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Marked Variation Exists Among Surgeons and Hospitals in the Use of Secondary Cleft Lip Surgery

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Abstract

Objective: To identify child-, surgeon- and hospital-specific factors at the time of primary cleft lip repair that are associated with use of secondary cleft lip surgery.

Design: Retrospective cohort study

Setting: Forty-nine pediatric hospitals

Participants: Children who underwent cleft lip repair between 1999 and 2015.

Main Outcome Measure: Time from primary cleft lip repair to secondary lip surgery

Results: By five years after primary lip repair, 24.0% of children had undergone a secondary lip surgery. In multivariable analysis, primary lip repair before 3 months had a 1.22-fold increased hazard of secondary surgery (95% CI, 1.02-1.46) compared to repair at 7-12 months of age, and children with multiple congenital anomalies had a 0.77-fold decreased hazard of secondary

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surgery (95% CI, 0.68-0.87). After adjusting for cleft type, age at repair, presence of multiple congenital anomalies, and procedure volume, there remained substantial variation in secondary surgery use among surgeons and hospitals ($p<0.01$). For children with unilateral cleft lip repaired at 3-6 months of age, the predicted proportion of children undergoing secondary surgery within five years of primary repair ranged 4.9% - 21.8% across surgeons and 4.5% - 24.7% across hospitals.

Conclusions: There are substantial differences among surgeons and hospitals in the rates of secondary lip surgery. Further work is needed to identify causes for this variation among providers.

Keywords

Health Services; Cleft Lip; Secondary Surgery

Introduction

The objectives of primary cleft lip repair are to restore the normal anatomic relationships and produce long-term symmetry of the lip and nose (Mulliken and LaBrie, 2012; Vyas and Warren, 2014). Ideally, every cleft lip repair would result in alignment of the vermillioncutaneous and vermillomucosal junctions, unification of the orbicularis oris muscle, and symmetry of the cutaneous lip height, vermillion height, nasal tip, nostril shape, and alar base position (Wilhelmsen and Musgrave, 1966; Stal and Hollier, 2002; Monson et al., 2014). Achieving each of these goals for every child can be complex, and may not be feasible for all children.

Patients, families, and surgeons recognize that imperfections can exist after primary cleft lip repair. Addressing these imperfections with secondary lip surgery is an important component of comprehensive cleft care. Secondary lip surgery can substantially improve the appearance of the lip and is pursued by many patients (Essick et al., 2007; Trotman et al., 2007a; Trotman et al., 2007b; Trotman et al., 2007c). In fact, secondary lip surgeries make up one-fourth of all cleft lip surgeries in the United States (Thompson et al., 2017). Secondary lip surgery does come with costs, however: surgeries require a general anesthetic, time off work and school for parents and children during the recovery period, and direct medical costs estimated at \$8,893 in 2009 (Thompson et al., 2017). Given the benefits of secondary lip surgery for improving appearance, its high prevalence, and its substantial cost, it is important for cleft providers and policy makers to understand how secondary surgery is utilized.

Prior studies suggest secondary surgery rates vary substantially across cleft teams (Semb et al., 2005b; Sitzman et al., 2015). A comparison across four cleft centers in North America found secondary lip surgery rates varied 5% to 60% (Sitzman et al., 2015), while a comparison across five centers in Europe found rates varied 4% to 69% (Semb et al., 2005b). Rates of secondary lip surgery varied even more broadly, from 0% to 100%, in a recent systematic review (Sitzman et al., 2016). Variation in secondary surgery has been attributed to patients' health literacy, socioeconomic status, cultural preferences, and insurance status (Cassell et al., 2007; Cassell et al., 2008; Trotman et al., 2010). Variation in secondary surgery has also been attributed to the technique of primary repair, skill of executing the primary repair, and use of pre-surgical orthopedics such as naso-alvolar molding (Stal and

Hollier, 2002; Crockett and Goudy, 2014). Further, the threshold for offering secondary surgery can vary among surgeons and cleft teams (Semb et al., 2005a), and the threshold for requesting secondary surgery can vary among patients and families. Determining the relative contributions of these patient- and provider-specific factors to the use of secondary surgery may provide insight into each patient's individual risk of secondary surgery and to changes that providers and hospitals can adopt to reduce the need for secondary surgery.

Previous studies evaluating the incidence of secondary lip surgery have included a limited number of cleft centers (Cohen et al., 1995; Friede and Enemark, 2001; Swennen et al., 2002; Semb et al., 2005b; Stein et al., 2007; Sitzman et al., 2015; Sitzman et al., 2016). Due in part to their small sample sizes, these studies have been unable to disentangle the effects of patient-, surgeon- and hospital-specific factors. The present study seeks to overcome both these limitations by providing estimates of secondary surgery at pediatric hospitals across the US and patient-specific estimates of secondary surgery that are adjusted for differences among surgeons and hospitals. The results will serve as a first step towards understanding the more specific factors that contribute to secondary lip surgery.

The present study evaluates the use of secondary cleft lip surgery across 49 free standing children's hospitals participating in Pediatric Health Information System (PHIS). These hospitals provide 85% of all pediatric specialty care delivered in the United States (Goodman et al., 2014), making this the largest longitudinal study of children undergoing cleft lip repair to date. The study has four distinct objectives: (1) to estimate the proportion of children undergoing secondary lip surgery during the first five years after primary lip repair, (2) to describe when after primary lip repair children undergo secondary lip surgery, (3) to identify child, surgeon, and hospital specific factors present at the time of the primary lip repair that are associated with secondary lip surgery, and (4) to estimate the magnitude of variation in secondary lip surgery attributable to differences among surgery and hospitals.

Methods

The Pediatric Health Information System (PHIS) is a comprehensive pediatric database containing administrative data from 49 children's hospitals across the United States. PHIS includes patient demographics, diagnoses and procedures for all inpatient stays, observation unit encounters, ambulatory surgeries, and emergency department visits at participating hospitals. Investigators can identify sequential hospital encounters for an individual patient using a hospital-specific identifier present in the database.

We used the PHIS database to perform a retrospective cohort study of children undergoing cleft lip repair. We included all children with cleft lip, with or without cleft palate, who underwent cleft lip repair between January 1, 1999, and September 30, 2015. We identified eligible children using *International Classification of Diseases, Ninth Revision (ICD-9) codes* for cleft lip (749.1), cleft lip and palate (749.2), and cleft lip repair (27.54). We excluded children older than 12 months at lip repair, as these children may have medical comorbidities or microform clefts that affect risk of secondary surgery. For children having only one cleft lip repair during their first year of life, this surgery was assumed to be their primary lip repair. We were unable to distinguish lip adhesion from primary lip repair using

the PHIS database, so if a child underwent cleft lip repair at an individual hospital twice during their first year of life, we assumed the first surgery was a lip adhesion and the second surgery was the primary lip repair.

For all children meeting inclusion criteria, we collected information on child demographics, medical conditions, and surgical care. Demographic data included gender, race, and median household income by ZIP code of residence. Race was included in the final model because prior research suggests variation among racial and ethnic groups in receipt of cleft surgeries (Cassell et al., 2009; Thompson et al., 2017). Median household income by ZIP code of patient residence was obtained from 2010 US Census data and split into four categories based on US federal poverty guidelines for a family of four, as previously described (Fieldston et al., 2013).

We identified each child's cleft lip type and medical comorbidities from their discharge diagnoses. We categorized cleft lip type as unilateral, bilateral, and unspecified, using *ICD-9* codes. We were unable to distinguish children with complete cleft lip and palate from children with incomplete cleft lip and Veau I or Veau II cleft palate using the PHIS database, so we did not include presence or absence of cleft palate in our analysis. Children with additional congenital anomalies that might influence treatment of their cleft lip were identified using previously validated *ICD-9* codes (Feudtner et al., 2001).

We extracted details of surgical care at each child's initial cleft lip repair. We categorized age at primary lip repair into three groups based on existing approaches to timing of primary lip repair: less than 3 months, 3 to 6 months, and 7 to 12 months. We determined postoperative antibiotic use, which we defined as receipt of any antibiotic on the first and/or second day after primary lip repair. We determined surgeon and hospital procedure volume for each child on the day of their lip repair by counting all cleft lip repairs (*ICD-9* codes 27.54) performed by that surgeon or hospital, respectively, in the prior 365 days. This approach decreases exposure misclassification compared to annual procedure volume for the same year the procedure was performed (McAteer et al., 2013). Counts included all cleft lip repairs in patients younger than 4 years of age as all lip repairs (primary and secondary) were predicted to increase the surgical team's experience with the procedure. For surgeons or hospitals who had reported in PHIS for fewer than 365 days at the time of a child's lip repair, procedure volume was calculated as the number of cleft lip repairs performed during their first 365 days of reporting. Procedure volume was categorized into tertiles.

For each included child, we identified all encounters at the hospital where primary lip repair was performed. We calculated time to secondary lip surgery for each child, which we defined as time from primary lip repair until the child underwent a second lip surgery. We defined receipt of secondary lip surgery as any hospital encounter after the primary lip repair that included an *ICD-9* code for correction of cleft lip (27.54). Children not undergoing secondary surgery during the observation period were censored on the date of their last encounter at their initial treating hospital. No information was available on receipt of secondary lip surgery at hospitals other than the child's initial treating hospital.

The Institution Review Boards at Phoenix Children's Hospital and Cincinnati Children's Hospital Medical Center reviewed this study and determined it was not human subjects research, as defined by the Common Rule (45CFR46.102[f]), because the dataset was deidentified.

Statistical Analyses

We plotted Kaplan-Meier time-to-event curves for each hospital and each surgeon in the cohort. We then fit a three-level mixed-effects parametric time-to-event model with clustering of patients within surgeons and clustering of surgeons within hospitals. We assumed a Weibull distribution for this model, after confirming appropriateness of this assumption by visual inspection of log-log plots of survival. Random effects were assumed to have normal distributions with zero means. We tested for all two-way interactions. We adjusted for year of primary lip repair in the model. After fitting the full model, we estimated the variability in time to secondary surgery attributable to patients, surgeons and hospitals (Yang et al., 2009). We then used the model to predict the proportion of children undergoing secondary surgery during the first five years after primary lip repair for each surgeon and each hospital (Yang et al., 2009). We presented the predicted proportion of children undergoing secondary surgery in funnel plots with 99.8% (~3 sigma) control limits (Spiegelhalter, 2005). Hospitals outside the control limits can be interpreted as deviating significantly from the overall rates.

Sensitivity analyses were conducted to evaluate whether the length of follow up time imposed bias through right censoring. Sensitivity analyses included: (1) censoring children two years after their last encounter; (2) censoring children four years after their last encounter; (3) censoring children at the end of the study observation period; (4) excluding children with less than one year of follow-up; and (5) excluding children with less than four years of follow up. Results of all sensitivity analyses were nearly identical to those of our main analysis and are available from the authors upon request.

Statistical analyses were performed using Stata version 14 (StataCorp, College Station, Texas). Statistical significance was set at $p < .05$.

Results

A total of 14,798 children underwent primary cleft lip repair at participating hospitals during the seventeen-year observation period. Right censoring occurred for 86.7% of the observations (N= 12,823). Characteristics of the study population are shown in Table 1.

Using a mixed-effects time-to-event model, we investigated the relationship between demographic, medical, and surgical factors and the use of secondary surgery (Table 2). Among the demographic and medical factors evaluated, median family income for ZIP code of residence, cleft type, and presence of additional congenital anomalies were associated with time to secondary lip surgery ($p < 0.05$). Children with a bilateral cleft lip had lower hazard of secondary lip surgery (HR 0.88, 95% CI 0.78-0.98) compared to children with a unilateral cleft lip. Children with additional congenital anomalies and/or syndromic diagnoses had a lower hazard of secondary lip surgery (HR 0.77, 95% CI 0.68-0.87).

Child's age at primary lip repair was associated with time to secondary lip surgery ($p=0.03$). Children who had primary repair before three months of age had a 1.22-fold increased hazard of secondary surgery (95% CI, 1.02-1.46) compared to children who underwent repair at 7-12 months of age. The hazard of secondary surgery for children who underwent primary lip repair at 3-6 months of age was not significantly different than the hazard for children who underwent repair at 7-12 months of age (HR 1.05, 95% CI 0.89-1.23).

Among the other features of primary lip repair investigated, postoperative antibiotic use, surgeon procedure volume, hospital procedure volume, and year of repair were not associated with time to secondary surgery. While child's race was associated with time to secondary surgery ($p=0.03$), only children who race was classified as 'other' had a hazard of secondary surgery significantly different than the baseline risk (HR 1.18, 94% CI 1.02-1.36).

From the mixed-effects model, we estimated the remaining variation in time to secondary surgery attributable to surgeons, hospitals, and children (Yang et al., 2009). Differences in care delivery between surgeons accounted for 13.6% of variation in secondary surgery ($p<0.01$); differences in care delivery between hospitals accounted for 17.0% of the variation in secondary surgery ($p<0.01$); and differences between children accounted for 69.4% of variation in secondary surgery ($p<0.01$).

Variation Among Surgeons

For each surgeon performing at least ten repairs during the study period, the proportion of each surgeon's patients undergoing secondary lip surgery is shown in Figure 1A. These curves display the raw, unadjusted values as a function of time from primary lip repair. As time from primary lip repair increased, the proportion of children who have undergone secondary surgery also increased. Among the entire study population, 24.0% of children underwent secondary surgery by five years after primary lip repair. However, this ranged from 0% to 100% across surgeons.

Using results from the mixed-effects time-to-event model described above, adjusted time to secondary surgery curves were created for each surgeon (Figure 1B). These curves show the predicted proportion of children undergoing secondary surgery for white males with an isolated unilateral cleft lip who undergo primary lip repair at 3-6 months of age at a medium-volume hospital. The curves were adjusted for hospital performance, such that each surgeon's curve was generated assuming the primary lip repair was performed at a hospital whose hazard of secondary surgery was at the median of all hospitals in the study. Using these adjusted values and the standardized patient as described, the proportion of children undergoing secondary surgery by five years ranged from 4.9% to 21.8% across surgeons (Figure 1C). For the median surgeon in the study, the predicted proportion of children undergoing secondary surgery by five years was 10.6%.

Variation Among Hospitals

For each hospital in the study cohort, the proportion of their patients undergoing secondary lip surgery is shown in Figure 2A. These curves display the raw, unadjusted values, as a function of time from primary lip repair. The proportion of children undergoing secondary

surgery by five years after primary lip repair ranged from 0% to 49.2% across the forty-nine hospitals.

Adjusted time to secondary surgery curves were created for each hospital (Figure 2B) using methods analogous to those described above for surgeons. These curves were adjusted for surgeon performance, such that each hospital's curve was generated assuming the surgeon performing the primary repair had a hazard of secondary surgery at the median of all surgeons in the study. Using these adjusted values and the standardized patient as described, the proportion of children undergoing secondary surgery by five years ranged from 4.5% to 24.7% across the forty-nine hospitals (Figure 2C). At the median hospital in the study, the predicted proportion of children undergoing secondary surgery by five years was 10.6%.

Figure 3 presents a funnel plot of the predicted portion of children undergoing secondary surgery within five years of primary lip repair against the number of children undergoing primary lip repair at each hospital during the observation period. In total, 37 (75.5%) hospitals fall within the 99.8% control limits, seven (14.3%) above the upper, and five (10.2%) below the lower 99.8% control limits.

Discussion

In this study, we found broad variation in the use of secondary lip surgery. Use of secondary lip surgery was associated with a child's cleft type, presence of multiple congenital anomalies, and a child's age at primary cleft lip repair. However, the largest variation in secondary surgery use was attributable to unexplained differences among the surgeons and hospitals performing primary lip repair: the proportion of children receiving secondary lip surgery varied four-fold across surgeons and it varied five-fold across hospitals. Of the 49 hospitals in this sample, seven hospitals (14.3%) had an incidence of secondary lip surgery substantially higher than the group as a whole and five hospitals (10.2%) had an incidence substantially lower than the group as a whole. These results suggest that surgeon- and hospital-factors have substantial impact on a child's risk of secondary lip surgery.

The findings in this study are consistent with and extend those from prior reports. We found that children who underwent primary cleft lip repair before 3 months of age had an increased hazard of secondary lip surgery, compared to repair at 7-12 months of age. This is consistent with the study by Weatherley-White et al. (1987) who reported greater than 40% of patients undergoing repair before three months of age required revision surgery, and with a recent study by Lee et al. (2018) which found that cleft lip repair in neonates was associated with an increased length of stay and an increased risk of complications. The specific cause for an increased hazard of secondary surgery in children receiving primary cleft lip repair before 3 months of age remains unclear, although it may be due to difficulty aligning anatomic landmarks on smaller children. Given the consistent finding that lip repair before three months of age is associated with more peri-operative complications and higher secondary surgery rates, surgeons and hospitals may want to consider delaying primary repair until children are at least three months of age.

The variation in secondary lip surgery usage across surgeons and hospitals observed in this study is similar to prior reports. The Americleft group reported secondary lip surgery rates varied 5% to 60% across four centers (Sitzman et al., 2015), while the Eurocleft group reported rates varied 4% to 69% across five centers (Semb et al., 2005b). A systematic review of secondary lip surgery found rates varied 0% to 100% among published reports (Sitzman et al., 2016). This review found that in large cohort studies with at least one year of follow-up, secondary surgery rates ranged 24% to 36% (Abyholm et al., 1981; Henkel et al., 1998; Mackay et al., 1999; Bongaarts et al., 2006), and that is consistent with the present study's finding that the overall secondary surgery rate was 24.0%. These results suggest that the variation in secondary lip surgery identified in smaller cohorts does indeed represent widespread variation in secondary lip surgery across all US cleft teams.

The variation in secondary lip surgery found in this study is similar in magnitude to the five-fold variation in secondary palate surgery across surgeons and hospitals (Sitzman et al., 2018). Variation across providers and hospitals in secondary surgery occurrence is also consistent with previous studies reporting variation among cleft centers in their facial appearance, facial growth, and speech outcomes (Asher-McDade et al., 1992; Mars et al., 1992; Brattstrom et al., 2005; Semb et al., 2005b; Daskalogiannakis et al., 2011; Britton et al., 2014; de Agostino Biella Passos et al., 2014; Lithovius et al., 2014). These reports suggest that understanding and reducing variation in treatment outcomes and care delivery may be an effective approach for improving cleft care.

Differences in the use of secondary lip surgery among surgeons and hospitals are multifactorial. At the surgeon level, both surgical technique and surgeon expertise may influence the occurrence of facial asymmetry necessitating secondary surgery. The evidence for an association of surgical technique is limited. There is a paucity of randomized controlled trials on cleft lip repair technique, likely due to the challenges in executing such a trial (Hardwicke et al., 2017; Bekisz et al., 2018). There have been numerous prospective and retrospective studies comparing techniques for primary cleft lip repair, but they have all had substantial methodologic limitations such as comparing surgical techniques applied over two distinct time periods (Reddy et al., 2010; Zaleckas et al., 2011), techniques applied by different surgeons (Adetayo et al., 2018), and changes in technique concomitant with changes in use of pre-surgical nasolabial orthopedics. Given the limited evidence to support one technique of cleft lip repair over another, it seems likely that the surgeon-level variation in secondary surgery is due to more than general technique of lip repair.

In the present study, we evaluated the effect of individual surgeons without separating the effect of surgical technique from surgeon expertise. This is perhaps the most appropriate level for analysis, since surgeons routinely individualize techniques based on personal experience. We found that use of secondary lip surgery varied four-fold among surgeons – far outpacing the effect of cleft type, presence of additional anomalies, and age at lip repair. Future studies are necessary to identify the sources for this variation, in particular whether the variation is due to differences in outcomes of primary repair or differences in thresholds for offering secondary surgery. Much of the decision making regarding secondary lip surgery can be affected by a surgeon's threshold to offer secondary surgery, and it is critical to recognize that variation among surgeons in use of secondary surgery may be due to

differences in threshold for offering secondary surgery rather than differences in outcomes of primary surgery.

The differences in secondary lip surgery observed across hospitals may be attributable to perioperative items such as anesthesia expertise, nursing care, or post-operative care instructions. Differences across hospitals may also be attributed to more long-range components of care, such as treatment protocols or hospital-specific thresholds for offering secondary lip surgery. Given the present study's finding that 24.5% of hospitals perform secondary surgery at a rate substantially different from the group as a whole, it is imperative that further research be conducted to identify the causative factors of between-hospital differences and then use this knowledge to spread best practices.

The present analysis found no association of secondary surgery with the use of post-operative antibiotics after primary lip repair, year of primary lip repair, and procedure volume of the surgeon and hospital. The absence of an association between postoperative antibiotic use and secondary surgery in this study, when combined with findings from Schonmeyr et al. (2015) that wound infection remained low even without postoperative antibiotic therapy, suggests that postoperative antibiotics may not be necessary following primary lip repair. The absence of association with year of primary lip repair suggests there has been no substantial change in use of revision surgery for children treated over the study period of 1998 to 2015. The absence of a volume-outcome relationship may be due to the paucity of very-low-volume operators included in this study (Williams et al., 1999; McAteer et al., 2013). These null findings suggest that among surgeons and hospital who routinely perform cleft lip repair, a deeper exploration of care delivery is necessary to understand the factors contributing to the broad variation in surgical outcomes.

The major implication of this work is that surgeons and hospitals may be able to reduce the need for secondary lip surgery by decreasing variations in care. We found that differences in care delivery between surgeons accounted for 13.6% of the variation in secondary surgery and differences in care delivery between hospitals accounted for 17.0% of variation in secondary surgery. As a result, we suggest that surgeons and hospitals could improve patient outcomes by identifying the causes of variation among surgeons and hospitals, establish evidence-based interventions and achievable targets for performance, and then implementing these interventions in clinical practice (Crandall et al., 2011; Anderson et al., 2014; Britto et al., 2018). One immediate example for this approach is provided by this study's finding that performing primary cleft lip repair before three months of age is associated with a 1.22-fold increase in the hazard of secondary lip surgery. Surgeons and hospitals could use this information to delay primary lip repair until at least three months of age, without negatively impact aesthetic outcomes (Goodacre et al., 2004).

While differences among surgeons and hospitals contribute to a substantial proportion of variability in second lip surgery, this study also found that 69.4% of variation in secondary lip surgery was due to differences among patients. Because many patient-specific factors are not modifiable, such as cleft type and socioeconomic status, efforts to improve cleft care delivery may be more effective if they focus on modifiable system characteristics such as differences in care delivery among surgeons or hospitals (Institute of Medicine, 2001).

Limitations

These results must be interpreted in the context of the study design. No conclusions can be drawn about aesthetic, functional, or quality of life outcomes from this study because these data are not present in the dataset. Unfortunately, no publicly available dataset in the US contains this data. Misclassification bias from coding inaccuracies in cleft type may bias results towards the null hypothesis. The presence or absence of a concomitant cleft palate could not be reliably determined from the dataset, preventing the comparison of secondary surgery between children with isolated cleft lip and those with cleft lip and palate. However, it is worth noting that two previous studies have failed to identify a difference in secondary lip surgery incidence between these two types of cleft (Mackay et al., 1999; Heliovaara and Rautio, 2011). In addition, no information was available on the extent of revision – complete takedown and re-repair versus limited vermilion adjustments or scar revisions.

Because the PHIS database does not distinguish cleft lip adhesion from cleft lip repair, we assumed if a child underwent cleft lip repair twice during their first year of life that the first surgery was a lip adhesion and the second surgery was the primary lip repair. This assumption may not be accurate in all cases. If a child had surgery twice in their first year of life but the first surgery was actually a lip repair rather than an adhesion, then we underestimated time to secondary surgery. If a child had a lip adhesion in the first year of life but the primary lip repair was not performed until after the first year of life then we overestimated time to secondary surgery for the child. The overall effect of this assumption is difficult to judge, although the effect should be small based on prior research that found only 4% of surgeon routinely perform lip adhesion in patient with complete unilateral cleft lip (Sitzman et al., 2008).

Referral of more complex patients to specific hospitals or surgeons within the study cohort could explain between-hospital and between-surgeon variation, although that is unlikely given the non-overlapping referral patterns of most participating hospitals (Fieldston et al., 2013). Finally, we may have underestimated time to secondary surgery for censored individuals. We classified children as lost to follow-up at their last visit to the initial treating hospital. If these children went on to receive secondary lip surgery at another institution after this visit, then results in the present study may be biased toward the null hypothesis. As a whole, these limitations may have led to underestimation of secondary surgery risk in this study.

An additional limitation to this study is that it does not identify the factors responsible for variations across surgeons and hospitals. The PHIS database, from which the data for this analysis were obtained, does not provide sufficient granularity on individual procedures or surgeons to evaluate the effect of surgical technique, treatment protocols, surgeon training, surgeon experience, or surgeon skill. Further, data was not available to determine whether variation in secondary lip surgery was due to differences in outcomes of primary surgery or differences in threshold for offering secondary lip surgery. This work is just a first step toward understanding the use of secondary surgery, and further research is still necessary to identify the additional factors that contribute to variation in use of secondary surgery.

Conclusions

Twenty-four percent of children with cleft lip undergo secondary lip surgery. However, there is substantial variation in secondary surgery based on child-, surgeon-, and hospital-specific factors. After controlling for cleft type and age at primary repair, there remains a four-fold difference in secondary surgery use across surgeons, and an independent five-fold difference in secondary surgery use across hospitals. These results suggest that there is substantial opportunity to reduce the use of secondary surgery by identifying the causes of variation among surgeon and hospitals, establish evidence-based interventions and achievable targets for performance, and then implementing these interventions in clinical practice.

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Figure 1. Time to revision lip surgery for individual surgeons.

Each line represents the proportion of a surgeon's patients who have undergone revision surgery. A, Unadjusted Kaplan-Meier curves for time until revision surgery. Solid black line indicates outcome for all children in the cohort. B, Adjusted time to revision surgery curves for white males with an isolated unilateral cleft lip who undergo primary lip repair at 3-6 months of age at a medium-volume hospital. Solid black line indicates result for median surgeon in the cohort. C, Proportion of children who undergo revision surgery within five years of their primary lip repair, adjusted for as in panel B. Box represents median and interquartile range, whiskers represent upper and lower adjacent values as defined by Tukey (1977)

Figure 2. Time to revision lip surgery for individual hospitals.

Each line represents the proportion of a hospital's patients who have undergone revision surgery. A, Unadjusted Kaplan-Meier curves for time until revision surgery. Solid black line indicates outcome for all children in the cohort. B, Adjusted time to revision surgery curves for white males with an isolated unilateral cleft lip who undergo primary lip repair at 3-6 months of age by a medium-volume surgeon. Solid black line indicates result for median hospital in the cohort. C, Proportion of children who undergo revision surgery within five years of their primary lip repair, adjusted for as in panel B. Box represents median and interquartile range, whiskers represent upper and lower adjacent values as defined by Tukey (1977)

Figure 3. Standardized rates of revision lip surgery for each hospital.

Rates are the proportion of children undergoing revision surgery within five years of their primary lip repair. Rates were adjusted for patient and surgeon risk factors, and represent results for white males with an isolated unilateral cleft lip who undergo primary lip repair at 3-6 months of age by a medium-volume surgeon. Dashed lines indicate 99.8% (~3 sigma) control limits. Hospitals outside the control limits can be interpreted as deviating significantly from the overall rates.

Table 1.

Characteristics of patients and the care delivered at their initial lip repair.

Characteristic	n (%)
Total	14,798
Male gender	9,323 (63.0)
Race	
White	10,278 (69.5)
Black	1,222 (8.3)
Asian or Pacific Islander	542 (3.7)
Other	2,023 (13.7)
Not specified	733 (5.0)
Median annual household income of postal code	
\$33,525 or less (<1.5 FPL ¹)	3,235 (21.9)
\$33,526 - \$44,700 (1.5-2 FPL)	3,845 (26.0)
\$44,701 - \$67,050 (2-3 FPL)	3,884 (26.2)
\$67,051 or more (>3 FPL)	1,117 (7.5)
Not specified	2,717 (18.4)
Cleft lip type	
Unilateral	10,662 (72.1)
Bilateral	2,977 (20.1)
Not specified	1,159 (7.8)
Additional congenital anomalies present	3,968 (26.8)
Patient age at repair	
<3 months	3,120 (21.1)
3-6 months	10,093 (68.2)
7-12 months	1,585 (10.7)
Postoperative antibiotic use	
None	8,086 (54.6)
Yes	6,712 (45.4)
Surgeon procedure volume (on day of repair)	
Low (<10 repairs in preceding 12 months)	4,559 (30.8)
Medium (10-25)	6,993 (47.3)
High (>25)	3,246 (21.9)
Hospital procedure volume (on day of repair)	
Low (<25 repairs in preceding 12 months)	4,096 (27.7)
Medium (25-50)	8,270 (55.9)
High (>50)	2,432 (16.4)

¹FPL, US Federal Poverty Level for a family of four

Table 2.

Adjusted hazard ratios for secondary lip surgery.

Risk Factor	<u>Secondary Lip Surgery¹</u>	
	Hazard Ratio	p-value
Gender		0.11
Male	Reference	
Female	1.08 (0.98-1.18)	
Race		0.03
White	Reference	
Black	0.87 (0.73-1.04)	
Asian or Pacific Islander	1.20 (0.92-1.57)	
Other	1.18 (1.02-1.36)	
Median family income		<0.01
\$33,525 or less (<1.5 FPL ²)	Reference	
\$33,526 - \$44,700 (1.5-2 FPL)	0.94 (0.81-1.08)	
\$44,701 - \$67,050 (2-3 FPL)	1.05 (0.91-1.21)	
\$67,051 or more (>3 FPL)	1.09 (0.88-1.34)	
Not specified	1.66 (1.26-2.19)	
Cleft lip type		0.05
Unilateral	Reference	
Bilateral	0.88 (0.78-0.98)	
Not specified	0.90 (0.76-1.07)	
Additional anomalies present		<0.01
None	Reference	
Yes	0.77 (0.68-0.87)	
Patient age at repair		0.03
<3 months	1.22 (1.02-1.46)	
3-6 months	1.05 (0.89-1.23)	
7-12 months	Reference	
Postoperative antibiotic use		0.06
None	Reference	
Yes	1.03 (0.92-1.15)	
Surgeon procedure volume ³		0.68
Low (<10)	Reference	
Medium (10-25)	0.97 (0.85-1.10)	
High (>25)	1.03 (0.85-1.24)	
Hospital procedure volume ⁴		0.07
Low (<25)	Reference	
Medium (25-50)	0.97 (0.84-1.12)	
High (>50)	1.25 (0.97-1.61)	

¹Model accounts for clustering of patients within surgeons and clustering of surgeons within hospitals; $p < 0.001$ for likelihood-ratio tests of $\theta = 0$ for both surgeon and hospital

²FPL, US Federal Poverty Level for a family of four

³Procedure volume is the number of cleft palate repairs performed by that surgeon (or hospital) in the preceding twelve months

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