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The Causal Impact of Cleft Lip and Palate Surgery on Psychological Wellbeing

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Abstract: Cleft lip and/or cleft palate (CL/P) are birth conditions in which a newborn has an opening in the roof of their mouth or a slit in their upper lip. Left untreated, CL/P can lead to speech impediments and an unsightly orofacial deformity. However, a sequence of simple, common surgical procedures can restore function with minimal scarring. To date, the psychological effects of being born with CL/P have been investigated in the literature, but the restorative impact of CL/P surgery on psychological wellbeing has not been rigorously evaluated. This study uses a quasi-experimental approach to estimate the impact of being born with CL/P on outcomes including anxiety, depression, self esteem, hope and an index of overall psychological well-being, as well as the impact of receiving reparative surgery prior to one's teenage years. The results show that a one-level increase in the severity of a child's CL/P is associated with a .074 standard deviation loss in overall psychological wellbeing. The study's key finding is that the average CL/P surgery improves psychological wellbeing by an estimated .237 standard deviations, more than 310% of the loss associated with increasing CL/P severity.

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1. Introduction

An estimated 5 billion people around the world lack access to adequate surgical care, the majority of whom live in low and middle income countries and suffer from other socio-economic vulnerabilities (Meara et al. 2015). A number of previous studies demonstrate the large potential economic returns to surgical care (Brazier and Johnson 2001; Meara et al. 2015; Debas et al. 2015) and several studies estimate the economic value of cleft lip and/or palate (CL/P) surgery for recipients (B. Alkire et al. 2011; Muntz and Meier 2013; Corlew et al. 2016; Poenaru, Lin, and Corlew 2016). These studies are critical to an informed investment case for expanding access to surgical care for CL/P patients, but lack insight on a potentially important component of CL/P surgery: the impact of surgery on psychological wellbeing. Understanding the causal impact of CL/P surgery on the psychological wellbeing of patients will add to the evidence base on the comprehensive returns to CL/P surgery, allowing for better informed public health policy and donor investment decisions.

Understanding the relationship between CL/P and psychological outcomes has been the subject of a broad body of literature. In general, this literature has found that the presence of CL/P is correlated with long-term social and psychological issues (Hunt et al. 2005, 2006) and that even after receiving surgical care, those born with CL/P still fare worse than their non-cleft counterparts in terms of a range of psychological outcomes including depression, anxiety, self-esteem, and suicide rates (Marcusson, Paulin, and Östrup 2002; T. Ramstad, Ottem, and Shaw 1995; Christensen et al. 2004; Millar et al. 2013). However, research to date has not compared changes in psychological outcomes after receiving treatment to a plausible counterfactual measure of the changes that would have been realized for the same CL/P patient had they never received treatment.

Clearly, there are many issues that make estimating a counterfactual difficult. The risk of selection bias discredits impact estimates achieved by comparing changes in outcomes for treated versus untreated patients, since there are a number of reasons that those patients who select into surgery may be systematically different than those that do not, and these differences could plausibly impact psychological outcomes. Furthermore, comparing those born with CL/P to non-cleft counterparts will also yield biased estimates of the impact of surgery, since there are a number of potential influences that CL/P will have on the patient's psychological outcomes that non-cleft individuals will not be susceptible to.

This study attempts to address this issue of endogeneity by using a quasi-experimental, cross-sectional difference-in-difference method that compares outcomes of treated patients to those of their non-cleft nearest-age siblings, and then compares that

difference to the difference in outcomes between untreated patients and their non-cleft, nearest age siblings. This method (explained further in section 4) will allow for an unbiased estimate of the *causal* impact of both being born with CL/P on psychological outcomes including anxiety, depression, self-esteem, and hope, as well as the impact of receiving reparative surgery on these outcomes.

This study takes place in West Bengal, India where surveys were administered with treated and untreated CL/P patients, their parents, and their nearest-age siblings in order to collect data on the psychological outcomes mentioned above. Preliminary results on a sample of 112 CL/P patient families show that a one-level increase in the severity of a child's CL/P at birth causes a .074 standard deviation loss in overall psychological wellbeing. Furthermore, the average CL/P surgery improves psychological wellbeing by an estimated .237 standard deviations, more than 310% of the loss associated with increasing CL/P severity. This impact may be driven by the effect of surgery on reducing the amount of bullying experienced by CL/P patients during their childhood, but with the current sample size this link is not conclusive. Data collection will continue until at least February 2019 when more robust conclusions will become feasible.

The remainder of this paper will be organized as follows: Section 2 provides a background on cleft lip and palate, as well as a review of previous literature on the impact of CL/P and CL/P surgery on psychological outcomes. Section 3 details the sampling strategy and data used for this study. Section 4 describes the empirical strategy used to identify causal impacts. Section 5 presents the results, followed by a brief summary and conclusion in section 6.

2. Literature Review

2.1 Background on Cleft Lip and Palate

More than 500,000 CL/P surgeries were performed across the globe during the last decade (Poenaru, Lin, and Corlew 2016). However, in South Asia, where this study takes place, over 90% of the population does not have access to safe, affordable surgical care (B. C. Alkire et al. 2015). Policy-makers and donors have competing priorities in terms of health care delivery spending, and this study aims to inform decision-making by providing an estimate of the psychological returns to CL/P surgery in India, where an estimated 27,000 – 33,000 children are born with CL/P annually (Mossey and Little 2009).[1]

Not all orofacial clefts are the same. Clefts of the lip and palate can vary in terms of type and severity (see Figure 1) and can have a number of associated complications. In order to properly interpret the impact estimates presented in this paper, it is important to understand the range of CL/P conditions experienced by patients and to conceptualize how

differences in CL/P type and level of severity will lead to heterogenous impacts on psychological outcomes. Section 3 of this paper includes a detailed discussion of how each different CL/P condition in the study's sample is coded so that it is possible to control for varying levels of CL/P severity.

CL/P arises in an estimated 1.7 per 1000 liveborn babies, with ethnic and geographic variation in prevalence (Mossey et al. 2009). In low- and middle-income countries, estimated prevalence is 1.38 per 1000 births (Kadir et al. 2017). There is some evidence suggesting that several genetic loci could play a causal role in CL/P prevalence, but study results are inconsistent and difficult to interpret with confidence given small sample sizes, potential publication biases, and a lack of common global standards (Prescott et al. 2000; Zeiger et al. 2003; Ioannidis et al. 2005; Mossey et al. 2009). In terms of environmental factors, maternal exposure to tobacco smoke appears to be the strongest correlate with CL/P incidence. The proportion of orofacial clefts attributable to maternal smoking may be as high as 22% (Little, Cardy, and Munger 2004; Little et al. 2004; Honein et al. 2007). Several other potential environmental factors including maternal alcohol consumption, exposure to toxins and agricultural chemicals, and nutrient and vitamin intake have been investigated but have yielded inconsistent results (Gordon and Shy 1981; García 1998; Dixon et al. 2011).

2.2 Cleft Lip and Palate as a Determinant of Psychological Well-Being

There are a number of pathways through which CL/P could impact psychological well-being. Research has shown that children with CL/P are perceived as less intelligent and less social than others (Richman 1976, 1978). Repeated studies have also concluded that those with CL/P experience significantly increased teasing and bullying compared to non-cleft counterparts (Heller, Tidmarsh, and Pless 1981; Noar 1991; Lorot-Marchand et al. 2015). This type of stigmatization and differential treatment from others can then influence how individuals with CL/P perceive themselves, creating a self-fulfilling prophecy cycle where self-perception and social behavior are shaped by the expectations of others, and those expectations and actions then further reinforce self-perceptions and behavior (Merton 1948; Darley and Fazio 1980). As such, both external (treatment from others) and internal (self-perceptions) mechanisms may drive psychological challenges related to self-esteem, depression, anxiety and hope.

Depression

Self-reported facial appearance is the number one correlate with depression amongst CL/P patients, even those who have been previously treated (Marcusson, Paulin, and

Östrup 2002). One study found that anxiety and depression were twice as prevalent in adults with CL/P, even after receiving reparative surgery, compared to non cleft counterparts (T. Ramstad, Ottem, and Shaw 1995a). Higher levels of Major Depressive Disorder are significantly more common in people born with CL/P as well (Demir et al. 2011). Two studies identified that teasing about an individual's facial appearance was significantly related to depression among those with CL/P, although no causal relationship has been established (Demir et al. 2011; Lorot-Marchand et al. 2015). In Denmark, Christensen et al. (2004) found that adults who were born with CL/P have a higher suicide rate than non-cleft adults. Similarly, in Nigeria, the prevalence of psychiatric morbidity was found to be significantly higher among CL/P patients than non-cleft controls (Yunusa and Obembe 2013). One case-control study with 122 children and adolescents aged 7-17 years in Brazil observed depression symptoms in the CL/P group, but no statistically significant difference between these patients and the non-cleft controls (Lima et al. 2015). Taken as a whole, the body of literature on CL/P and depression suggests a strong correlation between the two. [2]

Anxiety

Multiple studies that compare Chinese adults with CL/P to non-cleft counterparts have found that the presence of CL/P is correlated with higher levels of social anxiety, less social engagement, and lower self-esteem (Cheung, Loh, and Ho 2007; Berk et al. 2001). One study found that 24% of children aged 4-9 with CL/P screened positive for Separation Anxiety Disorder (SAD), significantly higher than the United States population estimate of 3-5% (Tyler et al. 2013). The same study estimated that cleft-related challenges related to speaking and eating were associated with a 100% increase in the risk of SAD. Other work has found that parents of CL/P children report higher levels of anxiety in their children compared to the parents of non cleft-controls (Hunt et al. 2007).

Self-esteem

Research on the relationship between CL/P and self-esteem has yielded somewhat mixed results, but overall the presence of CL/P appears to be correlated with lower levels of self-esteem, largely due to patients' displeasure with their facial appearance and a high prevalence of teasing or bullying. In one qualitative study with 60 teenage CL/P patients in Malaysia, a majority of patients reported that their self-confidence had been damaged due to teasing that they experienced (Noor and Musa 2007). In two other studies that compared CL/P adults (Cheung, Loh, and Ho 2007) and children aged 5-6 (Kramer et al. 2008), to non-cleft comparison groups, the presence of CL/P was associated with lower levels of self-

esteem. However, a study with 4-7 year olds in Germany found that levels of self-esteem were not significantly different for children with CL/P compared to their non-cleft counterparts (Sagheri et al. 2009).

Hope

There are no empirical studies that have estimated a correlation or causal relationship between being born with CL/P and hope. This is likely due to the nascence of hope as a quantifiable outcome in global health and development research. Only recently was hope introduced to the development economics literature as a measurable outcome within the scope of empirical impact evaluations (Glewwe, Ross, and Wydick 2014; Lybbert and Wydick 2016). The concept of hope as it relates to psychological well-being, economic development, and CL/P surgery is further explored in the following section.

2.3 The Impact of Reparative Surgery on Psychological Well-Being

While many empirical studies in development economics focus on the impacts of relieving external constraints to growth and development (capital for example), recent work has started to shed light on the impact that internal constraints such as agency, self-efficacy, hope, and aspirations play in human development. Amartya Sen may have been the first [3]to explicitly distinguish between internal and external constraints in his seminal book *Development as Freedom*, which provides a useful framework from which to consider the impact of internal constraints on development (Sen 2001). In *Development as Freedom*, Sen emphasizes one's ability to define and pursue their goals as a pillar of the development process. Crucial to this theory of development is the concept of individual agency: one's level of control over actions and their consequences (Moore 2016). CL/P surgery can be thought of as a mechanism for relieving a physical health constraint, which in turn improves psychological well-being, hence relieving a range of internal constraints including expanding one's perceived self-agency and ability to pursue their own independently defined goals.

By relieving these internal constraints, CL/P surgery could theoretically increase hope and aspirations for patients who receive reparative treatment. Appadurai (2004) introduces the concept of aspirations in development theory and highlights how one's own aspirations are never individual in nature, but are rather a function of their interaction with others and with the social norms in the environment that surrounds them. Building on this, the development economist Debraj Ray (2006) introduced the idea of an "aspirations window" formed by one's surrounding influences. Ray theorizes that one's aspirations window is dictated by the outcomes achieved by the individuals around them who they see

as similar in capacity. She posits that aspirations can cause a development trap for the poor who suffer from an aspirations failure. This could be the case when an individual's "aspirations gap" (the difference between the standard of living that one aspires to and the standard of living that one already has) is too large (i.e. fulfilling their aspiration would require too costly of an initial investment, which when not made induces a development trap).

Ray's formal description of the aspirations gap can help us understand how CL/P surgery could theoretically be a way to relieve internal psychological constraints. An individual's aspirations gap $g(a, s)$ is a function of a, the standard of living that is aspired to, and s, an individual's current standard of living. An individual then maximizes the difference between benefits (a reduced aspirations gap) and costs (lower current standards): $\max\left\{\frac{a-s}{a}, 0\right\}$. Thus, someone with very high aspirations relative to their current standards will have an aspirations gap approaching 1. Someone with very small aspirations will have $a \cong s$ and their aspirations gap will approach zero. An investment of i comes at a cost to an individual, but could be made to raise future standards of living to s' (hence lowering the aspirations gap, assuming that aspirations are held constant) resulting from a function $\omega(i, s)$. If the cost of this investment is denoted as $c(i)$ then an individual will choose i in order to minimize the sum of the resulting aspirations gap and the cost of the investment: $g(a, s') + c(i)$ subject to the constraint $s' = \omega(i, s)$. If we think of CL/P surgery as an investment i that raises future standards of living, then under this framework it is clear how making such an investment would theoretically have a positive impact on the aspirations of an individual. It is easy to apply this theoretical framework to other psychological outcomes as well, where an investment in CL/P surgery could act to shut down the self-fulfilling prophecy cycle that may influence levels of self-esteem, social anxiety, and depression.

Alternatively, the expectation of future surgery for untreated patients could plausibly drive increases in hope and aspirations for the future, even if they had never received surgery in the past. This is especially possible for the sample in this study, considering that all untreated patients were surveyed in the 6-week window prior to their first scheduled surgery. While this alternative mechanism may apply to hope and aspirations, it is less likely that the expectation of surgery in the future would have a similar effect on pre-surgery levels of self-esteem, depression, or anxiety.

To date, the empirical literature on the impact of CL/P surgery on the key psychological outcomes measured in this study (anxiety, depression, self-esteem, and hope) is sparse. Several studies have examined psychological outcomes of patients post-surgery (Marcusson, Paulin, and Östrup 2002; T. Ramstad, Ottem, and Shaw 1995b; T. Ramstad,

Ottem, and Shaw 1995a; Christensen et al. 2004; Millar et al. 2013), including one study that finds a positive correlation between the level of post-surgery scarring and levels of anxiety, depression, and self-esteem (Millar et al. 2013), but none have estimated the isolated causal impact of reparative surgery on these outcomes. This may be due to the difficulty of dealing with selection bias issues that make it challenging to create a valid counterfactual for individual CL/P patients who have received surgery. Section 4 provides a detailed explanation of how this study attempts to estimate a counterfactual using quasi-experimental methods.

3. Data

3.1 Description of the Data

Data for this study comes from surveys conducted with cleft patients between the ages of 11-19, their nearest-age siblings (aged 7 or older), and one of their parents. Two survey instruments are used in this study: One for cleft patient respondents and their nearest-age siblings, and one for the parents of cleft patients.

The survey with cleft patients and their siblings collects basic demographic data including age, gender, and birth order, as well as answers to survey questions that measure self-esteem, anxiety, depression, and hope. The survey with parents of cleft patients collects socio-economic data about the household including parental occupations and levels of educational attainment, materials that the house is constructed with, dummy variables for having electricity and a toilet within the home, age of the parents, and religion. The parents are also asked to answer a set of questions about their cleft child and each of their non-cleft siblings. These questions have to do with the previously mentioned key outcome indicators for psychological well-being (anxiety, depression, self-esteem and hope). Furthermore, the parents are asked to provide information about the cleft child's type of cleft at birth, and surgical history. Pictures of the patient's orofacial area are taken to validate the information provided by the parent regarding treatment status and original cleft condition.

The survey questions for patients and their siblings that were used as inputs to each psychological index are previously validated questions, some of which were slightly adapted for the Bengali context after several weeks of piloting. Three questions from the Patient Health Questionnaire Screening Instrument for Depression were used to measure depression. Three questions from the General Anxiety Disorder Screening Questionnaire were used to measure anxiety. Five questions from the Rosenberg Self-Esteem Scale were adapted after piloting to measure self-esteem. Finally two questions directly related to

future aspirations for employment and overall quality of life were taken from previous survey instruments used by (CITE WYDICK PAPER) to measure hope. The parent surveys used one question each to measure the main psychological outcomes. See Appendix 1 for the English translation of each psychological survey question in both surveys.

3.2 Description of the sample to be used in the study

Control patients and their siblings

To identify those individuals who made 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) the study used a roster of patients who were scheduled to get surgery at Operation Smile's surgery missions in August and November 2017. These respondents were 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 nearest-age siblings were surveyed in the same location to ensure that the location of the survey did 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 that is at least 7 years old[1], and they must have no other health conditions that make them ineligible for reparative CL/P surgery. In the event that a cleft patient either did not have a sibling aged 7 or older or if the sibling was unreachable, a patient's nearest-age cousin was surveyed in the sibling's place if that cousin lived or was raised in the same household as the patient. This is true for both control and treatment groups of patient-sibling pairs.

Treated patients and their siblings

The sample of those who make up the treatment group come from a roster of 282 cleft patients that the international NGO Operation Smile previously treated in West Bengal, India between 2004 and 2017. Other treated patients were those who had been previously operated on, but who appeared at screening camps for further follow-up surgeries. To be included in the sample, these previously treated patients were required to be between the ages of 11-19 at the time of surveying and they must have at least one sibling aged 7 or above.

A three-step process was followed to identify these patient-sibling pairs. First, the roster was narrowed to patients who had a family member's phone number listed in

[1] This is an arbitrary cutoff that the research team decided on to ensure that the nearest-age siblings of patients were old enough to accurately respond to survey questions.

Operation Smile's database. Second, several phone calls were made over the course of three days to all available phone numbers for that patient's family. If the patient's family was reached, they were screened over the phone for eligibility and invited to attend a survey day at a central location convenient for numerous families. Families were offered a financial incentive that ranged from Rs. 1000 to Rs. 2000 to attend the survey event, depending on how far they needed to travel. Third, if a family was unreachable over the phone, patient mobilizers that were previously contracted by Operation Smile to identify CL/P patients were hired to find them based on the address/village/town on file in the Operation Smile database. Families that were successfully located were then screened for eligibility and offered the same financial incentive to attend a survey event at a central location.

3.3 Variable Construction

Variables are taken from primary survey data. Data on psychological outcomes collected from the parent surveys is treated as one separate dataset on which hypotheses can be tested, and the patient/sibling survey data is treated as another separate dataset. Since multiple survey questions from the patient/sibling survey are used to measure each psychological outcome, Anderson indices (Anderson 2008) are constructed to represent each outcome as a whole, including overall psychological wellbeing, which is measured in this study as a combination of anxiety, depression, and self-esteem. A secondary psychological wellbeing index is created that also includes hope.

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 outcome group.[2] 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. The Anderson index assigns weights to variables such that a variable within the outcome group that exhibits lower covariance with the other variables becomes weighted proportionally higher in the index because it contains more independent information.

Clefts of the lip and palate can vary in terms of type and severity (see Figure 1) and can have a number of associated complications. Intuitively, it would make sense that the negative impact of a cleft would be positively correlated with the severity of the cleft. For this reason, this study carefully distinguishes between different levels of CL/P severity, rather than treating the presence or non-presence of CL/P as a binary variable. In this study, the degree of CL/P severity is measured by the average number of expected

[2] The outcome groups for this study are 1. Psychological wellbeing, without hope 2. Psychological wellbeing, with hope 3. Depression 4. Anxiety 5. Self-esteem 6. Hope

surgeries the child needs at birth to restore functioning and appearance to near normalcy.[3] Each cleft patient in the sample is placed in one of seven categories (listed below) that correspond to the estimated number of surgeries needed to restore a child born with the corresponding condition to near normalcy. These categories were established in consultation with the Chief Medical Officer at Operation Smile and two Orofacial surgeons at the Mahatma Ghandi College of Medicine in Mumbai.[4]

Average surgery scenarios

1. Incomplete unilateral or bilateral cleft lip, but no cleft palate: 2 surgeries

2. Incomplete unilateral or bilateral cleft palate, but no cleft lip: 3 surgeries

3. Complete unilateral or bilateral cleft lip: 4 surgeries

4. Incomplete cleft lip (bilateral or unilateral) and incomplete cleft palate (bilateral or unilateral): 5 surgeries

5. Complete unilateral cleft lip and palate: 6 surgeries

6. Complete bilateral cleft lip and palate: 7 surgeries

7. Complete bilateral cleft lip and palate with deviated premaxilla: 8 surgeries

3.4 Attributes of the Sample

Tables 1 and 2 show the key descriptive statistics of the sample. Table 1 shows unweighted sample averages for key demographic and outcome variables for patients and siblings in households in which the patient received one or more surgeries and households in which the patient did not receive any surgery prior to survey. Patients in no surgery households and surgery households were older than their nearest age siblings by just over half a year on average (0.615 years in no surgery households and 0.570 in surgery households). Patients in no surgery households were more likely to be male than their nearest age siblings (53.8% vs. 42.3%) while the opposite was true for surgery households (55.8% of siblings were male vs. 45.3% of patients). Patients in surgery households had a lower average birth order than patients in no surgery households (1.709 in no surgery households vs. 2.385 in no surgery households). On the whole, the table shows that siblings and patients in surgery households appeared to fare worse in terms of psychological

[3] Defined as normal orofacial functioning, fully recovered speech, and no visible cleft aside from minimal scarring. The average number of required surgeries is estimated assuming that patients will receive any required speech therapies.

[4] Dr. Ruben Ayala, MD, Operation Smile CMO; Dr. Gaurav Deshpande, MD, MGM Medical College Mumbai; Abhishek Das, candidate for MD, MGM Medical College Mumbai

outcomes than those in households where the patient never received surgery. This suggests that children in households who select into surgery are more likely to suffer from anxiety, depression, a lack of self-esteem, or a hope deficit.

The far right column of Table 1 shows the difference-in-difference of means in the sample for the (column 6 minus column 3). For the psychological outcome variables in the table, this column shows the expected direction of impact due to receiving one or more surgeries without controlling for household characteristics (via a household fixed effect) and without controlling for any covariates. It is important to note that the signs of all psychological variables align with the *direction of the impact* on the outcome. For example, a 0.167 standard deviation difference-in-difference estimate for the depression index suggests that surgery has a *positive* impact on depression (i.e. depression decreases due to surgery). The signs of the estimates for depression, anxiety (.382 standard deviations), and self-esteem (.639 standard deviations) fit the study's broad theory of change – that surgery will improve psychological outcomes. However the negative estimate for hope suggests a different mechanism may be at play for this variable.

ADD DISCUSSION OF FIGURES 2-3 AND TABLES 2-3 AND A GENERAL DESCRIPTION OF THE SAMPLE IN TERMS OF DESCRIPTIVE STATISTICS, DIFFERENCES BETWEEN HOUSEHOLDS, AND DIFFERENCES BETWEEN SIBLING PAIRS WITHIN TREATED AND UNTREATED HOUSEHOLDS.

4. Empirical Strategy

This study uses a cross-sectional difference-in-difference method with a household level fixed effect to estimate the impact of having a cleft of varying levels of severity on key outcome variables, as well as the impact of having subsequent surgeries. The first difference will be between each patient and their nearest age-sibling in terms of each outcome index and the second difference will be between the sibling-level differences in sibling pairs where the cleft patient was treated (to varying degrees) and the difference in outcomes between sibling pairs where the cleft patient was never treated.

The main specification will be as follows:

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

where y_{ij} is outcome index y for person i in household j, C_i is a variable representing the level of CL/P severity for individual i, S_i is the number of reparative cleft surgeries

performed on the child prior to the survey date, OS_i is a dummy variable equal to one if individual i received any surgeries from Operation Smile prior to the survey date, $\mathbf{X_i}$ is 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.

This specification allows for an estimate of the impact of being born with increasing cleft severity, β, the impact of surgeries, τ, and the added impact of having received any Operation Smile surgeries, 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 outcomes in when Operation Smile has performed at least one of the cleft surgeries.

Theoretically, it is plausible that both the degree of CL/P severity as well as surgeries have diminishing returns—that increasing levels of severity impact psychological outcomes less than simply having a cleft at all, or that the first surgery has the biggest effects on these outcomes and subsequent surgeries have lesser effects. Therefore, a second estimation will be carried out as follows:

$$y_{ij} = \alpha + \mathbf{C_i'\beta} + \mathbf{S_i'\tau} + \omega OS_i + \mathbf{X_{ij}'\theta} + \mu_j + \varepsilon_{ij} \qquad (2)$$

where $\mathbf{C_i}$ in (2) represents a vector of dummy variables for cleft severity that range from requiring at least two surgeries to requiring seven or more surgeries, $\mathbf{S_i}$ represents a vector of dummy variables indicating whether a child has had one CL/P surgery, two CL/P surgeries, three CL/P surgeries or four or more surgeries.

Additionally, a simplified specification will be used to measure the average impact of receiving any surgery at all. This specification in equation (3) does not control for variations in cleft severity or for the fact that some surgery recipients received more surgery than others.

$$y_{ij} = \alpha + D_i\beta_1 + K_i\beta_2 + \omega OS_i + \mathbf{X_{ij}'\theta} + \mu_j + \varepsilon_{ij} \qquad (3)$$

where $D_i$is a dummy variable for being born with CL/P of any severity or type and K_i is a dummy variable for receiving one or more surgeries of any kind.

ADD IN EXPLICIT DISCUSSION ABOUT THE MAIN IDENTIFYING ASSUMPTION, AND WHY IT IS VALID (I.E., THAT ON AVERAGE, THE DIFFERENCES BETWEEN CLOSEST AGE SIBLINGS IN TERMS OF PSYCHOLOGICAL OUTCOMES WILL BE THE SAME ACROSS FAMILIES

WHEN CONTROLING FOR AGE, GENDER, BIRTH ORDER, AND UNOBSERVED FAMILY CHARACTERISTICS).

5. Results

5.1 The restorative impact of the average CL/P surgery on psychological outcomes

Table 4 below shows the key empirical results of the study. Table 4 provides coefficient estimates for equation (1) using data from surveys with teenage patients and their nearest-age siblings. The estimates reveal that for each one-surgery increase in the number of surgeries a CL/P patient requires at birth (the measure of cleft severity), overall psychological wellbeing decreases by .07 standard deviations (significant at the 10% level). Receiving one additional *average* surgery leads to a .24 standard deviation increase in overall psychological wellbeing. This suggests that an average CL/P surgery can lead to psychological wellbeing gains that are more than three times the magnitude of what is lost due to increasing CL/P severity. Operation Smile surgeries did not have any significant additional effect on the impact of surgery on psychological wellbeing. **ADD FURTHER DISCUSSION HERE ON HOW THESE IMPACTS APPEAR TO BE DRIVEN BY THE IMPACT ON SELF ESTEEM, ESPECIALLY FOR THOSE WHO WERE BORN WITH SEVERE CASES BUT RECEIVED SEVERAL SURGERIES.**

Table 6 provides coefficient estimates for equation (1) using data from the surveys with parents of teen cleft patients. As parents were asked to give their observations about their children in terms of psychological characteristics, intuitively these impact estimates will be less efficient than those that use survey data collected directly from patients and siblings. The results suggest that for each one-surgery increase in the number of surgeries a CL/P patient requires at birth, overall psychological wellbeing decreases by .125 standard deviations (significant at the 1% level). Interestingly, the increasing cleft severity has a positive impact on hopefulness, which may be explained by the fact that most surveys are conducted in the days and weeks prior to surgery missions and patients could very likely be hopeful that surgery will have transformative impacts for them. Receiving one additional *average* surgery does not lead to any significant gains in the psychological wellbeing of patients, according to parental observation. Comparing the results from Table 1 and Table 2 may suggest that parents have a stronger perception of the negative impacts of cleft severity (and a weaker perception of the impact of surgery) than the impacts that their children actually realize.

5.2 The restorative impact of receiving at least one surgery, regardless of CL/P severity

ADD DISCUSSION OF TABLE *3*, WHICH SHOWS THE RESULTS FROM EQUATION (*3*)

5.3 The functional form of the relationship between the number of CL/P surgeries received and the restorative impact of an additional surgery

Table 5 provides insight on the impact of each specific level of cleft severity and each specific additional surgery (as opposed to an average surgery). The results do not show an easily interpretable relationship between cleft severity or number of surgeries received and psychological outcomes. It appears as though the negative impact of requiring three surgeries is larger than the negative impact of requiring two, both of which are dwarfed by the impact of the most severe cleft cases (Requiring 7+ surgeries), which causes overall psychological wellbeing to fall by .62 standard deviations. Furthermore the results in Table 5 show that those patients who received four or more surgeries realized a much greater positive change in psychological wellbeing than those who received one, two, or three surgeries.

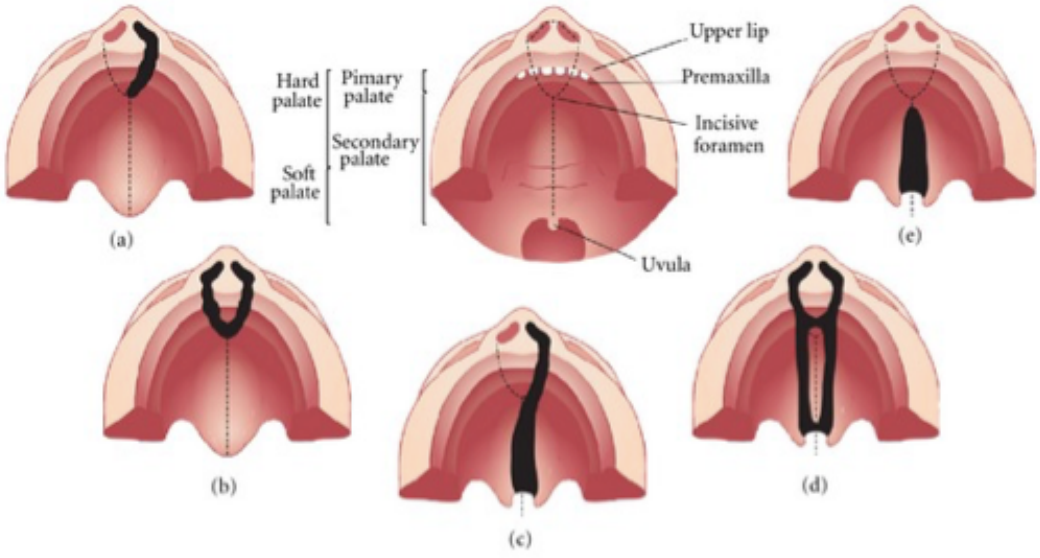
5.4 Limitations to the results presented in this study

INSERT DISCUSSION RELATED TO LIMITED STATISTICAL POWER AND THE INABILITY OF THIS STUDY TO IDENTIFY THE DIFFERENTIAL IMPACT OF CLEFT LIP VERSUS PALATE SURGERY. ALSO ADD A DISCUSSION ABOUT THE EXTERNALY VALIDITY OF THE RESULTS.

7. Summary and Conclusion

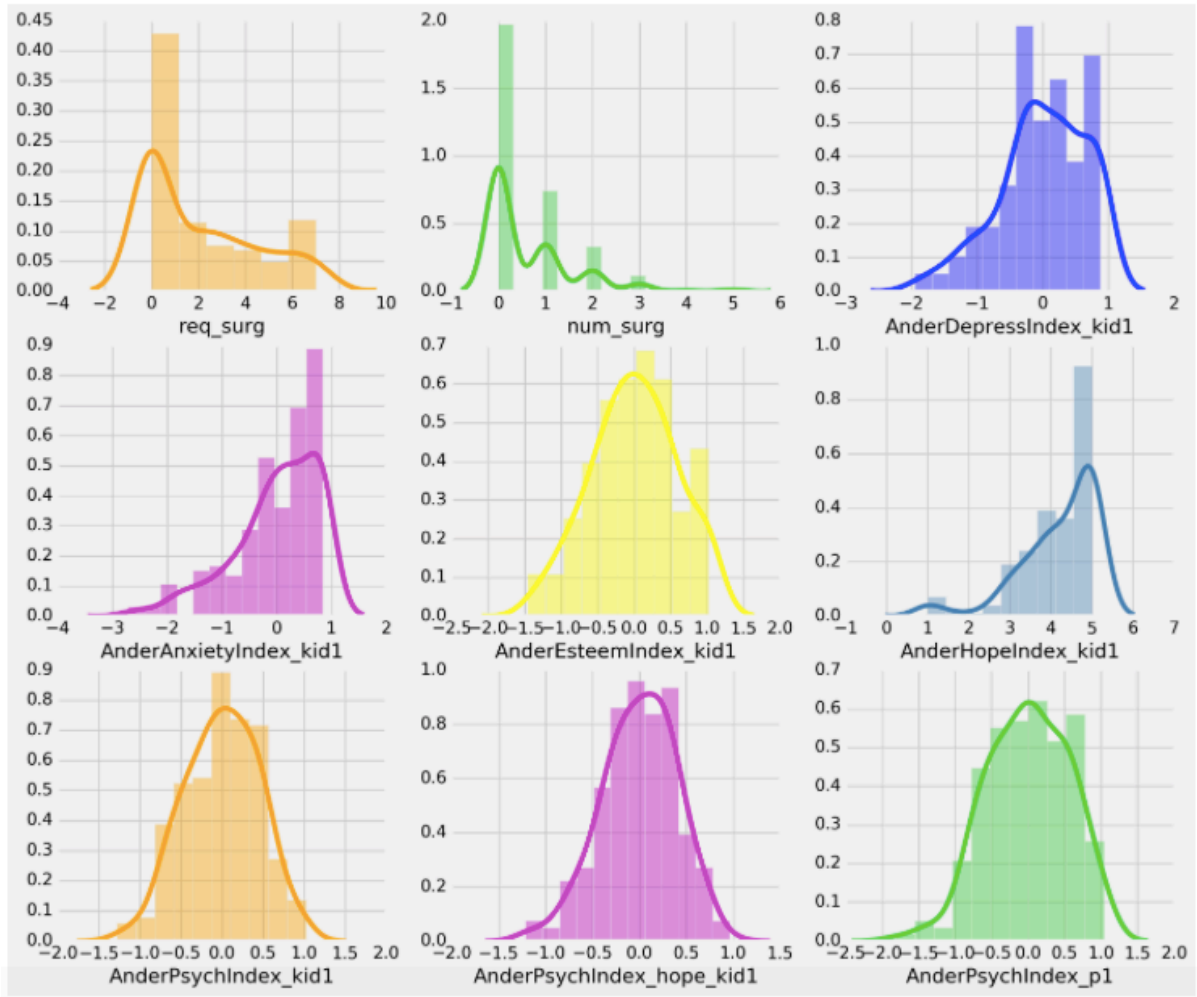
Tables and Figures

Figure 1: Visual Representation of Common CL/P Conditions



Notes: (a) Complete unilateral cleft lip (b) Complete bilateral cleft lip (c) Complete unilateral cleft palate and cleft lip (d) Complete bilateral cleft palate and lip (e) Incomplete cleft palate. Copyright Brito et al. 2012

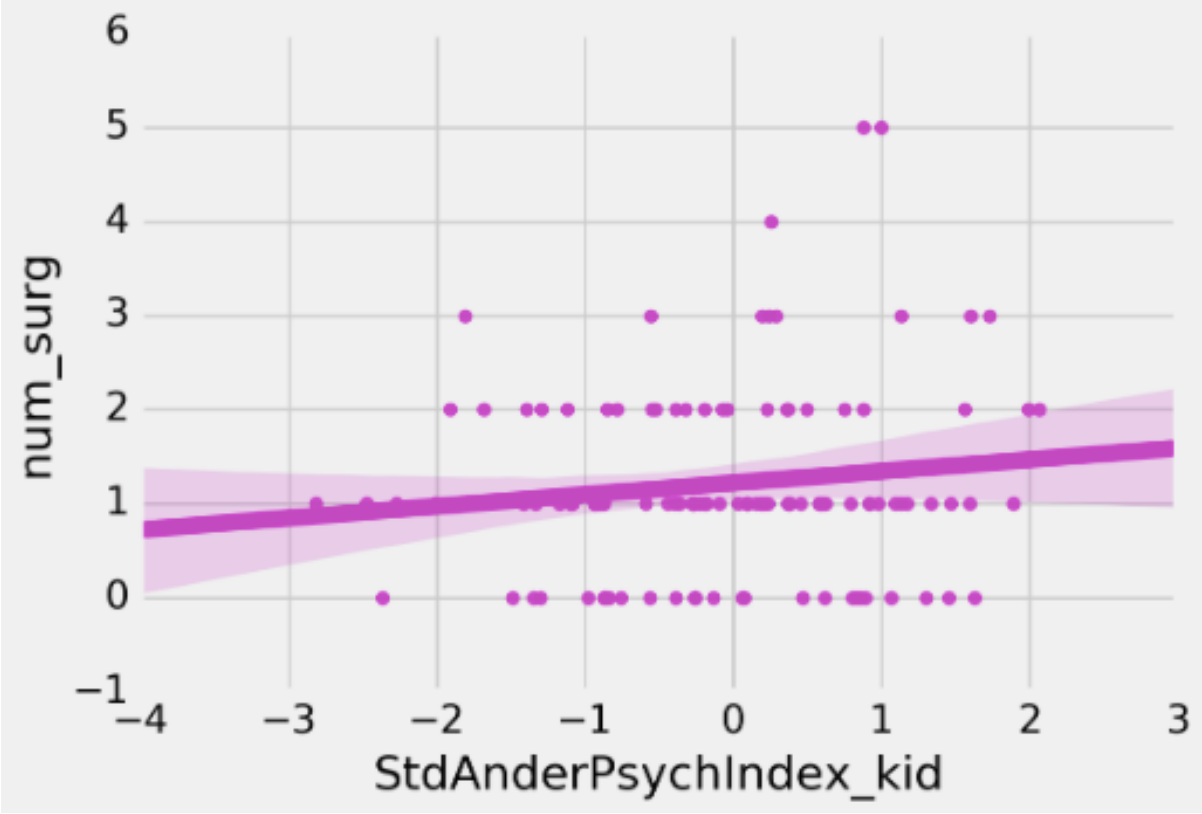
Figure 2: Distributions of Key Treatment and Outcome Variables – Full Sample



Notes: Variables listed from left to right: (Top Row) The average number of required CL/P

surgeries at birth, the total number of CL/P surgeries received prior to the survey date, Anderson index of all depression questions in the patient/sibling survey. (Middle Row) Anderson index of all anxiety questions in the patient/sibling survey, Anderson index of all self esteem questions in the patient/sibling survey, Anderson index of all hope questions in the patient/sibling survey. (Bottom Row) Anderson index of all psychological questions other than those measuring hope in the patient/sibling survey, Anderson index of all psychological questions in the patient/sibling survey, Anderson index of all psychological questions in the parent survey (only for patient-sibling pairs). All indices are oriented so that positive outcomes (i.e., lower depression, or higher self-esteem) are represented by higher positive values, and worse outcomes represented by lower values.

Figure 3: The Relationship between Number of Surgeries Received and Psychological Wellbeing



Notes: Y-axis is the number of surgeries received prior to the survey date. X-axis is the value on an Anderson index of all psychological questions (other than those measuring hope) in the patient/sibling survey.

Table 1: Descriptive Statistics - Within Households

	No Surgery Households			1 or More Surgery Households			
	Patient	Sibling	Patient-Sibling	Patient	Sibling	Patient-Sibling	Diff-in-Diff, no FE[4]
Age	14.423	13.808	0.615	15.430	14.860	0.570	-0.045
	(0.437)	(0.734)	(0.854)	(0.274)	(0.537)	(0.602)	
Male	0.538	0.423	0.115	0.453	0.558	-0.105	-0.22
	(0.100)	(0.099)	(0.140)	(0.054)	(0.056)	(0.078)	
Birth order	2.385	2.500	-0.115	1.709	2.058	-0.349	-0.234
	(0.229)	(0.186)	(0.295)	(0.111)	(0.096)	(0.146)	
Depression Index	0.271	0.499	-0.228	-0.147	-0.086	-0.061	0.167
	(0.204)	(0.134)	(0.244)	(0.105)	(0.113)	(0.154)	
Anxiety Index	-0.190	0.214	-0.403	-0.014	0.007	-0.021	0.382
	(0.200)	(0.177)	(0.267)	(0.112)	(0.106)	(0.155)	
Self Esteem Index	-0.319	0.300	-0.619	0.013	-0.007	0.020	0.639
	(0.191)	(0.186)	(0.267)	(0.118)	(0.098)	(0.153)	
Hope Index	0.051	0.052	-0.001	-0.075	0.044	-0.119	-0.118
	(0.176)	(0.196)	(0.264)	(0.117)	(0.102)	(0.156)	
Psych Wellbeing Index (no hope)	-0.090	0.426	-0.516	-0.035	-0.067	0.032	0.548
	(0.202)	(0.172)	(0.266)	(0.113)	(0.103)	(0.153)	
Psych Wellbeing Index (including hope)	-0.048	0.407	-0.456	-0.067	-0.042	-0.025	0.431
	(0.207)	(0.148)	(0.255)	(0.111)	(0.108)	(0.155)	
N	26	26	52	86	86	172	224

Notes: Unweighted sample averages reported with standard errors in parentheses. Male is a dummy variable for gender with male=1 and female=0. Each outcome index is an Anderson Index standardized to have a mean of 0 and standard deviation of 1. The far right column shows the difference of the difference in means presented in columns 3 and 6. The numbers in this column for each of the psychological outcomes (rows 4-9) should be interpreted as standard deviations.

Table 2: Descriptive Statistics - Across Households

	1 or More OS Surgeries	1 or More Surgeries	No Surgery	OS Surgery - Other Surgery	OS-No Surgery	Other Surgery - No Surgery
Housing Quality Index	0.011	0.057	-0.188	-0.110	0.198	0.244
	(0.088)	(0.077)	(0.130)	(0.157)	(0.154)	(0.158)
Parent Education	-0.100	0.057	-0.189	-0.377**	0.089	0.247
	(0.104)	(0.079)	(0.117)	(0.158)	(0.168)	(0.158)
Electricity Dummy	0.840	0.884	0.902	-0.104**	-0.062	-0.018
	(0.037)	(0.025)	(0.042)	(0.049)	(0.060)	(0.051)
Toilet Dummy	0.720	0.663	0.538	0.137*	0.182**	0.124
	(0.045)	(0.036)	(0.070)	(0.073)	(0.080)	(0.076)
Religion	0.300	0.314	0.500	-0.033	-0.200**	-0.186**
	(0.046)	(0.035)	(0.070)	(0.072)	(0.081)	(0.075)
N	100	72	52	172	152	124

Notes: Unweighted sample averages reported with standard errors in parentheses. Housing Quality Index is a standardized Anderson Index of wall, roof, and floor materials. Parent education is the average of a categorical variable for all parents in the household: None=0, Primary=1, Secondary=2, University=3. It is standardized so that it can be interpreted as standard deviations in this table. Electricity Dummy is a dummy variable for having electricity in the house. Toilet is a dummy for having a toilet or latrine at the house. Religion is a categorical variable where 0=Hindu and 1=Muslim. *=p<.1 **=p<.05 ***p<.01

Table 3: The Impact of Receiving 1 or More Surgeries, Regardless of CL/P Severity

	Psych Wellbeing Index, No Hope	Psych Wellbeing Index, w/ Hope	Depression	Anxiety	Self Esteem	Hope
CL/P Dummy (Any Severity)	-0.474**	-0.407*	-0.217	-0.307	-0.577***	0.0319
	(0.203)	(0.222)	(0.226)	(0.243)	(0.192)	(0.221)
Dummy for Receiving 1 or More Surgeries	0.484	0.423	0.00374	0.146	0.601*	-0.0249
	(0.323)	(0.311)	(0.343)	(0.350)	(0.316)	(0.271)
OS dummy	0.0469	-0.0386	0.241	0.132	0.0601	-0.149
	(0.290)	(0.290)	(0.292)	(0.309)	(0.294)	(0.284)
Birth order	0.0480	0.0503	-0.104	-0.115	0.193	0.0639
	(0.138)	(0.144)	(0.139)	(0.154)	(0.146)	(0.124)
Age	-0.0205	-0.0358	-0.0401*	-0.0675**	0.00806	-0.0361
	(0.0261)	(0.0281)	(0.0230)	(0.0290)	(0.0253)	(0.0288)
Male	-0.210	-0.181	0.0163	-0.592***	-0.217	-0.0301
	(0.168)	(0.182)	(0.157)	(0.187)	(0.175)	(0.224)
_cons	0.365	0.572	0.853	1.603**	-0.355	0.451
	(0.611)	(0.650)	(0.545)	(0.647)	(0.618)	(0.598)
N	224	224	224	224	224	224

OLS with fixed effects at the household level. Robust standard errors clustered at the household level are in parentheses. Dependent variables are all standardized Anderson Indices and are listed across the top row. OS dummy is a dummy variable where 1=Received at least one Operation Smile surgery and 0=Has not received any OS surgeries. Male is a dummy variable for gender where male=1 and female=0.

* $p < 0.10$, ** $p < 0.05$, *** $p < 0.01$

Table 4: The Impact of an Average Surgery on Psychological Variables - Patient and Sibling Data

	Psych Wellbeing Index, No Hope	Psych Wellbeing Index, w/ Hope	Depression	Anxiety	Self Esteem	Hope
Cleft severity	-0.0758*	-0.0464	-0.0651*	-0.0349	-0.0821*	0.0413
	(0.0395)	(0.0426)	(0.0385)	(0.0513)	(0.0475)	(0.0444)
No. of surgeries received	0.236**	0.131	0.0902	0.0466	0.209	-0.182
	(0.109)	(0.0980)	(0.106)	(0.146)	(0.154)	(0.130)
OS dummy	-0.00813	-0.0424	0.160	0.0453	0.0858	-0.0248
	(0.225)	(0.255)	(0.224)	(0.251)	(0.258)	(0.264)
Birth order	0.0525	0.0530	-0.100	-0.104	0.189	0.0459
	(0.138)	(0.145)	(0.135)	(0.152)	(0.146)	(0.127)
Age	-0.0212	-0.0367	-0.0393*	-0.0680**	0.00724	-0.0365
	(0.0269)	(0.0289)	(0.0237)	(0.0294)	(0.0260)	(0.0295)
Male	-0.229	-0.200	0.0311	-0.594***	-0.239	-0.0309
	(0.166)	(0.179)	(0.155)	(0.191)	(0.167)	(0.220)
_cons	0.339	0.565	0.815	1.553**	-0.348	0.499
	(0.617)	(0.656)	(0.538)	(0.639)	(0.618)	(0.616)
N	224	224	224	224	224	224

OLS with fixed effects at the household level. Robust standard errors clustered at the household level are in parentheses. Dependent variables are all standardized Anderson Indices and are listed across the top row. Cleft Severity is proxied by the number of surgeries that the patient needs at birth to be restored to near normalcy. OS dummy is a dummy variable where 1=Received at least one Operation Smile surgery and 0=Has not received any OS surgeries. Male is a dummy variable for gender where male=1 and female=0.

* $p < 0.10$, ** $p < 0.05$, *** $p < 0.01$

NEED TO ADD IN TEST FOR WHETHER ABS. VALUE OF THE SURGERY COEFFICIENT IS LARGER THAN THE ABS. VALUE OF THE SEVERITY COEFFICIENT

NEED TO ADD IN ROBUSTNESS CHECK WITH THE FIXED EFFECTS FIX FROM THE DUKE PAPER AND BERK OZLER'S DEVELOPMENT IMPACT BLOG POST

Table 5: The Impact of Specific Surgeries on Psychological Variables – Patient and Sibling Data[5]

	Psych Wellbeing Index	Depression	Anxiety	Self Esteem
Need 2 Surgeries	-0.431	-0.103	-0.0430	-0.434*
	(0.293)	(0.369)	(0.308)	(0.252)
Need 3 Surgeries	-0.591**	-0.254	-0.645*	-0.778***
	(0.293)	(0.292)	(0.359)	(0.276)
Need 4 Surgeries	-0.403	-0.287	0.253	-0.353
	(0.433)	(0.480)	(0.371)	(0.390)
Need 5 Surgeries	0.210	-0.299	0.161	0.445
	(0.384)	(0.400)	(0.508)	(0.486)
Need 6 Surgeries	0.112	-0.236	0.0228	0.0336
	(0.418)	(0.394)	(0.427)	(0.493)
Need 7+ Surgeries	-0.617*	-0.524	-0.258	-0.579
	(0.322)	(0.375)	(0.480)	(0.360)
Received 1 Surgery	0.297	-0.0417	-0.185	0.392
	(0.359)	(0.393)	(0.389)	(0.316)
Received 2 Surgeries	0.0817	0.270	0.229	-0.0952
	(0.492)	(0.509)	(0.546)	(0.520)
Received 3 Surgeries	0.325	0.265	-0.334	-0.0142
	(0.484)	(0.491)	(0.666)	(0.545)
Received 4+ Surgeries	0.850*	0.282	0.293	0.725
	(0.432)	(0.571)	(0.731)	(0.876)
Any OS surgeries?	0.145	0.206	0.0540	0.184
	(0.323)	(0.340)	(0.343)	(0.302)
Birth Order	0.0813	-0.0892	-0.102	0.210
	(0.139)	(0.138)	(0.152)	(0.144)
Age	-0.0122	-0.0377	-0.0658**	0.0174
	(0.0273)	(0.0244)	(0.0274)	(0.0256)
Male	-0.161	0.0202	-0.570***	-0.141
	(0.173)	(0.158)	(0.201)	(0.177)
Constant	0.131	0.770	1.527**	-0.564
	(0.631)	(0.561)	(0.619)	(0.613)
N	224	224	224	224

Robust standard errors clustered at the household level are in parentheses.
* $p < 0.10$, ** $p < 0.05$, *** $p < 0.01$

Table 6: The Impact of an Average Surgery on Psychological Variables – Parent Data

	Psych Wellbeing Index	Depression	Anxiety	Self consciousness	Hopefulness	Extent to which child's health causes emotional distress
Cleft severity	-0.125***	-0.272***	-0.234***	-0.227***	0.114***	-0.200***
	(0.0416)	(0.0641)	(0.0655)	(0.0630)	(0.0356)	(0.0555)
No. of surgeries received	0.112	0.115	0.265	0.0585	0.0179	0.159
	(0.144)	(0.235)	(0.199)	(0.274)	(0.122)	(0.200)
Any OS surgeries?	-0.194	0.523	-0.274	0.350	-0.467*	-0.508
	(0.247)	(0.353)	(0.308)	(0.342)	(0.269)	(0.345)
Birth Order	0.0606	0.245**	0.146	-0.0122	0.00415	-0.00656
	(0.0820)	(0.117)	(0.115)	(0.138)	(0.0954)	(0.140)
Age	-0.00923	0.0700**	0.0138	0.00887	-0.0430	-0.0175
	(0.0227)	(0.0309)	(0.0291)	(0.0327)	(0.0284)	(0.0418)
Male	0.169*	0.0778	-0.0294	0.219	0.0883	0.222
	(0.100)	(0.130)	(0.143)	(0.151)	(0.161)	(0.140)
Constant	0.0832	2.082***	3.389***	3.379***	4.184***	4.066***
	(0.517)	(0.716)	(0.685)	(0.784)	(0.605)	(0.912)
N	308	308	308	308	308	308

Robust standard errors clustered at the household level are in parentheses.
*p < 0.10, ** p < 0.05, *** p < 0.01

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