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The Melbourne Classification of the Complete Unilateral Cleft Lip Based on Hypoplasia

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Background: The hypoplastic lateral lip element within the cleft lip presentation is a recognized entity that has been recently shown to be more common on the right side. The spectrum of such change is yet to be defined. The authors propose the Melbourne classification system of cleft lip hypoplasia and see it as an important step towards discerning the relevance of these anatomical observations to the management of cleft lip/palate patients.

Methods: This is a retrospective observational study of patients with complete unilateral cleft lips treated by the senior author (DKC) at the Royal Children's Hospital, Melbourne. Patient charts were retrospectively reviewed and patients were classified into different degrees of hypoplasia based on preoperative, intraoperative, and postoperative photography. Data was reported using descriptive statistics.

Results: Fifty-nine patients with complete unilateral cleft lip deformity were grouped according to lateral lip element hypoplasticity. Twenty patients had right-sided clefts and 39 patients had cleft lips on the left side. Of those with right-sided clefts, 18 patients had evidence of hypoplasia (90%). Three patients had Type 1 deformities, 3 patients were Type 2, and 12 patients were Type 3. Patients with left-sided clefts were found to have hypoplasia less frequently with 15 patients showing evidence (38.5%).

Conclusions: The authors report a classification system of hypoplasia involving the lateral lip element in complete unilateral cleft lip. The authors propose this classification system as a new measure of cleft severity that will have implications for patient expectations, surgical planning, and future outcome studies.

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Received August 30, 2021.

Accepted for publication September 26, 2021.

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This work has not been previously presented at a public forum.

The authors report no conflicts of interest.

Supplemental digital contents are available for this article. Direct URL citations appear in the printed text and are provided in the HTML and PDF versions of this article on the journal's Web site (www.jcraniofacialsurgery.com).

ISSN: 1049-2275

DOI: 10.1097/SCS.0000000000008277

Key Words: Classification, hypoplasia, sidedness, unilateral cleft lip

(*J Craniofac Surg* 2022;33: 380–384)

Hypoplasia of the lateral lip element within the cleft lip deformity is a recognized entity.[1–6] The hypoplastic lateral lip element has been shown to be more common with right-sided cleft lips, but the spectrum of such hypoplasia is yet to be defined.[2]

The emphasis of planning in unilateral cleft lip repair has been directed towards the medial lip element and the various designs required to balance the Cupid's bow.[7–10] Minimal attention has been afforded to incisional designs for the lateral lip element before the paper by Fisher.[9] Equally, an approach to addressing the spectrum of hypoplasia of cleft lips is missing.

The aim of this paper is to propose a descriptive system for the range of hypoplasia seen in the lateral lip element including the nasal construct. Encouraged by our recent anthropometric study, we test our findings with a classification for complete unilateral left and right-sided clefts.[2] Our goal is to facilitate the cleft surgeon to perceive the lateral lip element in its various presentations.

METHODS

This is a retrospective observational study of patients with complete unilateral cleft lips treated at the Royal Children's Hospital by the senior author (DKC). These observations are based on the senior author's experience treating cleft lip and palates at the Royal Children's Hospital from 2008 to 2021 and augmented by his experience from over 40 international cleft missions. A classification was proposed, and then patient charts were retrospectively reviewed by the authors to classify patients based on preoperative, intraoperative, and postoperative photography. Patients were considered to have adequate photography if they had preoperative photos taken after 2 months of age, had immediate perioperative photos on table including anterior view and worm's eye view, and at least a 1-year postoperative photograph. Patients who were animating in their preoperative and postoperative photographs were excluded as this prevented accurate assessment of alar or vermilion thickness.

All patients included in this series had unilateral complete cleft lips. Patients were excluded if they had bilateral cleft lip, incomplete cleft lip, Simonart's band, presence of a syndrome, or if there was inadequate photographic evidence to assess lip or nose hypoplasia. Patient records were retrospectively reviewed for: age at time of surgery, gender, type of cleft lip, side of cleft lip deformity, and presence of a syndrome. Data was reported using descriptive statistics and the observations made by the authors were classified.

This study was approved by the Ethics Committee at the Royal Children's Hospital, Melbourne (HR32690). Patient consent for photography was obtained.

RESULTS

Patient Demographics

Over the study period, a total of 67 patients with complete unilateral cleft lip deformities underwent cleft lip repair by the

senior author at the Royal Children's Hospital, Melbourne. Of these 67 patients, 5 patients had a cleft lip as part of a syndrome and 3 patients had incomplete photography for evaluation purposes and were excluded. Fifty-nine patients with complete unilateral cleft lip deformity met inclusion criteria for the study. Our study population consisted of 23 females and 36 males. There were 20 patients with right-sided complete cleft lip deformities, and 39 patients with cleft lips on the left side.

The Melbourne Classification of the Hypoplastic Cleft Lip

The proposed classification can be divided into 3 anatomical components. Our system is based on assessment of the height of the skin lip, the volume of the red lip on the cleft side (vermilion and mucosa) compared to the non-cleft side, and the thickness of the alar base and nostril sidewall on the cleft side compared to the non-cleft side.

The classification system can be thought of as generally additive, meaning that a Type 2 lip has the features of a Type 1 lip, and a Type 3 lip has the components of a Type 1 and 2 lip. Type 1 is the least severe and Type 3 represents the most severe form of cleft lip hypoplasia.

The Type 1 deformity is schematically depicted in Figure 1A. Type 1 hypoplastic cleft lip is characterized by a short vertical height of the skin lip compared to the non-cleft side, as measured from the peak of the Cupid's bow to the alar base on the non-cleft side and Noordhoff's point to the alar base on the cleft side. Visual clues include the observation that Type 1 hypoplastic lips are more vertically oriented in reference to the horizontal plane of the lip (Fig. 1A). Figure 1B shows representative photos of a patient with a Type 1 cleft lip deformity. Here, the skin lip is deficient in height, and the lateral lip element is oriented vertically in relation to the horizontal plane of the lip. The volume of the red lip and the alar thickness on the cleft side are not obviously hypoplastic relative to the non-cleft side.

The Type 2 hypoplastic cleft lip is illustrated in Figure 1C. In addition to the findings described in the Type 1 cleft lip deformity, Type 2 lips are characterized by hypoplasticity of the red lip. The overall bulk of the red lip of the lateral lip element is reduced compared to the same region on the non-cleft side. Visual clues include observation of the vermilion just lateral to the oral commissure and comparing it to the opposite side. Representative photos of a patient with a Type 2 cleft lip deformity are shown in Figure 1D. This patient has a short skin lip as described for a Type 1 deformity, and the observed hypoplasia of the vermilion and mucosa on the cleft side relative to the non-cleft side.

Figure 1E demonstrates the Type 3 hypoplastic cleft lip deformity. On top of the findings described for the Type 1 and 2 hypoplastic lips, patients with Type 3 deformities also have a hypoplastic ala observed on the cleft side. This is best appreciated on the worm's eye view comparing the alar base and side wall to the opposite side. Figure 1F shows representative photos of a patient with a Type 3 cleft lip deformity. This patient has the findings of a Type 1 and 2 cleft lip, but also has the hypoplastic ala characteristic of a Type 3 deformity. The stigmata of hypoplasia is also noticed to persist postoperatively. Figure 2 shows representative photos of an early postoperative result of a 10-month-old boy with a Type 3 deformity who is 2 months post cleft lip repair; asymmetry in nasal alar thickness and vermillion bulk on the cleft side remain a feature.

Supplementary Digital Content, Table 1, http://links.lww.com/SCS/D430 shows the distribution of cleft lip hypoplasia based on the Melbourne classification in our study population. Of the 20 patients with right-sided cleft lip deformities, 2 patients had no evidence of hypoplasia, 3 patients had Type 1 deformities, 3 patients were Type 2, and 12 patients were Type 3. Vermilion hypoplasticity (Types 2 and 3) was, therefore, seen in 15/20 patients (75%) and some form of alar hypoplasticity (Type 3) was seen in 12/20 patients (60%).

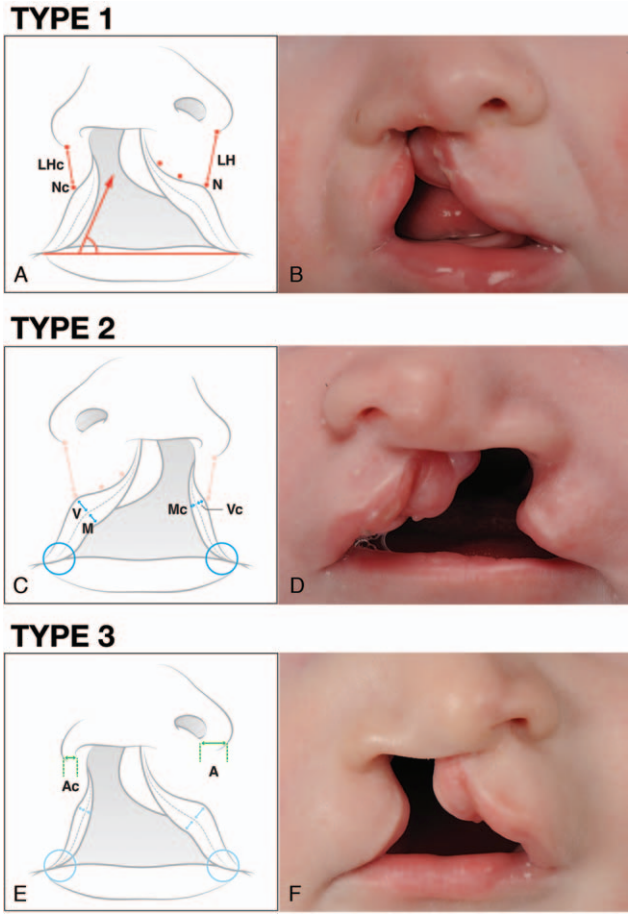

FIGURE 1. The Type 1 deformity. (A) Schematic showing the Type 1 deformity. Vertical lip height on the cleft side (LHc) as measured from the alar base to Noordoff's point (N) is shorter than that on the non-cleft side (LH). Illustration also depicts the more vertically oriented lateral lip element in reference to the horizontal plane of the lip. (B) Representative patient with a Type 1 cleft lip deformity. **The Type 2 deformity**. (C) Schematic showing the Type 2 deformity. Hypoplasticity of the red lip is seen in both the vermilion (Vc) and the mucosa (Mc) on the cleft side as compared to the non-cleft vermilion (V) and mucosa (M). This deformity is often apparent when observing the vermilion near the oral commissure. (D) Representative patient with a Type 2 cleft lip deformity. **The Type 3 deformity**. (E) Schematic showing the Type 3 deformity. Hypoplasticity of the ala is present on the cleft side (Ac) as compared to the non-cleft nasal ala (A). (F) Representative patient with a Type 3 cleft lip deformity.

Hypoplasia was less frequent in patients with left-sided cleft lips, as 24 patients (61.5%) did not have evidence of hypoplasia of the lip or nose. As shown in Supplementary Digital Content, Table 1, http://links.lww.com/SCS/D430, 2 patients had Type 1 deformities, 6 patients were Type 2, and 5 patients were Type 3. There were 2 additional patients that did not fit the classification system as they only had evidence of nasal ala hypoplasia. Obvious vermilion hypoplasticity was, therefore, seen in 11/39 patients (28.2%) and alar hypoplasticity in 7/39 patients (17.9%).

DISCUSSION

Lateral lip hypoplasia as part of the cleft lip deformity has been identified, both on a histological and anatomical basis.[1–6] Little

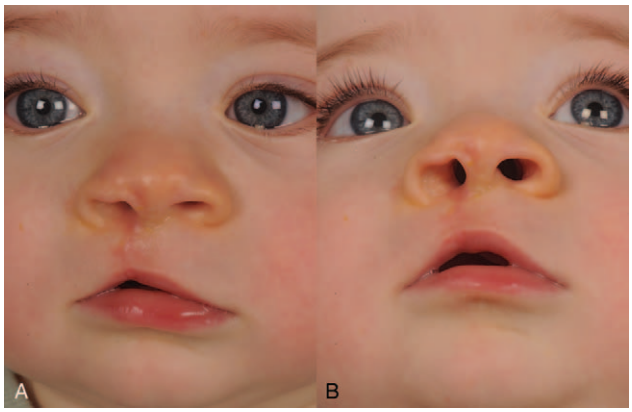

FIGURE 2. Type 3 deformity early postoperative result. (A-B) Postoperative photographs of a 10-month-old boy with a Type 3 cleft lip deformity 2 months after repair demonstrating persistence of vermillion hypoplasia and hypoplastic alar rim on the cleft side.

attention has been given to the spectrum of severity of the lateral lip element. Most surgical techniques emphasize the levelling of the Cupid's bow of the medial lip element and the various incisional designs to "rotate" the medial lip down.[7,8] Less attention has been given to the incisional design of the lateral lip element and the varying effect of the extent of hypoplasia.

We propose the Melbourne classification of hypoplasticity for the cleft lip deformity to assist in the evaluation of the unilateral cleft lip. The Type 1 short lip presentation has been noted in the literature previously.[9] The challenge of a Type 1 lip lies with recognizing its presence and planning the lateral element incision accordingly.

The Type 2 deformity is characterized by the decrease in overall bulk of the vermilion and mucosa on the cleft side. This has been further described in a recent anthropometric study.[2] Follow-up postoperative photographs, particularly the worm's eye view, demonstrate the persistence of the hypoplastic vermilion compared to the non-cleft side (Fig. 3). This finding may be less evident in the immediate postoperative period due to surgical swelling or local anesthetic infiltration.

The Type 3 deformity represents the most severe hypoplastic entity. The nasal observations warrant further discussion. Although

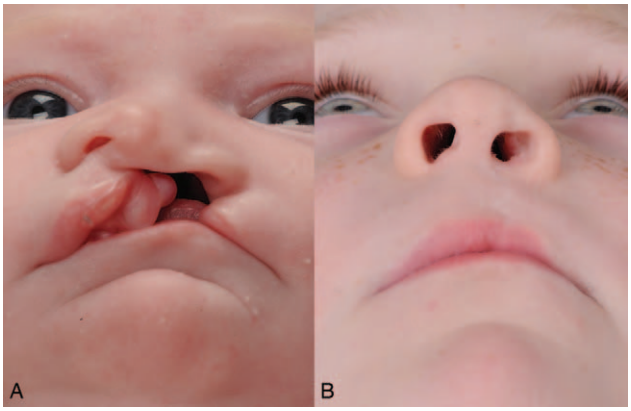

FIGURE 3. Type 2 deformity long-term postoperative result. (A) Preoperative photographs of a patient with a Type 2 cleft lip deformity at 4 months of age. (B) Postoperative photographs of the same patient 6 years after repair demonstrating persistence of a hypoplastic vermilion and normal alar thickness.

FIGURE 4. Type 3 deformity long-term postoperative result. (A) Preoperative photographs of a 4-month-old girl with a Type 3 cleft lip deformity. (B-D) Postoperative photographs of the same patient 2 months (B), 3 years (C), and 9 years (D) after repair demonstrating persistence of the hypoplastic vermilion and a hypoplastic alar rim, with evolving ramifications on facial symmetry preoperatively (A), postoperatively (B), at short-term follow-up (C), and at long-term follow-up (D).

it is possible that the ala is "thinner" as the sequelae of a stretching process from a wider cleft, the observation of hypoplastic alae without elongation in some patients, would suggest it is likely a separate process rather than a secondary phenomenon. This is further supported by patients like the Type 2 cleft seen in Figure 3. If a thin ala were the result of a stretching process, a patient with a wide Type 2 cleft would also present with a "thinned out" ala. As evidenced in Figure 3, there are wide clefts observed where the thickness of the ala remains equivalent to the other side. We postulate it is the hypoplasticity of the ala itself, which is the primary event.

Type 3 deformities may have increased potential for adverse aesthetic and functional outcomes. One such consequence is satisfaction in facial appearance. Regardless of the prowess of the operating surgeon, the likelihood of persisting asymmetry is greater in these more severe presentations. Figure 4 demonstrates a right-sided Type 3 cleft lip patient. As shown through the series of follow-up photographs, the hypoplasticity of the ala and red lip can be recognized preoperatively and in the early postoperative result, with evolving ramifications on facial balance as the child ages.

The identification of hypoplasia reinforces the concept that not all clefts are the same. Traditionally the difficulty of a cleft has been related to the width of the cleft. We propose that a better indicator of severity is the extent of lateral element hypoplasia. The extent of involvement of the various structures discussed has importance preoperatively, intraoperatively, and postoperatively.

Preoperatively, identifying the extent of hypoplasia will guide parental counselling and surgical planning. Observations of the

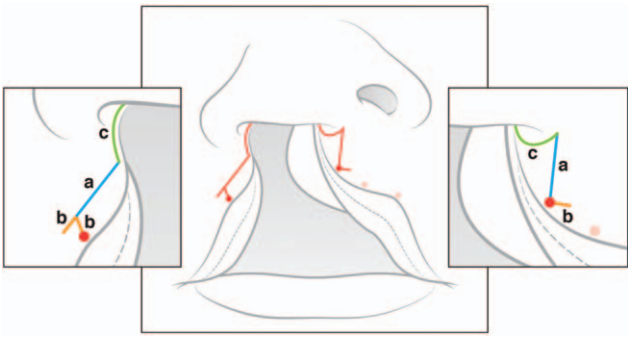

FIGURE 5. Type 1 deformity solution. Schematic showing the solution to the Type 1 deformity as originally described by Fisher[9] and modified by the senior author. The triangle (B), which represents the lesser height of the lip is designed within the greater height of the lip (A). Color coding on the diagram and letters represent corresponding points of closure.

various structures before surgery will assist with expectations of the postoperative result as well as the expected difficulty of surgery.

Intraoperatively, the extent of hypoplasia will have further considerations. The Type 1 deformity will need to incorporate an incisional design that recognizes the reduced lip height as described by Fisher.[9] Careful geometric evaluation will ensure that the correct size triangle is drawn within the constraints of the reduced height of the lip (Fig. 5).

The Type 2 deformity provides the surgical challenge of reduced vermilion volume on the cleft side. It will aid surgical planning to ascertain the difficulty to achieve a symmetrically distanced commissure-to-Cupid's bow point compared to the non-cleft side, due to the reduced size of the vermilion. A further surgical challenge will be the balanced union between the thinner cleft vermilion and the fuller medial lip vermilion.

Type 3 differences are the most surgically challenging. As well as the challenges already discussed, the hypoplastic ala is more likely to result in buckling and asymmetry of the nose post repair. Inserting primary cartilage grafts has been described,[11] but we are yet to elucidate a satisfactory surgical solution at primary repair. The identification of the hypoplastic ala may be more to temper surgeon expectations.

Postoperatively, the groupings may assist in evaluation of outcomes. Research endeavors will be precipitated by the improved stratification of severity. Reporting on the long-term effects of hypoplasia will become more evident if the surgeon is aware of its presence.

The functional repercussions of lateral element hypoplasia also warrant further investigation. The extent of tissues involved may include skin, fat, muscle, cartilage, teeth, and bone. We are intrigued with the link between cleft lip hypoplasia and the presence of dental anomalies and maxillary growth restriction.[3–5] Antonarakis and Fisher[3] have retrospectively shown that children with a short vertical lip height are more likely to present with lateral incisor agenesis on the cleft side. Further retrospective studies from the same group have shown an association between lateral lip height deficiency and maxillary growth restriction.[4,5] As such, the functional consequences of a shorter lateral lip height on maxillary growth and dental relationships already holds merit. The stratification of varying extents of hypoplasia on maxillary growth and dentition may further define this relationship. Associations between lateral element hypoplasia and palatal morphology or speech outcomes are yet to be explored.

With applying the Melbourne classification to our patient population, we also confirm that the incidence of hypoplasia appears much higher on the right side.[2] This adds to the body of evidence that suggest that right-sided clefts may present as a distinct entity with real "physiognomic asymmetries" and warrants further investigation into the aetiology of sidedness and its phenotypic effect.[12,13]

Previous studies have noted the influence of laterality of clefts on facial appearance. Feragen et al[12] published findings of 160 patients with cleft lips who were rated for facial disfigurement and found that right-sided cleft patients were consistently judged to be more affected than their left-sided counterparts. This observation was confirmed even when facial photos were converted to their mirror image to blind the observer to sidedness. They concluded that the influence of cleft laterality on facial disfigurement was real rather than biased by the perception of the rater.

Bella et al[14] had 76 images of cleft patients reviewed by 29 UK-based surgeons and found a similar preference for the raters to rank the facial appearance of right-sided cleft patients more poorly. Asymmetry measured via a computer program, where the same facial images were reflected on themselves, did not reveal a laterality difference, leading the authors to conclude there was no objective evidence to support the surgeon's preference for left-sided repairs. They conclude that the phenomenon could be a result of perceptual bias rather than an inherent true difference. However, the objective 2D analysis technique used by Bella et al[14] would not have perceived the hypoplasia we postulate is responsible for the judgements observed by the human raters.

This study has several limitations. The classification is based on the senior author's observations. The anthropometric measurements of the cleft lip have been previously published on a separate dataset of patients and confirmed measurements supporting the classification of Types 1 and 2 deformities.[1–5] The nasal contribution to the classification is based on observation alone and not by objective measure. We recognize the potential for observer bias and more objective investigations are required to confirm this aspect of our classification and to help qualify "obvious" hypoplasia. We believe the role of 3D photography will help to refine our observations further. Challenges to this goal include the use of 3D photography in infants, the confounding factor of facial animation, and the difficulty of objectively measuring hypoplasia.

The Melbourne classification is applicable to left and right-sided complete clefts, with an increased incidence of severe forms of hypoplasia (Type 3) noted on the right side. By proposing a new classification system, we suggest that the measure of cleft severity may be more accurately reflected by extent of tissue hypoplasia rather than from cleft width alone. This has implications for parental expectations, surgical planning, as well as future outcome studies. We hope that this will encourage other units to explore the observations of hypoplasticity in their patient cohort, with investigation of their long-term ramifications.

ACKNOWLEDGMENTS

The authors thanks Mr. Bill Reid for his detailed illustrations and Mr. David McCombe for his guidance in developing this classification.

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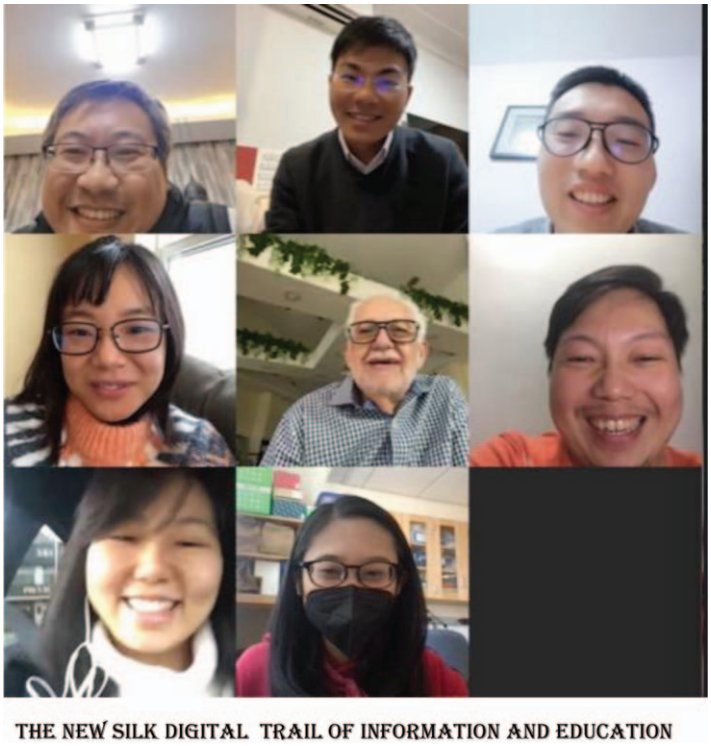

The China virtual craniofacial conference.