Long-term Clinical and Holistic Outcomes in Children with Cleft Lip and/or Palate: A Multidisciplinary, Mixed-Methods Approach

Irene J. Pien, MD; Danielle L. Sobol, BS; Anna R. Carlson, MD; Alexander C. Allori, MD, MPH; Stephanie Watkins, PhD, MSPH; Arthur S. Aylsworth, MD; Robert Meyer, PhD, MPH; Luiz Pimenta, DDS, PhD; Ronald Strauss, DMD, PhD; Barry Ramsey, BS; Jeffrey R. Marcus, MD

INTRODUCTION: The purpose of this study was to evaluate long-term holistic outcomes for children with cleft lip and/or palate (CL/P) in North Carolina using a multidisciplinary, mixed-methods approach. This entailed cross-sectional evaluation of population-based data, longitudinal evaluation of clinical, and qualitative/semi-quantitative survey-based evaluation of outcomes from the perspective of the patient/parent. Domains assessed include health, appearance, academic performance, behavior, school life and social life, economics, and "burden"/process of care.

MATERIALS AND METHODS: 712 children with isolated CL/P born in North Carolina between 1997-2003 were identified using the NC Birth Defects Monitoring Program; a random sample from 6,822 controls (children without structural birth defects) was also identified. A 200-question survey was prepared by way of expert panel with input from patient focus groups and advisory board and was completed by parental proxy. Corresponding data on academic performance were collected from NC Department of Public Instruction end-of-3rd-grade test scores. Complementary longitudinal clinical data were collected by way of clinical chart review of 167 children with isolated CL/P treated from birth between 2002-2012 at a single institution. Phenotypic classification, surgical interventions, clinical and non-clinical outcomes, and process metrics were evaluated.

RESULTS: Epidemiologic data demonstrated statewide prevalence of cleft lip (CL) to be 27%, cleft palate (CP) 35%, and cleft lip with cleft palate (CL+P) 39%; these results were similar to the experience of the representative institution (CL=25%, CP=29%, CL+P=46%). Survey completion rate was 32% for CL/P and 27% for controls. Nasal congestion and obstruction were significantly more severe and frequent in CL+P compared to control, CL, and CP ($p<0.0001$); unilateral CL+P was more significantly affected. Obstructive sleep apnea was significantly more frequent in CP and CL+P compared to controls and CL ($p<0.0001$). Significant differences existed in patient concern regarding appearance of teeth/smile, nose, and upper lip, but not other parts of the face or body. Clinically, the overall rate of adverse events was 9.4% for minor events, 5.1% for moderate, and 0.9% for severe. Specific findings related to the process of care and other domains will also be presented.

CONCLUSION: This is the first mixed-modality study to provide both clinical and population-based data demonstrating significant functional and aesthetic concerns extending through adolescence. These results offer broad insight into long-term holistic outcomes for children with CL/P.

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