

Pre-Analysis Plan: Comprehensive Impact of Cleft Lip and Palate Surgery on Teenage Life Outcomes

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Fieldwork Location and Dates of Fieldwork:

West Bengal, and Andhra Pradesh, India May 2017 to August 2018

IRB Approval:

IRB Protocol #822 with the project title “The Comprehensive Impacts of Receiving Cleft Surgery” has been approved by the IRB Chair at the University of San Francisco under the rules for expedited review on 05/11/2017.

Abstract

Cleft palate and cleft lip are birth defects in which the newborn has an aperture in the roof of the mouth and/or the upper lip. Left untreated, a cleft can lead to speech impediments, problems with eating, and an unsightly facial deformity. A sequence of simple, common surgical procedures can restore function with minimal scarring, but the social, educational, physical, and psychological impacts of receiving this surgery have not been rigorously evaluated. This study uses a difference-in-difference identification strategy with a household level fixed effect to compare teenage life outcomes with treated cleft lip and palate to the life outcomes of their nearest-age sibling, and then compares that difference to the difference between the teenage life outcomes of untreated cleft patients and those of their own nearest-age sibling. This strategy allows estimation of the impact of being born with a cleft, as well as the impact of receiving a full or partial course of reparative surgeries early in life. Key outcomes of interest include a range of indicators that measure social integration, education and learning, psychological well-being, behavioral problems, levels of parental support, and physical health.

This pre-analysis plan is submitted as data collection is approximately half completed, but researchers will not have examined or analyzed the data until after the filing of the plan. Understanding that we are using a quasi-experimental method, for the remainder of this pre-analysis plan, we follow David McKenzie’s (2012) checklist of articles for a pre-analysis plan suggested for field experiment studies:

1. Description of the sample to be used in the study:

A. Treated Patients and their siblings

The sample of those who will make up our treatment group will come from a roster of 282 cleft patients that our partner organization, Operation Smile, previously treated in West Bengal, India between 2004--2015. Other treated patients will be those who have been operated on, but who appear at screening camps for further surgeries. To be included in the sample, these previously treated patients must be between the ages of 11-19 at the time of surveying and they must have at least one sibling aged 7 or above.

B. Control Patients and their siblings

To identify those individuals who will make up the comparison group (i.e., respondents between the age of 11-19 that have a cleft, and have not yet received surgery and their nearest-age siblings) we will use a roster of patients who are scheduled to get surgery at Operation Smile's surgery missions in August and November 2017 and in the spring and summer of 2018. These respondents will be surveyed either on the pre-surgery screening day immediately before to the start of each surgery mission, at screening camps held by Operation Smile during the months prior to each surgery mission, or at respondent's homes. Patients and their siblings will be surveyed in the same location to ensure that the location of the survey does not have any confounding influence on differences in survey responses among siblings. To be included in the sample, these teens (11-19 years old) must have at least one sibling, and they must have no other health conditions that make them ineligible for reparative cleft surgery. In the event that a cleft patient either does not have a sibling, or the sibling is unreachable, a patient's nearest-age cousin will be surveyed in the siblings place if that cousin lives or was raised in the same household as the patient. This is true for both control and treatment groups of sibling pairs. Other data will be taken from the mother that will include information from all siblings in the family.

C. Sample Size

Our target sample size will be approximately 250 children born with cleft lip and cleft palate who have had no surgery, or one or more surgeries. Assuming that these patients have 2.5 siblings on average (the estimated fertility rate in India for 2017 is 2.43 (U.S. Central Intelligence Agency, 2017), and our sample projects to be above the mean), this will give us a total sample of approximately 500 treated and control observations in our estimations using only the nearest-age sibling and approximately 875 observations on estimations that use a more restricted data set based on parent interviews that gather information on all of the cleft patient's siblings.

2. Key data sources:

Data for this study will come from surveys conducted with cleft patients, their nearest-age siblings, and one of their parents. Two survey instruments are used: One for cleft patient respondents and their nearest-age siblings, and one for the parents of cleft patients.

The survey that we use with cleft patients and their siblings collects the following information:

- A. Basic Demographic Data: Age, gender, and birth order.
- B. Observed Physical Health Data: Height, Weight, Grip Strength, Voice Recordings (to evaluate speech outcomes).
- C. Observed cognitive function/education data: Digit-span memory test, [ASER](#) math and reading tests, counting from 0 to 20, educational attainment.
- D. Drawings: Respondents are asked to draw a picture of themselves with their immediate family and to label themselves and their parents. These drawings will be coded and used to evaluate psychological outcomes following Wydick, Glewwe, and Rutledge (2013).
- E. Survey questions about social inclusion, self-esteem, anxiety, depression, hope and future aspirations, parental support, educational attainment, and physical health.

The survey that we use with parents of cleft patients collects the following information:

- A. Socio-economic data about the household: Parental levels of education and occupations, materials used in home construction, dummy variables for having electricity and a toilet within the home, age of the parents, and religion.
- B. The parents are asked to answer a set of 26 questions about their cleft child and each of their non-cleft siblings. These questions have to do with the previously mentioned key outcome indicators: education and learning, psychological well-being (anxiety, depression, self-esteem), social inclusion, hope and aspirations, each child's behavior, and each child's physical health.
- C. The cleft child's type of cleft, and surgical history. We will independently rank the quality of previous surgeries received on a 0-1 scale for cleft palate and a 0-8 scale for cleft lip, following Campbell et al. (2017).

3. Hypotheses to be tested from the impact of the being born with a cleft and from the impact of having reparative cleft surgery(s):

Family 1: Physical and Health Outcomes

- I. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of overall physical health and well-being (composed of weight for height, grip strength, and responses to questions about physical health and independence).
- II. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of physical strength (composed of weight for height and grip strength).
- III. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of general health and physical independence (composed of questions related to general health and physical independence).
- IV. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on observed speech clarity and hypernasality.

Family 2: Social Inclusion, Family Relationships, and Behavior

- V. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of social inclusion, behavior, and parental relationship.
- VI. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of social inclusion.
- VII. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of 5 questions related to parental support for and relationship with their cleft and non-cleft children.
- VIII. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of 5 questions about behavior of the patient and their siblings at school and at home.

Family 3: Psychological Outcomes:

- IX. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of overall psychological well-being (composed of questions about anxiety, depression, self-esteem, aspirations, and hope).
- X. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of questions related to anxiety.
- XI. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of questions related to depression.
- XII. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of questions related to self-esteem.
- XIII. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of questions related to hope and aspirations for the future.

Family 4: Cognition, Education, and Learning

- XIV. H_0/H_a : No impact/positive impact of the surgery on an index representative of overall education, cognitive function and learning.
- XV. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on cognitive function (as measured by a digit span memory test).
- XVI. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on educational attainment.
- XVII. H_0/H_a : No impact/positive impact of being born with a cleft (and subsequent surgery) on an index of performance on math and reading tests.
- XVIII. A Human Dignity Index will consist of an equally weighted measure of each of our four family indices.

4. How variables will be constructed:

Variables will be taken from survey data. Aside from the data collected in the parent survey on cleft condition, surgical history, and socio-economic variables that describe the household, we will treat data collected from the parent surveys as one separate dataset on which we can test our hypotheses, and we will treat the patient/sibling survey data as another separate dataset that we will use to test our hypotheses.

Indices will be constructed from an index suggested in Kling et al. (2007), which calculates a simple average of normalized variables. As a check on these we will also use an Anderson Index (Anderson 2008). The Anderson Index is created by orienting variables in a single direction of impact, de-meaning and normalizing each of the dependent variables in the respective group j . The Anderson Index assigns a weight on each impact variable by the sum of its row entries across the inverted variance-covariance matrix of the impact variables in the group j . Specifically, each variable i in group j receives a weight, or index score, of $\bar{s}_{ij} = (1' \Sigma^{-1} 1)^{-1} (1' \Sigma^{-1} y_{ij})$, where 1 is a $m \times 1$ column vector of 1's, Σ^{-1} is the $m \times m$ inverted covariance matrix, and y_{ij} is the $m \times 1$ vector of outcomes for individual i . The Anderson Index assigns weights to variables such that a variable within the family that exhibits lower covariance with the other variables becomes weighted proportionally higher in the index because it contains more independent information.

Cleft birth defects appear in the lip, the palate, or both and may be unilateral or bilateral, incomplete (less severe) or complete (more severe). We will measure the degree of the cleft abnormality by the number of expected surgeries on average the child should be expected to have on average to restore functioning and appearance to “near normalcy” if surgery were to be accompanied by the requisite therapies. Each cleft child will be placed in one of nine categories, with the estimated number of surgeries needed to restore a child born in the corresponding condition to “near normalcy.” These were established in consultation with the Operation Smile medical team and are as follows:

AVERAGE SURGERY SCENARIOS (Drs. Rueben Ayala, Gaurav Deshpande, Abhishek Das)

1. Incomplete unilateral cleft lip, but no cleft palate: 2 surgeries (lip repair)
2. Incomplete bilateral cleft lip, but no cleft palate: 2 surgeries (lip repair)
3. Incomplete unilateral cleft palate, but no cleft lip: 3 surgeries (palate closure)
4. Incomplete bilateral cleft palate, but no cleft lip: 3 surgeries (palate closure, fistula and/or VPI, and ABG)
5. Complete unilateral cleft lip: 4 surgeries (2 lip surgeries and 2 nose)
6. Complete bilateral cleft lip: 4 surgeries (2 lip surgeries, 1 jaw, and 1 nose)
7. Incomplete cleft lip (bilateral or unilateral) and incomplete cleft palate (bilateral or unilateral): 5 surgeries
8. Complete unilateral cleft lip and palate: 6 surgeries (lip primary repair, lip secondary repair, palate primary and secondary repair, alveolus, nose)
9. Complete bilateral cleft lip and palate: 7 surgeries (lip primary repair, lip secondary repair, palate primary and secondary repair, 2 alveolus, nose)
10. Complete bilateral cleft lip and palate with deviated premaxilla: 8 surgeries (lip primary repair, lip secondary repair, palate primary and secondary repair, 2 alveolus, nose, and jaw)

In the event that children present cleft conditions not fitting into one of these categories, other categories may be added. These categories may evolve as a result of further consultation with cleft experts.

5. Specify the equation to be estimated:

We will use a cross-sectional difference-in-difference estimation with household level fixed effects to estimate the impact of having a cleft of varying levels of severity on our outcome variables, as well as the impact of subsequent surgeries.

Our main specification will be as follows:

$$y_{ij} = \alpha + \beta C_i + \tau S_i + \omega OS_i + \mathbf{X}_{ij}'\boldsymbol{\theta} + \mu_j + \varepsilon_{ij} \quad (1)$$

where y_{ij} is outcome index y for person i in household j , C_i is a variable representing the severity of a cleft lip in terms of number of surgeries required for repair to “near normalcy,” S_i are the number of reparative cleft surgeries performed on the child, OS_i is a dummy variable representing whether Operation Smile performed at least one of the surgeries, $\mathbf{X}_i'\boldsymbol{\theta}$ are a

vector of control variables including gender, age, and birth order that will be used to distinguish a child within the household, μ_j is a household level fixed effect, and ε_{ij} is the error term. Our ability to include the OS_i dummy variable will depend on whether project resources allow us to obtain a large enough sample of patients who were previously treated by Operation Smile.

Using this specification, we will estimate the impact of being born with increasing cleft severity, β , the impact of surgeries, τ , and the added impact of Operation Smile surgery, ω .

For example, assuming $\beta < 0$, and $\tau > 0$, then $\frac{\tau}{\beta} \times 100$ gives us a measure that indicates what percent cleft surgery restore losses in life outcomes from a cleft birth abnormality. Similarly, $\frac{\tau + \omega}{\beta} \times 100$ yields the percent that cleft surgery restores child outcomes in the dependent variable when Operation Smile has performed at least one of the cleft surgeries.

It may be that both the degree of cleft severity as well as surgeries have diminishing returns—that increasing levels of severity matter less than simply having cleft at all, or that the first surgery has the biggest effects on life outcomes and subsequent surgeries have lesser effects. Therefore, a second estimation will be carried out that estimates the following:

$$y_{ij} = \alpha + \mathbf{C}_i' \boldsymbol{\beta} + \mathbf{S}_i' \boldsymbol{\tau} + \omega OS_i + \mathbf{X}_{ij}' \boldsymbol{\theta} + \mu_j + \varepsilon_{ij} \quad (2)$$

Where \mathbf{C}_i in (2) represents a vector of dummy variables for cleft severity that range from requiring at least 2 surgeries to 7 or more surgeries, \mathbf{S}_i represents a vector of dummy variables indicating whether a child has had 1 cleft surgery, 2 cleft surgeries, 3 cleft surgeries or 4 or more cleft surgeries.

Additionally, we reserve the right to test subsets of our sample using identical variable construction to examine potential heterogeneous treatment effects. For example, we will explore whether the impact of the surgery is different for patients who had their first surgery earlier in life than others.

6. Plan for how to deal with multiple outcomes and multiple hypothesis testing:

We have several instances in which we have a family of outcomes that can be tested individually and jointly. When testing individually, we will control the family-wise error rate using the Holm-Bonferroni Step-Down procedure. When testing jointly, all variables in a family, we will use summary indices over all of the variables in our survey of the same family created in the manner of Kling et al. (2007) and Anderson (2008).

7. Procedure to be used for addressing missing data:

For missing right-hand-side variables, we will replace these values with a zero and use a missing variable indicator or drop the control variable. If a RHS control has more than 15% missing values and is not deemed essential to the regression, the RHS variable will be dropped.

8. Outcomes with limited variation:

We may omit dependent and independent variables from our analysis if one standard deviation in the distribution of the variable is within 5% of the mean in either direction.

References

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