

# Strategy for Correction of the Whistling Deformity in Secondary Cleft Lip Reconstruction

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**Background:** Following primary repair of a cleft lip, patients present with many facial deformities. One of the commonly observed sequelae of cleft lip repair is a whistling deformity. This retrospective study was carried out to evaluate the outcomes following correction of whistling deformities in secondary cleft lip reconstruction.

**Methods:** We retrospectively reviewed the hospital records of patients with various whistling deformities who underwent repair from April 1989 to March 2018; 2 surgeons performed the repair using either the double movable mucomuscular complex flaps technique, modified Abbe flap technique, or Abbe flap technique. The postoperative anatomical structure and aesthetic effects of the surgery were evaluated.

**Results:** In total, 136 patients were included in this study. Among these patients, 60 (44.2%) had a grade I whistling deformity and 47 (34.5%) had a grade II deformity and repair was performed using the double movable mucomuscular complex flaps technique and modified Abbe flap transfer technique, respectively, whereas the Abbe flap transfer technique was used in 16 patients (11.8%) and 13 patients (9.5%) with a grade III and grade IV whistling deformity, respectively. All patients were found to have normal postoperative anatomical structures and aesthetic effects of the upper lip, with all patients experiencing mild to moderate postoperative edema of the upper lip, and 29 cases (21.3%) developed an inconspicuous scar.

**Conclusion:** The repair technique should be chosen based on the type of whistling deformity. (*Plast Reconstr Surg Glob Open* 2020;8:e3156; doi: [10.1097/GOX.0000000000003156](https://doi.org/10.1097/GOX.0000000000003156); Published online 24 September 2020.)

## INTRODUCTION

Patients with a unilateral cleft lip or bilateral cleft lip and palate develop a variety of secondary deformities after primary cheiloplasty. Patients with secondary deformities, after primary repair of a bilateral cleft lip or bilateral cleft lip and palate, who were treated at the Department of Oral and Maxillofacial Surgery of Xiangya Hospital, Central South University, were observed, and 19 secondary deformities of the nose, lip, occlusion, and jaw were found.<sup>1</sup> These secondary deformities consisted of both hard and soft tissue

deformities. The hard tissue deformities included reversed overbite of the anterior teeth, displaced or missed canines, cleft alveolar bridge, small dental arch, disorder of the dentition, no anterior alveolar bridge, and a protruded anterior maxillae. The soft tissue deformities included a flattened nasal tip, displacement of the alar cartilages, asymmetric nostrils, absent nostril sill, short columella, tight upper lip, loose upper lip, no Cupid's bow or a Cupid's bow peak, no median tubercle, and a whistling deformity.<sup>1-5</sup>

Defects of the labial tubercle often occur after cleft lip repair due to improper treatment of the vermilion tissue. Absence of the labial tubercle, Cupid's bow, Cupid's bow peak, and Cupid's concave are often accompanied by exposure of the upper anterior crown or even the anterior crown and gums of the upper incisors. The second-stage repair of the vermilion of the lip after bilateral cleft lip surgery is more complicated than unilateral cleft lip surgery. Considering the particular position of the labial tubercle, its shape and fullness are essential to the overall repair effect.

Over the last 28 years, we have classified the different whistling deformities into 4 grades, according to the size of the exposure of the upper anterior crown or even the anterior crown and gums of the upper incisors. We aimed

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to determine the outcomes of the 3 different surgical techniques that are used in the repair of whistling deformities, which are selected based on the grade of the whistling deformity.

## METHODS

### Clinical Data

We reviewed and analyzed the data, including preoperative symptoms and postoperative effects, of patients with different whistling deformities following primary repair of a unilateral cleft lip or bilateral cleft lip at the Department of Oral and Maxillofacial Surgery of Xiangya Hospital, Central South University, between April 1989 and March 2018. The secondary repair of these deformities were performed by 2 surgeons. Patients were included in this study if they underwent repair of their whistling deformity. Patients were excluded if there was insufficient surgical and clinical data regarding the repair or they were lost to follow-up.

In this study, data collected for analysis included age, medical history, medicine hypersensitivity, defect size, and degrees of gum and crown exposure of the anterior upper incisors. Different techniques had been selected to repair the various whistling deformities. Elements used in the evaluation of the surgical repair included the Cupid's bow, Cupid's bow peak, the vermilion tubercle, the height and width of the upper lip, and scarring at the base of the nose.

### Classification of the Different Whistling Deformities

Patients' whistling deformities were graded according to the degree of exposure of the crown and the gums of the anterior upper incisors. The grade of each patient's deformity was evaluated. The authors classified these whistling deformities into 4 grades as follows: grade I, the crowns of the left and the right upper incisors are exposed above the first and second incisor; grade II, exposure of the crowns of the upper incisors and/or the crowns of the medial sagittal portion of the left and the right upper lateral incisors; grade III, exposure of the crowns of the two upper incisors and half of the gums over the two upper incisors or the crowns and half of the gums over the two upper incisors and more than two-thirds of the transverse crowns of the lateral incisors; grade IV, exposure of the crowns of the upper incisors and their attached gums and over two-thirds of medial sagittal crowns of the upper lateral incisors (Table 1).

### Surgery Selection

The surgical techniques used were selected according to the grade of whistling deformity or the absence of the

vermilion tubercle. Double movable mucomuscular complex flaps were used for repair of grade I whistling deformities (Fig. 1A), the modified Abbe flaps were used for repair of grade II and minor grade III whistling deformities (Fig. 1B), and grade III and grade IV whistling deformities were repaired using the Abbe flap (Fig. 1C).

### Surgical Techniques

#### The Double Movable Mucomuscular Complex Flaps for Grade I Whistling Deformities

In this group, 60 patients, with a grade I whistling deformity after a cleft lip repair, underwent the double movable mucomuscular complex flaps repair. Two planes of dissection were necessary. The anterior plane divided the lip coronally with approximately one-third of the orbicularis oris muscle mass anterior to it; the posterior plane was behind the labial vessels, deep in the submucosal layer. This dissection was performed to create pyramid flaps, with the two lateral islands of tissue still attached to the upper part of the lip, while being swung together as pendulums to meet in the prolabium. A 4-0 fine silk line was used to suture the layers (Figs. 1A and 2).

#### Modified Abbe Flap for Repair of Grade II Whistling Deformities

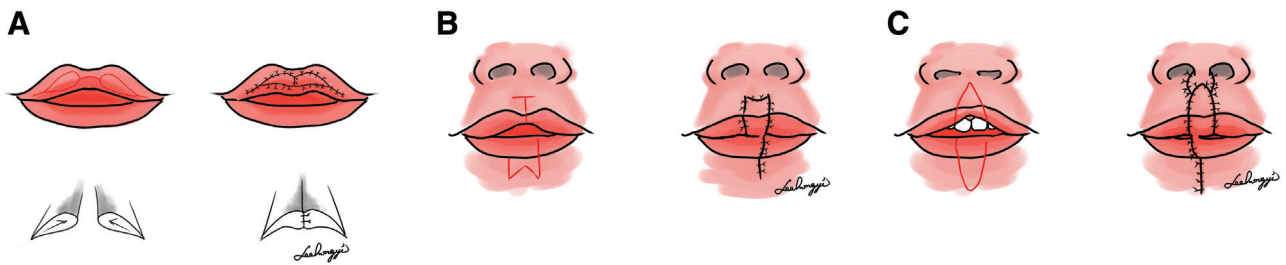
A modified Abbe flap was performed in the repair of grade II whistling deformities. With this technique, the pocket was creased for the vermilion tubercle by splitting the lip. The skin was incised several millimeters beyond the vermilion-skin border and was moved laterally for a distance equal to the breadth of the vermilion tubercle. The orbicularis oris muscle and the full length of the upper lip vermilion were incised. By releasing the tension, the modified Abbe flap that was taken from the central portion of the lower lip vermilion was designed to repair the vermilion tubercle and the Cupid's bow. The flap was approximately 8mm in width. The full length of the lower lip vermilion and the orbicularis oris muscle were incorporated. A thin portion of skin was included, which facilitated closure of the donor site. The flap, based on the labial vessels, was rotated 180° and sutured into the created defect of the upper lip. The pedicle was divided 10 days after surgery (Figs. 1B and 3).

#### Abbe Flap for Repair of Grade III and Grade IV Whistling Deformities

The shape of the defect depended on how far upward the lip had to be split to produce a normal appearance. It may be necessary to split the lip as far as the columellar base, creating a defective shape of an inverted V.

**Table 1. Whistling Deformity Grades**

Grade	Criteria
Grade I	The crowns of the left and the right upper incisors are exposed above the first and second incisor
Grade II	Exposure of the crowns of the upper incisors and/or the crowns of the medial sagittal portion of the left and the right upper lateral incisors
Grade III	Exposure of the crowns of the two upper incisors and half of the gums over the two upper incisors, or the crowns and half of the gums over the two upper incisors and more than two-thirds of the transverse crowns of the lateral incisors
Grade IV	Exposure of the crowns of the upper incisors and their attached gums and over two-thirds of medial sagittal crowns of the upper lateral incisors



**Fig. 1.** Diagram showing different whistling deformity repair methods. A, Double movable mucomuscular complex flaps repair. B, Modified Abbe flap transfer method. C, Abbe flap transfer method.

When the defect had been fully displayed, the flap was drawn up to the lower lip, and the flap was symmetrical on either side of the midline of the lower lip. The flap, based on the labial vessels, was rotated 180° and sutured into the created defect of the upper lip. The pedicle was divided 14 days after surgery (Fig. 1C).

### RESULTS

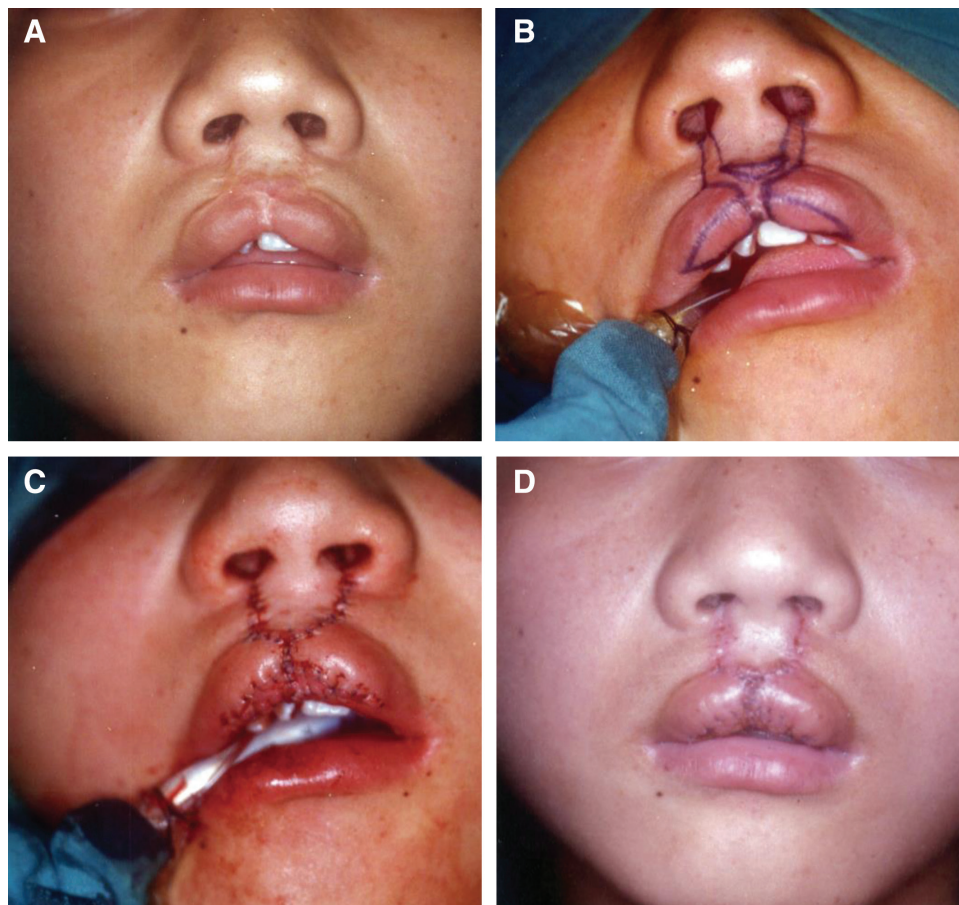
Of the 136 patients included in this study, 98 were men (72.0 %) and 38 were women (28.0 %). Their ages ranged from 9 to 34 years, and the average age was 15.6 years.

Among the total patients, 60 (44.2%) were classified as having a grade I whistling deformity, 47 patients (34.5%) had

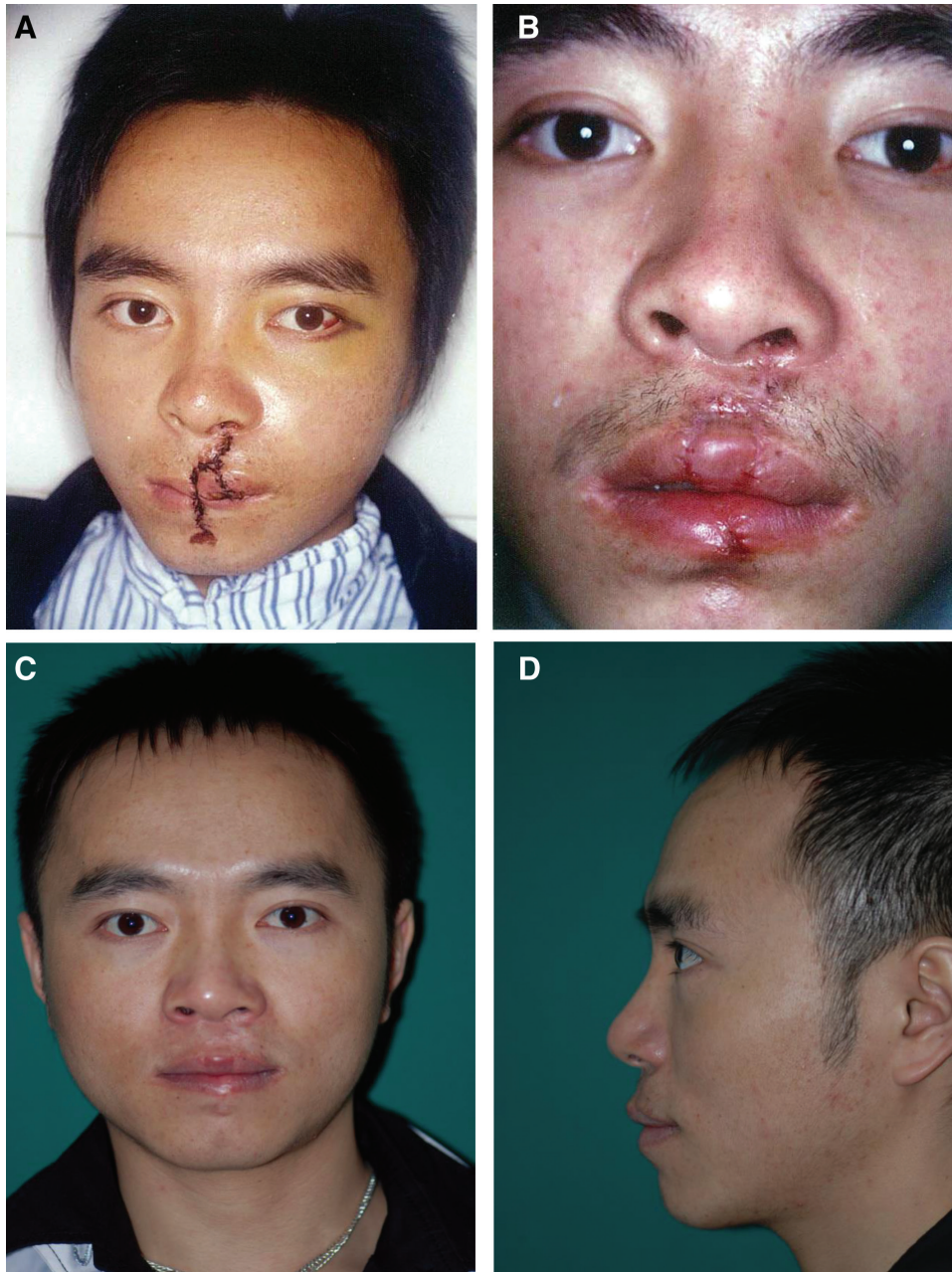
a grade II whistling deformity, 16 patients (11.8%) had a grade III deformity, and 13 patients (9.5%) had a grade IV whistling deformity. The results showed that grade I and grade II whistling deformities (107 cases, 78.7%) occurred significantly more frequently (about 3.7 times) than grade III and grade IV whistling deformities (29 cases, 21.3%) (Table 2).

Sixty patients (44.1%) with a grade I whistling deformity underwent the double movable mucomuscular complex flaps procedure, 47 patients (34.6%) underwent the modified Abbe flap transfer procedure, and 29 patients (21.3%) underwent the Abbe flap transfer procedure (Table 3).

Postoperative follow-up occurred from 6 months to 28 years. All patients showed an obvious median tubercle, and



**Fig. 2.** Effects after repair using the double movable mucomuscular complex flap technique. A, Preoperative frontal view. B, Frontal view after incisive design. C, Postoperative frontal view. D, Frontal view of the postoperative effect.



**Fig. 3.** Effects after repair using the modified Abbe flap transfer technique. A, Frontal view after modified Abbe flap transfer. B, Frontal local view after modified Abbe flap transfer. C, Frontal view, 5 years after surgery. D, Lateral view, 5 years after surgery.

the height and width of the upper lip, Cupid's bow, and Cupid's bow peak were within the normal range. Scarring of the base of the nose was not obvious (Fig. 3C, D). The complications of bleeding, infection, or dehiscence did not occur in any of our cases. All patients developed mild to moderate postoperative edema of the upper lip, with 29 cases (21.3%) developing conspicuous scars. Other cases showed inconspicuous scars.

### DISCUSSION

The whistle deformity is defined as a central vermilion notching due to complex causes.<sup>6</sup> The typical clinical

findings of the whistle deformity include upper lip notching, bulging on the lateral lip segment, volume deficiency, and a defect of the upper lip.<sup>7</sup>

This deformity may occur after a unilateral cleft lip repair or unilateral cleft lip and cleft palate repair and may also occur after bilateral cleft lip repair and bilateral cleft lip and cleft palate repair. The main cause of a whistling deformity<sup>8</sup> is the tissue deficiency of the orbicularis oris muscle fibers in the prolabium. The vertical shortage of the upper lip in the mid-line, especially when the prolabium is small, can also lead to a whistle deformity after a bilateral cleft lip repair (Fig. 1).<sup>9</sup>

**Table 2. Different Grades of the Whistling Deformities of the Upper Lip (n = 136)**

Classifications	Male (%)	Female (%)	Total (%)
Grade I	46 (33.8)	14 (10.3)	60 (44.2)
Grade II	38 (28.0)	9 (6.6)	47 (34.5)
Grade III	9 (6.6)	7 (5.2)	16 (11.8)
Grade IV	5 (3.6)	8 (5.9)	13 (9.5)
Total	98 (72.0)	38 (28.0)	136 (100.0)

**Table 3. Selection and Contribution of Different Surgical Techniques of Different Whistling Deformities (n = 136)**

Classifications	Double Movable Mucomuscular Complex Flap (%)		Abbe Flap (%)
	Modified Abbe Flap (%)	Abbe Flap (%)	
Grade I	60 (44.1)	0	0
Grade II	0	45 (33.1)	2 (1.5)
Grade III	0	2 (1.5)	14 (10.3)
Grade IV	0	0	13 (9.5)
Total	60 (44.1)	47 (34.6)	29 (21.3)

Currently, preoperative objective evaluation of the extent of the whistling deformity is difficult due to a lack in supporting literature. We divided the whistling deformities into 4 different grades according to the degree of exposure of the gums and the crown of the anterior upper incisors and upper lateral incisors. The deformities were then repaired based on this grading using the double movable mucomuscular complex flap,<sup>10</sup> modified Abbe flap,<sup>11</sup> or Abbe flap.

For repair of a small vermilion deficiency, local flaps such as the Kapetansky flap,<sup>12</sup> mucosal transposition flap,<sup>13</sup> rectangular mucosal flap,<sup>14</sup> vermilion double V-Y advancement, and mucosal V-Y advancement<sup>15</sup> can be used. According to our classification, we selected the double movable mucomuscular complex flap<sup>12</sup> for the treatment of grade I whistle deformities or upper lip notching and volume deficiency. With this surgical method, the orbicularis oris muscle, which bundles on both sides of the whistle deformity, was dissected from the subcutaneous tissue and freed up to the nasolabial line. By lateral mobilization and midline suturing of the corresponding muscle segments, the continuity of the orbicularis oris muscle and median tubercle was restored. This surgical technique has advantages such as less tissue injury, good blood supply, less scarring, and infrequent relapse.

For repair of a defect of the medium tubercle or grade II whistling deformity, a modified Abbe flap<sup>11</sup> was used to reconstruct the medium tubercle of the upper lip and partial upper lip skin. In this series, we demonstrated that this procedure may also be used to treat minor grade III whistle deformities after the primary repair of a bilateral cleft lip. This method is safe, useful, and effective to correct whistle deformities with central deficiency.

The Abbe flap repair can be used to treat grade III and grade IV whistling deformities. This is because a large vermilion defect requires tissue transfer, such as island flaps from the lower lip or an Abbe flap. The Abbe flap is a musculocutaneous flap from the central portion of the lower lip, which can be used for the correction of middle defects of the upper lip and Cupid’s bow in cleft lip patients; thus, the transposition of full thickness parts of the lower lip is still a common procedure used for building up the missing muscle in the prolabium in bilateral clefts.

## CONCLUSIONS

In this study, the whistle deformities were divided into 4 different grades; different surgical techniques may be selected to repair different grades of whistle deformities in patients with unilateral cleft lip or unilateral cleft lip and palate and bilateral cleft lip or bilateral cleft lip and palate. We reviewed and analyzed 3 effective techniques for repairing the different whistle deformities. The different repair methods chosen to repair the different grades of whistling deformities are scientific methods that have an important clinical impact.

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