Bilateral Cleft Lip Repair: Lessons from History

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Learning Objectives: After studying this article, the participant should be able to: 1. Describe the bilateral cleft lip and nasal deformity and associated anatomical variations. 2. Understand the history and evolution of the bilateral cleft lip repair. 3. Understand the key principles of the surgical repair.

Summary: This article describes characteristics of the bilateral cleft lip and nasal deformity and its management, including presurgical orthopedics, operative techniques, and postsurgical care. (*Plast. Reconstr. Surg.* 150: 201e, 2022.)

…a baby born with a congenital bilateral cleft… has…the clefts gaping between him and his share of happiness. This has challenged thousands of surgeons over many centuries to surpass the feats of previous surgeons in the evolution of cleft craft.

—D. Ralph Millard, Jr.

The incidence of orofacial cleft varies depending on geography and ethnicity, ranging from 0.1 to 2.3 per 1000 live births.[1] Parts of Asia and Latin America demonstrate higher rates, and parts of Africa and Europe demonstrate lower rates.[2] Within the United States, Asian and Native American populations demonstrate a higher incidence, one in 450 births, whereas White people are affected one in 1000 births and African Americans one in 2000.[1] Comparing to unilateral cleft lip, bilateral cleft lip is notably less common, with a bilateral-to–right unilateral–to–left unilateral cleft ratio of 1:3:6 ratio.[3] Cleft lip is associated with other birth defects 30 percent of the time.[4–7] Approximately 20 percent of the nonsyndromic cases are familial in inheritance, and 80 percent are sporadic.[8]

Cleft lip with or without cleft palate has been linked to more than 200 specific genetic syndromes,[2,9] the most notable of which include van der Woude (*IRF6*),[10–13] Kallmann (*FGFR1*),[14] X-linked cleft and ankyloglossia (*TBX22*),[15] and Gorlin (*PTCH1*)[16] syndromes, which typically follow mendelian inheritance. Mutations linked to orofacial cleft encompass a wide variety, from transcription factors (*IRF6*, *TBX22*, and *MSX1*), growth factors (*TGFA* and *TGFB3*), and metabolic factors (*CYP1A1* and *MTHFR*), to immune modulators (*IRF6* and *PVRL1*).[2] Many of these genetic findings also have close environmental interactions. Methylenetetrahydrofolate reductase is a key enzyme in folic acid metabolism, and mutations are found at a higher frequency in the setting of maternal hyperhomocysteinemia, leading to syndromic cleft lip and palate.[17] *MSX1*, a muscle segment homeobox gene on chromosome 4, has been associated with nonsyndromic cleft lip[18] in the setting of prenatal maternal alcohol consumption and cigarette smoking.[19,20] Other interactions under investigations include *TGFA* with smoking[21,22] and vitamin supplementation,[23] *TGFB3* with smoking and alcohol,[20,24] occupational exposure,[25] and maternal medication exposure.[26] None of the studies are currently conclusive.

Bilateral complete cleft lip, involving the lip, nasal sill, and nasal floor (Fig. 1), occurs when the medial nasal prominence failed to fuse with the maxillary prominence bilaterally, which normally occurs between the fourth and seventh weeks of gestation.[2,3] This process occurs by an intricate series of interactions between neural crest cells and mesenchymal tissue, which modulate formation and fusion of the prominences.[2] Just before completion, there is a peak in cellular division, thereby increasing susceptibility to teratogenic insults, leading to failure of the fusion mechanism.[2] Variation in severity and/or timing of the insult may explain the variations in the deformity. The complete cleft demonstrates no orbicularis oris muscle in the prolabium, which may suggest a failure of muscle ingrowth toward the midline because of the bony discontinuity.[27]

From Texas Children's Hospital, Baylor College of Medicine.
Received for publication February 2, 2021; accepted March 4, 2022.

DOI: 10.1097/PRS.0000000000009241

Disclosure: *Dr. Hollier serves as Chair of the Global Medical Advisory Board for Smile Train.*

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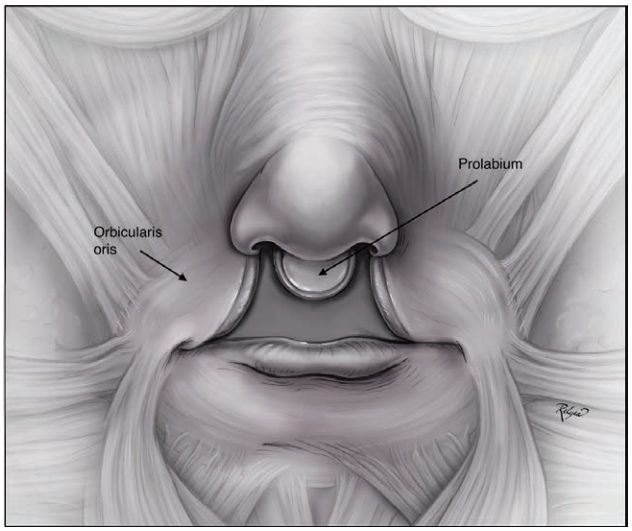

Fig. 1. Bilateral complete cleft lip. Note the discontinuity of orbicularis oris muscle across the cleft, and that the prolabium does not contain muscle.

The bilateral incomplete cleft lip, in contrast, has intact nasal sills, and may demonstrate muscle continuity, although it is frequently atrophic. Minor form, microform, or "forme fruste" applies to less severe presentations, frequently manifest as vermilion notching. A notch greater than 3 mm is classified as minor form, which is accompanied often by a vertical depression extending from the notch to the nasal sill.[1,3,28] Alternatively, a notch less than 3 mm in height is considered microform. For bilateral cleft, any combination of the above subtypes can occur, each with variable degrees of nasal deformity and asymmetry.

BILATERAL CLEFT LIP DEFORMITY AND REPAIR

The bilateral cleft lip deformity has an entirely different configuration from the unilateral cleft, and the evolution of its repair is demonstrative of the recognition of its unique challenges. The hallmark of the complete unilateral cleft lip is the asymmetry of the lip, nasal position, and inherent nasal structures, deriving from the asymmetric growth of the premaxilla, maxilla, and orbicularis oris muscle. The unilateral cleft repair focuses on correction of these anatomical asymmetries. The hallmark of the bilateral complete cleft, in contrast, is a premaxilla that is unattached to the palatal shelves and maxilla, and as a result advances unrestrained during development. Frequently, it is also rotated outwardly, well beyond the level of the alveolar arch. Latham postulated that in normal early development, the nasal septum serves as a key determinant of midfacial growth, drawing the maxilla forward as it grows.[29] The failure of fusion between the premaxilla and maxilla leads to a growth discrepancy. Millard wrote that the cause is most likely "an interrelation of many—lack of bony continuity, growth at the [vomeropremaxillary] suture, cleft of the orbicularis oris muscle, forward growth of the cartilaginous septum and expansion of the alveolar process."[27] The result of this unchecked forward growth leads to a prolabium that appears to attach almost directly to the nasal tip with a diminutive and sometimes missing columella.

Hypotheses regarding the embryologic cause of the columellar deformity are similarly varied. Latham and Workman described the unchecked abnormal growth of the premaxilla, leading to the lack of differential development between the septum and the premaxilla, as a cause of columellar loss.[27] McComb later postulated that the retroposition and lateral displacement of the lower lateral cartilages result in flattening of the nasal tip and shortening of the columella.[30,31] The reality is likely a combination of several different factors.

The treatment of the bilateral cleft lip evolved to address three specific concerns: (1) how to use the prolabium, (2) how to recreate the missing columella, and (3) how to manage the protruding premaxilla. As described previously, the actual cleft presentation may vary widely, depending on the severity of the deformity. The recognition of the deformity and its variations is a key driving force for the evolution of surgical techniques.

Prolabium

The prolabium (Fig. 1) challenge has been a multifaceted one. Early surgeons recognized the variation in prolabial size, its often far and asymmetric distance from the lateral lip elements, and its potential paucity of blood supply (Fig. 2) and growth capacity. As a result, many opted for a staged repair, particularly in those cases with marked asymmetry. The aim was to convert a bilateral cleft into a unilateral cleft. Proponents included Veau, Cronin, Bauer, Trussler and Tondra, Slaughter, Henry, and Berger, and Monroe.[27] Millard quoted Slaughter, Henry, and Berger over the concern about blood supply and the need for a "revascularization of the area in a manner compatible with accepted plastic surgical procedures." By the time *Cleft Craft*[27] was published in 1976, the tide was already swinging toward the single-stage repair, because of concern for introducing further asymmetry and anatomical distortion.

Similar to the unilateral cleft, the technique of the bilateral repair underwent many variations

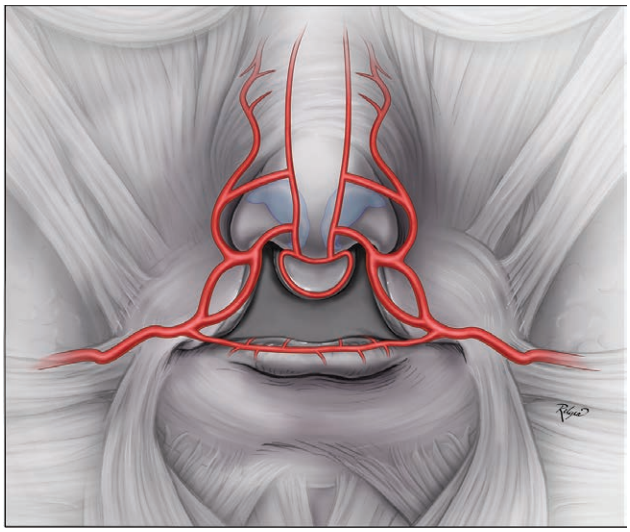

Fig. 2. Blood supply of complete cleft lip. Note the superiorly based nutrient arteries supply the prolabium, making possible full elevation of tissue during repair.

over the years and continues to build on the knowledge of the predecessors. This progression largely mirrored that of the unilateral cleft techniques, from the Rose/Thompson[3,32] straight-line repairs over a century ago, to techniques designed to lengthen the philtrum as in the repairs proposed by Randall-Tennison,[33] Le Mesurier and Hagedorn[34] using geometric flaps, and Z-plasty as designed by Berkeley and Bauer[35] and Skoog.[36] More modern variations are created based on the success and pitfalls of these techniques. Many of these repairs, including the popular Manchester repair,[37] which uses the prolabial vermilion to reconstruct the Cupid's bow and tubercle, did not restore the continuity of the orbicularis muscle, because of concern for excessive tension. Manchester repaired the muscle edges to the subcutaneous tissue instead.

Millard introduced his rotation-advancement technique in 1957, which served as a major paradigm shift to approaching the unilateral and bilateral cleft repair. The essence of the technique relies on rotation to correct asymmetry of the philtral complex, and advancement of the lateral lip tissue to fill defects created by the rotation. For the bilateral cleft, the prolabium was elevated completely to allow for restoration of muscle continuity across the premaxilla; the Cupid's bow white roll was created by bilateral advancement flaps from the lateral lip elements, and bilateral fork flaps created from lateral prolabial tissue were rotated superiorly and banked for secondary columellar lengthening. Millard wrote about the "destiny" of the prolabial tissue, which went from neglect and full excision, to complete use to recreate the central lip or the nasal columella, to his prolabial sharing for both lip and nose[27] (Fig. 3). Modern techniques, although derived directly from Millard's work, now rarely use the

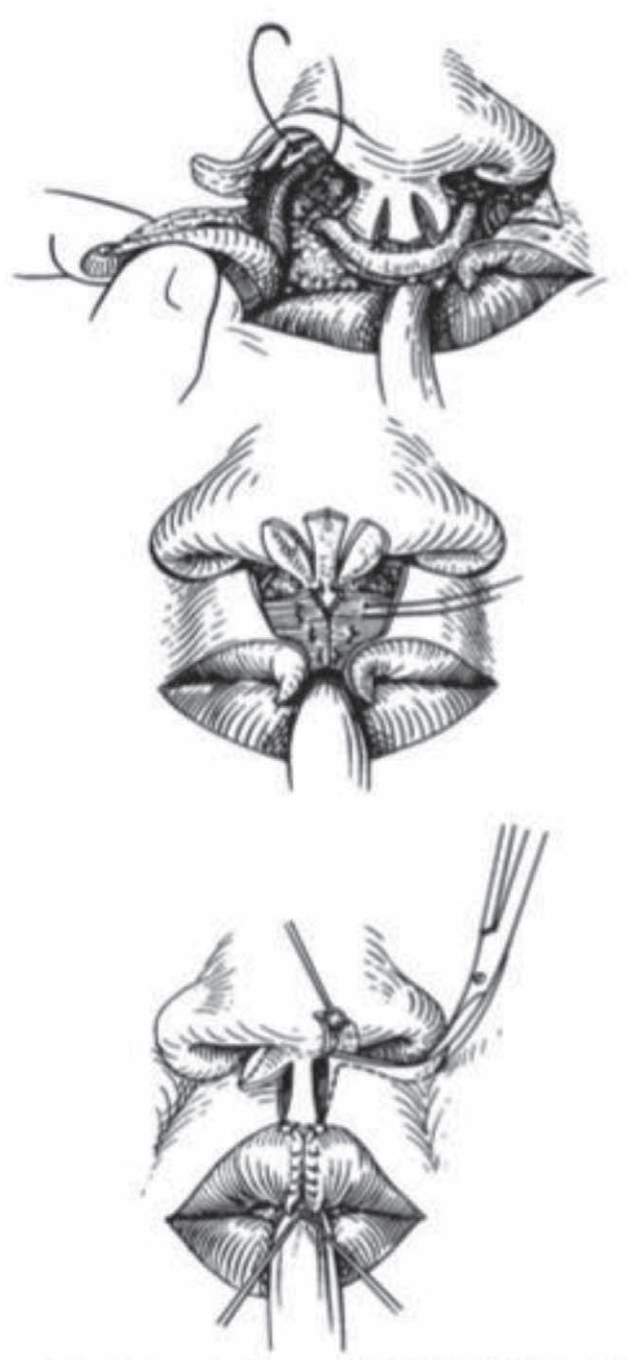

Fig. 3. Original 1985 primary lip repair with fork flaps banked at the columellar base for planned secondary columellar lengthening. (Reprint with permission from Mulliken JB. Principles and techniques of bilateral complete cleft lip repair. *Plast Reconstr Surg*. 1985;75:477–486.)

fork flap because of the resultant nonanatomical placement of the lower lateral cartilage footplates and the columellar scar burden, both leading to secondary distortion attributable to the "fourth dimension" of growth, as famously coined by Mulliken.[38] Regardless, it is at this point that the focus of the bilateral cleft repair shifted from repairing only the lip to the need to address the nose as well, recognizing that the two deformities are linked.

Columella

Historical cleft lip repairs often did not address the bilateral cleft nose deformity in the primary stage. It was widely believed that early nasal surgery would interfere with nasal growth.[3] This belief was a steadfast one until disproven in the 1980s.[39] The impetus toward early operation, however, stemmed from the persistent dissatisfaction in the nasal outcome, beginning with the columella. Earlier surgeons recognized the deformity as a deficiency in length, and when combined with a hesitance to explore the intrinsic nasal structures, their solutions to this problem expectedly lied outside of the nose—frequently in the repaired upper lip. As early as the nineteenth century, vertical V-Y advancement from the central lip into the columella had been described.[27] Over the next 100 years, various modification were published, including trefoil and fleur-de-lis designs.[40] Converse rotated tissue from the nasal sills[41]; Millard banked prolabial fork flaps for secondary use. Unfortunately, none of these techniques withstood the test of time, as they introduced additional scar burden to the columella and caused secondary distortions.

Broadbent and Woolf[42] first described a case of primary nasal repair using medial advancement of alar domes combined with skin excision in a broad tip. In 1990, McComb published his 15-year review of the forked flap, and found that with adolescent development, the reconstructed columella is longer than normal, leading to enlarged nostrils; the nasal tip broadened because of persistent separation of the alar domes; and the columellar base drifted inferiorly because of scarring.[31] He further described the anatomical findings of the bilateral cleft nasal deformity based on stillborn infants—separation of alar domes and medial crura—as a columella that has been "unzipped and its component parts lie within the broad nasal tip" (Fig. 4). He modified his own technique (Fig. 5) to achieve columellar lengthening by suturing together the alar domes and medial crura by means of direct

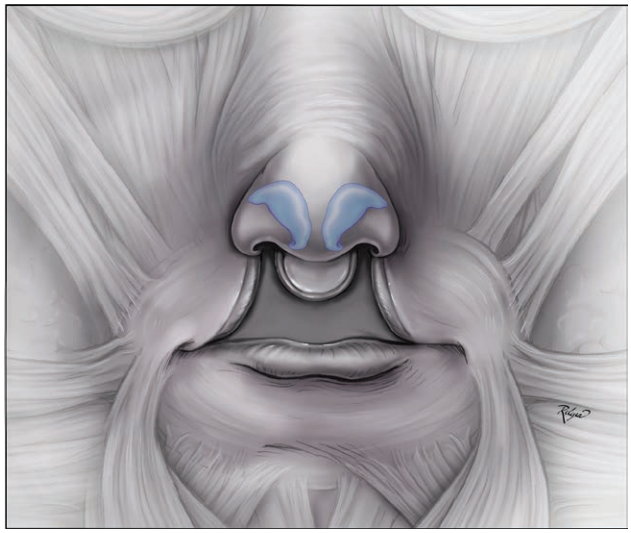

Fig. 4. Bilateral complete cleft lip, demonstrating nasal deformity, marked by separation of alar domes and medial crura.

exposure through a combined alar rim and columellar incision at the time of primary repair. Trott and Mohan[43] described similar suturing techniques under direct visualization by elevating the entire prolabium and columella en bloc. Because of concern for disruption of blood supply of the prolabium, this exposure was rarely performed.[44] Mulliken's[45] well-documented evolution of technique (Fig. 6), from staged modified Millard with forked flaps (Fig. 3) to single-staged nasolabial repair, included bilateral alar rim incisions to allow dissection of the nasal tip and use of interdomal sutures. It is important to point out that although primary cleft nasal repair frequently improves the cleft nasal deformity, most children with bilateral cleft lip, particularly those with severe findings, will need formal secondary cleft septorhinoplasty at skeletal maturity.

Premaxilla

In 2001, Mulliken[46] named two important advances in the repair of the bilateral cleft: (1) the evolution of single-staged nasolabial repair with columellar lengthening through restoration of intrinsic nasal anatomy; and (2) improvement in techniques for presurgical maxillary alignment. The latter is essential to the success of the former; thus, it is no surprise that both developed around the same time.

The protruding premaxilla has long plagued the cleft surgeon since the first reported cases centuries ago. Early surgeons thought of it as a mechanical hindrance to lip closure; thus, many opted for excision. The more astute surgeons recognized its teeth-bearing function, and many

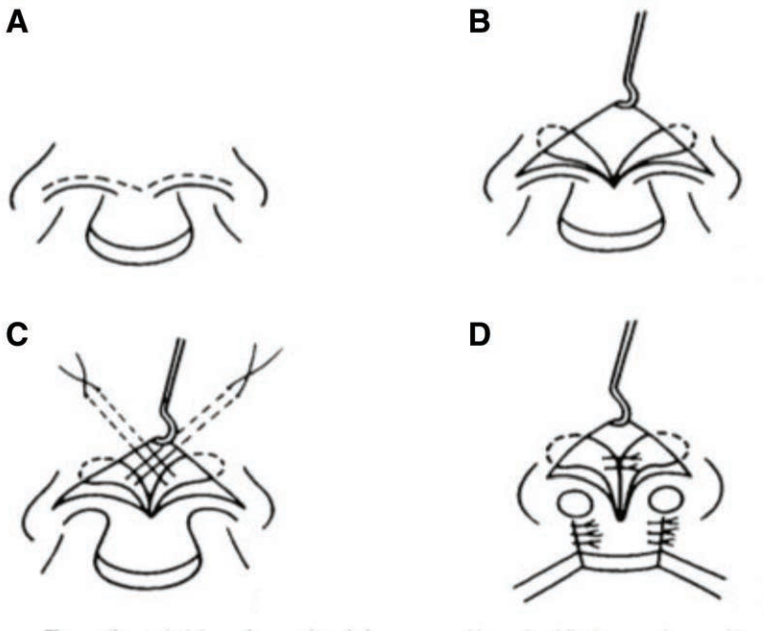

Fig. 5. McComb primary cleft nose repair. The access incision is a long alar rim and nasal tip incision. Lower lateral cartilages are dissected free and brought to the midline using interdomal sutures. (Reprinted with permission from McComb H. Primary repair of the bilateral cleft lip nose: A 15-year review and a new treatment plan. *Plast Reconstr Surg*. 1990;86:882–889.)

came up with elaborate systems of compression. Many of these contraptions undoubtedly paved the way toward modern day presurgical orthopedics. Millard described the eighteenth century surgeon P. J. Desault, who placed his patient in compressive headgear to set back the premaxilla before surgical repair.[27] Conceptually, this is remarkably similar to modern practice.

Nonsurgical management of the premaxilla in bilateral cleft is somewhat similar to the unilateral cleft. The goal is to improve the discrepancy between the alveolar and lip segments, and improve nasolabial symmetry. Lip taping is the simplest form of presurgical orthopedics, by providing sustained external pressure. Although its effect in severe bilateral cases is limited, it can be a potential option for children who are not candidates for anything else because of medical comorbidities.

The Latham device is an active presurgical device that uses a custom-made appliance pinned to the maxillary shelves. A looped wire passes through the neck of the premaxilla just behind the alveolar segment and retracts the premaxilla as the device is turned.[47,48] Nasoalveolar molding uses an intraoral acrylic appliance with a nasal stent. The device is reshaped to apply the tension needed to push back the premaxilla and reshape the nose.[49,50] Both require close follow-up and frequent adjustments. Nasoalveolar molding has the added benefit of reshaping the nose and lengthening the columella (Fig. 7). In 1998, Cutting et al. first described adopting nasoalveolar molding as an adjunctive method for primary columella lengthening.[51] A subsequent 12-year anthropometric evaluation of these patients demonstrated nearly normal proportional growth in nasal tip protrusion, columellar length, and width.[52] A recent survey demonstrated that approximately half of cleft teams offer presurgical infant orthopedics, and nasoalveolar molding is the most common method (88.2 percent versus 14.7 percent using the Latham device).[53]

In severe cases and those not amenable to more conservative treatment, lip adhesion (Figs. 8 through 10) with or without premaxillary setback may be needed. Tissue conservation is an essential component of lip adhesion, particularly in complex bilateral cleft, which has a high tendency toward dehiscence. Wide undermining may be necessary to reapproximate the orbicularis oris muscle under the prolabium. This provides the appropriate restriction needed on the premaxilla without placing undue tension on the skin closure.

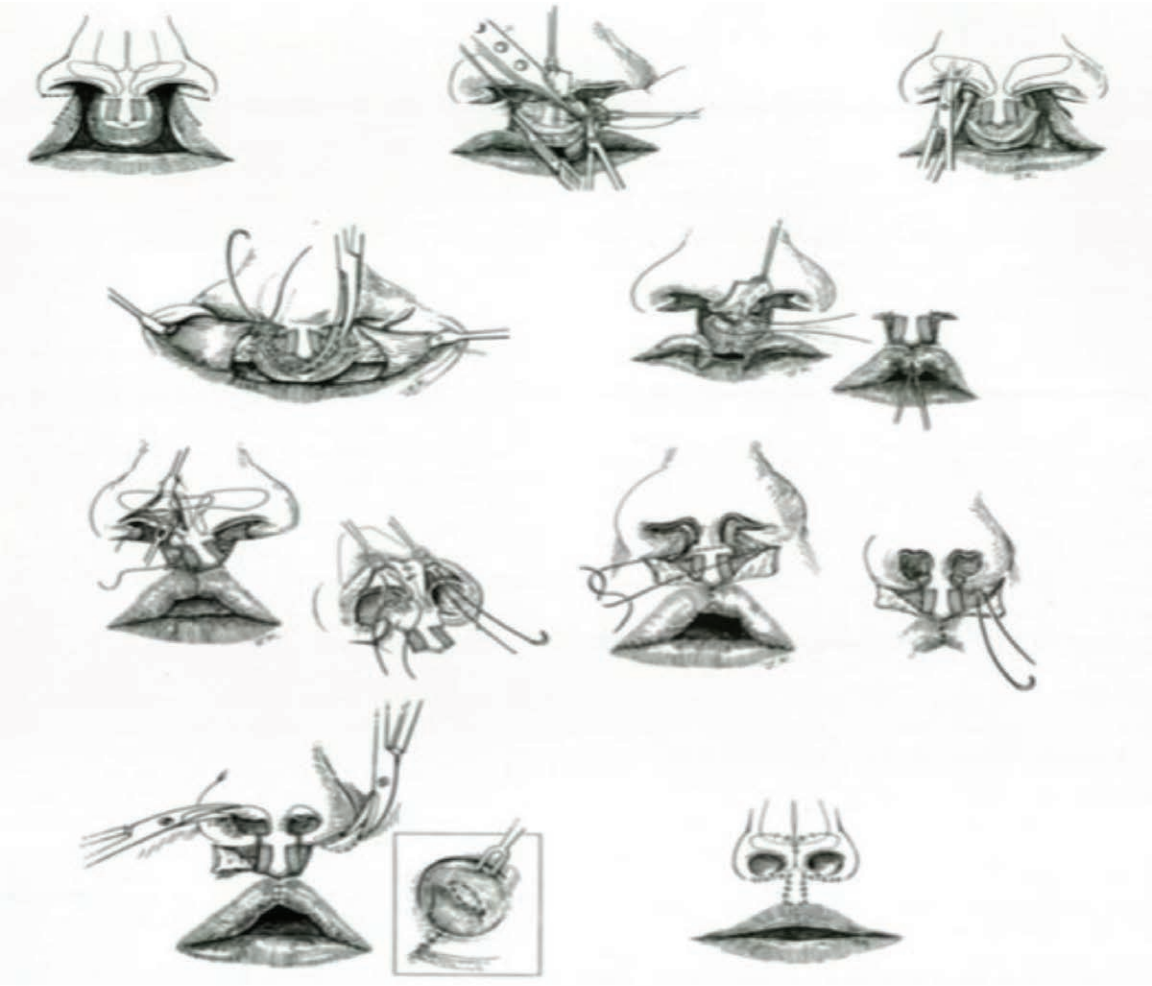

Fig. 6. The 2001 primary lip and nasal repair, in which the fork flaps are discarded and two small alar rim incisions are used to provide access to the nasal cartilage for reshaping. (Reprinted with permission from Mulliken JB. Primary repair of bilateral cleft lip and nasal deformity. *Plast Reconstr Surg*. 2001;108:181–194.)

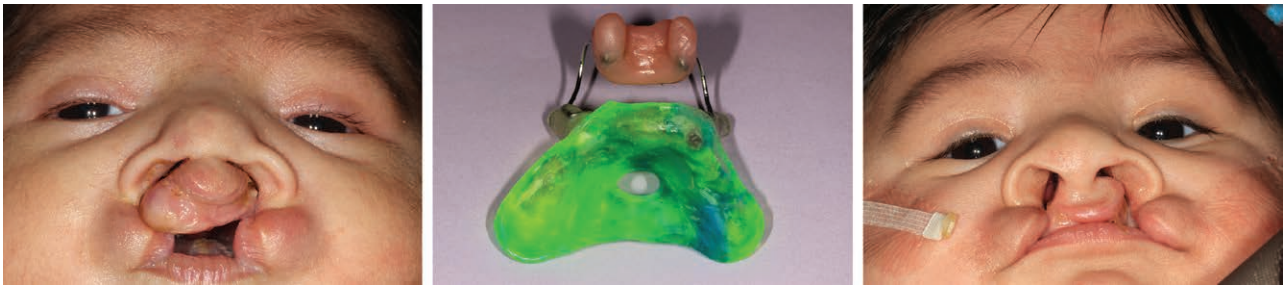

Fig. 7. A 4-week-old girl with bilateral complete cleft lip and palate. (*Left*) Before placement of nasoalveolar molding. Note protruding premaxilla and diminutive columella. (*Center*) The nasoalveolar molding appliance. (*Right*) After 2 months of nasoalveolar molding, just before primary repair. Note the improvement in alar position, elongation of the columella and retrusion of the premaxilla/prolabium tissue. (Courtesy of John Wirthlin, D.D.S., M.S.D.)

Fig. 8. A 2-month-old girl with bilateral complete cleft lip and palate. (*Above*) Preoperative views. Note the premaxillary protrusion and complete lack of columella. She underwent 1 month of nasoalveolar molding without significant improvement. (*Below*) Postoperative views after bilateral lip adhesion

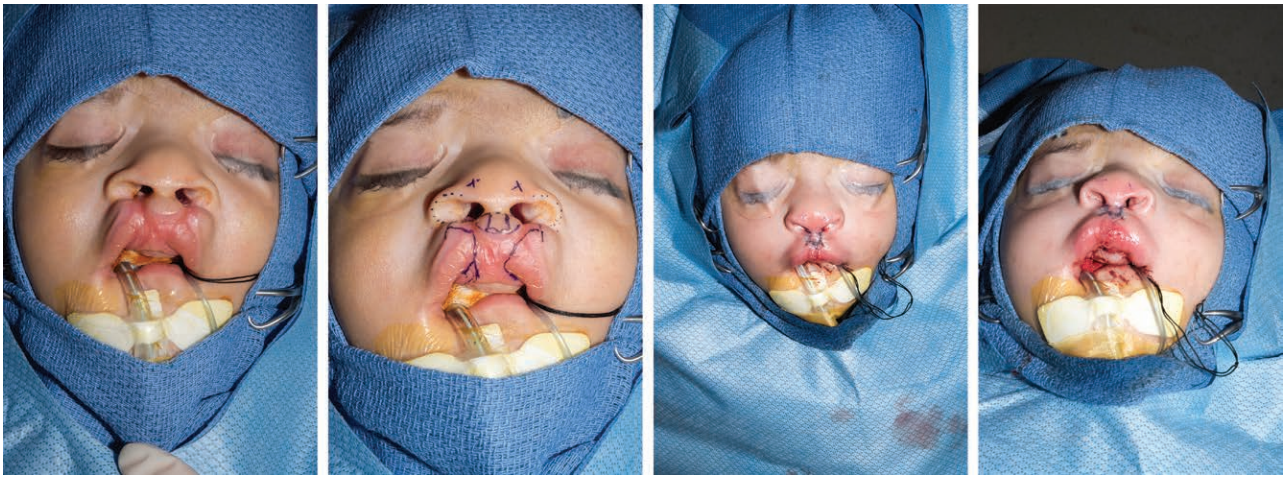

Fig. 9. Intraoperatively, during definitive lip repair.

Fig. 10. Postoperatively, 18 months after lip repair.

Table 1. Multidisciplinary Cleft Team

Pediatric plastic and reconstructive surgery
Craniofacial orthodontics
Otolaryngology
Pediatrics
Speech pathology
Pediatric dentistry
Audiology
Pediatric neuropsychology
Genetics
Advanced practice providers
Nurse coordinators

MODERN APPROACH

The care of patients with cleft lip with or without cleft palate needs to be performed by a multidisciplinary team (Table 1). As many of these children may present with other congenital anomalies, the cleft surgeon is likely far from being the most important provider during the first 3 to 6 months of life before repair. In our own practice, cleft care begins in the prenatal period for those with known prenatal diagnosis. This is particularly useful to provide information and counseling for parents, who are frequently overwhelmed (Fig. 11). Visits in the first months of life focus on general health, feeding, and growth. From the cleft perspective, they are also assessed for nasoalveolar molding (Fig. 12), which can be helpful in management of prolabial deficiency, premaxilla protrusion, and shortened columella. Nasoalveolar molding may not be successful in the most severe cases, which may require lip adhesion and premaxillary setback before definitive repair. A survey study in 2012 demonstrated that more than 70 percent of cleft surgeons use some variation of presurgical orthopedics, with nasoalveolar molding being the most common. The most common techniques used are the Millard (38 percent), Mulliken (26 percent), and

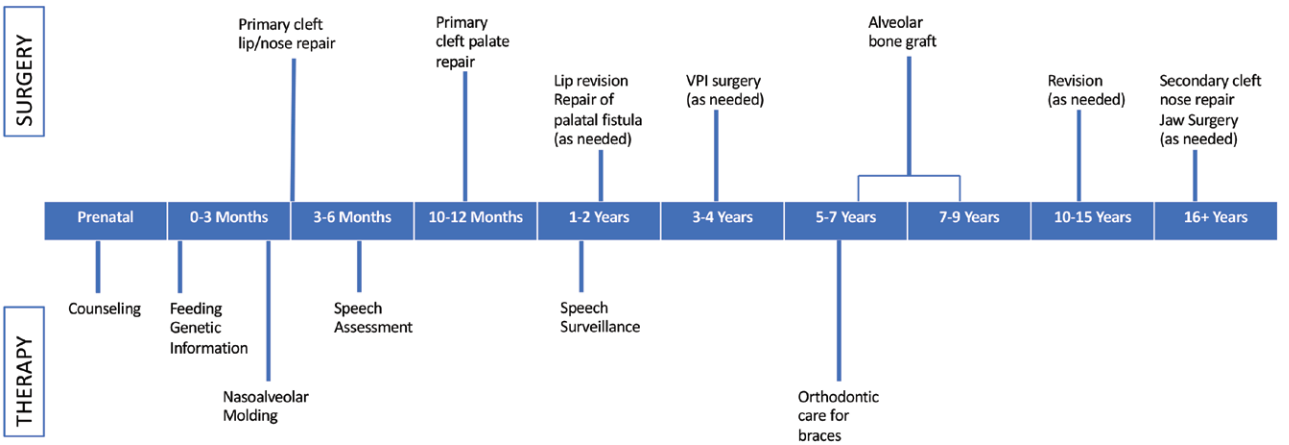

Fig. 11. Timeline for standard cleft and palate repair. *VPI*, velopharyngeal insufficiency.

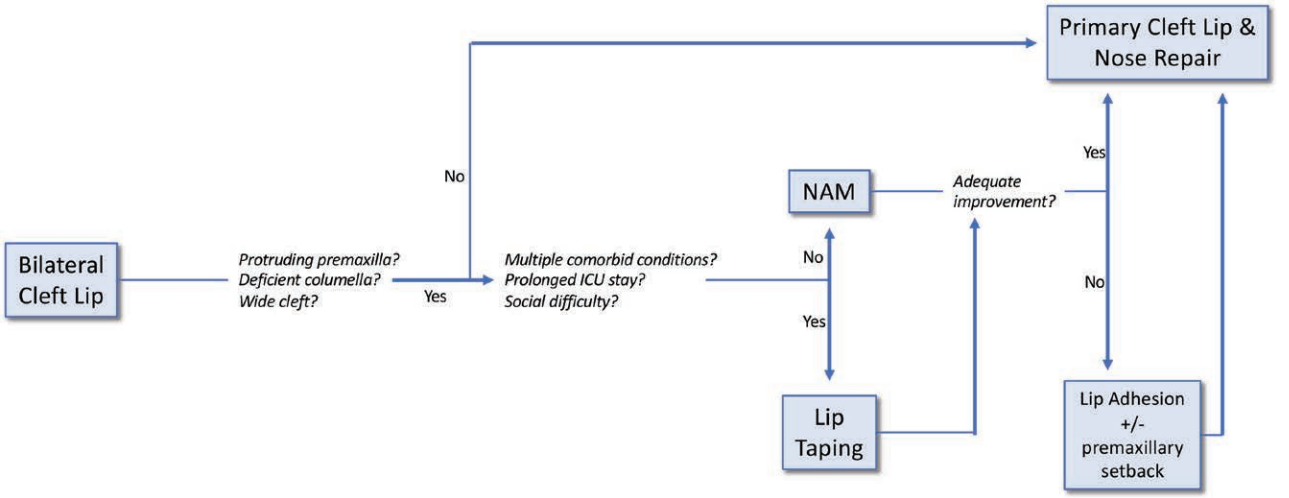

Fig. 12. Algorithm for repair of bilateral cleft lip and palate. *ICU*, intensive care unit; *NAM*, nasoalveolar molding.

Manchester (12 percent) techniques.[54] This trend is largely preserved.

Surgical Technique

The bilateral cleft lip deformity demonstrates a wide variety of presentations, and the cleft repair similarly has undergone many variations over the past centuries. From these variations, along with their successes and failures, arose a series of important lessons, as detailed by Mulliken[45]: (1) preserve symmetry; (2) restore muscle continuity; (3) use prolabium judiciously to recreate anatomical philtrum; (4) use lateral lip tissue to recreate median tubercle and mucocutaneous ridge; and (5) reconstruct the nasal tip and columella by restoring alar cartilage anatomy (Figs. 8 through 10). [**See Video 1 (online)**, which displays a 5-month-old girl with bilateral complete cleft lip and palate. She underwent previous lip adhesion, as nasoalveolar molding was not an option because of distance. The Mulliken technique was used for this repair. Marking is demonstrated. **See Video 2 (online)**, which displays the Mulliken technique used for this repair. Repair technique is demonstrated.]

Postoperatively, although the families are given the option to stay or go home, most choose to stay overnight to ensure adequate oral intake. Most children easily meet oral intake criteria, and rarely require more than acetaminophen for pain control. Before discharge, parents are educated in proper care of incisions and stent. Antibiotic ointment is used on all incisions until 1 week postoperatively. Nasal stents are cleaned daily using half-strength hydrogen peroxide solution, and are used for 3 months, during which time they are up-sized as needed.

Scar care is discussed during the first postoperative visit. Sunblock and scar massage are the essential components during the early postoperative period. Silicone-based scar ointment can be used in conjunction. Aggressive scar therapy with silicone sheet, or intralesional steroid injection, may be implemented with evidence of hypertrophic scarring.

CONCLUSIONS

Brown et al. wrote back in 1947 that the bilateral cleft lip deformity is twice as difficult to repair, and the results are only half as good.[55] This sentiment has been repeated by cleft surgeons since then. Looking back through history, however, demonstrates how far cleft care for children with bilateral cleft lip has evolved. The earlier repairs identified the challenges of the deformity, and the innovative techniques of the time served as strong frameworks for subsequent improvements. The advent of presurgical orthopedics, particularly nasoalveolar molding, made possible better surgical outcome for the most severe clefts. The modern multidisciplinary team model shifts the focus from a surgery-centric approach to one that is more comprehensive, thereby optimizing patient outcome. In many ways, the bilateral cleft repair is a victory for modern plastic surgery.

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ACKNOWLEDGMENTS

The authors thank Dr. John Wirthlin for sharing his patient photographs. In addition, the authors thank Katherine Relyea Tompkins for graphic assistance during the preparation of this manuscript.

PATIENT CONSENT

Parents or guardians provided written informed consent for use of the patients' images.

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