage of tissue, by using combination of V-Y advancement of labial mucosa , with providing additional tissue to the deficient area with a cellular human dermis or temporoparetal fascia. Another combination is rectangular mucosal flap with artificial dermis graft for vermilion deformity described by S. Wakami et al ⁽²¹⁾.

Song T et al. developed new technique to address triplex of deformities (Upper lip whistle deformities, asymmetric upper lip thickness and insufficient vermilion tubercle) simultaneously ⁽²²⁾.

In our study, we evaluated the versatility of the procedure described by Song T. and this technique can be combined with the other procedure to correct severe type of whistle deformity.

PATIENTS AND METHODS

From January 2012 to July 2013, 9 patients (5 males and 4 females; ages at operation ranged from 5-32 years; mean 14 years) were enrolled in our study (table 1). We selected those cases that they presented with notching of upper lip, insufficient tubercle and the bulging on the lateral lip segment. None of these patients had undergone previous attempt to correct the whistling deformity. Cleft lip repair was unilateral in all cases.

All patients were assessed preoperatively with appropriate clinical examination, taking into consideration the previous surgery and its details. Examination of the lip for symmetry, volume deficiency, scar, and associated nose deformity were done. Severity of the defects, categorized in to mild, moderate and severe through calculated defect ratio for labial width and labial height, and then defined the product of both ratios as a defect score . Photographs were taken before and six month after operation to evaluate the out comes, and this was through comparing the ratio of five parameters as shown in (Figure 2). Informed consent was obtained from all the patients or their parents if the patient was younger than 18 years before their participation in the study.



Figure 2. The five parameters are, AA': vertical Height at non-affected Cupid's bow peak. BB': vertical Height at affected Cupid's bow peak. CC' is the vertical lip heights at the mid-point between the Cupid's bow peak and oral commissure on the non-affected side. DD': is the vertical lip height at the mid-point between the Cupid's bow peak and oral commissure on the affected side. OO': is the vertical lip height at the philtrum point.

Table 1. Case summary *

No	Age(yr)/Sex	Type /Site	Severity by defect ratio†	Type of operation	F/U	Complications
1	5/female	Complete/Lt.	Moderate	Bilobed mucosal flap	16 month	Small notch at the base of the flap revised after 3 month.
2	24/female	Complete/Lt.	Moderate	Bilobed mucosal flap	14 month	Small notch at the base of the flap revised after 3 month.
3	11/male	Complete/Lt.	Moderate	Bilobed mucosal flap + upper lip scar revision + correction of muscular diastases	12 month	Color mismatch
4	15/female	Complete/Lt.	Sever	Bilobed mucosal flap + derma fat graft	11 month	Non
5	32/male	Complete/Lt.	Sever	Bilobed mucosal flap + derma fat graft	10 month	Non
6	14/female	Complete/Lt.	Mild	Bilobed mucosal flap + upper lip scar revision	10 month	Erythematic upper lip scar in early post operative period, treated conservatively
7	6/male	Complete/Lt.	Moderate	Bilobed mucosal flap + upper lip scar revision + correction of muscular diastases	8 month	Color mismatch
8	7/male	Incomplete /Rt.	Mild	Bilobed mucosal flap	5 month	Non
9	18/male	Microform/Lt.	Moderate	Bilobed mucosal flap + scar revision + correction of muscular diastases	5 month	Non

* Yr, year; Lt., left; Rt., Right; F/U, follow-up; †, product of defect ratio in height and width.

Surgical technique

All patients underwent surgery under general anesthesia. The deficient area was evaluated and bilobed mucosal flap was marked by methylene blue marker (figure 3). The first lobe (Lobe A) is originally located at the oral mucosa of the prolabium as described by Song T. Here we put this lobe on the mucosa of the affected side in the manner that, the base and medial incision of the lobe is located lateral to the deepest point of the notch, and it reached the labiokingival groove. The second lobe

(Lobe B) located at the vermilion mucosa on the lateral lip segment, and the length and width were adjusted according to discrepancies of vermilion thickness between the affected and normal side.

The operative area was infiltrated with 1% lidocaine and 1:200,000 epinephrine. Incisions were made with no. 15 scalpel, after seven minutes from the time of infiltration, the mucosal flaps were raised off the orbicularis oris muscle. Both of the two lobes were rotated inward (Figure4).

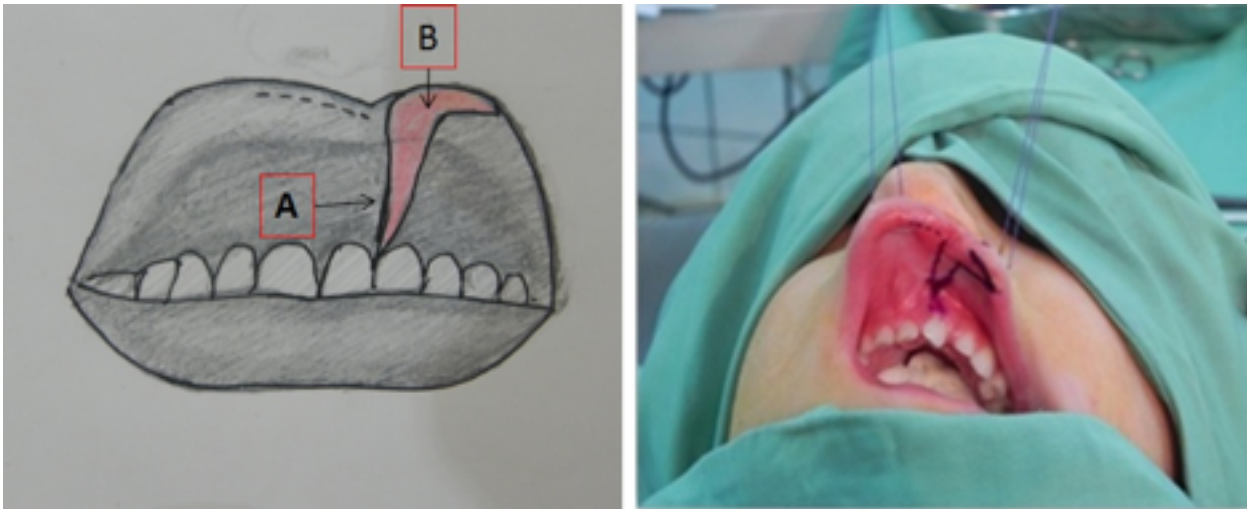


Figure 3 . The first lobe (Lobe A) is located at the oral mucosa of the lateral lip. The second lobe (Lobe B) is located at the vermilion mucosa on the lateral lip segment. The dotted line indicates an incision between dry and wet vermilion on the whistle area to house the inseting of (Lobe A).

The operative area was infiltrated with 1% lidocaine and 1:200,000 epinephrine. Incisions were made with no. 15 scalpel, after seven minutes from the time of infiltration, the mucosal flaps were raised off the orbicularis oris muscle. Both of the two lobes were rotated inward (Figure 4).

Lobe A was used to correct whistle deformity and augment the tubercle. Lobe B was rotated to repair the oral mucosal defect created by Lobe A. Through harvesting the Lobe B, the thick affected side becomes thinner and more vermilion symmetry was achieved. Thus it corrects three deformities simultaneously (Figure 5).

In patients that were repaired by dermal fat graft in combination with bilobed mucosal flap, the procedures were done in the same manner like above. Both A and B flap were raised and the incision between dry and wet vermilion through the whistle area was done. Through the same incisions, undermining of mucosa just above the orbicularis muscle was done then graft was harvested from groin area through an elliptical incision in the direction of skin creases, after proper trimming, the graft is placed in the area with dermis side inward,

facing the muscle. The graft is fixed through both ends by 0.4 absorbable sutures (figure 6).

In cases of upper lip scar and muscular diastases with whistle deformity, they were submitted to scar revision and repair of diastases first, then reconstruction of whistle lip by bilobed flap in the same section. Closures of donor site of Lobe B, and inseting of Lobe A were done by 0.6 polyglycolic acid suture. The upper lip skin was sutured by 0.5 prolene, and the donor area of dermal fat graft sutured in layers. Wound covered by application of antibiotic ointment.

Patient's first visit is after five days, to check the surgical site, oedema, and cleaning of the wounds. Within five to seven days suture are removed, with prevention of direct sun light were taught. The third follow up visit is at the fourth week. Early assessment is at three month, here, patients asked whether they are satisfied with the result, check for correction of triplex deformities (whistle deformity, deficient tubercle and lateral lip thickness), and clinical photographic follow-up. Final assessment was at six month.

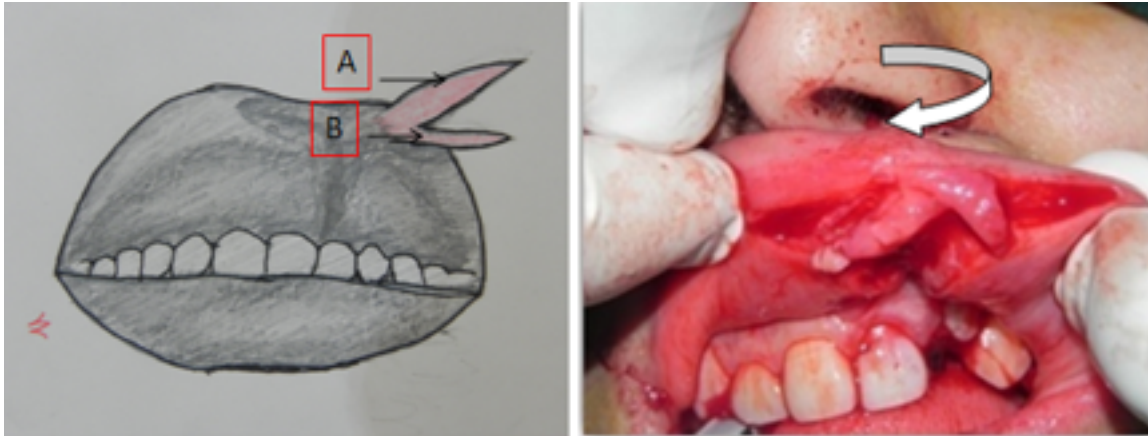


Figure 4. The mucosal flaps were raised from the orbicularis oris muscle, and they were rotated inward.

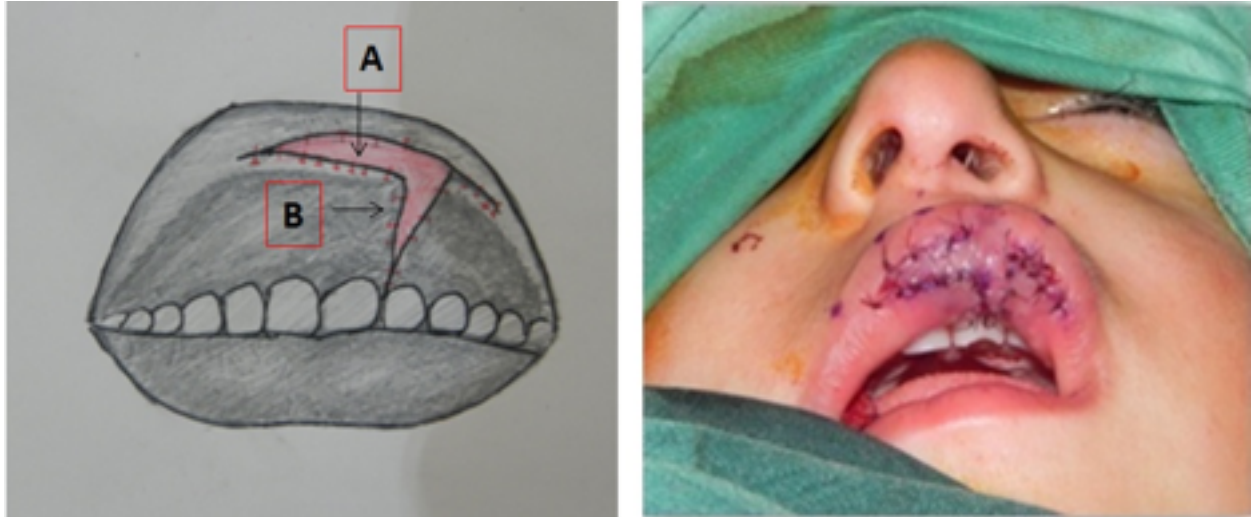


Figure 5. Lobe A was used to correct whistle deformity and reconstruct the vermilion tubercle. Lobe B was used to repair the oral mucosal defect.

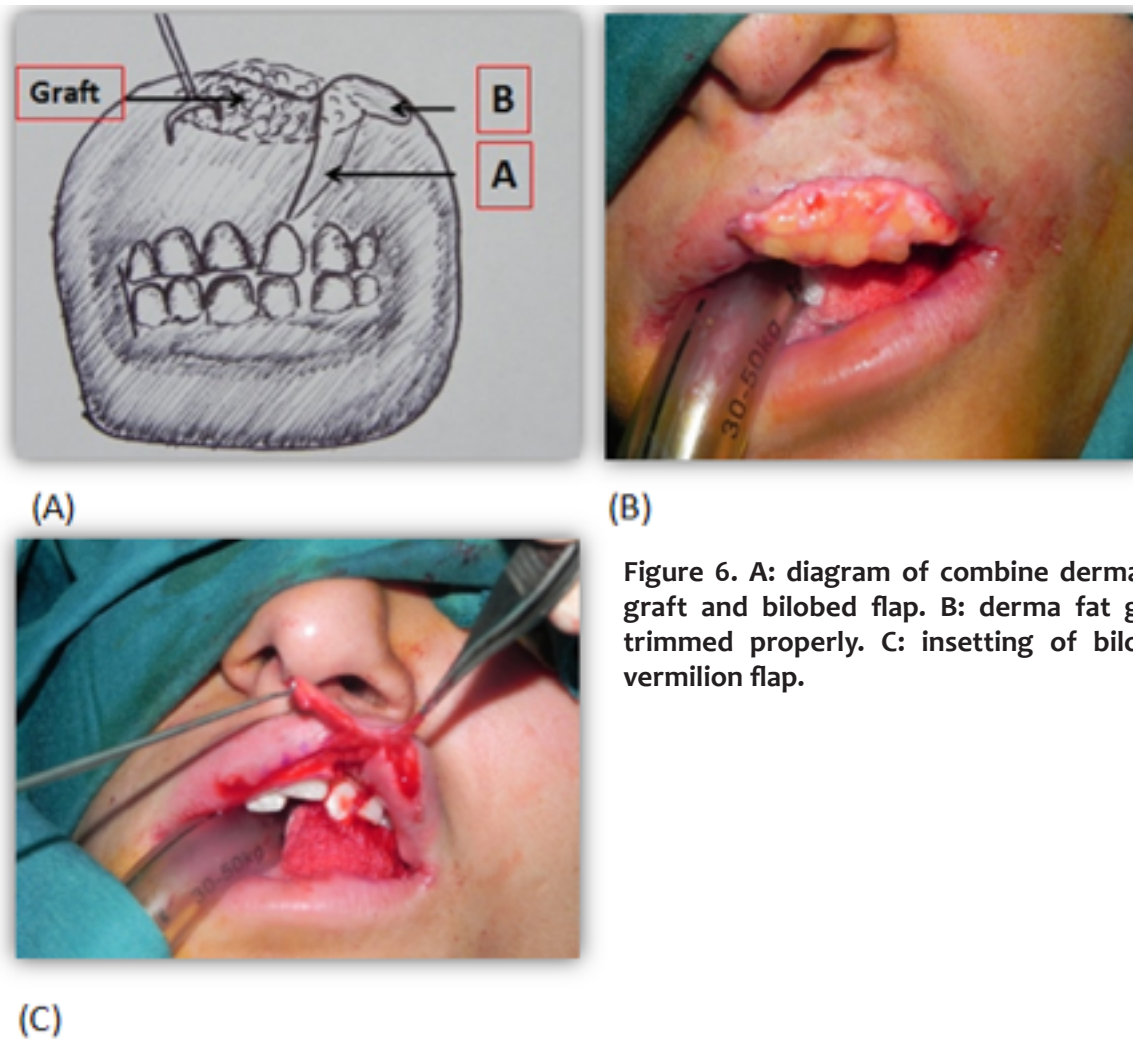


Figure 6. A: diagram of combine derma fat graft and bilobed flap. B: derma fat graft trimmed properly. C: inseting of bilobed vermilion flap.

RESULTS

Assessment of the results was evaluated after 6 month, through the clinical examination and statistical analysis using the statistic package for Social Science (SPSS) software. Student's t-test was used to assess differences between the measurements. (Table 2) shows four vertical lip height ratios before and after operation of each case. (Table 3), showing mean of the four height ratio pre- and post operatively. Preoperatively, the vermilion on the affected side was thicker than that on the unaffected side (DD'/AA' bigger than CC'/AA'). And tubercle is inadequate in the affected patients as determined by small OO'/AA' (Figure 7).

After 6 month, there was improvement of both DD'/AA' and OO'/AA' . CC'/AA' was nearly equal to DD'/AA' , this mean that whistle deformity were corrected and the vermilions on either sides became more symmetrical in thickness (Figure 8).

All patients were satisfied with results. In all of the patients, there were no major complications such as hematoma, infection, partial or total flaps loss. The patients had no complaint of dryness or crusting in their lips mucosa. The vermilions displayed total closure at rest.

Minor complication like color mismatch notable in two cases. Small notch at the base of flap were seen in two cases; therefore, revision were performed after 3 month. Erythematic upper lip scar is seen in one case, which was treated by topical steroid ointment for two weeks and then applications of silicone scar gel for three month. In cases of derma fat graft, patients felt upper lip firmness in the first few weeks only; there was no infection or cyst formation seen, donor area healed well and free from hypertrophy or scar widening. Patients showed good movement and good shape of upper lip without any tightness and recurrent whistling seen.

Table 2. Ratio of preoperative and postoperative vertical measurements at four different points on the upper lip.

case	Height ratios comparison before operation				Height ratios comparison after operation			
	OO/AA	BB/AA	CC/AA	DD/AA	OO:AA	BB/AA	CC/AA	DD/AA
1	0.667	0.917	0.568	1.083	0.944	1.000	0.600	0.488
2	0.643	0.857	0.610	1.014	0.959	0.993	0.632	0.517
3	0.800	0.867	0.592	0.967	1.000	0.955	0.583	0.636
4	0.267	0.733	0.622	0.933	0.833	0.889	0.614	0.722
5	0.625	0.750	0.612	0.938	0.996	0.835	0.603	0.588
6	0.800	0.800	0.613	1.200	1.083	1.000	0.656	0.600
7	0.667	0.883	0.513	0.979	0.997	0.942	0.548	0.582
8	0.902	0.900	0.566	1.377	1.105	1.036	0.573	0.703
9	0.699	0.756	0.609	0.946	0.909	0.922	0.612	0.653

Table 3. Average of height ratios comparison before and after operation.

	OO'/AA'	BB'/AA'	CC'/AA''	DD'/AA'
Before op	0.671	0.838	0.587	1.061
After op	0.990	0.956	0.601	0.605

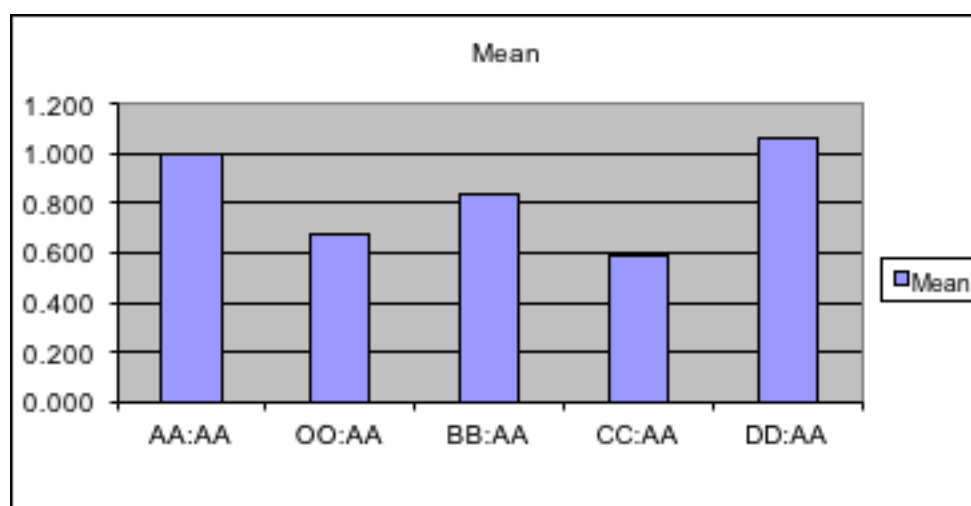


Figure 7. Height ratio comparison before operation. CC'/AA' was smaller than DD'/AA' (P< 0.05) and OO'/AA' was inadequate.

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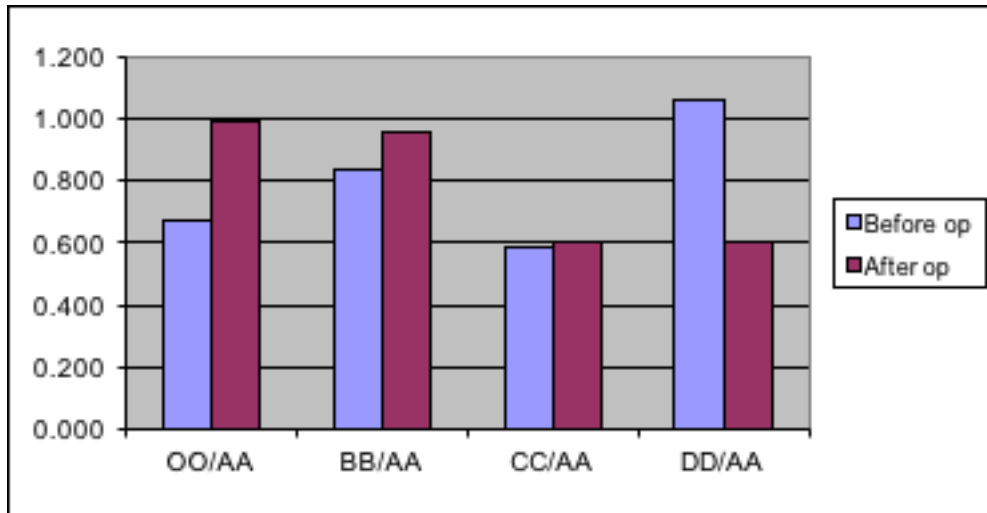


Figure 8. Marked improvement in DD'/AA' and OO'/AA' after operation ($P < 0.05$)
OO'/AA' rose approaching 1.000.

CASE EXAMPLE NO. 1



Figure 9. A 5-year old female underwent primary repair of a complete cleft lip on the left side at the age of 4 month. (A,B) preoperative image showing, whistle deformity, deficient vermilion tubercle and vermilion thicker on the left side. (C,D) postoperative images after 6 month showing correction of three deformities by bilobed mucosal flap technique.

CASE EXAMPLE NO. 2



Figure 10. A 13-year old female underwent primary repair of a complete cleft lip on the left side at the age of 6 month. (A,B) preoperative image showing, upper lip scar on the affected side, whistle lip deformity and deficient vermilion tubercle. (C,D) post operative image at 6 month showing correction of whistle deformity by bilobed flap and upper lip scar revision at the same time

CASE EXAMPLE NO. 1



Figure 11. A 15-year old female underwent primary repair of a complete cleft lip on the left side at the age of 18 month. (A,B) preoperative image, showing whistle deformity, vermilion tubercle severely deficient and lateral lip thicker than the non-affected side. (C,D) postoperative image at 6 month later showing correction of triplex deformity by the use of combined dermal fat graft and bilobed mucosal flap in the same section under general anesthesia.

DISCUSSION

Cleft lip patients have a variety of secondary deformities after primary cheiloplasty. Whistle deformity is one of the common deformities and usually requires surgical correction⁽²³⁾. Aiming to gain functionally and aesthetically proper vermilion border without any irregularities, with sufficient tubercle bulk tapered with vermilion of lateral lip element, and allows the patient to close the mouth completely⁽²⁴⁾.

This deformity usually present with asymmetric upper lip thickness and insufficient vermilion tubercle, usually it is not possible to correct all three deformities with a single surgical technique by previous described technique⁽²²⁾.

Many techniques have been described for correcting whistle deformities, their uses are depend on the severity of defect and the presence of local tissue. If the vermilion deficiency is mild to moderate, the upper lip itself can be used for reconstruction, such as V-Y advancement, Z-plasty^(3, 8) are performed both the most frequently and easily, but they are used for the mild deficiency and they will not correct the lateral lip excess. In bilateral lateral vermilion border transposition flaps described by the Matsuo K⁽⁹⁾, tension was reduced on the upper lip and deepens the labiogingival sulcus; however, the use of this method can impair the continuity of orbicularis oris muscle because this flap contains only a thin layer of orbicularis oris muscle. The double V-Y advancement of vermilion mucosal flaps, described by Robinson⁽⁴⁾, result in multiple intersecting scars at the junction of the four flaps. Mucosal, as well as intramuscular, scarring is also a possible complication of 'double pendulum' flaps described by Kapetansky⁽¹⁰⁾. In some patients, however, rearranging local flaps from the lateral area sacrifices the horizontal width of the upper lip, which worsens the existing asymmetry of the upper lip and result in an unnatural outcome. And they are not enough to overcome severe deficiency⁽²⁰⁾.

Distant flaps have also been used, including Abbe⁽²⁵⁾ and tongue flaps⁽¹²⁾. Although these flaps provide constant volume, Abbe flap require secondary divisional procedures and tongue flap often do not match the color and texture of the lip. Also these flaps have donor site morbidities which make them undesirable to the patient. The use of tongue graft⁽¹⁸⁾ also is not without complication, it is used only for small defect; as well as graft failure, color miss match, and donor site morbidities are other drawback.

Finally, autograft or allograft material for reconstruction can be used. The use of dermal fat graft and autologous fat graft, alone^(14, 16), are simple and easy techniques, but they are not increasing the mucosal surface and it is difficult to predict the rate of absorption, as well as upper lip firmness.

Craig staebel⁽¹⁷⁾, showed the fewer rate of lip firmness, by positioning the dermal fat graft in a manner, with dermis side inward facing the muscle and the fat side outward facing mucosal surface. With the new study of KN Lee and ks Koh⁽²⁰⁾, they combined local tissue flap with acellular human dermis, they gained lower absorption rates and better projection of the graft due to decreased surface tension on the graft. Through reviewing these two studies, we combined dermal fat graft with bilobed mucosal flap for repairing severe degree of whistle deformities.

Song T. developed new technique, the bilobed mucosal flap. It consist of two lobes (A and B), to address the triplex deformities (upper lip whistling, lateral vermilion excess and inadequate vermilion tubercle) in single stage, which has not been corrected by the other previous conventional procedure. Also he claimed that this flap has a good vascularity, so the chance of flap necrosis is very unlikely. He did operation on a 30 patients, their ages ranged from 10 to 35 years. All healed well with no complications of the flaps. The gross appearance of the vermilion was significantly improved; normal tubercle and good symmetry were achieved⁽²²⁾.

This flap is an excelent choice for cases that they have whislte deformity with a lateral bulging segment on the affected side, we could lessen the lateral bulging segment on the affected side and augment the defective central tubercle and correct the lip notching deformity at the same time. However, small notching at the base of flap were seen in the first our two cases, and this could be because of, First, the location of flaps were too medial on the prolabium in a manner that it is base located on the original notch. Second, inadequate lateral vermilion excess at all. for these reasons we have decided to move the flap lateraly putting on the mucosa of the affected lip.

Although correction of the severe degree of whistle deformity was mentioned by Song T. through demucosalysing the distal part of the lobe A and folding it inward, but in our opinion this part of flap

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is insufficient in correcting this problem as it tapered when approaching labiogingival groove. Even with folding, it doesn't reach an adequate bulk. As described by Patel, absolute shortage of tissue in the free border of the lip necessitates use of other tissue to achieve an aesthetic pleasing result ⁽¹⁴⁾.

We realized that the complication of dermal fat graft in combined with bilobed flap was much less as compared with use of graft alone because this combination augments the lip as well as increase mucosal surface, at the same time combined technique decreases the absorption rate of fat, as bilobed flap has a good vascularity, and it will decrease tension on the graft by donating more tissue.

In comparing our study with the original study of Song T. we have seen nearly the same result regarding correction of whistle deformity and decreasing the lateral excess and the P-value is significant ($P < 0.05$) in both.

We found that this technique can be combined with; First, upper lip scar revision and correcting vermilion border irregularity. Second, muscular diastases repair. Third, derma fat grafting. We added some extra point on the advantage and disadvantage on this technique which was mentioned previously by Song T.

The advantages of bilobed flap are:

- 1- It is an easy procedure.
- 2- It can provide both subcutaneous bulk and surface mucosa at the same time.
- 3-Enough tissue can be provided to avoid upper lip tightness.
- 4- Little tissue is wasted with this technique.
- 5- Through rebalancing of the tissue, horizontal vermilion symmetry were achieved using this technique.

The disadvantages of this procedure are:

- 1- This technique is not enough to correct the severe deficiency if it is alone, although we improved this drawback by combined it with dermal fat grafting with the acceptable results.
- 2- This technique leaves additional scars in the vermilion.
- 3- It will not correct the upper lip vermilion irregularity alone.
- 4- It show slight color mismatch between the donor and mucosal flap (figure 12).



Figure 12. Color mismatch between dry and wet vermilion.

In conclusion, the bilobed mucosal flap is shown to be effective in simultaneous repair of, whistle lip deformity, lateral vermilion excess and inadequate tubercle, in unilateral cleft lip patient. It is an easy technique and free from major complication. It can be combined with derma fat graft to repair severe whistle deformity.

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