

Research Article

Evaluation Of The Double Unilimb Z-Plasty Technique For Whistle Deformity Repair In Unilateral Cleft Lip Patients.

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Abstract

Background: The whistler deformity is characterized by a notching of the upper lip at the site of the lip repair. The incisors are usually exposed, and the patient lacks lip competence.

Objective: The purpose of this study was to evaluate the symmetry in lip and vermilion height after using the double unilimb Z-plasty method for Whistler deformity repair. Patients: from January 2017 to August 2018, 12 patients with unilateral whistle deformity were operated by using the double unilimb Z-plasty.

Results: The study found no significant differences between the right and left side in lip and vermilion height after lip repair using the double unilimb Z-plasty technique measured at least 6 months postoperatively.

Conclusions: The results suggest that the double unilimb Z-plasty technique is a good alternative to address the whistler deformity related to the unilateral cleft lip primary repair. This is a simple method, easy to perform.

Keywords: Whistler Deformity, Secondary Unilateral, Cleft Lip.

INTRODUCTION

The whistle deformity is characterized by a notching of the upper lip at the site of the lip repair. The incisors are usually exposed, and the patient lacks lip competence [1]. Secondary deformities of the unilateral cleft lip are diagnosed on physical examination. They include the deficient tubercle, vermilion deficiency and irregularities, the short upper lip, long upper lip, tight upper lip, and unfavorable scars. The most common irregularity is a notch or whistle deformity [2]. Secondary deformities of the lip after primary repair are spectrum of anomalies that range from superficial scar abnormalities to full thickness derangement of the skin, muscle, and mucosa. The most frequent secondary anomalies after primary repair include vermilion deficiency contributing to the "whistles" deformity, lip skin excess (long lip), lip skin deficiency (short lip), orbicularis disorientation and vermilion misalignment. Modalities for preventing secondary cleft lip deformities stem from appropriate presurgical treatment, choice of

operative technique, and meticulous attention to details during operative repair [3]. Vermilion deformities include thin or thick lip segments, Vermilion mismatches (between the wet and dry vermilion), Vermilion notching or border malalignment (between the white roll and vermilion), and the whistle deformity (median tubercle paucity resulting in non-apposition of upper and lower lip segment at rest) [4]. Vermilion deficiency occurs in unilateral patients in different forms. In unilateral deformity, deficiency of the vermilion on the medial lip is a common problem. This causes an apparent fullness on the vermilion of the lateral lip element, the "whistle" deformity or vermilion notch [5]. A short lip may result from straight line closure techniques or from inadequate rotation and advancement techniques or adequate rotation advancement technique used in unilateral cleft lip with short lateral segment, a dehiscence, scar disorder, or failure to approximate the lower portion of the orbicularis oris muscle during primary repair (resulting in an upward pull on the central tissue) may also cause the whistler deformity

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[5], It also can occur secondary to scar contracture across the vermilion, failure to fill the central tubercle with lateral vermilion tissue or combination of these.

The whistle deformity can be classified into mild, moderate and severe, according to the degree of tissue deficiency on each side as follows: Mild deformity, the difference between the right side and left side vertical height is less than 3 mm. In moderate deformity, the difference between the right side and left side vertical height is between 3 and 6 mm. In severe deformity, the difference between the right side and left side vertical height is greater than 6 mm. [5]. Careful analysis of the deformity may reveal misalignment of the muscle, vermilion, or labial mucosa as well as fullness or hypoplasia of the medial or lateral segment. Malalignment without significant tissue deficit can be corrected by readjustment of the available local tissue. This may involve complete revision of the lip with mobilization and realignment of the superficialis and profundo's components of the muscle. Realignment of the vermilion red line can be achieved by advancing the lateral segment excess into a medial segment red-line incision or back-cut. Hypoplasia or tissue deficiency can be noted in either medial or lateral segment [6].

Many surgical techniques have been described before to address these secondary deformities. These methods may be

selected in relation to the severity of the deformity. For severe deformities, an Abbe flap or lip reoperation is necessary. For mild to moderate deformities, many options may be used with good results. These include local flaps, Z-plasties, fillers and free grafts can be used with relatively good outcomes. The double unilimb Z-plasty method described by Mulliken for primary microform cleft lip repair is an operative correction which includes a double-limb Z-plasty at the vermilion-cutaneous and vermilion-mucosal junctions. [7]. Percy Rossell-Perry used this method for whistler deformity repair in 52 unilateral cleft lip adult patients (2009-2012) [8]. In this study we utilized the double unilimb z-plasty technique for correction of whistle deformity in unilateral cleft lip patient.

PATIENT AND METHODS

Design of the Study

Twelve patients were presented to the outpatient clinic with mild to moderate post cleft lip repair whistle deformity (unilateral cleft lip) at least one year postoperatively from the primary cleft surgery. Those patients were surgically treated for their whistle deformity by using double unilimb Z-plasty technique at Al-Wasity hospital between the periods of January 2017 to August 2018. Patient's data are shown (**Table 1**).

Table 1. patients' data.

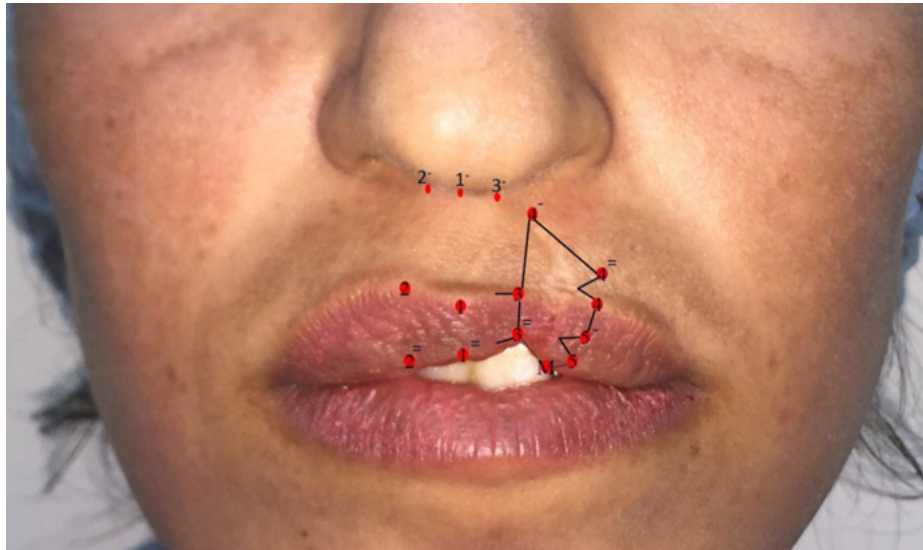
| Patient | Sex | Age | Site of unilateral C.L | Vertical difference between RT and LT. vertical height | Type of whistle deformity |
|---------|--------|---------|------------------------|--|---------------------------|
| 1 | Male | 37 year | Lt. | 3mm | mild |
| 2 | Male | 32 year | Lt. | 2mm | mild |
| 3 | Female | 28years | Rt. | 5mm | Moderate |
| 4 | Male | 6 years | Lt. | 3mm | mild |
| 5 | Female | 13years | Lt. | 4mm | Moderate |
| 6 | Male | 17years | Lt. | 4mm | Moderate |
| 7 | Female | 7 years | Lt. | 2mm | mild |
| 8 | Male | 4 years | Rt. | 3mm | mild |
| 9 | Female | 7 years | Lt. | 5mm | Moderate |
| 10 | Male | 5 years | Rt. | 3mm | mild |
| 11 | Female | 14years | Rt. | 3mm | mild |
| 12 | Male | 18years | Lt. | 3mm | mild |

Those patient with severe form of whistle deformity (> 6mm discrepancy between Rt. and Lt. Vertical height) were excluded from the study. Preoperative measurement and examination were done, measurement was done using caliper to measure both Rt. and Lt. Side vertical height of the lip and calculated the difference between them. Physical examination was done to assess the vermilion deficiency, asymmetry of vermilion and teeth appearance. Our assessment was also based on comparison of both pre- and post-operative photographs which was taken to the patient both in frontal and lateral profile. Routine preoperative investigation was done for all of our patient, and informed consent was obtained from them preoperatively.

Operative technique

All operations were done under general anesthesia. After antisepsis and drapping, the procedure begins by marking both lateral and medial lip as follows (**Figure 1**);

Figure 1. The double-limb Z-plasty technique markings Medial lip.



At the vermilion-cutaneous junction point 1 is marked which represents the middle point of the Cupid's bow. Point 2 at the peak of cupid's bow on normal side and point 3 at the peak of cupid's bow on the cleft side. Then, at the base of the columella (mid-point of lip -columellar junction) point 1' is marked, point 2' and 3' are marked at the lip-columellar crease on both sides. The distances 2-2' , 3-3' will represent the height of philtral columns on the normal and cleft sides respectively. The Degree of vertical lip deficiency will be calculated by the difference between 2-2' and 3-3'. At vermilion mucosal junction point 2'', 1'' and 3'' which are mirror image of point 2, 1 and 3 are marked respectively.

Lateral lip segment of the cleft side

Point 4 is chosen along vermilion-cutaneous junction where the cutaneous roll (white roll) end at the level of cleft lip repaired scar. Then, at the level of intersection of vertical scar with subalar crease we mark point 4'. Sometimes 4'-point and 3' may be located at same plane. Then according to the difference between 2-2' and 3-3', point 4' is chosen above point 4. Then point 5 is marked at the red line, Below point 4 point 5' is marked which is located at equal length to line 3-3' (i-e: 4-5' =3-3'). And the distance from point 5 to point 5' is equal to the difference between 2-2' and 3-3'. On the oral mucosa point M is marked in a distance from point 5 similar to the distance between point 5 and point 4.

Distance 4-4' will represented the lip height on lateral side of the cleft, which sometime equals to 3-3' (which represents lip height on the medial cleft side). The difference between 2-2' and 3-3' (H-value) will represent the amount of tissue deficiency and determines the necessary amount of rotation of the medial cleft side. While the difference between 2-2' and 4-4' will represent the amount which is needed for lengthening the lateral side of the defect. So the amount of tissue deficiency in medial segment and the vertical height of

the vermilion should be:

$$2-2'' = (3-3'') + (5-5') + 1\text{mm (Rose - Thomson effect)}$$

Then we draw two triangles, the 1st on the vermilion cutaneous junction in which there sides equal to (difference between 2-2' and 3-3') started from point 4 or 1mm above it (Rose - Thomson effect) to point 4' and medially to cleft scar . From point 3 we draw line medially equal to (H-value) above the white roll. The 2nd triangle on the vermilion mucosal junction in which three sides equal to (difference between 2-2' and 3-3') starts from point 5 to point 5' superiorly and medially to cleft mucosal scar. From point 3' we draw line medially equal to (difference between 2-2' and 3-3') above the red roll.

Surgical operation

All marked points are tattooed by using methylene blue dye. Local infiltration is used with 2% xylocaine mixed with 1:200,000 adrenaline. The surgical operation starts by using No. 15 blade. First the incision is done starting from 4- to point 4' and point 3 downward (laterally and medially respectively) ending to point M, to creating 1st triangle of skin and subcutaneous tissue of the (vermilion cutaneous flap) and creating lower vermilion advancement flap . The incision is done through skin and subcutaneous tissue with Deepithelialization of scar, then the pars marginalis of orbicularis oris muscle is freed completely from overlying and underlying skin and mucosa respectively. Then downward rotation of Cupid's bow is done. After that lateral segment incision is done to create upper and lower advancement flap. The orbicularis oris muscle is freed from mucosa and skin by using blunt scissors (**Figure 2**).

After the completion of rotational and advancement flaps on both sides of cleft, closure of the orbicularis muscle is started by using 5/0 polyglactine suture, the same suture then used to repair the mucosa. Then careful skin approximation is done by using 6/0 polyglactine suture so that both point 3, 4 are

approximated together. Then vermilion repair is done by same suture. The closure of flaps would be like as (V in slit). After finishing the operation, light dressing is applied on the lip which consists of antibiotic impregnated gauze and patients were kept on oral antibiotic for 5-days postoperatively, the patients were instructed to avoid hot food and excessive chewing for 1st 2 week postoperatively. The Patient were kept on regular follow up period for at least 1 year postoperatively.

Ethical Considerations

The study was conducted in compliance with ethical standards. Permission to access patient data and perform diagnostic evaluations was obtained from the respective hospitals. All patient data were anonymized to maintain confidentiality. If required by the institutional review boards, verbal or written consent was obtained prior to inclusion.

Figure 2. Intraoperative view of the medial and lateral segment rotation and lengthening.



RESULTS

12 patients with post unilateral cleft lip repair whistler deformity were operated on by using double unilimb Z- plasty technique. The whistle deformity was mild (<3 mm difference between right and left side line in 7 cases (**Figures 3-8**), and in 4 cases the deformity was moderate (between 3-6 mm). postoperative results showed little or no difference both in cleft and non- cleft side lip and vermilion height, with achievement of good symmetry in vermilion on both sides with smooth, adequate fullness of cupid bow and no teeth show. The Patients were satisfied with postoperative result and they regarded the operation offered them excellent aesthetic result regarding their vermilion appearance. No postoperative complication like wound infection or dehiscence were seen in the immediate period and on long term follow up the resulting scars were barely visible with time, and no hypertrophic scars were encountered. No recurrence of the whistle deformity was seen.

Figure 3. Case NO. 2: Preoperative and 6 months Postoperative.



Figure 4. Case NO. 3: Preoperative and 1 month Postoperative.



Figure 5. Case NO. 4: Preoperative and 3 months Postoperative.



Figure 6. Case NO. 5: Preoperative and 3 months Postoperative.



Figure 7. Case NO. 11: Preoperative and 2 months Postoperative



Figure 8. Case NO. 12: Preoperative and Postoperative.

DISCUSSION

The Z-plasty is a frequently used procedure in plastic surgery. Classically this technique involves construction a Z-shape with 3 limbs of equal length that form 2 triangular flaps of equal length and equal angle. The two triangular represented flaps are interchanged with each other by both pivotal and tissue advancement movement [8]. The exact origin of the Z-plasty concept is unclear, but most likely dates back several centuries ago. The first documentation of this procedure, however, does not appear until early 1800 with firick and horner where they described single transposition flaps [9].

Patients with cleft lip typically will undergo a different reconstructive surgical procedure throughout their life, yet they are still left with stigmata of residual tissue defects. Most commonly, these tissue deficiencies of the upper lip lead to many abnormalities such as indistinct philtral column, upper lip flatness and deficiency of vermilion or what is called whistle deformity. These problems are usually worsened by the postoperative labial scarring [10].

Whistle deformity is the condition where the free margins of upper and lower lip did not meet together in the center when lips are in repose position so that give whistling appearance. It is a common secondary deformity of post-cleft lip repair, and its more frequently occur in bilateral cleft lip [11]. In this study double unilimb Z-plasty was evaluated for treatment of 12 patients who were presented to us with mild to moderate post unilateral cleft lip repair whistle deformity. All of our patients were satisfied with their postoperative result. Double unilimb Z-plasty technique was used by Percy Rossell-Perry, successfully for treatment of 52 patients with whistle deformity, their study showed no statistically difference in right and left side lip height and height of vermilion postoperatively. The double unilimb procedure for correction of the whistle deformity, had been described by Mulliken for correction of microform cleft lip, where he was used unilimb double z-plasty at the vermilion cutaneous and vermilion - mucosal

junction to correct the vertical asymmetry in microform cleft lip and in same time he limited the resultant scar to the lower one half of upper lip. The main idea of using double unilimb Z-plasty in correction of the whistle deformity is to equally distributing the tissue and so that symmetry is maintained. In whistle deformity there is discrepancy in vertical height between cleft and non-cleft side, so using of unilimb double Z-plasty will equalize the vertical height of both the lip and vermilion,

Double unilimb Z-plasty offer a simple method for correction of mild to moderate whistle deformity. First, it addresses the causes of the whistle deformity, namely, short vertical height of the lip and muscle deficiency, by using of double -unilimb Z-plasty both the short vertical height of the lip and vermilion can be corrected, and in addition to that, by bringing muscle fibers of parsmarginlis of the orbicularis muscle together, this will regain muscle continuity along the vermilion and will fill tissue deficiency across the vermilion which aids in correction and prevents the recurrence of whistle deformity. Second, double unilimb Z-plasty can correct the whistle deformity with minimum violation or distortion of lip tissue, since it depends on mathematically measured deficient tissue value, so that the rotation and advancement of the tissue is calculated according to the previously deficient value. Third, the double unilimb Z-plasty technique will end with small incision at the muco-cutaneous and vermilion-mucosal junction, these small incisions at the anatomical junctional side will leave barely visible scar with time as it was noticed in our patients.

Z-plasty may be modified by using flaps that have unequal angles. In such case, the flap which have more acute angle is pivoted in greater arc than that of larger angle degree flap. Minimal gain in the central limb will occur with this type of Z-plasty [12]. Because secondary cleft lip deformities are in most of the cases are asymmetrical, this will necessitate to use a symmetrical Z-plasty as was done successfully in this procedure. Various techniques have been used for surgical correction of the whistle deformity. In general, there is no

single technique used for all types of whistle deformity. One of the simplest approaches to correct mild whistle deformity is direct excision of the notch, this approach can increase tension in the area of paucity and can aggravate the condition [13]. Vaughn and Cronin have emphasized on the importance of using of vermilion flap to augment the problem during primary cleft repair to avoid whistle deformity. Crick Clair and thickly and Avons had described using of Z-plasties and other form of tissue arrangement of local Vermilion tissue for correction of whistle deformity [14]. The most frequently used operation for correction of mild whistle deformity is the Z-plasty and V-Y plasty. They seldom provide enough tissue for significant tissue deficit, and one of the disadvantages of these methods i.e Z- plasty and V-Y advancement is leaving the wet vermilion take a crusty appearance with chapped dry mucosa because of transfer of wet mucosa up on exposed site [15]. Although V-Y plasty is one of the most frequently used procedures for correction of whistle deformity, one of criticism of this technique is for shortening of the horizontal length of the lip, resulting in fishlike appearance of the lip [16]. In this procedure the transvers length of the lip was maintained.

Moreover, labial mucosa can be utilized as bipedicle or island pedicle myomucosal flap for correction of whistle deformity. Nevertheless, aesthetic result is usually less satisfactory, due to difference in texture, color and light reflection [17].

For many years, dermal fat grafting used with limited success. It is useful in those patients with available mucosa but with paucity of the underlying tissue. However, its survival is unpredictable and in addition to that it can result in lip firmness. Filler can be used to correct minor to moderate whistle deformity, but its cost, durability and tissue reaction still a matter of debate.

A number of authors have stressed on the importance of orbicularis muscle repair during primary cleft lip repair. The important of muscle repair has also been emphasized for correction of secondary deformity namely whistle deformity. Although successful correction of whistle deformity can be achieved without touching of the orbicularis muscle, it has been stressed that isolated scar revision of the lip will often follow by recurrence of deformity. In this procedure adequate approximation of the orbicularis muscle was done which gave satisfactory postoperative result, and prevented the recurrence of whistle deformity.

CONCLUSION

Double unilimb Z-plasty is simple method that can be used for surgical correction of mild to moderate whistle deformity without distortion of lip tissue with minimal incision. It depends on using of a symmetrical Z-plasty technique to redistribute tissue of cleft and non-cleft side together with

appropriate orbicularis muscle approximation, this procedure cannot be used with severe tissue deficit i.e. severe form of whistle deformity, where the difference between right and left side vertical height is greater than 6 mm.

Conflict of Interest

The authors declare no conflict of interest

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