Incidence of secondary surgery in children with CLP treated before and after standardization of cleft treatment regimes

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Scientific outline

Study Population/Data Source: The data for this retrospective cohort study will be assembled from two cross-sectional studies of all five-year-old children with nonsyndromic complete unilateral CLP in the UK. The pre-standardization cohort is from CSAG and includes all children born 4/1/89 - 3/31/91. The post- standardization cohort is from Cleft Care UK and includes all children born 4/1/05 - 3/31/07. These studies collected information on burden of care and clinical outcomes through direct interview and clinical exam conducted when the children were five years old.

Approach*:* First, we will describe the number of children who received secondary surgeries in each cohort. We will then compare the proportion of children undergoing secondary surgery pre- and post- standardization using a multivariable logistic regression model to adjust for patient gender and race. We will also perform a subgroup analysis comparing the effect among children with good clinical outcomes, defined according to accepted standards as good appearance (Asher-McDade score <3 in all categories) and absence of consistent hyper nasality in speech.

Sample Size and Power: There are 239 children in the pre-intervention cohort and 268 children in the post-intervention cohort. Setting the Type I error rate at 0.05, we have 80% power to detect a 45% reduction in the proportion of children undergoing secondary surgery by age five from an estimated baseline of 20%. This reduction in secondary surgery would create a strong incentive to adopt standardized care in the United States and other developed countries.

Anticipated Results*:* This analysis will determine if standardizing care led to an increase or decrease in the use of secondary surgery. If there was a decrease in secondary surgery and an increase in the odds of achieving a good clinical outcome without secondary surgery, this will provide both financial and clinical incentives to implement standardized care. By

contrast, if the odds of achieving a good clinical outcome without secondary surgery decreased after standardization of care delivery, this would suggest the need for substantive modifications to care delivery protocols in the UK.

Cleft care delivery in the United Kingdom underwent a substantial reorganization during the 1990s, with consolidation of treatment delivery to high-volume surgeons with extensive cleft-specific training, a reduction in the number of cleft teams, implementation of standardized care delivery pathways, and mandatory outcome reporting from each team. Results of the recent CCUK study, when compared to the pre- transformation CSAG study, demonstrated that this service delivery reorganization was associated with overall improvements in facial appearance, speech outcomes and dentoalveolar relationships. While these improvements are laudible, it remains unclear if they are due to improvements in primary cleft surgery, which would suggest improved technical performance by surgeons as the principal causative agent, or to increased used of secondary surgery, which would suggest that increased outcome monitoring and care coordination were essential to the improvements in outcome. To answer this question, we propose a study to compare the proportion of children undergoing secondary lip surgery and secondary palate surgery pre-reorganization (CSAG) and post-reorganization (CCUK). We will also perform a subgroup analysis to determine if the odds of achieving good clinical outcomes, defined as both good appearance and absence of consistent hypernasality in speech, increased following the reorganization. Our results will provide important insights about which factors within the cleft care reorganization led to improvements in clinical outcomes and generate new knowledge about the effect of reorganization on the number of surgical services delivered to each child. These insights will influence efforts to standardize and improve cleft care delivery in other developed countries.