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# The Americleft Project: Burden of Care from Secondary Surgery

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**Background:** The burden of care for children with cleft lip and palate extends beyond primary repair. Children may undergo multiple secondary surgeries to improve appearance or speech. The purpose of this study was to compare the use of secondary surgery between cleft centers.
**Methods:** This retrospective cohort study included 130 children with complete unilateral cleft lip and palate treated consecutively at 4 cleft centers in North America. Data were collected on all lip, palate, and nasal surgeries. Nasolabial appearance was rated by a panel of judges using the Asher-McDade scale. Risk of secondary surgery was compared between centers using the log-rank test, and hazard ratios estimated with a Cox proportional hazards model.
**Results:** Median follow-up was 18 years (interquartile range, 15–19). There were significant differences among centers in the risks of secondary lip surgery ($P < 0.001$) and secondary rhinoplasty ($P < 0.001$). The cumulative risk of secondary lip surgery by 10 years of age ranged from 5% to 60% among centers. The cumulative risk of secondary rhinoplasty by 20 years of age ranged from 47% to 79% among centers. No significant differences in nasolabial appearance were found between children who underwent secondary lip or nasal surgery and children who underwent only primary surgery ($P > 0.10$).
**Conclusions:** Although some cleft centers were significantly more likely to perform secondary surgery, the use of secondary surgery did not achieve significantly better nasolabial appearance than what was achieved by children who underwent only primary surgery. *(Plast Reconstr Surg Glob Open 2015;3:e442; doi: 10.1097/GOX.0000000000000415; Published online 6 July 2015.)*

Outcomes of cleft lip and palate care can be divided into clinical, psychosocial, and systems of care.[1] Systems of care include the burden of care experienced by patients and the costs of delivering care. Unlike cost of care, which has been extensively studied in children with clefts,[2–5] less is known about the patients' burden of care.

Burden of secondary surgery is an important measure for patients, families, and health-care payers. Secondary surgery causes substantial pain and fear for children and time off work for families. Secondary surgery also leads to direct costs of $6000–$12,000 per surgery.[2,4,5] For these reasons, information on the use of secondary surgery is highly relevant.

Primary cleft surgery and secondary cleft surgery are distinct categories of surgical interventions.[6,7]

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**Table 1. Patient Characteristics and Treatment Center Protocols**

| | Center | | | | |
|---|---|---|---|---|---|
| **Characteristic** | **A (N = 20)** | **C (N = 39)** | **D (N = 35)** | **E (N = 36)** | ***P*** |
| Male, % | 55 | 59 | 71 | 61 | 0.59 |
| Follow-up duration, y | | | | | |
| Median | 13 | 19 | 18 | 17 | <0.001 |
| Interquartile range | 10–17 | 16–21 | 17–19 | 14–19 | |
| Presurgical orthopedics | No | No | Yes* | Yes* | |
| Lip repair | | | | | |
| Age, m | Varied | 3 | 3 | 3 | |
| Technique | Varied | Tennison | Millard | Millard | |
| Primary bone grafting | No | No | No | No | |
| Hard palate repair | | | | | |
| Age, m | 9–12 | 12 | 12 | 12–14 | |
| Technique | Varied | Vomer flap | Vomer flap | Vomer flap | |
| Soft palate repair | | | | | |
| Age, m | 9–12 | 18 | 12 | 12–14 | |
| Technique | Varied | Median suture with intravelar veloplasty | Wardill | Veau pushback | |
| Surgeons (No.) | 2 | 1 | 4 | 4 | |

Treatment center labels follow the convention of prior Americleft publications. Center B in the initial study did not participate. *P* values are for the comparison across centers and were calculated with $\chi^2$ test for ordinal outcomes and the Kruskal–Wallis test for continuous outcomes.
*Infant presurgical orthopedic treatment was done using a modified McNeil technique with extraoral taping.

Primary surgery includes the initial lip repair, with or without rhinoplasty, and the initial palate repair. Secondary surgery includes lip revision to improve a child's appearance; fistula closure, palate re-repair, or pharyngeal surgery to improve speech; and rhinoplasty performed after the initial lip repair to improve breathing or appearance. When it is possible to achieve the same or better outcomes through primary surgery alone, secondary surgery can be seen as an avoidable burden on patients and their families.

The objective of this study is to describe the burden of secondary surgery among children with cleft lip and palate who participated in the Americleft Intercenter Outcome Study.[8] This group of children was chosen for the following reasons: all participating children had nonsyndromic complete unilateral cleft lip and palate, thus minimizing variation because of differences in the initial cleft deformity; within each center, children were treated according to consistent protocols, minimizing variation because of changes in treatment protocol; lastly, dental, aesthetic, and facial growth outcomes of these patients have been previously described using objective measures.[8–12] The central hypothesis of this study was that the cleft center where a child was initially treated predicts whether or not that child subsequently had secondary surgery.

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## METHODS

### Participants

This retrospective cohort study included 130 Caucasian children with nonsyndromic complete unilateral cleft lip and palate (CUCLP) who received all of their primary cleft treatment at one of the 4 cleft centers in North America. All children were consecutively treated at the participating centers according to the centers' established protocols (Table 1). All patients were followed to at least 6 years of age.

All children were previously enrolled in the Americleft Intercenter Outcome Study.[8] Participating centers were labeled following the convention of the Americleft Study. Center B in the initial study did not participate because of unavailable hospital records; children treated at the remaining 4 institutions did participate. Institutional review board approval was obtained from all participating centers.

### Data Collection

Data were obtained from a review of existing medical records at each center. The patient's age at all surgical procedures was recorded, including every procedure involving the lip, palate, and/or nose. Follow-up duration was also recorded; this was defined as the child's age at their last clinical encounter at the treating center. Frontal and profile images of each subject between the ages of 6 and 12 years were obtained from existing records. Images were not available for children treated at center A.

### Endpoints

There were 3 primary outcomes. Time to secondary lip surgery was defined as the time (in years) from primary lip repair to secondary lip repair. Time to secondary palate surgery was defined as the time (in years) from primary palate repair to secondary palate repair, including closure of a fistula in the secondary palate, palate re-repair, pharyngeal flap, or sphincter pharyngoplasty. Time to secondary rhinoplasty was defined as the time (in years) from primary rhinoplasty to secondary rhinoplasty. For children who underwent no secondary surgery during the period of observation (ie, censored observations), observation time was defined as time from primary repair to last clinical encounter at the treating center. Incorporating observation time into the analysis prevents the bias that occurs if children are assumed to be incapable of receiving secondary surgery once they leave the initial treatment center.

Secondary surgery was defined as any procedure that occurred after the initial lip, palate, and/or nasal repairs in infancy. For centers performing lip adhesion and then primary repair, the primary repair was not classified as secondary surgery. For centers performing two-stage palate repair, the second stage was not classified as secondary surgery.

Financial support for surgical care at centers in Canada was provided by public payers, whereas surgical care at centers in the Unites States was supported by a combination of public payers and private insurance.

### Nasolabial Appearance

Nasolabial appearance was previously rated for study participants during the initial Americleft study.[11] Photographic records were not available for children treated at center A; this subgroup was not included in the analysis of nasolabial appearance. Ratings were assigned according to the system of Asher-McDade et al[13] using cropped frontal and profile images of each subject obtained between the ages of 6 and 12 years. Ratings were individually assigned to nasal form, symmetry, profile of the upper lip, and the shape of the vermillion border by 5 independent raters. Intrarater and interrater reliability was evaluated using weight kappa scores. The median score among the 5 raters was reported for each nasolabial feature. The rating protocol and reference pictures are available from the authors upon request.[14]

### Analysis

The $\chi^2$ test and Kruskal–Wallis test were used for among-center comparisons of discrete and continuous variables, respectively. The Kaplan–Meier product limit estimator was used to construct a time to event curve for the primary outcomes. The log-rank test was used to compare survival between centers. A Cox proportional hazards model was then constructed to estimate hazard of secondary surgery at each center. The proportional hazards' assumption was assessed using the score test, and no significant departures from the proportional hazards assumption were found.[15] To compare nasolabial appearance between children who had and those who had not undergone secondary lip or nasal surgery, scores for each feature of nasolabial appearance were compared using the Fisher exact test. Analyses were performed using JMP (11.0.0) and R (3.0.3).

## RESULTS

### Patient Characteristics

The study participants consisted of 130 children from 4 cleft centers in North America. The majority of children were male (N = 81, 62%). All children were born with complete unilateral cleft lip and palate. The median follow-up was 18 years (interquartile range, 15–19). Patient characteristics and treatment protocols at each center are described in Table 1.

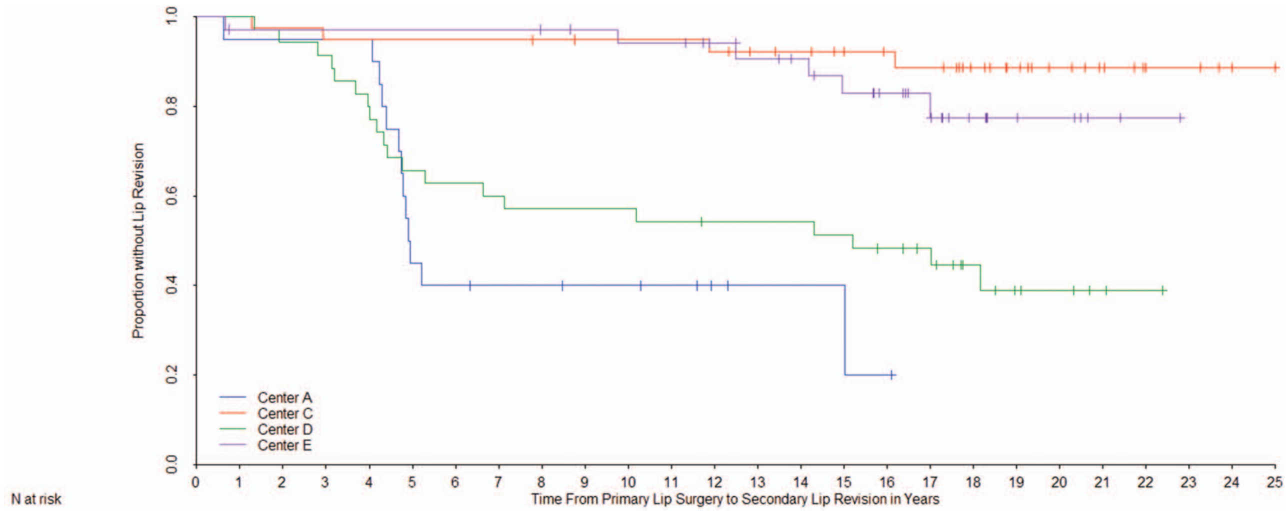
### Secondary Lip Surgery

Forty-three children (33%) underwent secondary lip surgery during the period of observation. The risk of secondary lip surgery at each center is shown in Figure 1. The estimated cumulative risk of secondary lip surgery by the age of 10 years was 60% for children treated at center A, 43% for children treated at center D, 6% for children treated at center E, and 5% for children treated at center C ($P < 0.001$).

The Cox regression analysis quantified the risk of secondary lip surgery between centers (Table 2). The analysis revealed that children treated at center C had a 12-fold lower risk of secondary lip surgery than children at center A. Children treated at center E had a 7-fold lower risk of secondary lip surgery than children at center A. The risk of secondary lip surgery was not statistically different between center A and center D.

### Secondary Palate Surgery

Twenty-five children (19%) underwent secondary palate surgery during the period of observation. The risk of secondary palate surgery at each center is shown in Figure 2. The estimated cumulative risk of secondary palate surgery by the age of 10 years was 26% for children treated at center E, 14% for children treated at center D, 13% for children treated at center C, and 6% for children treated at center A ($P = 0.057$). The Cox regression analysis revealed no significant differences in the risk of secondary palate surgery between centers (Table 2).




| N at risk | 0 | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | 17 | 18 | 19 | 20 | 21 | 22 | 23 | 24 | 25 |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Center A | 20 | 19 | 19 | 19 | 19 | 9 | 8 | 7 | 7 | 6 | 6 | 5 | 3 | 2 | 2 | 2 | 1 | | | | | | | | | |
| Center C | 39 | 39 | 38 | 37 | 37 | 37 | 37 | 37 | 36 | 35 | 35 | 35 | 34 | 32 | 31 | 29 | 27 | 26 | 21 | 17 | 13 | 10 | 7 | 6 | 4 | 3 |
| Center D | 35 | 35 | 33 | 32 | 28 | 23 | 22 | 21 | 20 | 20 | 20 | 19 | 18 | 18 | 18 | 17 | 15 | 13 | 8 | 5 | 4 | 2 | 1 | | | |
| Center E | 36 | 34 | 34 | 34 | 34 | 34 | 34 | 34 | 33 | 32 | 31 | 31 | 28 | 26 | 24 | 21 | 18 | 15 | 9 | 6 | 5 | 2 | 1 | | | |

**Fig. 1.** Kaplan–Meier curves of time to secondary lip surgery. Censored data are depicted by +'s. Log-rank test $P < 0.001$.

**Secondary Rhinoplasty**

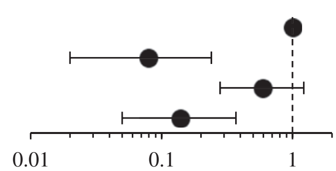
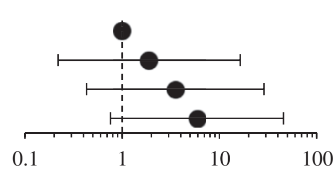
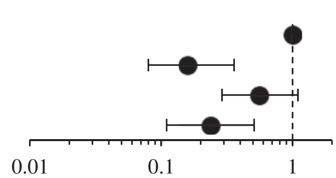
Sixty-nine children (53%) underwent secondary rhinoplasty during the period of observation. The risk of secondary rhinoplasty at each center is shown in Figure 3. The estimated cumulative risk of secondary rhinoplasty by the age of 20 years was 79% for children treated at center D, 75% for children treated at center A, 63% for children treated at center E, and 47% for children treated at center C ($P < 0.001$).

The Cox regression analysis quantified the risk of secondary rhinoplasty between centers (Table 2). The analysis revealed that children treated at center C have a 6-fold lower risk of secondary rhinoplasty than children at center A. Children treated a center E had a 4-fold lower risk of secondary rhinoplasty than children at Center A. The risk of secondary rhinoplasty was not statistically different between center A and center D.

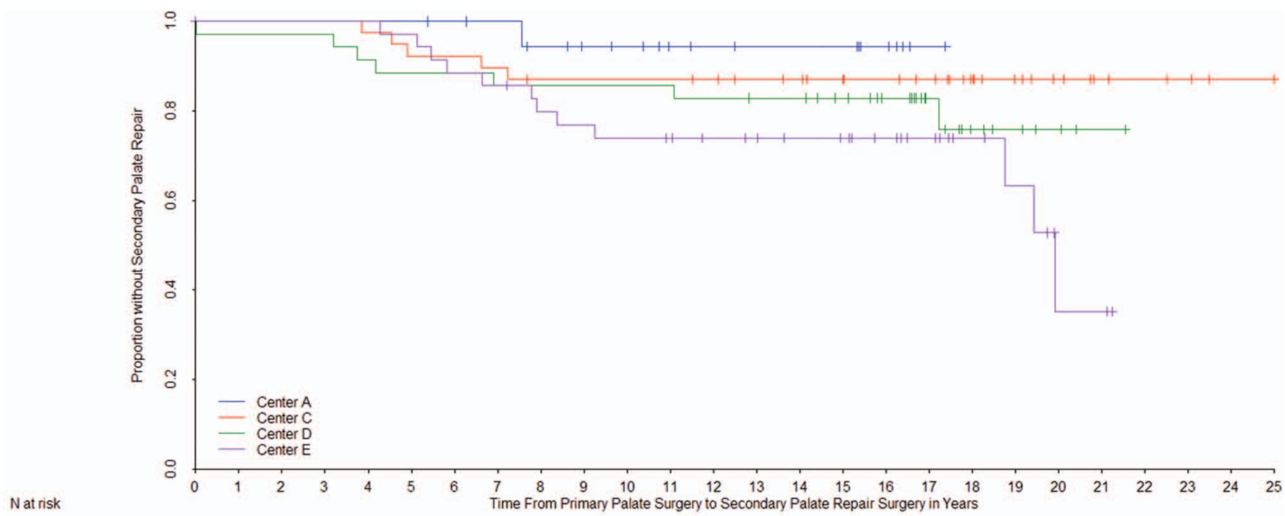
**Effect of Secondary Surgery on Nasolabial Appearance**

Nasolabial appearance was assessed for children treated at centers C, D and E; photographs for chil-

**Table 2. Risks of Secondary Surgery as a Function of Treatment Center**

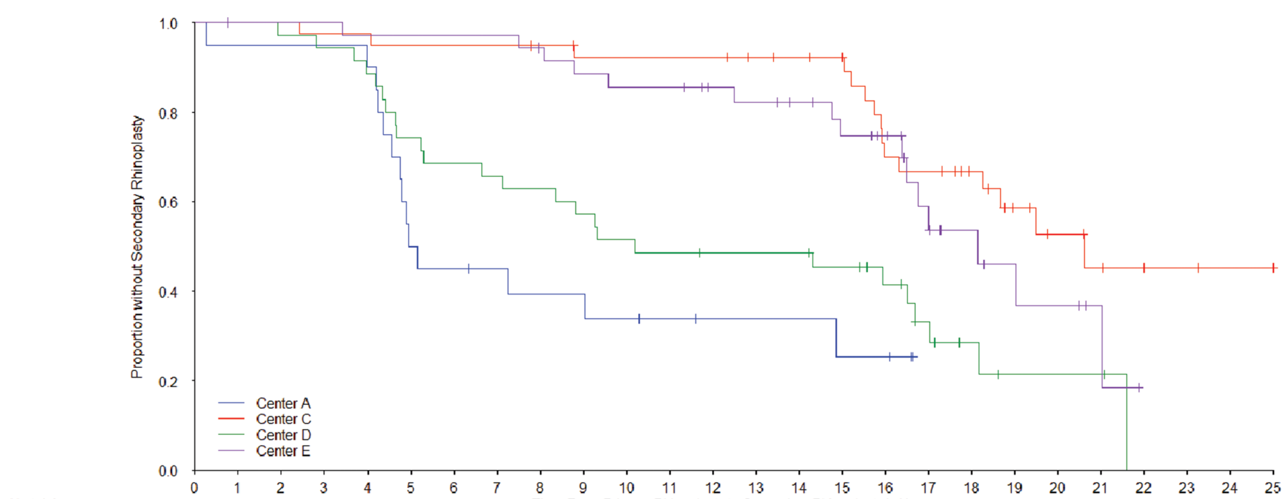
| Center | No. Participants | No. Events | Hazard Ratio (95% CI) | | *P* |
|---|---|---|---|---|---|
| Risk of secondary lip surgery | | |  | | |
| A | 20 | 13 | | 1.00 | <0.001 |
| C | 39 | 4 | | 0.08 (0.02–0.24) | |
| D | 35 | 20 | | 0.59 (0.28–1.22) | |
| E | 36 | 6 | | 0.14 (0.05–0.37) | |
| Risk of secondary palate surgery | | |  | | |
| A | 20 | 1 | | 1.00 | 0.057 |
| C | 39 | 5 | | 1.88 (0.22–16.37) | |
| D | 35 | 7 | | 3.52 (0.43–28.78) | |
| E | 36 | 12 | | 5.90 (0.76–45.65) | |
| Risk of secondary rhinoplasty | | |  | | |
| A | 20 | 14 | | 1.00 | <0.001 |
| C | 39 | 15 | | 0.16 (0.08–0.36) | |
| D | 35 | 25 | | 0.56 (0.29–1.10) | |
| E | 36 | 15 | | 0.24 (0.11–0.51) | |

Hazard ratios are on a log scale. *P* values are for log-rank tests.
CI, confidence interval.




| N at risk | 0 | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | 17 | 18 | 19 | 20 | 21 | 22 | 23 | 24 | 25 |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Center A | 20 | 20 | 20 | 20 | 20 | 20 | 19 | 18 | 16 | 14 | 13 | 10 | 9 | 8 | 8 | 8 | 5 | 1 | | | | | | | | |
| Center C | 39 | 39 | 39 | 39 | 38 | 36 | 36 | 35 | 33 | 33 | 33 | 33 | 32 | 30 | 29 | 27 | 24 | 22 | 17 | 13 | 10 | 7 | 6 | 5 | 3 | 3 |
| Center D | 35 | 34 | 34 | 34 | 32 | 31 | 31 | 30 | 30 | 30 | 30 | 30 | 29 | 28 | 28 | 25 | 20 | 12 | 7 | 5 | 3 | 1 | | | | |
| Center E | 36 | 35 | 35 | 35 | 35 | 34 | 31 | 30 | 27 | 26 | 25 | 24 | 22 | 21 | 19 | 18 | 15 | 12 | 8 | 6 | 2 | 2 | | | | |

**Fig. 2.** Kaplan–Meier curves of time to secondary palate surgery. Censored data are depicted by +'s. Log-rank test $P = 0.057$.




| N at risk | 0 | 1 | 2 | 3 | 4 | 5 | 6 | 7 | 8 | 9 | 10 | 11 | 12 | 13 | 14 | 15 | 16 | 17 | 18 | 19 | 20 | 21 | 22 | 23 | 24 | 25 |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Center A | 20 | 19 | 19 | 19 | 18 | 10 | 9 | 8 | 7 | 7 | 6 | 5 | 4 | 4 | 4 | 3 | 3 | | | | | | | | | |
| Center C | 39 | 39 | 39 | 38 | 38 | 37 | 37 | 37 | 36 | 34 | 34 | 34 | 34 | 32 | 31 | 30 | 22 | 21 | 17 | 11 | 8 | 6 | 5 | 4 | 3 | 3 |
| Center D | 35 | 35 | 34 | 33 | 31 | 26 | 24 | 23 | 22 | 20 | 18 | 17 | 16 | 16 | 16 | 14 | 11 | 7 | 4 | 2 | 2 | 2 | | | | |
| Center E | 36 | 35 | 35 | 35 | 34 | 34 | 34 | 34 | 32 | 30 | 29 | 29 | 26 | 25 | 23 | 20 | 17 | 11 | 7 | 5 | 4 | 2 | | | | |

**Fig. 3.** Kaplan–Meier curves of time to secondary rhinoplasty. Censored data are depicted by +'s. Log-rank test $P < 0.001$.

dren treated at center A were not available for rating. Sixty-six children had received only primary surgery at the time of their assessment, whereas 19 children had undergone both primary surgery and a secondary surgery on the lip or nose by the time of their assessment. Intrarater reliability for each of the 5 raters was good to very good (mean weighted kappa, 0.739; range, 0.638–0.823). Interrater reliability was moderate to good for each component of nasolabial appearance (mean weighted kappa, 0.645; range, 0.587–0.708).

Nasolabial appearance scores were compared between these 2 distinct groups of children. Scores for each of the 4 components of nasolabial appearance are shown in Figure 4. For all of the 4 components, scores were not significantly different between children who underwent both primary and secondary surgeries and children who underwent only primary surgery ($P > 0.40$ for each component).

## DISCUSSION

The principal finding of this study is that use of secondary lip surgery and secondary rhinoplasty is dependent upon the center where a child is treated. The risk of secondary lip surgery varied 12-fold across the 4 participating centers. The risk of secondary rhinoplasty varied 6-fold across centers.

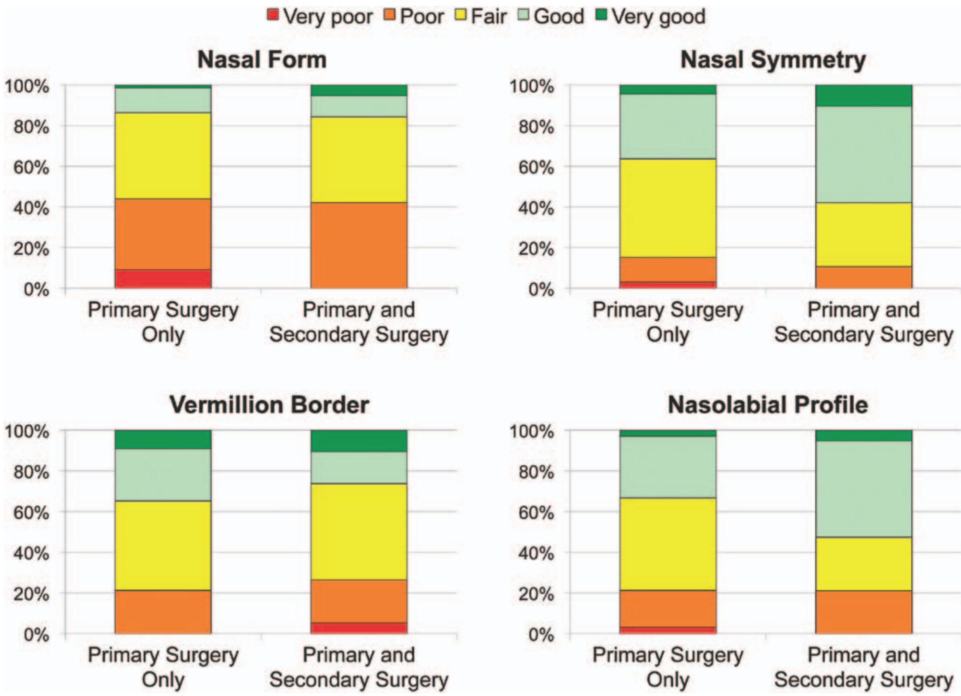



**Fig. 4.** Nasolabial appearance among children who had primary surgery only (N = 66) versus children who had primary surgery and secondary lip or nasal surgery (N = 19). There were no significant differences in the distribution of ratings for any component of nasolabial appearance ($P > 0.40$, Fisher exact test).

Although use of secondary lip or nasal surgery may have improved results from primary surgery for individual patients, as a group, children receiving both primary and secondary surgeries did not achieve significantly better nasolabial appearance scores than children who received only primary surgery. These findings are of clinical importance because they show that use of secondary surgery is highly variable across cleft centers and suggest that undergoing secondary lip or nasal surgery does not lead to a nasolabial appearance beyond what many children receive from primary surgery alone.

These finds are consistent with and extend those from Eurocleft. Eurocleft investigators compared outcomes of cleft care across 6 European centers. They found the incidence of secondary surgery varied 4–69% for secondary lip surgery, 15-42% for pharyngoplasty, and 15–65% for secondary rhinoplasty.[7] Eurocleft investigators found the incidence of secondary surgery varied 4–69% for secondary lip surgery, 15–42% for pharyngoplasty, and 15–65% for secondary rhinoplasty. Reports of secondary lip surgery from North American centers have suggested similar variation, with reported revisions rates of 29%, 36%, and 64%.[16–18] This study, which is the first to directly compare burden of secondary surgery between centers in North America while controlling for variation in the cleft deformity, found secondary surgery rates similar to those from Eurocleft. Another similarity between Eurocleft and this study is that centers with a single surgeon using a consistent surgical technique achieved the lowest incidence of secondary lip surgery. These similarities suggest that the variation in care delivery and outcomes found among European cleft centers is also present in North America.

This study raises the important question of why variation exists between centers in the use of secondary surgery. One hypothesis is that clinical outcomes of primary surgery differ between centers, thus leading some centers to perform more secondary surgery in an attempt to achieve the same results other centers reach from primary surgery alone. Under this hypothesis, outcomes of primary surgery could differ because of variation in treatment protocol, technical skill, or differences in the type or severity of cleft deformity. This hypothesis is supported by the Eurocleft studies, which found both treatment protocol and surgeon volume were associated with differences in outcomes of primary surgery.[19] An alternative hypothesis is that all centers achieve the same clinical outcomes from primary surgery, but centers differ in their thresholds for further improving on results of primary surgery by application of secondary surgery. This difference in threshold could be because of differences among providers, patients, or local customs. This study did not collect the data on pre-revision clinical outcomes that would be necessary to test these 2 hypotheses. Rather, this study demonstrates that broad variation in the use of secondary surgery exists, and that testing these hypotheses through longitudinal clinical studies could improve our understanding of when patients should be offered secondary surgery.

**Timing of Secondary Surgery**

This study extends the current understanding of secondary cleft surgery by describing when each type of secondary surgery occurs in a child's life. The analysis reveals that secondary lip and nasal surgery are performed at 2 time points:[1] 4–6 years of age at some centers and[2] after 15 years of age at all centers. Secondary palate surgery appears to occur at 4–7 years of age at all centers. This information suggests that comparing outcomes between centers is best performed either before age 4, to evaluate results of primary surgery, or delayed until age 7 when the second wave of surgical interventions is complete. Centers appear to concentrate secondary cleft surgery within the 4–7 years age group, making this a time of rapid change in patient outcomes.

The interaction between secondary surgery and growth is complex. Nasolabial features change as the child grows.[20] Centers that evaluate patients for secondary surgery at 4 years of age may see fewer indications for revision than centers evaluating patients for secondary surgery at 6 years of age. Furthermore, secondary surgery may negatively influence subsequent facial growth.[21] To improve our understanding of how growth impacts need for secondary surgery and how secondary surgery impacts growth, additional research is necessary using standardized records at fixed time points from birth through skeletal maturity.

**Benefits of Secondary Lip or Nose Surgery**

As discussed above, one explanation for variation in rates of secondary surgery may be the existence of a lower threshold for improving results of primary surgery at some centers. If this is true, the results of this study suggest that caution may be necessary when applying a lower threshold to lip or nasal surgery because this study failed to demonstrate superior nasolabial appearance outcomes for children who underwent secondary lip or nasal surgery when compared with children who underwent primary surgery alone. This result adds to an existing controversy on the effectiveness of secondary lip surgery. Trotman et al[22] found that fewer than half of patients undergoing lip revision achieved improved nasolabial appearance. These same investigators also found no consistent improvement in facial movements following lip revision.[23] However, other investigators have reported small improvements in some aspects of nasolabial appearance following lip revision.[24] These discrepancies highlight the need for further evaluation of the effectiveness of secondary lip surgery.

**Limitations**

The data in this study must be interpreted in the context of the study design. All children had nonsyndromic complete unilateral cleft lip and palate, and results may not generalize to children with other cleft types or those with additional medical anomalies.[5] Confounding may exist because of unmeasured differences in the severity of the cleft deformity treated at each center, differences in the socioeconomic status of children at each center that limited ability to present for evaluation or treatment, and/or differences in the ages at which subjects' photographs were obtained for rating nasolabial appearance. The authors are not aware of evidence to suggest confounding by any of these mechanisms occurred in this study, but they cannot be definitively excluded. Clustering of results by surgeon may explain part of the between-center differences in secondary surgery, but unfortunately the sample sizes in this study were insufficient to test this hypothesis. Care delivery at the 4 participating centers is unlikely to represent care at all cleft centers in North America, and there is evidence that the use of secondary surgery is substantially different at other centers.[25–28] The Asher-McDade system for evaluating nasolabial appearance has limitations, including subjectivity and imperfect agreement among clinicians.[11,13,22,29] This study's limited sample also reduced the ability to detect small differences in nasolabial appearance. Whether the study's results reflect cleft care delivery across North America will need to be determined in future studies with a larger, more diverse sample of patients.

**Conclusions**

In conclusion, the use of secondary lip and nasal surgery varies significantly between cleft centers. Although the sources of this variation remain unclear, the effect is broad differences in a child's burden of surgical care depending upon where they are treated. These results highlight the need for additional studies evaluating the efficacy and effectiveness of secondary surgery for children with cleft lip and palate.

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