

Geospatial Analysis of Risk Factors Contributing to Loss to Follow-up in Cleft Lip/Palate Care

Banafsheh Sharif-Askary, BS*
 Peter G. Bittar, BS, BA*
 Alfredo E. Farjat, PhD||
 Beiyu Liu, PhD†
 Joao Ricardo Nickenig Vissoci,
 PhD‡§¶
 Alexander C. Allori, MD, MPH*

Background: Multidisciplinary cleft care depends on follow-up at specified time points to monitor and address functional or aesthetic concerns that may arise during a child's development. However, loss to follow-up (LTFU) is common and can lead to missed opportunities for therapeutic and surgical intervention. This study explores clinical, demographic, and geographic determinants of LTFU in cleft care.

Methods: Medical records were retrospectively evaluated for 558 pediatric patients of a single mid-volume cleft team. The primary outcome was LTFU. Spatial dependency was evaluated using variograms. The probability of LTFU was assessed using a generalized linear geostatistical model within a Bayesian framework. Risk maps were plotted to identify vulnerable communities within our state at higher risk of LTFU.

Results: Younger age at last encounter was a strong predictor of LTFU ($P < 0.0001$), even when ignoring spatial dependency among observations. When accounting for spatial dependency, lower socioeconomic status [OR = 0.98; 95% CI = (0.97–0.99)] and cleft phenotype [OR = 0.55; 95% CI = (0.36, 0.81)] were significant predictors of LTFU. Distance from the cleft team and rural/urban designation were not statistically significant predictors. Cartographic representation of predicted probability of LTFU revealed vulnerable communities across our state, including in the immediate vicinity of our cleft center.

Conclusions: Geostatistical methods are able to identify risk factors missed by traditional statistical analysis. Knowledge of vulnerable populations allow a cleft team to allocate more resources toward high-risk areas to rectify or prevent deficiencies in care. (*Plast Reconstr Surg Glob Open* 2018;6:e1910; doi: 10.1097/GOX.0000000000001910; Published online 14 September 2018.)

INTRODUCTION

Cleft lip and/or palate is one of the most common birth defects and, due to its complexity, best cared for by a multidisciplinary team dedicated to cleft care.¹ Team care

is largely protocol-based and includes a series of diagnostic and therapeutic interventions at prescribed time points that span longitudinally from birth to young adulthood (ie, a “care plan”). Many necessary operative interventions are coordinated so as to minimize the number of anesthetic events required (eg, myringotomy and tympanostomy tube placement at the time of cleft palatoplasty).

In a preliminary quality-improvement project,² we identified several factors associated with deviation from the ideal care plan: (1) transfer to the team at older age (eg, migration or adoption); (2) medical or surgical complication necessitating an alternative care pathway; (3) poor communication between team members, leading to uncoordinated services; (4) patient/parent noncompliance with instructions; and (5) missed/cancelled appointments resulting in loss to follow-up (LTFU). Additional stressors were postulated to predispose to these points of failure, including low socioeconomic status (SES), rural isolation, and distance from our cleft team.^{3–5}

Based on these preliminary findings, we designed the present study to explore the factors contributing to LTFU in cleft care. We hypothesized that poor SES would be

From the *Department of Surgery, Division of Plastic, Maxillofacial and Oral Surgery, Duke University Medical Center, Durham, N.C.; †Department of Biostatistics and Bioinformatics, Duke University Medical Center, Durham, N.C.; ‡Duke Global Health Institute, Duke University, Durham, N.C.; §Division of Emergency Medicine, Department of Surgery, Duke University Medical Center, Durham, N.C.; ¶Duke Global Neurosurgery and Neuroscience, Department of Neurosurgery, Duke University Medical Center, Durham, N.C.; and ||Thrombosis Research Institute, London, United Kingdom.

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the principal risk factor for LTFU. Given prior work examining geospatial dependency of health outcomes, we hypothesized that the relationship between LTFU in cleft care would be spatially dependent.⁶ We also hypothesized that distance from our cleft team would be positively correlated with LTFU, that is, patients living further from our team were at greater risk of LTFU. To test these hypotheses, we used geostatistical methods to investigate the degree to which location and other geospatial characteristics are related to LTFU. This approach enabled a population-based appraisal of team performance in a manner specific to the team’s geographic region. Ultimately, this analysis will inform cleft teams on how to better serve their regions by allocating resources to high-risk areas so that deficiencies in care provision may be rectified.

METHODS

Study Population

This study was approved by the institutional review board at Duke University. Cleft patients diagnosed and treated at Duke University Hospital between 1998 and 2013 were eligible for inclusion. Exclusionary criteria were Robin sequence, craniofacial syndromes, and other major comorbidities or congenital abnormalities (ie, cardiac, urogenital, etc.) that required operative care beyond standard cleft care.⁷ As this study focused on cleft-care provision within our home state, patients living outside of North Carolina were excluded from analysis. There were no exclusionary criteria based on age, race, phenotype, or SES.

Primary Outcome: LTFU

The primary outcome of interest was LTFU. This was defined using the definition provided by the International Consortium for Health Outcomes Measurement Standard Set of Outcome Measures for Cleft Lip/Palate: either as 3 consecutively missed (“no-show”) appointments (despite documented attempts at reestablishing follow-up

via telephone call or letter) and/or greater than 2.5 years without returning to see the team. This definition was compatible with the practice of our team as we see all active patients at 6-month, 12-month, 18-month, or 24-month frequencies, depending on clinical context. This plan is clearly documented in the chart by the team coordinator, as are cancelled/rescheduled and missed/“no-show” appointments. Patients were not considered LTFU if they cancelled and rescheduled an appointment within the next 6 months of an intended encounter, if they were officially discharged from team care, or if they transferred care to other cleft teams.

Independent Variables (Predictors)

The electronic health record for each subject was retrospectively reviewed by 2 independent clinical reviewers and abstracted for clinical, demographic, and geographic variables (Table 1).

Clinical phenotype was designated using 4 categories: cleft lip only (CL); cleft palate only (CP); cleft lip and alveolus (CLA); and cleft lip, alveolus, and palate (CLAP).^{8,9}

Age at first and last clinical encounter with the cleft team was calculated from administrative data and represented in years.

The following geographic variables were collected: patient residential street address, city, state, primary postal code, and county. The latitude and longitude of an address were determined using the World Geodetic System 84 DATUM. Rural versus urban designation was determined at the census-block level based on U.S. Census classification and coded as a binary variable.¹⁰ Distance from the Duke cleft team was measured as Euclidean distance in kilometers.

SES index was a representative assignment based upon the patient’s address and the Agency for Healthcare Research and Quality algorithm.¹¹ This index is a weighted aggregate of metrics describing the region surrounding the subject’s home address: percentage of households containing 1 or more person per room; median value of owner-occupied homes; percentage of

Table 1. Demographics and Clinical Characteristics of Cleft Lip and/or Palate Patients

Variable	Total (n = 502)	Lost to Follow-up*		P
		No (n = 338)	Yes (n = 164)	
Sex, n (%)				0.923
Female	214 (42.6)	145 (42.9)	69 (42.1)	
Male	288 (57.4)	193 (57.1)	95 (57.9)	
Race, n (%)				1.000
White	354 (70.5)	238 (70.4)	116 (70.7)	
Non-White	148 (29.5)	100 (29.6)	48 (29.3)	
Socioeconomic index	52.2 (±9.3)	52.8 (±8.4)	51.0 (±10.7)	0.059
Age at first encounter (y)	1.6 (±2.7)	1.8 (±3.0)	1.2 (±1.9)	0.835
Age at last encounter (y)	5.0 (±3.7)	5.8 (±3.8)	3.4 (±2.9)	< 0.0001
Phenotype, n (%)				0.0001
CL	64 (12.7)	37 (10.9)	27 (16.5)	
CLA	37 (7.4)	23 (6.8)	14 (8.5)	
CLAP	221 (44.0)	172 (50.9)	49 (29.9)	
CP	180 (35.9)	106 (31.4)	74 (45.1)	
Distance to Duke (km)	114.8 (±103.1)	117.2 (±104.0)	109.7 (±101.4)	0.677
Location type, n (%)				0.619
Rural	176 (35.1)	116 (34.3)	60 (36.6)	
Urban	326 (64.9)	222 (65.7)	104 (63.4)	

*Three consecutive missed appointments or 2.5 years without seeing the team despite attempts at reestablishing follow-up. Patients who transferred care to other teams were not considered LTFU.

Table 2. Results of a Multivariate Logistic Regression Analysis for LTFU as a Function of Selected Predictors (Ignoring Spatial Dependency)

Effect	Estimate	SE	OR	2.5%	97.5%	P
Intercept	1.294	0.663	3.648	0.996	13.370	0.051
Sex						
Male	0.149	0.211	1.161	0.768	1.754	0.479
Race						
Non-White	-0.117	0.236	0.890	0.560	1.413	0.620
Socioeconomic index	-0.016	0.011	0.984	0.963	1.006	0.146
Age at first encounter (y)	0.070	0.052	1.072	0.967	1.189	0.183
Age at last encounter (y)	-0.206	0.039	0.814	0.754	0.879	< 0.0001
Phenotype (reference: CL)						0.063
CLA	-0.062	0.449	0.939	0.390	2.264	0.889
CLAP	-0.572	0.319	0.564	0.302	1.056	0.073
CP	0.022	0.316	1.022	0.550	1.899	0.945
Distance to Duke (km)	-0.001	0.001	0.999	0.997	1.001	0.285
Location						
Urban	-0.094	0.222	0.910	0.590	1.405	0.671

persons below the federally defined poverty line; median household income; percentage of persons aged > 25 years with at least 4 years of college; percentage of persons aged > 25 years with less than a 12th grade education; and percentage of persons aged ≥ 16 years in the labor force who are unemployed.

Statistical Analyses

Continuous variables were summarized with means and SDs. Categorical variables were reported using frequency counts and percentages. Patient characteristics and demographics were compared using the Mann-Whitney *U* test (continuous variables) and the Fisher exact test (categorical variables).

First, traditional statistical modeling was performed, initially ignoring spatial dependency. Using multivariate logistic regression, the probability of LTFU was modeled as a function of the independent variables (eg, clinical, demographic, and geographical characteristics; Table 2). The CL phenotype served as the reference group in the multivariate logistic regression analyses. This was chosen because the CL phenotype requires the least follow-up, with most patients being formally discharged from team care approximately 3 years after cleft repair.

Next, empirical variograms were used to evaluate the spatial dependency of both the primary outcome (LTFU) and the explanatory or independent variables (ie, age, sex, race/ethnicity, cleft phenotype, SES, and rural/urban designation; Fig. 1).

Following evaluation of spatial dependency, a generalized linear geostatistical model (GLGM) was fit to the data to account for the spatial dependency among the observations through the inclusion of spatial covariate effects and an exponential correlation structure. A Bayesian approach was used to estimate the model parameters and to perform the subsequent statistical inference. The binary response variable was LTFU, and the predictors were the same as in Table 2. (SDC1) (See table, **Supplemental Digital Content 1** which describes the Generalized Linear Geostatistical Model. <http://links.lww.com/PRSGO/A902>).

Predicted probability maps of LTFU occurrence were produced based on the observed data considering SES index, age at last encounter, and cleft phenotype.

Predictions were obtained at a regular grid across North Carolina using the mean of the posterior distribution. To visualize the obtained predictions, a smooth surface was fit using a bivariate interpolation method.^{12,13–16}

All statistical analyses were conducted using R statistical software (R Core Team, 2016).¹⁷ Significance was assessed at level $\alpha = 0.05$.

RESULTS

Five hundred and two patients seen by the Duke cleft team between 1998 and 2013 were eligible for inclusion. Of these, 164 (32.7%) were classified as LTFU, whereas 338 (67.3%) were not LTFU. Patient clinical, demographic, and geographical characteristics were reported by LTFU status (Table 1). There was a significant difference in phenotypic distribution between the LTFU and non-LTFU groups. Otherwise, patients in the 2 groups did not differ by sex, race, age at first encounter, distance to Duke Hospital, or rural/urban designation.

LTFU Was Spatially Dependent within a Radius of 2 km

The geographic pairwise distance differences between observations suggested that the majority of neighbors were located within a radius of about 100 km from each other. The shape of the empirical variograms of LTFU status as a function of spatial lag indicated the existence of spatial dependency in the observations. The posterior mean of parameter range [$\varphi = 2.01$; 95% CI = (0.08–7.52)] is expressed in kilometers and indicates that correlation between observations considerably decreases after approximately 2 km (Fig. 1). The spatial variance [$\sigma = 0.61$; 95% CI = (0.26–1.41)] is associated with the spatial uncertainty around the observations.

LTFU Was Strongly Associated with Younger Age at Last Cleft Team Encounter

The average age at last cleft team encounter for patients in the non-LTFU group was 5.8 ± 3.8 years compared with 3.4 ± 2.9 years in the LTFU group. When ignoring spatial dependency, older age at last encounter was found to be significantly protective against LTFU [OR = 0.814; 95% CI = (0.75–0.88); $P < 0.0001$].

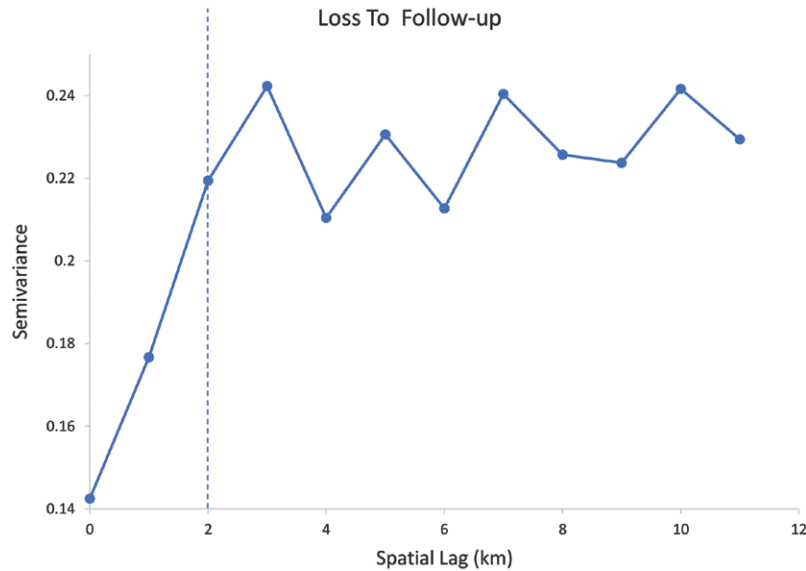


Fig. 1. Empirical variograms of LTFU as a function of spatial lag. The semivariance (vertical-axis) is a measure of spatial dissimilarity between all pairs of values. It may be understood as opposite to correlation, which measures the degree of similarity between observations. As the correlation between observed values decreases, the semivariance increases with increasing separation distance. The shape indicates the existence of a 2-km spatial lag in the observations. In other words, observations that are spatially closer within a range of 2 km are more similar than observations that are farther apart (> 2 km).

For the geostatistical analysis, posterior mean, SD, odds ratio, and 95% odds ratio credible set for the regression coefficients of the GLGM are shown in Table 3. The spatial model indicates that older age at last encounter remained a highly significant protective factor against LTFU [OR = 0.81; 95% CI = (0.77–0.84)]. In contrast, older age at first encounter was not associated with LTFU at 5% significance level [OR = 1.07; 95% CI = (1.01–1.15)]. Older age at last cleft team encounter was reflective of both later phases of treatment and a better track record of prior successful follow-up with the team (ie, pattern of successful annual visits in previous years).

LTFU Was Strongly Associated with Lower SES Index

Univariate analysis indicates that LTFU patients had lower SES index (51.0 ± 10.7 versus 52.8 ± 8.4 ; $P = 0.059$).

Multivariate logistic regression while ignoring spatial dependency showed an insignificant association between SES and LTFU [OR = 0.98; 95% CI = (0.96–1.01); $P = 0.15$]. However, when incorporating geospatial dependency in the model, SES index was noted to be significantly associated with LTFU [OR = 0.98; 95% CI = (0.97–0.99)]. This is in marked contrast with the weak association noted without consideration of spatial dependency.

LTFU Was Weakly Associated with Phenotype

Phenotypic distribution for patients in the non-LTFU group was as follows: 37 (10.9%) CL, 23 (6.8%) CLA, 172 (50.9%) CLAP, and 106 (31.4%) CP. In comparison, the LTFU group had the following phenotypic distribution: 27 (16.5%) CL, 14 (8.5%) CLA, 49 (29.9%) CLAP, and 74 (45.1%) CP. First, we performed an exploratory analysis

Table 3. Results of a GLGM for LTFU as a Function of Selected Predictors

Effect	Mean	SD	OR	2.5%	97.5%
Intercept	1.3584	0.4208	3.8901	1.7297	9.1228
Sex					
Male	0.1611	0.1328	1.1748	0.9068	1.5326
Race					
Non-White	-0.1226	0.1489	0.8846	0.6578	1.1855
Socioeconomic index	-0.0168	0.0070	0.9833	0.9696	0.9966
Age at first encounter (y)	0.0717	0.0325	1.0744	1.0085	1.1468
Age at last encounter (y)	-0.2145	0.0252	0.8070	0.7658	0.8457
Phenotype (reference: CL)					
CLA	-0.0619	0.2830	0.9399	0.5365	1.6349
CLAP	-0.5993	0.2013	0.5492	0.3651	0.8142
CP	0.0200	0.1991	1.0202	0.6866	1.5114
Distance to Duke (km)	-0.0012	0.0007	0.9988	0.9975	1.0001
Location					
Urban	-0.0977	0.1391	0.9069	0.6900	1.1908

to determine whether all-cause attrition (both planned discharges and unanticipated, unexplained LTFU) differed across phenotype (SDC2). (See figure, Supplemental Digital Content 2 which displays the Kaplan-Meier plot depicting all-cause attrition for each phenotype. <http://links.lww.com/PRSGO/A903>).

Next, we performed a standard multivariate regression, ignoring geospatial dependency, and found phenotype to be weakly predictive of LTFU ($P = 0.06$). However, when including spatial dependency, phenotype became a strong predictor: The CLAP phenotype was significantly protective against LTFU [OR = 0.55; 95% CI = (0.36, 0.81)], whereas the CLA and CP phenotypes were not significantly associated with LTFU [CLA: OR = 0.94, 95% CI = (0.54–1.63); CP: OR = 1.02, 95% CI = (0.69–1.51)].

Distance to the Hospital and Urban/Rural Environment Were Not Significant Predictors of LTFU

Interestingly, and contrary to expectations, patients living far from Duke did not have an elevated risk of LTFU compared with those living close to the hospital [OR = 1.00; 95% CI = (1.00–1.00)]. Urban designation exhibited slightly lower risk of LTFU, but the evidence was not statistically significant [OR = 0.91; 95% CI = (0.69–1.19)]. Geographic risk maps were created using the GLGM geostatistical model with 3 predictors (phenotype, age at last encounter, and SES index; Fig. 2). The maps reveal spatial clustering of LTFU in “pockets” (communities) of high and low risk. These communities existed both far from and close to Duke, emphasizing that while the outcome is geospatially dependent, it is driven by community-based factors and not by distance to the hospital.

DISCUSSION

Cleft lip and/or palate requires long-term, multidisciplinary care delivered by specialized cleft teams. In this

study, we chose to focus on LTFU as a marker of the success of a cleft team’s ability to provide important services to patients in its catchment area. The purpose of this study was to identify characteristics that predispose patients to a higher risk of LTFU, with the hope that this may lead to strategies for improving care to vulnerable populations.

Significance of Spatial Dependency

As depicted by the variograms in Figure 1, there was strong spatial dependency in our primary outcome, LTFU. The results of the standard multivariate regression were first presented to highlight that, when applying the more appropriate geostatistical approach, there are new findings that were not drawn out from standard statistical methods. This proved that standard multivariate logistic regression methods were insufficient (Table 2), and a geostatistical model was required (Table 3).

Previous epidemiological work has applied census-tract-level and postal codes to create neighborhood level representations of health outcomes.^{18–20} These methods, however, presume that spatial units are statistically independent of one another, and are therefore considered unrealistic.²¹ Modeling spatial dependency, on the other hand, examines both correlation between patients in a spatial unit and correlation between spatial units themselves in a region of interest. Positive spatial dependency indicates that health outcomes are more similar to each other in spatial units that are closer together, and less similar in spatial units that are farther apart.²² Intuitively, this makes sense as the spatial dependency of health outcomes, such as LTFU, in nearby spatial units can be attributed to homogeneity in factors such as SES among neighbors.⁶ Additionally, spatial analysis lends itself to practical application; the results directly inform researchers about geographic regions that are at high-risk for LTFU, allowing for community engagement and targeted interventions.

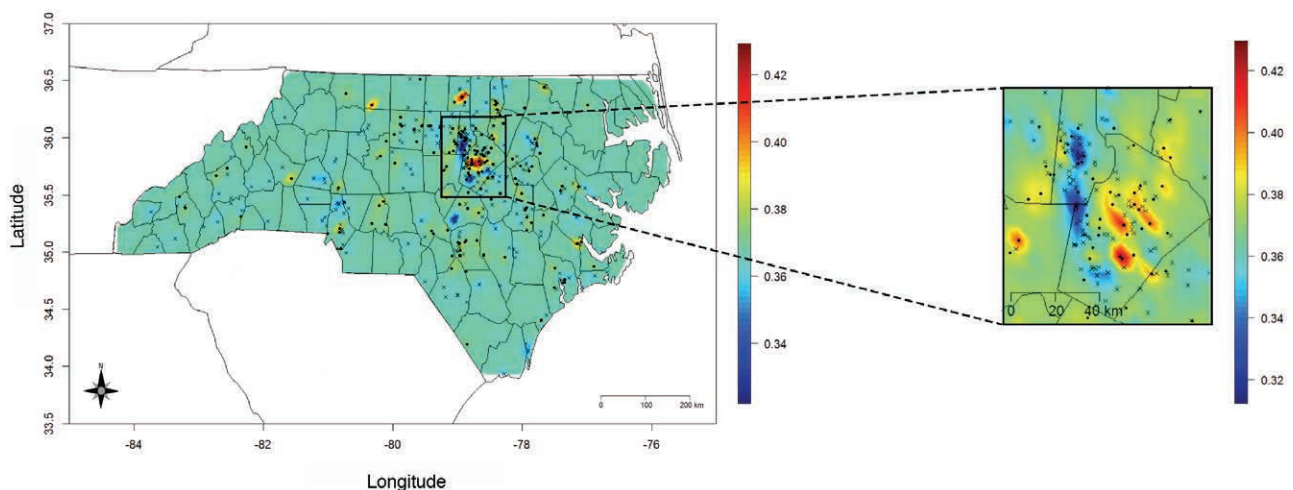


Fig. 2. Predicted probability map of LTFU occurrence across North Carolina (with detail around Duke Hospital) using a GLGM. Only 3 predictors were considered: SES index, age at last encounter, and phenotype. Patients across North Carolina appear to spatially cluster based on LTFU status. Detail: In the immediate vicinity of the Duke Hospital cleft team, there are clusters of both high risk of LTFU and low risk of LTFU patients. This suggests that proximity to Duke Hospital is not predictive of risk of LTFU. Dot, LTFU observation; Cross, No LTFU observation; Red, high risk of LTFU; Blue, low risk of LTFU.

“Spatial Dependency” Was Mostly Defined by Community Factors, Not by Distance

We discovered that areas at highest risk for LTFU existed throughout North Carolina and were even in the immediate vicinity of our cleft team. This was a surprising finding that refuted our hypothesis that risk for LTFU would increase as distance from our hospital increased. Prior studies report that distance to location of care is positively correlated with incidence of LTFU—a phenomenon known as distance decay. A systematic review reported that, of 108 studies, 77% identified evidence of distance decay.³ It is unclear why our data lack an appreciable distance decay, but it may be explained in part by our proximity to other cleft teams. Though we did not designate patients as LTFU if it was known that they were transferring care, not all patients may have disclosed this information. Another explanation may be that, after a certain distance, any additional distance traveled for follow-up care is inconsequential and likely does not affect a patient’s decision to make the trip. It has been shown that in sparsely populated communities, patients are more willing to travel longer distances to appointments,²³ a phenomenon that could apply to cleft care in North Carolina. Finally, it may be that distance is not as strong a determinant of LTFU as means of transportation to the hospital (eg, driving, public transport).

Significance of Socioeconomic Index

The finding that lower SES index was associated with higher risk of LTFU is consistent with existing literature.²⁴ The burdens associated with low SES can affect a patient’s decision to pursue follow-up care in various ways. Office visits can pose a considerable financial burden both directly, due to the cost of travel to the cleft center, and indirectly, due to salary lost from time off work and the cost of child care for siblings.^{25,26} Lower education, a component of the SES index, has also been shown to adversely affect compliance with care recommendations.^{27–30} Parents with less education may have more difficulty understanding the complexities of their child’s condition or need for strategic, long-term follow-up. This finding highlights both the need for more accessible educational materials and the importance of patient education during initial cleft team visits.³¹

Significance of Age

The finding that risk of LTFU is greatest earlier in the timeline of cleft care (eg, younger age at last encounter) was initially counterintuitive, as we originally expected to find LTFU to occur later in the course of care. However, it appears that families at risk for LTFU “declare themselves” quite early in the timeline of care. Another way of viewing this is that families who have followed up for years are more likely to continue doing so, and thus older children have a lower risk of LTFU. A similar phenomenon has been observed in HIV care programs.³² In this sense, older age at the last visit reviewed in this study is reflective of 2 things: (1) later phase of treatment; and (2) a longer track record of successful prior follow-up with the team. Thus, our observation that older age is “protective” against LTFU may be explained by the impact of time on the patient-clinician relationship: Families that successfully attend the

initial clinic visits may have built a robust relationship with the team, witnessed the benefits of longitudinal care, and integrated these visits in their routine. This suggests that the initial cleft team visits are most impactful, and that clinicians should stress the importance of follow-up during these early visits.

Significance of Phenotype

Although some intended differences in team protocols do exist based on phenotype (eg, earlier planned discharges for CL and CP, and longer follow-up for CLA and CLAP), as demonstrated by the all-cause attrition depicted by the Kaplan-Meier curves in Supplemental Digital Content 2, this study was able to clearly demonstrate how phenotype also affects unplanned LTFU. Specifically, CLAP phenotype exhibited the lowest risk of LTFU, which may be explained by the fact that it is the most severe presentation, both visually and in terms of functional impairment, <http://links.lww.com/PRSGO/A836>. Parents may witness higher rates of difficulty feeding in infancy, nasal regurgitation, speech impairment, otologic complications, dental malocclusion, and nasolabial aesthetic concerns.³³ As such, patients with CLAP may be more motivated to seek follow-up to address these concerns.

Limitations

The main limitation in this study is that we included data from patients seen only by 1 cleft team (Duke University Hospital) and not surrounding cleft centers in North Carolina. During the study period, there were 2 other cleft centers approved by the American Cleft Palate–Craniofacial Association, the University of North Carolina-Chapel Hill and Wake Forest hospitals. The exclusion of neighboring institutions makes it impossible to assess whether patients who were considered LTFU at Duke instead established care elsewhere, unbeknownst to our team. This is a topic of future collaborative investigation.

Another limitation is the use of Euclidean distance, rather than travel time, which may have been a more accurate indicator of the effect of distance on LTFU. Euclidean distance was used because it represents the most simple and intuitive approach. Additionally, the use of time traveled in spatial models is constrained by the effect of local features on its calculation.³⁴

CONCLUSIONS

In this exploratory study, LTFU for patients with cleft lip/palate was found to be associated with SES, duration of team contact, and phenotype. Geostatistical methods confirmed the presence of spatial dependency for this outcome and identified specific communities that were at greater risk for LTFU. Geospatial analysis is important to perform when studying provision and utilization of care at the population level.

Alexander C. Allori, MD, MPH
DUMC 3974 – Plastic Surgery
200 Trent Drive at Erwin Road
Durham, NC 27710
E-mail: alexander.allori@duke.edu

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