Reviewer’s report

Title: The Effects of Oral Clefts on Hospital Use throughout the Lifespan

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Reviewer: Syed Shahrukh Hashmi

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The manuscript titled "The Effects of Oral Clefts on Hospital Use throughout the Lifespan" by Wehby et al. reported the results from the analysis of a Danish population-based dataset to evaluate the effect of oral clefts on hospital admissions and length of stay once admitted. The authors concluded that based on the results, hospitalizations (both admissions and length of stay) are increased in patients with oral clefts compared to control patients. Overall, the manuscript was well written and the conclusions are supported by the data that has been appropriately analyzed. The study had obvious strengths in the population-based data used for analysis and the large sample size that the dataset provided.

Major Compulsory Revisions: None

Minor Essential Revisions:

1. Authors state in the Introduction that oral clefts are the MOST prevalent birth defect in the US. The MMWR publication that the statement is based on does not include certain defects such as ventricular septal defects, that have been reported elsewhere to be more common. Therefore, the statement should be corrected to reflect that oral clefts are one of the most common defect as opposed to the most common.

2. When describing the empirical models, the authors should clarify if adjustments for repeated measurements were included. For example, during a certain age group (eg. 10-19) one might assume that an affected individual might be hospitalized more than once in different years during that 10-year time frame. Although the "Year" is included in the models and would account for changes in health care over time, it is unclear if and how hospitalizations for the same individual over multiple years within a single age-group were considered.

3. Although the authors considered statistical significance at p<0.01, it should be noted that multiple comparisons were performed overall. In addition, the analysis included a large number of observations, which on its own would influence p-values as well. Together, these two factors may result in statistically significant results. It would be beneficial if the authors discuss the possible inflation of p-values and Type I error in their Discussion section. Also, any discussion should also consider the statistical significance in light of clinical significance. Although I'm convinced that oral clefts infer a burden on health care resources, a statistically significant but low magnitude increase, for example, of 0.4 days per
year for patients with cleft lip with palate in the 20-29 age group, may be a function of multiple comparisons and large sample size.

4. The authors mentioned the limitations due to censoring of data and discuss the implications on the results due to cases that die early. However, the data would also be censored by individuals who migrate out of Denmark. One might assume that those individuals who have higher health care needs due to oral clefts might be limited in their "migration" possibilities (either due to SES factors or due to the fact that health insurance would need to cover them in their new country). Therefore, a relatively "healthy" population (requiring less hospitalization) might be able to afford migration. Is migration out of Denmark an issue that should be considered? If so, a statement regarding the implications should be added.

5. It should be stressed that although "oral clefts" overall do result in an increase in health care utilization, the increase is predominantly due to "cleft lip with palate" and is probably a directly correlated with the severity of the "oral cleft".

6. The tables should reflect if the results reported therein were adjusted or crude. This has been mentioned in the title or footnote of some but not all tables.

7. The authors provide estimates of % changes in hospitalizations and length of stay in their tables. It would be informative for the reader if 95 or 99% confidence intervals for these changes were also included.

8. It was mentioned that detailed regression results are available from the authors upon request. However, the detailed regression results (for both the logistic and poisson models for all age groups) should be included as supplemental data rather than available upon request.

9. There are a few typos in the manuscript that would probably be missed by a spell check (eg. "clef" instead of "cleft"). Another read-over should catch them.

Discretionary Revisions: None.

Level of interest: An article whose findings are important to those with closely related research interests

Quality of written English: Needs some language corrections before being published

Statistical review: No, the manuscript does not need to be seen by a statistician.