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# The Impact of Being Born with Cleft and Cleft Reparative Surgery on Overall Health and Speech Outcomes

*JEL Classifications: I14, I15, I18, L31*

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Abstract: Orofacial cleft is one of the most common and treatable birth defects in the world. If left untreated, orofacial cleft can impair normal speech development, growth, and could lead to a number of health consequences later in life. The main motivation of the study is to measure the impact of being born with cleft and the cleft reparative surgery on overall speech and health cleft for teenagers in India using difference-in-differences approach along with household fixed effects method. An overall health outcome was measured using height, weight, grip strength and BMI, and the speech acceptability was measured using a “Universal Parameters of Speech Evaluation”. At the current sample size, the result suggests that there is no significant impact of being born with cleft and receiving cleft reparative surgery on the overall health outcome. However, being born with cleft decreases overall speech acceptability by 0.327 ( $p < 0.01$ ) standard deviations but I find no significant impact of receiving surgical treatment on overall speech quality.

I would like to thank my advisor, Professor Wydick, for all the time and feedback he provided for this study and Professor Stopnitzky for all the suggestions that helped improve this paper. Next, I thank my research partners Jeremiah Maller and Sam Manning, for being great team players, and Dr. Rashmi Rao for analyzing speech evaluations. Lastly, I would like to dedicate this study to our field coordinator, Saugata Gupta, who passed away without having the chance to finish the data collection and see the results of the study. He deserves an honorable mention for all his hard work, dedication and time he invested in quality data collection.

# 1. Introduction

Cleft lip and cleft palate are known as “orofacial cleft” and it happens when a child’s lip, and/or palate fail to form properly during early stages of pregnancy. It is one of the most common and treatable congenital birth defects caused by genetic and environmental factors. Currently, about every 1 in 700 babies are born with orofacial cleft, approximately 30% of the cleft is considered syndromic and 70% is non-syndromic<sup>1</sup> (WHO, 2002). If we assume that there are about 15,000 births per hour worldwide, a child is born with cleft every 2 minutes throughout the world (Mossey et al., 2009). The prevalence rate of such orofacial cleft varies from 1/500 to 1/2500 births depending on the geographic origin, race, socioeconomic status and ethnic backgrounds (Agbenorku, 2013). A study conducted in America shows that Asians have the highest risk of being born with any type of orofacial cleft, followed by Caucasians and the African Americans have the lowest risk of being born with cleft (S.K Das et al., 1995). An epidemiological study conducted by Panamonta et al. (2015) found that the descendants of American Indians had the highest prevalence rate of being born with cleft, followed by Japanese, Chinese and Caucasians.

The reasons as to why the prevalence rate varies among ethnicity is still unclear. The incidence of orofacial cleft is higher in developing countries than in developed countries (S. Ghani et al., 2004). This is because a majority of the cleft individuals in developing countries are unable to afford or do not have access to receive adequate cleft reparative surgery.

In developed countries, infants born with orofacial clefts receive reparative surgery within 3 years of age or earlier to maximize the benefits of cleft surgery, whereas in the developing countries infants born with cleft do not receive the necessary surgeries due to lack of resources or religious belief.<sup>2</sup> For instance, people who practice Islam and Hinduism consult with their religious healers before seeking Western practitioners for treatment intervention (Ross E., 2007). Muslim healers believe that orofacial cleft is god sent while in Hindu religion, it is seen as karma (Ross E., 2007). Also, it is challenging to obtain reliable data on orofacial cleft in developing countries due to limited resources (Mossey et al., 2012). Challenges to data

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<sup>1</sup> Syndromic cleft lip and palate is strongly characterized by chromosomal abnormality or monogenic diseases. While Non-Syndromic cleft lip and palate is associated with the interactions of the environment and genetics (Stupia et al., 2011).

<sup>2</sup> Muslim and Hindu traditional healers believe that orofacial cleft is God sent (Ross E, 2007)

collection in developing countries include: sensitivity to hierarchy, gender issues and distrust directed towards non-locals (Nori-Sarma et al., 2017).

If cleft is left untreated, the affected individual could suffer from various health complications such as impaired growth and weight gain, inability to feed, impaired speech development, hearing loss, upper respiratory infections, sleep apnea, impaired cognition, and psychosocial problems. Beyond aesthetics, psychosocial and health concerns, orofacial cleft has negative impact on education attainment which could hinder later life achievements (Persson M., 2012; Agbenorku, 2013)

To date, a majority of the existing literature focuses on health and speech outcomes for individuals who received cleft reparative surgery(s) at a very young age in developed countries. Few studies have been done in developing countries focusing on the physical and speech characteristics of cleft individuals who received the cleft surgery at a young age. To my knowledge, there are limited studies conducted on health outcomes for individuals with un-operated cleft or those who received cleft reparative surgeries later in life. Therefore, this study uses a new approach to measure impact of being born with cleft and receiving cleft surgery and serve as a contribution to existing wealth of literatures in craniofacial anomalies.

In order to study the impact of receiving cleft reparative surgery and how cleft severity affects speech and health outcomes; the study utilized simple difference-in-differences method combined with household fixed effects to measure the differential health and speech outcomes of the cleft individuals. The health outcomes are measured in terms of height, weight, BMI, and grip strength while the speech outcomes are measured using Universal Parameters of Speech Evaluation which is discussed in Section 4 of this paper. An Anderson Index was built to measure overall health outcome and speech acceptability parameter was used to determine overall speech outcome. We hypothesize that being born with cleft has negative impact on overall health and speech outcomes and receiving cleft reparative surgery has positive impact on overall health, and speech outcomes.

This study was conducted in West Bengal and Andhra Pradesh region of India in collaboration with Operation Smile Inc., an international non-profit organization specialized in providing reparative cleft surgeries for those who do not have access to such treatment. The study participants were cleft individuals between the ages of 11-19, their un-affected closest age sibling over the age of 7, and one of their parent/guardians. To date, we have surveyed 228 patient and sibling participants.

At the current sample size, the findings suggest that there is no statistically significant impact of being born with orofacial cleft and no significance in receiving a reparative surgery on overall health. However, the parents perceive that being born with cleft has a negative impact on overall health ( $p < 0.10$ ) but do think that cleft affects the ability to feed, hear or completing daily tasks. The parents' response to survey questionnaires may present social acceptability bias to avoid social stigmatization. The results for speech outcome suggests that overall speech suffers with increasing cleft severity but most importantly, speech acceptability decreases by 0.327 ( $p < 0.01$ ) standard deviations. However, the cleft restorative surgery does not have a significant impact on speech acceptability.

The paper is organized as follows: Section 2 of the paper provides background information on orofacial cleft; Section 3 covers findings from existing literatures on health and speech outcomes; Section 4 addresses methodology, data and empirical strategy; Section 5 contains the key results from the study; Section 6 provides discussion and concluding remarks.

## 2. Background on Orofacial Cleft

In 2005, *Jornal de Pediatria* defined cleft as "...congenital defect which can be defined based on their manifestations in terms of the discontinuity of structures of the lip, palate or both, with these lesions occurring at different locations and to a variable extent". Orofacial cleft involves the lip, hard roof of the mouth (hard palate) or soft tissue in the back of the mouth (soft palate) (Agbenorku, 2013). Orofacial clefts are classified based on the following characteristics:

- *Uni (one sided) or bilateral (two sided) cleft lip only (CLO)*: this can be characterized as one or more clefts on either side of the upper lip and it is often a result of not fusing properly during embryonic development.
- *Uni- or bilateral cleft palate only (CPO)*: cleft affecting either hard and/or soft palate of the mouth and caused by a failure to fuse properly during embryonic development.
- *Cleft lip and palate (CLP)*: cleft that affects both the lip and the roof of the mouth that is caused by a congenital fissure.

The functions of the lip, hard and soft palates are especially important in feeding and speaking. The primary lip functions include creating suction to drink, keeping food and water in, and helping to create different sounds for speech (Lip, P.C. 1999). The hard palate is important for creating suction to feed and it interacts with tongue for speech development, whereas the soft palate blocks the nasal passages during feeding and prevents regurgitation

(Lip, P.C. 1999). An individual born with cleft lip and/or palate won't be able to feed and speak properly due to lack of functions from the lip and palate.

Each type of anomaly has different degrees of cleft severity ranging from complete to incomplete cleft for both lip and palate (Figure 1), and the surgical outcomes are heterogeneous as a result depending on the timing of surgical intervention. It may require several surgeries and a number of follow-up treatments for the cleft to be restored to "near normalcy".

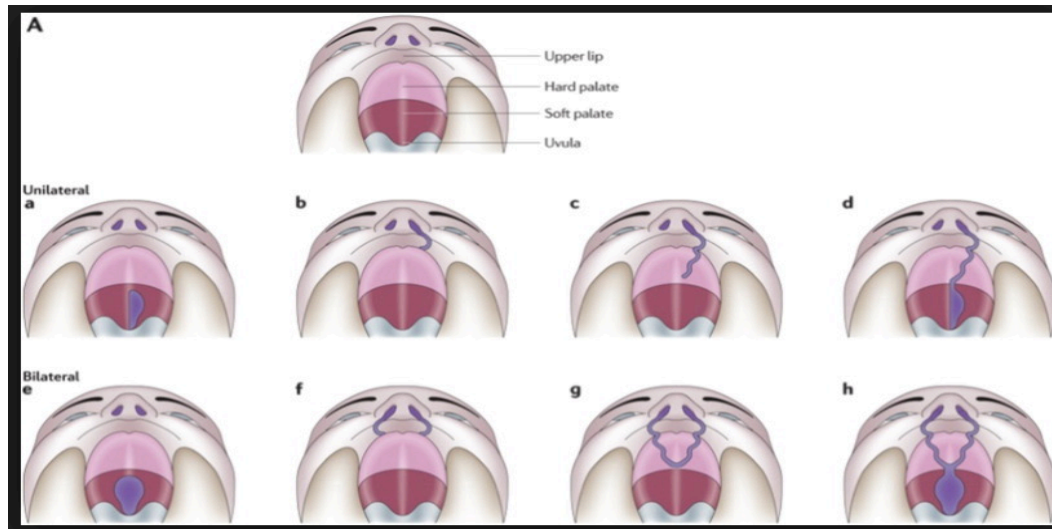


Figure 1. Characteristics of unilateral and bilateral cleft lip and/or cleft palate. Adapted from "Dental materials for cleft palate repair, by Sharif et al., 2015, *Science Direct*. Copyright 2015 Elsevier

On average, children with bilateral CLP receive about 10 surgeries, 5.3 surgeries for CLO and 5.9 for CLP throughout their lifetime (McIntyre, 2016). The study also concluded that cleft patients can receive more than 10 surgeries if they were operated on by different surgeons (McIntyre, 2016). It is highly recommended that the cleft lip be repaired within 12 months and the palate be repaired within 18 months of birth to optimize the benefits of the cleft surgery (CDC, 2006).

The cause of orofacial cleft is very complex and not well understood. There are multiple factors which could cause an individual to be born with cleft. This could range from chromosomal abnormalities, syndromes, genetics, smoking, environmental toxins, geographical locations, certain drugs the mother was taking during early stages of pregnancy, drug and alcohol abuse, socioeconomic status and nutrition deficiency (Agbenorku, 2013; WHO 2002; Mossey et. al., 2009; CDC 2006).

Being born with cleft can have many negative consequences for the affected individuals. Often cleft individuals have several different issues that can include aesthetic problems, congenital heart issues, feeding difficulty, hearing loss caused by infections in the ear, lag in speech development, growth impairment and psychosocial issues (Mohd et. al., 2015; Montagnoli et. al., 2005; Timmons et. al., 2001; WHO, 2002). The most common types of problems identified by parents were nasal regurgitation and vomiting (Lee et.al.,1997). Inability to feed could impair growth, weight gain and result in lower BMI; hence, the reason why early treatment intervention is recommended. It should be noted that children born with CPO have a higher frequency of genetic syndromes that put these children at higher risk of developmental abnormalities (Jones, 1988; WHO, 2002; Timmons et al., 2001).

### 3. Literature Review

#### *3.1 Theory of Change*

A “Theory of Change” addresses how a series of activities produce a result that can contribute to achieving an impact (UNICEF, 2014). The theory of change involves identifying the problem, opportunities to address the issue, intended outcome and the expected impact (UNICEF, 2014). The motivation of this theory of change is to address the burden of being born with orofacial cleft and the challenge of managing cleft in developing countries. This paper utilizes a result chain, a simplified version of obtaining the anticipated outcomes, but it plans out the important steps to achieve the desired impact.

There is shortage in adequately trained medical professionals in developing countries that can treat orofacial cleft and the patients often lack access to such resources due to accessibility and affordability (Ghani et al., 2004; O'Donnell, 2007). Non-profit organizations have made significant impact on the management of cleft patients, but addressing orofacial cleft still remains a big challenge in developing countries (Cubitt et al., 2014). There are about 4.8 billion people globally that lack access to surgical care (Scott Corlew et al., 2017) and one of which includes orofacial cleft surgery.

Non-profit organizations, such as Operation Smile, fly international medical professionals and surgeons to a central location where cleft surgeries can be performed on cleft individuals free of charge. This also allows local doctors and internationally trained medical professionals to exchange experience and knowledge. By conducting an impact evaluation study on the burden of being born with cleft, and the impact of receiving a cleft surgery will allow

international organizations to pool more resources to make more impact on managing orofacial cleft in developing countries. This will serve as the inputs in theory of change.

The importance of early surgical intervention is to restore orofacial cleft, prevent further health, speech, cognitive and psychological complications caused by cleft. A study was conducted using DALYs (disability life adjusted life years) to measure the impact of cleft restorative surgery. The results found that CL surgery averted 2.2 DALYs and CP surgery averted 3.3 DALYs on average (Scott Corlew et al., 2017). A similar study was conducted in Eastern and Central Africa and found that mean averted DALYs per patient were 5.6 and the surgical correction resulted in \$292 million in economic gains (Hamze et al., 2017). If a treatment intervention is delayed the cleft individual may suffer from multiple complications later in life. Therefore, the immediate outcome in the results chain is more surgical interventions to treat cleft patients to prevent future health complication and allow international professionals to provide positive spillovers through knowledge exchange with the local medical team. The overall impact is the reduced prevalence rate of orofacial cleft and cleft management in developing countries.

### *3.2 Physical Health*

There are several studies focusing on growth and weight gain patterns of cleft children to evaluate whether there is a normal growth pattern after receiving the surgical treatment. Children born with orofacial cleft experience difficulties in feeding due to lack of proper functions of the lip and palate. Feeding difficulties caused by orofacial cleft in the first months of life, as well as infections in the upper airways or in the middle ear, are factors that cause impaired growth (Montagnoli et al., 2005). Because of these difficulties, health professionals recommend early treatment intervention for any craniofacial anomalies<sup>3</sup> to maximize the benefits of the treatment for better health outcome later in life.

Different types of cleft (i.e., CLO, CLP, CLP)<sup>4</sup> can affect the child's growth pattern and ability to gain weight differently shortly after birth. A study conducted in Denmark found that infants born with orofacial cleft significantly showed reduced length and weight after 9 weeks (around the typical time when cleft lip surgery is performed) and 22 months of birth (around the typical time when palate surgery is performed) (Jensen BL et al., 1988). The researchers

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<sup>3</sup> Craniofacial anomalies refer to deformity in bones of the skull and face. There are diverse group of deformities and orofacial cleft is one of the widely known craniofacial anomaly (WHO, 2002).

<sup>4</sup> Refer to section 2



separated the cleft children into CLO, CPO and CLP groups and found varying growth and weight gain achievement among males and females. Both males and females with CLP and CP showed significant reduction in body length and weight. The result was consistent with similar studies done in different countries. For instance, in Brazil a study was conducted on children aged 1-24 months to evaluate impairment in growth and height for cleft children. The result was compared to the NCHS reference and concluded that CLP and CPO children showed severe impairment in growth and weight gain (Montagnoli et al., 2005). A study conducted at the University of Iowa aimed to explore the pituitary volume for cleft individuals in the age range of 7-22 years and found that there was no significant change in pituitary volume. However, the study noted that cleft males were shorter in stature and had lower BMI compared to non-cleft males. Additionally, cleft females had a slower growth rate, but their BMI did not differ from non-cleft females (Van Der Plas E et al., 2012). A similar study concluded that males with unilateral CLP had impaired growth when compared to their non-cleft counter-parts (Bowers et al., 1987). Another study found that adult males with CLO and CLP aged 19 years were shorter in height compared to unaffected individuals in the same age cohort (Persson M. 2017). It is important to note that the majority of these studies were conducted on individuals who received the cleft surgery at a young age. As far as growth and weight gain, studies suggest that being born with orofacial cleft has a negative effect on growth and weight gain patterns, though other studies suggest that the gap can be closed with early surgical intervention (Barakati et al., 2002; Ranalli et al., 1975; Frietas et al., 2012, Lee et al., 1997).

Children who received cleft lip surgery between 0-3 months of age and cleft palate surgery between 12-18 months of age showed no significant difference in growth pattern when compared to children born with different types of cleft and without any cleft (Barakati et. al., 2002). Cleft children tend to lag in growth and weight gain due to early life health complications shortly after birth, but by 3 years of age, the cleft individuals catch up to the normal height and weight after being treated (Ranalli et.al., 1975). However, these studies did not find a significant difference in growth and weight for children born with different types of cleft after being treated. These results are consistent with another study conducted in Brazil. The result of the study conducted in Brazil suggested that children born with cleft weigh less than unaffected children during the first months of life, but this gap closes by the end of the first year because of early life cleft treatment intervention (Frietas et.al., 2012). Cleft palate is associated with growth deficiency, but rapid recovery takes place following restorative surgery

which results in no residual growth impairment (Lee et.al., 1997). A majority of these studies were done on children who received the surgical treatment at a recommended age and the results confirm the importance of early life treatment intervention for cleft individuals.

There are limited studies done on individuals with un-operated cleft and those who received the treatment during adolescent years. It is difficult to find individuals with un-operated orofacial cleft for a study because a majority of the individuals receive the surgical treatment at a young age. However, this scenario can be completely different in developing countries because of lack of access to medical services that can treat craniofacial anomalies, inability to afford such care, and cultural influences (Ghani et al., 2004). For instance, in Bangladesh, there are approximately 300,000 people with orofacial cleft and a majority of these people are unable to afford cleft surgeries. Furthermore, there is a shortage of adequately trained surgeons (Ghani et al., 2004). The birth prevalence in India alone is about 27,000-33,000 per year (Mossey et al., 2009). This means there are a number of un-operated individuals in developing countries who are unable to afford treatment due to availability, geographic accessibility, affordability and acceptability (O'Donnell, 2007). Additionally, obtaining quality data is challenging in developing countries due to lack of resources, infrastructure, quality data collection, cultural constraints and lack of qualified statisticians (Asad Elahi, 2008).

To my knowledge, there are very few studies on measuring physical characteristics using grip strength of cleft individuals. One study conducted extensive research on physical characteristics of Swedish cleft men aged between 17-19 years, and the result suggested that there was no significant reduction in muscular strength for individuals born with CLO and CLP. However, significant reduction was observed for the CP group when compared to the unaffected individuals (Persson M., 2017).

### *3.3 Speech*

The important aspects of receiving a cleft surgery at a young age are the ability to feed and develop normal speech outcomes later in life. There are two important phases of speech development: *pre-linguistic* and *linguistic*. The pre-linguistic phase involves babbling and gestural communication, while the linguistic phase consists of true words and the development of spoken language (D'Antonio & Scherer, 2008). Early intervention is important for pre-linguistic vocalization and language development (Mitacek, 2014). One study found that babies with un-repaired cleft palates had a smaller canonical babbling ratio when compared to their age cohort; 57% of the babies with cleft palate achieved canonical babbling stage by 9 months

compared to 93% of the non-cleft babies (Chapman et al., 2001). At 12 months of age, children with CLP babbled less than un-affected children (Scherer et al., 2008). The children with CLP produced about 41% fewer babbled utterances than un-affected children (Scherer et al., 2008). Therefore, delays in speech development in cleft infants may lead to delays in language development later in life (Mitacek, 2012).

Cleft lips are surgically closed at three months of age while cleft palate repair is done between nine and twelve months or later (Kuehn & Henne, 2003). One study aimed to evaluate the speech outcomes for children who received a palate repair before the age of six months and found that 87 out of 100 subjects developed acceptable speech, and the remainder had unacceptable speech at age five (Copeland M., 1990). A similar study was conducted in Australia, but with a different approach to evaluate the speech outcomes of cleft children treated at three months of age. The study asked *non-trained* listeners (non-speech pathologists) to compare speech outcomes for those with and without cleft and found that overall, children born with cleft palate had less acceptable speech (A.D Bagnall & D.J David, 1988). Sometimes, even if there is an early intervention, some cleft children exhibit abnormal speech patterns (Nagarajan et al., 2009). Additional surgery, referred to as secondary surgeries, may need to be performed to treat any lingering speech problems related to the cleft palate condition (Kuehn & Henne, 2003). If speech and language problems persist after cleft surgery, then additional intervention may be necessary (Prathanee et al., 2016). It is important to note that cleft surgery alone may not be able to restore speech, speech therapy is highly recommended to fix pre-existing speech anomaly caused by cleft.

Existing literature results are mixed when it comes to later life speech development and the timing of the cleft palate surgery. For instance, a study conducted in India found that children who received primary palate repair after ten years of age showed significant reduction in articulation errors and improved resonance. However, nasal emission showed little improvement and very few achieved normal speech (Murthy 2009). One study noted that children made progress in their speech development during school age years (linguistic phase), but the progress was much more rapid for the younger children compared to older cleft children (D'Antonio & Scherer, 2008). In Sweden, speech pathologists assessed the impact of two-stage cleft palate surgery on children who received soft palate surgery at the age of seven months and hard palate surgery between 38-89 months to assess any correlation between speech outcome and timing of cleft palate surgery. The study concluded that earlier cleft palate

operation had no effect on speech complications up to age ten (Lohmander et.al., 2009). Another study conducted in Malaysia found that children who received cleft surgery around six months of age showed overall poor speech and hearing status, despite the fact that the surgery was performed during early infancy (Mohd et.al., 2015). Contradictory to the previous studies, a study done in Finland found that children who received cleft surgery as early as 12-18 months showed better speech outcomes than those who received the surgery later than 22 months (Haapanen, M.L & Rantala S.L 1992). If the palate repair is delayed, it may be difficult for a cleft child to integrate the pharyngeal muscle movements<sup>5</sup> for speech that has already been established, even if the current muscle movement do not result in acceptable speech (Jones & Jones, 2005).

The relationship between early speech, later speech and language development in cleft population has been a popular research area, but there is limited data available to study the differential outcomes of speech in cleft population (Mitacek, 2012). As noted in the previous section, there are very few studies done on cleft populations in developing countries. Sell and Grunwell published a study that had been conducted on un-operated individuals more than 11 years of age in Sri Lanka. They assessed speech outcomes prior to palate repair, eight months after the surgery before speech therapy, eight months after speech therapy and 12 months postoperatively. The results suggested that speech was severely impaired with late repair, but overall speech improved after speech therapy (Sell & Grunwell, 1990).

It is important to note that speech outcomes cannot be just evaluated using a single measurement; the trained professionals use different parameters to measure different aspects of speech outcomes. The speech pathologist is involved early on to identify children who are at risk of developing speech disorders to initiate early intervention to mitigate any speech disorders caused by cleft (Nagarajan et al., 2009). In order to evaluate speech disorders, speech pathologists use “Universal Parameters for Reporting Speech Outcomes” (Henningson et. al., 2008). The parameters include the following:

- *Hypernasality*: excessive nasal resonance heard on vowels and consonants.
- *Hyponasality*: decreased or insufficient nasal resonance heard on vowels and consonants.

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<sup>5</sup> The activities such as swallowing and speaking depend on the ability to obtain adequate closure of the velopharyngeal port. Velopharyngeal movements are quite distinct from those involved in swallowing and it is an important part of speech (Schloegel et al., 2015)

- *Voice Disorders*: a deviation in voice characteristics due to structural and functional level of the larynx.
- *Audible Nasal Air Emission and/or Turbulence*: audible escape of air through the nasal passage (non-turbulent). If air emission accompanies a snorting sound, it is considered turbulent.
- *Consonant Production Errors*: This is measured in single words and sentence level. Often times individuals with CP have difficulties producing consonants that require high amounts of intra-oral air pressures (Prandini et al., 2011)

In addition to the above 5 parameters, 2 other global ratings are also looked at when evaluating speech and the following are included:

- *Speech understandability*: Measures how well the speaker's message can be understood by the listener.
- *Speech Acceptability*: Measure how acceptable the speech is based on the acceptable range compared to the general population.

The communication disorders associated with different types of cleft can vary (D'Antonio & Scherer, 2008). Children born with CLP and CPO have negative speech outcomes in the linguistic phase. Several studies suggest that children with CLP, especially those with CPO, continue to show poorer language performance than un-affected individuals later in life (D'Antonio & Scherer, 2008). Children with CLP had the highest articulation errors, hypernasality, voice disorders and poor speech understandability (Prathanee et al., 2016). Similarly, children with unilateral CLP showed overall poor speech outcomes despite the fact that two-thirds of participants underwent speech therapy (Sell D et.al., 2001). Another study found that CPO patients had poorer results after receiving primary surgery than CLP patients (Timmons et al., 2001). They did not find any correlation between cleft severity and speech outcomes.

Overall the studies conducted on health and speech were conducted on small sample size and do not have strong comparison group. Majority of the existing literatures compared health and speech outcomes to age matched cleft counterparts born with different types of cleft. The next section will discuss the methods used that differentiates this paper from the rest of the existing studies.

## 4. Methodology

This study aims to provide more insights on health and speech outcomes for those born with orofacial cleft and measure the impact of receiving orofacial surgery on overall health and speech outcomes. Overall health outcome is being measured using height, weight, grip-strength and BMI parameters, and overall speech outcome is measured using hypernasality, hyponasality, audible nasal air emission, speech acceptability and speech understandability. Since multiple health hypotheses are being tested, an Anderson Index was built to pool these outcomes into one to measure overall health outcome. Anderson index is robust to over-testing, provides an overall general effect of the treatment, and is more efficient than testing multiple single hypotheses (Andersen, 2008). Speech acceptability is a measure of overall speech outcome and composed of five different speech parameters.

The research aims to answer the following questions:

- 1) What is the impact of being born with cleft on overall health and speech outcomes later in life?
- 2) What is the impact of receiving orofacial surgeries on health and speech outcomes later in life?

This study evaluated two treatment effects: the impact of being born with orofacial cleft and the impact of receiving a cleft surgery. The control group is the un-affected siblings to the cleft individuals and the counterfactual is the difference in health and speech outcomes between un-operated cleft individuals and their siblings. In other words, the counterfactual would be what would happen to the treatment group in the absence of treatment.

The primary data collection took place in the states of West Bengal and Andhra Pradesh in India, in collaboration with Operation Smile, an international non-profit organization whose mission is provide cleft reparative surgeries free of charge for those who are unable to afford it or have no access to such treatment.

The study was approved by the Ethics in Research Committee of the Mahatma Gandhi Mission's Dental College and Hospital (MGM) on July 4<sup>th</sup>, 2017 and the Institutional Review Board (IRB) at University of San Francisco.

### *4.1 Data*

The primary data comes from surveying the cleft individuals, their age-proximate sibling (greater than 7 years of age) regardless of their gender as well as one of their parents/guardians. Parents/guardians were surveyed separately to capture data on overall

health and well-being based on parental/guardian's observations. The closest age cousins who were raised in the same household were surveyed in the event if the age proximate sibling was not reachable and/or unable to participate due to geographical distance. Those with obvious syndromic cleft were excluded from this study.

The patient and sibling survey was designed to collect basic demographic information on cleft individuals and their age proximate sibling such as age, gender, number of siblings, and birth order as well as their weight (kg), height (cm), BMI and grip strength (kg) using a hand dynamometer. BMI is calculated using body weight (kg) divided by height in square meters (m<sup>2</sup>).

$$BMI = \frac{\textit{Weight in kilograms}}{\textit{Height in meters}^2}$$

The parental survey captures basic demographic information, number of children and socio-economic data including occupation and level of education, whether the household has electricity and a toilet/latrine, material the home is constructed of, and religion. The parent/guardian was asked a set of physical health questionnaires on a scale of 1-5 (1 being strongly disagree and 5 being strongly agree) about their cleft and non-cleft children (see Appendix I). In addition, the parents/guardians were asked about the details of their cleft children's past surgeries and cleft types (see Appendix I).

The treatment group consists of individuals born with cleft and individuals who received partial or full cleft surgery, and the control group consists of un-affected siblings to the cleft individuals. We assumed that the treatment is assigned at random because the deformity itself affects individuals randomly.

The un-operated cleft treatment group must be between the ages of 11-19, had not yet received any surgical treatment, and must have an age proximate sibling older than 7 years of age. A list of primary care patients<sup>6</sup> who were scheduled to be operated on at Operation Smile's surgery mission week in 2017 and 2018 was used for the study. The surgery mission usually lasts for a week, and days one and two are screening days where health professionals examine the cleft patients to ensure that they are good candidates for the surgery. The respondents were either surveyed during screening days or at the screening camps during the months prior to the surgery mission. Operation Smile set up screening camps months before the surgery

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<sup>6</sup> Primary care refers to individuals who are set to receive their 1<sup>st</sup> orofacial surgery.

mission to recruit and identify individuals who need primary/secondary cleft surgeries. The screening camps were also used to identify and survey eligible respondents for the study.

The treated cleft treatment group came from a list of 708 previously treated cleft children in West Bengal and Andhra Pradesh between 2004-2017. This group consisted of individuals who received at least one cleft reparative surgery in the past and needed secondary surgery<sup>7</sup> or who were fully treated cleft individuals. The treated treatment group was surveyed at the surgery mission week or central location for surveying.

For both groups to be eligible for surveying, the participants had to be between the ages of 11-19 years at the time of the surveying, had to have at least one sibling older than seven years of age, and had to come with one of their parents/guardians. The unaffected age proximate sibling is considered a control group. Prior to surveying, informed consent forms, translated in Bengali and Telugu, were provided to all participants (see Appendix II). Those who refused to sign the informed consent forms were not included in the study. A monetary incentive was given after the completion of surveying if all eligibility criteria were met.

#### *4.2 Variable Construction*

##### *4.2.1 Health Outcome*

The main dependent variables of interests are health and speech outcomes. Health outcomes were measured using height (cm), weight (kg), BMI and grip strength (kg). In order to use these factors to measure health outcomes as a whole, an Anderson index was used to pool these outcomes into one. Most studies use anthropometric measures to obtain useful insights on individual's nutritional intake and overall health and well-being (Gorstein et.al., 1988; CDC, 1988). At an individual level, anthropometric indicators can be used to assess overall health and nutrition well-being (WHO, 2006). Height and weight can be used to assess the adequacy of diet and growth for infants and children (WHO, 2006). BMI is a measure used to define overweight and underweight (WHO, 2006). Grip strength has been strongly related to muscle strength, related physical activity levels and used to predict other disabilities later in life (Eckman et al., 2014). Low grip strength is correlated with malnutrition, higher risk of mortality from conditions such as cardiovascular disease and respiratory diseases (Eckman et al., 2014), and can be a good indicator for health in cleft individuals.

For this study, a digital weight scale, standard measuring tape and hand dynamometer were used to capture data on weight, height and grip strength. The participants were asked to

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<sup>7</sup> Secondary surgery refers to follow up or revision surgery and can be considered partially treated.



squeeze the dynamometer using their weak hand three times with 10 to 20 seconds of rest period in between. If the difference in scores was not within 3 kg for each attempt, the respondents were asked to squeeze the instrument one more time. The mean scores were used for the analysis. All health parameters were standardized to have a mean score of zero and a standard deviation of one.

#### *4.2.2 Speech outcome*

The overall speech outcome was measured using five parameters: hypernasality, hyponasality, audible nasal air emission, speech acceptability and speech understandability. Only three parameters from Universal Parameters for Reporting Speech Outcomes and two global ratings were used to measure speech outcome. Voice disorders and consonant production was not measured due to background noise recorded in speech recordings. The speech acceptability parameter itself measures overall speech outcome as a whole. Therefore, an Anderson Index was not used for the speech outcome. At the time of surveying, voice recordings were obtained while the respondents were asked to count from 1 to 20, 60 to 70, and reading from short and long passages. Counting from 60-70 allows the speech pathologist to evaluate for hypernasality. Counting from 1-20 and reading tests allow the evaluation of hyponasality, nasal emission, speech acceptability and understandability. The voice recordings were then sent to a speech pathologist for evaluation. Since individuals with CLP and CPO have the worst speech outcomes compared to the CLO group<sup>8</sup>, the speech pathologist was not informed of the cleft severity and treatment status to avoid any bias in the speech measurement. Not all of the participants were able to read due to their young age or illiteracy, but the speech pathologist was able to evaluate all five parameters based on counting from 1 to 20 and 60 to 70.

Unlike the health parameters, the speech parameters were scored based on the severity of speech abnormality. Table 1 represents the speech scoring protocol used to measure the overall speech outcome in this study. Speech understandability and speech acceptability are scored based on 0-3, whereas hypernasality, hyponasality and audible air emission are scored 0 and 1. Any deviation from 0 indicates the presence of speech abnormality and its severity.

The cleft severity was measured using the number of expected surgeries the child should be expected to have, on average, to restore the deformity to “near normalcy”. The

higher the number of surgeries, the more severe the cleft characteristic would be. Table 1a represents average surgery scenarios and were established in consultation with the Operation Smile medical team.

Hypernasality- Single Words		Speech Understandability - Conversational Speech
0= Within Normal limits/None		0= Within normal limits, easy to understand
1= Present		1= Mild, occasionally hard to understand
		2= Moderate, hard to understand
		3= Severe, hard to understand
Hyponasality-Sentences		Speech Acceptability- Whole Speech Sample
0= Within Normal limits/None		0= Within normal limits, normal speech
1= Present		1= Mild, speech deviates from normal to a mild degree
		2= Moderate, speech deviates from normal to a moderate degree
Audible Air Emission and/or Nasal Turbulence- Single Words		3= Severe, speech deviates from normal to severe range
0= Within Normal limits		
1= Present		

Table 1: Scoring protocol for measuring speech outcomes for all five speech parameters.

Diagnosis	Average expected number of surgeries
Incomplete unilateral CLO	2
Incomplete bilateral CLO	2
Incomplete unilateral CPO	3
Incomplete bilateral CPO	3
Complete unilateral CLO	4 (2 lip and 2 nose repairs)
Complete bilateral CLO	4 (2 lip, 1 jaw and 1 nose)
Incomplete CLP (bilateral/unilateral)	5
Complete unilateral CLP	6 (primary & secondary lip & palate repair, alveolus <sup>9</sup> , nose repair)
Complete bilateral CLP	7 (primary & secondary lip & palate repair, nose, 2 alveolus repairs)
Complete bilateral CLP with deviated premaxilla <sup>10</sup>	8 (primary & secondary lip & palate repair, nose, 2 alveolus, & jaw)

Table 1a: Average expected number of surgeries and diagnosis.

<sup>9</sup> Alveolus is a defect in the bone around the teeth in front of the palate.

<sup>10</sup> Deviation of small cranial bones at the tip of the upper jaw or maxilla

### 4.2.3 Empirical Strategy

The study used simple difference-in-differences (DID) method along with family level fixed effect to measure the impact of being born with different types of cleft as well as the impact of receiving cleft reparative surgeries. The DID approach allows us to control for the unobserved differences between the two groups and fixed effect eliminates any time-invariant factors  $\alpha_i$  (e.g. age, education level, socioeconomic status etc.) between households.

We measured speech and health outcomes of the previously treated respondents to their age proximate un-affected siblings and compared that difference to the difference between the un-treated respondents and their un-affected age proximate siblings within household. The counterfactual in this study is the difference between health and speech outcomes of un-operated cleft teenagers and their un-affected siblings in India, which means the outcome of the treatment group in the absence of treatment would be similar to the control group. The sibling comparison strategy allows us to compare the health and speech outcomes for individuals (cleft child and their sibling) who were raised in the same environment and have biological similarities.

The key identifying assumption in DID is parallel trend assumption, which strictly assumes that there is no differential trend between the control and treatment groups in the absence of treatment. Any difference in the outcomes between the two groups must be caused by the treatment itself. However, this assumption only applies if there is time trend,  $T_1$  and  $T_0$ , but this study utilized cross sectional data where time-trend does not apply. Thus, the traditional identifying assumption of DID cannot be tested directly on this cross-sectional study. Instead of traditional time trend, the study has incorporated sibling trend; assuming that the difference in overall health and speech outcome between the control and treatment group is similar in the absence of treatment. However, the identifying assumption may be violated if the parents reallocate more time and resources to the cleft child at an expense of their un-affected child. If that is true, then this will lead an upward biased estimates of the impact. Additionally, there could be a favoritism bias among parents. If the cleft child is their least favorite because he/she was born with a deformity; they may neglect to care and nurture the child. In this case, the estimates for the impact will be biased downwards. In order to control for this, the model incorporates family level fixed effects.

The main regression equation is the following:

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

The dependent variable  $y_{ij}$  is the outcome of interest built using an Anderson Index for overall health and speech outcomes (standardized) for person  $i$  in household  $j$ ,  $C_i$  represents cleft severity measured in number of expected surgeries the child should be getting (Table 1),  $S_i$  is the number of surgeries received,  $OS_i$  is a dummy variable representing surgeries performed by Operation Smile,  $\mathbf{X}'_{ij} \boldsymbol{\vartheta}$  is the vector includes control variables such as birth order, gender and the child's age. The last term  $\mu_j$  denotes the family fixed effects. The main coefficients of interest are  $\beta C_i$  and  $\tau S_i$ , where  $\beta C_i$  allows us to measure the impact of being born with different types of cleft, while  $\tau S_i$  allows us to measure the impact of receiving cleft surgery on health and speech outcomes. Standard errors are adjusted for clustering at the family level.

Additionally, an assumption has been made that an increasing the number of surgeries will have diminishing returns on health and speech outcomes. The first and second surgery (i.e., primary surgery) may have the biggest impact on all individuals with different cleft types. The impact of number of surgeries after the primary surgery may have a heterogeneous effect on the outcomes depending on the cleft severity and the timing of the surgery. Therefore, our second regression model includes number of surgeries ( $\mathbf{S}'_i \boldsymbol{\tau}$ ) the cleft child has had.

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

The main coefficients of interest in model two are  $\beta C_i$  and  $\mathbf{S}'_i \boldsymbol{\tau}$ . It should be noted that the cleft surgery cannot completely restore aesthetics and functioning of the lip and palate, but the restoration can be “near normalcy”. Therefore, the model allows us to evaluate the degree of restoration the cleft surgery provides.

## 5. Results

### 5.1 Descriptive Statistics for Health Parameters

Table 2 presents summary statistics on key variables for operated and un-operated cleft treatment group and their un-affected sibling control group. The table is divided into two panels. The first panel shows summary statistics for the un-operated and operated cleft treatment group between the ages of 11 and 19 years, and the second panel shows summary statistics for the

control sibling group. The table presents an unweighted sample mean on the key outcomes of interest, standard deviations, and minimums and maximums for each group. The summary statistics table was used to detect the presence of any outliers in the sample. There are a total of 228 patient and sibling pairs. Out of 228 observations, 60 are the un-operated cleft treatment group and their unaffected siblings, and 168 out of 228 observations are treated cleft treatment group and their un-affected siblings.

The overall mean for the key variables for the un-operated cleft treatment group are greater than the un-operated control sibling group, because there are 10 siblings under the age of eleven. The age range of the control group is from age 7 to age 33, while the treatment group is restricted to ages 11 to 19. In our sample there are 37 control siblings over the age 19 and 32 siblings are under the age of 11. However, it can be noted that the mean grip strength is lower in the un-operated cleft treatment group when compared to their age proximate siblings. The differences in key variables are driven by age differences between treatment and control group in Table 2.

Figures 1 to 4 present standardized bar graphs with error bars on weight, height, mean grip strength and BMI for un-treated, partially treated and fully treated cleft treatment group. However, no statistically significant differences can be detected among different cleft treatment groups by gender. To test for statistical significance, one-way ANOVA tests have been performed on all four health parameters by gender and the p-value suggested that there is no statistically significant difference between the different cleft groups on four health parameters.

### *5.2 Descriptive Statistics for Speech Parameters*

Table 2a represents the summary statistics on all five speech parameters by group. Not all sibling pairs had matching voice recordings, and the voice recordings with no sibling pair was dropped from the study and total of 186 observations were used for the analysis. Prior to the speech evaluation, the speech pathologist was not told of the cleft severity and treatment status to prevent upward biased estimates on speech parameters. Majority of the treated treatment group is clustered around mild to moderate severity on speech understandability and speech acceptability and existing hypernasality. This could mean that the hypernasality, speech understandability and speech acceptability were severe for the treated treatment group prior to the surgical treatment. There are total of 40 un-treated treatment and sibling control pairs. It can be noted that majority of the treatment and control group fall under no hyponasality and audible air emission.

### 5.3 Results

Table 3 presents the main DID regression output on the impact of being born with cleft and cleft reparative surgery on overall health using equation (1). The main output and coefficient of interests are presented in column (1). Surprisingly, the estimates suggest counterintuitive results. One unit increase in cleft severity increases overall health by 0.07 standard deviations while cleft surgery decreases overall health by 0.09 standard deviations. Columns (2) and (3) regression outputs are consistent with results from column (1). It can be noted that grip strength decreases by 0.18 standard deviation and BMI decreases by 0.13 standard deviation with 1 unit increase in cleft severity. However, cleft surgery coefficients for grip strength and BMI are negative.

Since the variable BMI was constructed using height and weight, I have removed the variable to reduce redundancy and test for significant difference. The result did not make a significant difference to the main dependent variable of interest. At the current sample size, the main coefficients  $\beta C_i$  and  $\tau S_i$  on all outcomes returned statistically insignificant results.

Table 4 presents the impact of varying cleft severity and each additional surgery outcomes on overall health. We measured cleft severity using the average number of cleft surgeries the affected individual is expected to have<sup>11</sup>. The assumption was that severe cleft has severe health impact for the affected individuals. Additionally, we assumed that each additional surgery after the primary surgery has diminishing returns on overall health. Column (1) presents the impact of different types of cleft severity and number of surgeries received on overall health impact. The estimated  $\beta$  coefficients for cleft severity are positive but statistically not significant and there seems to be no trend in the estimated  $\beta$  coefficients for cleft severity. The regression coefficients for number of surgeries received in column (1) are highly positive for the first two surgeries (i.e., primary surgery) the cleft individuals received on overall health but are not statistically significant. In our data, individuals who received one surgery had their cleft lip repaired and the second surgery was performed to repair their palate. Therefore, first and second surgeries are considered primary care. Column (1) results present that the first surgery improves overall health by 0.22 standard deviations while the second reparative surgery improves overall health by 0.50 standard deviation. The standard procedure is that the first surgery is performed to repair cleft lip to restore lip functions and support growth of the facial

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<sup>11</sup> Refer to Table 1a on page 17.

skeleton (Lip, P.C., 1999) and the second surgery is performed on cleft palate to create normal speech development and minimize maxillary<sup>12</sup> disturbance caused by cleft (Agrawal, 2009). The impact of receiving the third surgery decreases overall health by 0.01 standard deviations but the fourth surgery increases overall health by 0.19 standard deviations. The impact definitely seems to be lower in secondary surgeries and higher in primary surgery.

Table 5 presents regression output results using parental data. The survey questions<sup>13</sup> were answered based on parent's perception of their cleft child's physical ability and well-being. Column (1) represents the main results on overall health and the overall health index was built using dependent variables in Columns (2) to (6). The DID estimates suggest that being born with orofacial cleft decreases overall health by 0.10 ( $p < 0.10$ ) standard deviations and cleft surgery improves overall health by 0.04 standard deviations. However, the cleft surgery does not have a statistically significant impact on overall health. Columns (4) to (6) presents counterintuitive results for difficulty in feeding, hearing and executing daily tasks. The estimated  $\beta$  coefficients show that trouble eating decreases by 0.21 ( $p < 0.01$ ) standard deviations, while trouble hearing and trouble completing tasks decrease by 0.10 ( $p < 0.10$ ) standard deviations with increasing cleft severity. The estimates for  $\tau S_i$  implies that cleft surgery decrease difficulty in feeding and hearing but increases the difficulty in completing tasks. However, cleft surgery does not seem to have statistically significant impact on trouble eating, hearing and completing tasks. The parents of the cleft individuals perceive that cleft severity has negative impact on overall health but do not think that cleft affects the ability to feed, hear and complete daily tasks. The results in column (4) to (6) could be reversed due to multiple reasons. Majority of the parent's answer clustered around "strongly disagree" when asked whether there is a difficulty eating, drinking and hearing. It is possible that the parents may have misinterpreted the questions or there is a social desirability bias. In other words, the parents do not want to agree with the fact that their children suffer physically even if they do.

Table 6 represents the impact of varying cleft severity and number of surgeries on overall health using parental data. Column (1) shows that bilateral CLP decreases overall health by 0.819 ( $p < 0.10$ ) standard deviations which is expected since bilateral CLP is considered to be a severe cleft condition. However, Column (2) shows that good health increases by 1.175 ( $p < 0.10$ ) standard deviations if the cleft child has incomplete CLP and Columns (4), (5) and (6)

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<sup>12</sup> Maxillary refers to upper jaw while mandible refers to lower jaw.

<sup>13</sup> Parental questionnaire can be found in Appendix I.

presents the same counterintuitive results as Table 5. It is noted that the parents tend to agree with positive questions and disagree with negative questions. Nevertheless, the parents seem to think that severe cleft hinders overall health.

Table 7 Column (1) shows the results for speech acceptability and subsequent columns represent that results from four other speech parameters. Speech acceptability is a global measurement for reporting overall speech outcome. Column (1) shows that an increase in cleft severity decreases speech acceptability by 0.327 ( $p < 0.01$ ) standard deviations and speech understandability decreases by 0.347 ( $p < 0.01$ ) standard deviations. Hyponasality, hyponasality and audible air emission increases with increasing cleft severity at the 1% significance. However, cleft surgery does not have any significant impact in correcting speech outcomes. It is important to note that cleft surgery alone cannot not fix speech anomalies (Nagararajan et al., 2009) and any lingering abnormal speech pattern must be re-evaluated. Some individuals will need speech therapy after having their palate repaired to correct abnormal speech pattern caused by cleft. Also, speech acceptability may differ depending on the timing of cleft palate surgery.

In contrast, Table 8 provides detailed information on how different types of cleft severity and number of surgeries affect speech acceptability. Column (1) shows that speech acceptability decreases when an individual has varying degree CPO and CLP severity. The results from subsequent columns are consistent with the results from Column (1). These results are consistent with other studies as well (Mohd et.al., 2015; Timmons et. al., 2001). Similar to Table 7, the cleft restorative surgeries have no significant impact on speech acceptability. Column (3) shows that hyponasality decreases by 1.650 ( $p < 0.10$ ) standard deviations with the second surgery.

In our data, there are sibling pairs with age gap of more than 5 years. Therefore, a similar analysis was done using patient and sibling data but with age restriction of 5 years between the sibling pairs to test for difference in outcomes. Upon enforcing age restriction, 60 observations were dropped from the analysis, resulting in 168 observations for analysis. Tables 9 and 10 represents regression output on age restricted sibling pairs. Interestingly, the results are consistent with the results from Table 3 except for cleft reparative surgery. Results from Table 9 suggests that increasing cleft severity increases overall health by 0.02 standard deviations and cleft surgery improves overall health by 0.09 standard deviations but not at a statistically significant level. Table 10 provides more detailed information on the impact of varying cleft



severity and each additional surgery on overall health. The cleft restorative surgeries have positive impact but not at a statistically significant level.

Analyses were done using condensed version of cleft severity and cleft management care. The parents were asked to describe the cleft condition at birth and number surgeries the cleft child has gotten to date. If the child received 1 surgery on lip and one surgery on palate, then the child has completed primary cleft surgery. Subsequent number of surgeries after the initial cleft surgery is considered secondary surgery. A cleft individual can get multiple secondary surgeries. Medical literatures focus on primary surgery more than secondary surgery because it allows the cleft child to feed properly and allows for proper tongue and palate interaction to create normal speech pattern. Thus, Table 11 includes three different cleft categories: CLO, CLP and CLP and different cleft management: primary and secondary care. The overall health index was constructed using height, weight and grip strength. BMI variable was excluded from this analysis since it is calculated using height and weight. Column (1) results show that CPO and CLP have negative impact and CLO has positive impact on overall health.

In general individuals with CPO and CLP tend to have shorter stature and suffer with other congenital health problems (Persson M., 2012). Individuals with CLO are able to feed better compared to CPO and CLP individuals. As expected, primary care increases overall health by 0.15 standard deviations while secondary surgery decreases overall health by 0.09 standard deviations, however the results are not significant.

A similar analysis was done using the same variables on parental data shown on Table 12. The overall health index is built using the dependent variables from Columns (1) to (6). Column (1) estimates show that children born with CPO and CLP have the worst health outcome. Being born with CLP decreases overall health by 0.669 ( $p < 0.10$ ) standard deviations. However, parents think that secondary surgery has overall positive health outcome compared to the primary surgery. It is possible that parents could think that receiving multiple surgeries can help improve child's overall health. However, primary and secondary care estimates are not statistically significant.

Lastly, Table 13 represents speech acceptability for CLO, CPO and CLP groups. The results show that overall speech acceptability decreases with different cleft anomalies, but this decrease is significant for CPO and CLP. The speech acceptability and speech understandability decrease even more for individuals with CPO at the 1% significance. Individuals with CPO are vulnerable to syndromic cleft and often times experience developmental delay (Timmons et. al.,

2001; WHO, 2002). Overall, there is no significance in receiving cleft surgeries on overall speech. However, Column (2) suggests that hypernasality increases by 0.813 ( $p < 0.10$ ) standard deviations with secondary surgery. Existing studies suggest that secondary revision surgeries can improve speech acceptability and hypernasality if the velopharyngeal<sup>14</sup> flap is closed correctly (Kummer W., 2014). Also, the data consists of few number of individuals with more than three surgeries with existing hypernasality.

## 6. Discussion and Conclusion

Through this study, I have attempted to answer the following questions:

- What is the impact of being born with orofacial cleft on health and speech outcomes?
- What is the impact of receiving cleft reparative surgery on health and speech outcomes?

In order to answer these questions, the study utilized two different survey instruments: cleft treatment group and sibling control group pairs, as well as one of their parents/guardians. The overall health outcome was measured using height, weight, grip strength and BMI parameters, and the speech outcome was measured using hypernasality, hyponasality, audible air emission, speech acceptability and speech understandability. The identifying strategy is DID method using cross-sectional data. The method itself mimics random assignment, where the cleft condition is assumed to happen at random and DID controls for permanent differences between the treatment and control group. The study used an age proximate sibling comparison strategy, since siblings are a strong control group for the study. However, there could be a potential weakness in using fixed effect at a family level because cleft is considered to be genetic.

The main motivation of the study was to measure whether there is a negative impact of being born with cleft and if there is, what would be the degree of surgical restoration if the surgery was performed to correct it. The results demonstrated that there is no statistically significant impact of being born with orofacial cleft and impact of receiving cleft restorative surgery on overall health at the current sample size ( $N=228$ ). Although, there are number of medical studies evaluating the importance of early treatment intervention, there have not been

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<sup>14</sup> Velopharyngeal muscle is a soft tissue on your soft palate and it is important for speaking, drinking and feeding.

studies conducted that evaluate the health outcome of un-operated cleft individuals over the age of 10, besides one case study done in Japan on an un-operated cleft adult to evaluate for maxillary growth (Sakuda et. al., 1985).

The existing literature is not consistent when it comes to overall health outcomes after being surgically treated. Cleft conditions can have heterogenous outcomes on overall health and speech outcomes. For instance, individuals born with CPO and CLP are shorter in stature and weigh less compared to their non-cleft age cohort group (Bowers et. al., 1987; Montagnoli et. al., 2005; Persson M. 2017; Van Der Plas E et. al., 2012). Other studies have made counter arguments and concluded that early surgical intervention has a significant impact on overall health of cleft individuals and the surgical impact was not heterogenous among different cleft groups (Barakati et. al., 2002; Frietas et. al., 2012, Lee et. al., 1997; Ranalli et. al., 1976).

This study has a few shortcomings. In order to draw a causal inference, a larger sample size is needed for un-operated and operated cleft treatment groups. The study had 30 un-operated cleft and 84 treated treatment groups, which could explain why some variables display no significance. Also, the study tried to exclude individuals with obvious syndromes in the treatment group, but some may have been included in the study since the participants were not evaluated by a medical professional to rule out any syndromes associated with cleft. During Operation Smile mission week, international medical professionals evaluate cleft individuals to determine whether they are good candidates for the surgery, but do not exclude people with syndromic cleft from being treated. Individuals born with CPO are vulnerable to syndromes and developmental abnormalities (Jones, 1988; WHO, 2002, Timmons et al., 2001), and if these individuals were included in the study; then the estimated results would be biased downwards. There are other unobserved factors that could affect overall health, particularly weight and height, for cleft individuals such as genetic makeup, environmental factor and lifestyle. Also, it is possible that the parents may have misinterpreted the questions regarding their children's ability feed, hear and complete tasks. The parents may have had difficulties understanding the questionnaires because they are unaccustomed to answering questions based on an agreement scale<sup>15</sup>. Also, as mentioned in Section 5, social desirability bias may affect have caused the results to be biased downwards. Thus, future studies should change the way the questions are worded to capture an unbiased result.

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<sup>15</sup> See Appendix I, parental questionnaire

In order to study the impact of surgical treatment, future studies need to incorporate timing of the surgery. It is recommended that the cleft child should be treated early to be able to feed normally and develop normal speech. There are limited to no studies on the impact of late cleft treatment on overall health. Thus, incorporating timing of the surgery is important in determining the surgical impact on overall health and speech outcomes.

Some children were not able to read or count during the audio recording for speech evaluation. These children were asked to repeat when the enumerators counted from 1 to 20 and 60 to 70. The cleft children may have been able to manipulate their normal speech pattern to match with the enumerator's speech pattern. This could cause the speech results to be bias downwards. Also, the scoring of speech parameters are based on perceptual analysis of the speech therapist. The voice recordings were evaluated by one speech pathologist and may not be as robust as having multiple speech pathologists evaluate the samples for data integrity. Future studies should have more than one speech pathologists to evaluate the speech parameters.

In conclusion, there is wealth literature on orofacial cleft and the impact of surgery for those who were treated. However, the studies lack uniformity and the results are inconsistent. The results of this study suggested that both cleft and cleft surgery have no significant impact on health. The cleft severity has significant negative impact on speech acceptability but there is no significant impact of cleft restorative surgery on speech. The results of this study cannot be taken at face value due to number of limitations discussed above. However, the empirical approach used in this study can be applied to future studies with larger sample size to capture true causal effect of surgical impact on cleft individuals.

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TABLE 2: SUMMARY STATISTICS FOR PATIENT-SIBLING PAIR ON HEALTH PARAMETERS

--Means with Standard Deviation--

Group	Statistics	Age	Weight	Height	Grip Strength	BMI
<b>Un-operated</b>	N	30	30	30	30	30
	mean	14.67	38.36	146.06	18.42	17.74
	SD	2.19	8.97	9.50	6.92	2.89
	min	11.00	20.00	125.00	5.50	12.78
	max	19.00	50.90	165.30	29.50	25.20
<b>Partial Treatment</b>	N	39	39	39	39	39
	mean	15.18	40.33	149.17	17.59	17.98
	SD	2.59	10.00	13.35	7.96	3.30
	min	11.00	22.20	120.00	6.73	13.83
	max	19.00	62.15	181.00	41.67	30.42
<b>Full Treatment</b>	N	45	45	45	45	45
	mean	15.62	39.12	150.98	19.63	16.99
	SD	2.46	8.94	10.98	9.00	2.63
	min	11.00	19.50	118.00	6.80	11.68
	max	19.00	58.90	175.10	43.23	22.75
<b>Sibling Un-Operated</b>	N	30	30	30	30	30
	mean	13.00	34.78	140.75	18.87	16.69
	SD	4.32	15.00	18.34	11.52	3.78
	min	7.00	13.30	110.00	3.90	7.63
	max	21.00	70.70	178.20	50.25	24.75
<b>Sibling Partial Treatment</b>	N	39	39	39	39	39
	mean	14.33	36.72	144.04	16.39	17.04
	SD	5.17	13.38	17.73	7.18	3.22
	min	7.00	13.90	101.00	5.63	12.04
	max	24.00	65.10	171.90	34.25	24.77
<b>Sibling Full Treatment</b>	N	45	45	45	45	45
	mean	15.44	40.32	148.04	19.50	17.63
	SD	5.93	15.57	16.76	9.60	3.96
	min	7.00	15.00	108.00	2.78	10.14
	max	33.00	74.60	175.00	41.13	27.40
<b>Total</b>	N	228	228	228	228	228

NOTE: Summary statistics on key outcomes by operated and un-operated cleft treatment group and control sibling groups. Mean, standard deviation, minimum, maximum and sample size are reported for each group.

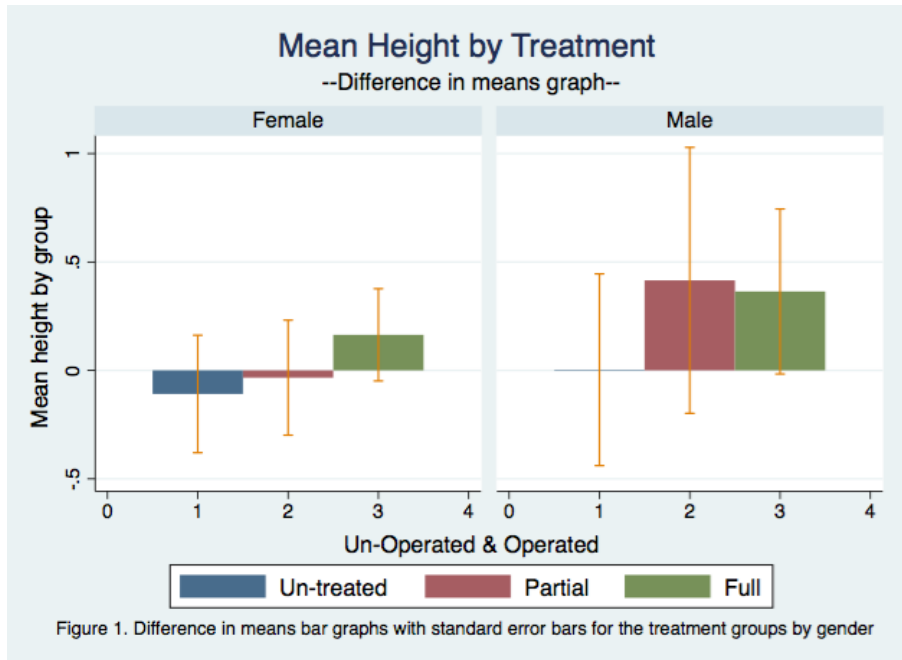


Figure 1. Mean height by different cleft treatment groups and gender

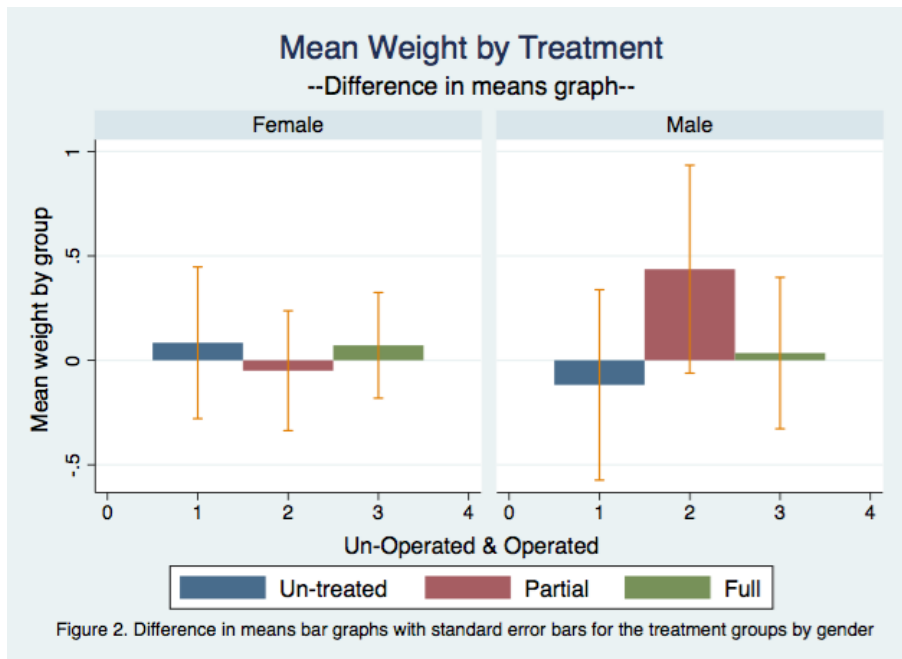


Figure 2. Mean height by different cleft treatment groups and gender

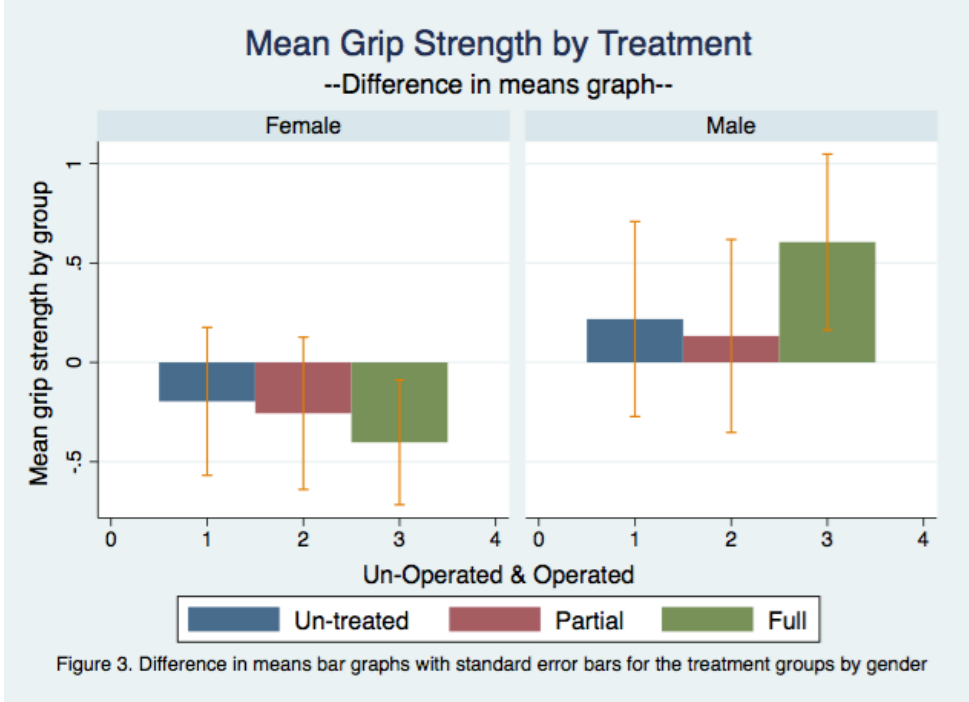


Figure 3. Mean Grip Strength by different cleft treatment groups and gender

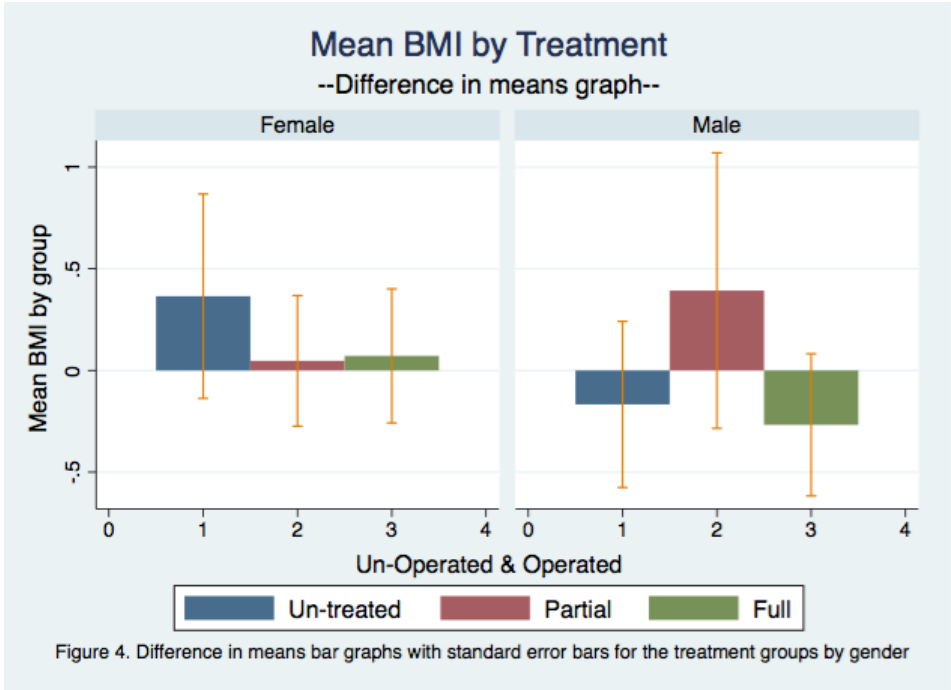


Figure 4. Mean BMI by different cleft treatment groups and gender

TABLE 2a: SUMMARY STATISTICS FOR SPEECH PARAMETERS  
 --Overall Speech Parameters measurement by group--

<b>Hypernasality</b>			
<b>Group</b>	<b>None-0</b>	<b>Present-1</b>	<b>Total</b>
<b>Un-Operated Cleft</b>	4	16	20
<b>Partially treated</b>	5	27	32
<b>Fully Treatment</b>	7	34	41
<b>Sibling Un-Operated</b>	16	4	20
<b>Sibling Partial Treatment</b>	32	0	32
<b>Sibling Full Treatment</b>	38	3	41
<b>Total</b>	102	41	186

<b>Speech Understandability</b>					
<b>Group</b>	<b>None-0</b>	<b>Mild-1</b>	<b>Moderate-2</b>	<b>Severe-3</b>	<b>Total</b>
<b>Un-Operated Cleft</b>	7	1	10	0	20
<b>Partially treated</b>	6	13	11	2	32
<b>Fully Treatment</b>	13	16	10	2	41
<b>Sibling Un-Operated</b>	20	0	0	0	20
<b>Sibling Partial Treatment</b>	31	1	0	0	32
<b>Sibling Full Treatment</b>	38	3	0	0	41
<b>Total</b>	115	36	31	4	186

<b>Speech Acceptability</b>					
<b>Group</b>	<b>None-0</b>	<b>Mild-1</b>	<b>Moderate-2</b>	<b>Severe-3</b>	<b>Total</b>
<b>Un-Operated Cleft</b>	3	6	6	5	20
<b>Partially treated</b>	3	9	15	5	32
<b>Fully Treatment</b>	9	16	11	5	41
<b>Sibling Un-Operated</b>	12	8	0	0	20
<b>Sibling Partial Treatment</b>	24	8	0	0	32
<b>Sibling Full Treatment</b>	27	14	0	0	41
<b>Total</b>	78	61	32	15	186

<b>Hyponasality</b>				<b>Audible Air Emission</b>			
<b>Group</b>	<b>None-0</b>	<b>Present-1</b>	<b>Total</b>	<b>Group</b>	<b>None-0</b>	<b>Present-1</b>	<b>Total</b>
<b>Un-Operated Cleft</b>	16	4	20	<b>Un-Operated Cleft</b>	11	9	20
<b>Partially treated</b>	24	8	32	<b>Partially treated</b>	20	12	32
<b>Fully Treatment</b>	33	8	41	<b>Fully Treatment</b>	23	18	41
<b>Sibling Un-Operated</b>	19	1	20	<b>Sibling Un-Operated</b>	20	0	20
<b>Sibling Partial Treatment</b>	32	0	32	<b>Sibling Partial Treatment</b>	32	0	32
<b>Sibling Full Treatment</b>	41	0	41	<b>Sibling Full Treatment</b>	41	0	41
<b>Total</b>	165	21	186	<b>Total</b>	147	39	186

TABLE 3: IMPACT OF UN REPAIRED CLEFT AND CLEFT SURGERY ON OVERALL HEALTH

--Dependent Variable: Overall Health--

	(1)	(2)	(3)	(4)	(5)
	<b>Overall Health</b>	<b>Weight</b>	<b>Height</b>	<b>Grip Strength</b>	<b>BMI</b>
<b>Cleft Severity</b>	0.07	0.00	0.07	-0.02	-0.04
	(0.06)	(0.03)	(0.04)	(0.06)	(0.04)
<b>Surgery Received</b>	-0.09	-0.07	-0.11	-0.02	-0.02
	(0.15)	(0.10)	(0.12)	(0.14)	(0.17)
<b>Operation Smile Surgery</b>	0.44	0.15	0.11	0.24	0.24
	(0.28)	(0.19)	(0.22)	(0.25)	(0.33)
<b>Birth Order</b>	-0.13	-0.01	-0.03	-0.02	-0.02
	(0.16)	(0.09)	(0.11)	(0.12)	(0.14)
<b>Age</b>	0.115**	0.181***	0.146***	0.131***	0.160***
	(0.05)	(0.02)	(0.02)	(0.03)	(0.04)
<b>Male</b>	0.12	0.15	0.323**	0.650***	-0.13
	(0.21)	(0.11)	(0.15)	(0.19)	(0.18)
<b>Constant</b>	-1.668*	-2.745***	-2.355***	-2.225***	-2.228***
	(0.99)	(0.46)	(0.52)	(0.59)	(0.72)
<b>N</b>	228	228	228	228	228

Standard errors in parentheses

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

NOTE: Overall health is the main dependent variable of interest and is constructed using 4 parameters of interest reported on top row. Column (1) regression output reports counter-intuitive and statistically insignificant results for main coefficients of interest, cleft severity and surgery received. It is true for weight and height. Grip strength and BMI show negative results for both cleft severity and surgery received. These results are not significant.

TABLE 4: IMPACT OF CLEFT SEVERITY AND CLEFT SURGERIES RECEIVED ON OVERALL HEALTH

--Dependent Variable: Overall Health--

	(1)	(2)	(3)	(4)	(5)
	<b>Overall Health</b>	<b>Weight</b>	<b>Height</b>	<b>Grip Strength</b>	<b>BMI</b>
<b>Require 2 Surgeries Incomplete CLO</b>	0.27	0.13	0.28	-0.18	0.04
	(0.44)	(0.19)	(0.27)	(0.30)	(0.27)
<b>Require 3 Surgeries Incomplete CPO</b>	0.53	-0.12	0.08	-0.21	-0.07
	(0.43)	(0.22)	(0.26)	(0.32)	(0.35)
<b>Require 4 Surgeries Complete CLO</b>	0.01	0.04	0.46	0.05	-0.40
	(0.57)	(0.30)	(0.41)	(0.46)	(0.38)
<b>Require 5 Surgeries Incomplete CLP</b>	0.24	0.05	0.47	-0.07	-0.30
	(0.61)	(0.39)	(0.42)	(0.48)	(0.53)
<b>Require 6 Surgeries Complete CLP</b>	0.12	0.07	0.41	-0.786*	-0.13
	(0.53)	(0.33)	(0.35)	(0.47)	(0.56)
<b>Require 7+ Surgeries Bilateral CLP</b>	0.04	-0.08	0.51	-0.18	-0.58
	(0.63)	(0.31)	(0.40)	(0.57)	(0.49)
<b>Received 1 Surgery</b>	0.22	0.05	-0.09	0.30	0.20
	(0.45)	(0.21)	(0.28)	(0.33)	(0.30)
<b>Received 2 Surgeries</b>	0.50	-0.07	-0.30	0.43	0.29
	(0.60)	(0.37)	(0.37)	(0.51)	(0.64)
<b>Received 3 Surgeries</b>	-0.10	-0.25	-0.55	-0.09	0.12
	(0.62)	(0.42)	(0.54)	(0.49)	(0.67)
<b>Received 4+ Surgeries</b>	0.19	-0.16	-0.10	0.24	-0.12
	(0.86)	(0.46)	(0.53)	(0.54)	(0.59)
<b>Birth Order</b>	-0.12	-0.01	-0.04	0.00	-0.02
	(0.16)	(0.10)	(0.11)	(0.12)	(0.14)
<b>Age</b>	0.115**	0.179***	0.143***	0.134***	0.160***
	(0.05)	(0.02)	(0.02)	(0.03)	(0.04)
<b>Male</b>	0.14	0.15	0.333**	0.713***	-0.14
	(0.21)	(0.11)	(0.16)	(0.19)	(0.20)
<b>Constant</b>	-1.73	-2.713***	-2.296***	-2.366***	-2.238***
	(1.05)	(0.49)	(0.51)	(0.58)	(0.76)
<b>N</b>	228	228	228	228	228



Standard errors in parentheses

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

NOTE: This is a regression output representing the effect of being born with different types of cleft and the number of surgery outcomes on overall health. Surprisingly, the overall health shows positive trend throughout different types of cleft severity and surgeries received. Overall health seems to decrease when a cleft individual receives 3 surgeries but increases if operated on for more than four times. These results are not statistically significant.

TABLE 5: OVERALL HEALTH OUTCOME USING PARENTAL DATA  
 --Dependent Variable: Overall Health--

	(1)	(2)	(3)	(4)	(5)	(6)
	<b>Overall Health</b>	<b>Good Health</b>	<b>Fitness</b>	<b>Trouble Feeding</b>	<b>Trouble Hearing</b>	<b>Trouble Completing Task</b>
<b>Cleft Severity</b>	-0.10*	0.11	-0.06	-0.21***	-0.10*	-0.10*
	(0.05)	(0.07)	(0.07)	(0.08)	(0.05)	(0.05)
<b>Surgery Received</b>	0.04	0.13	0.03	-0.05	-0.13	0.10
	(0.17)	(0.21)	(0.23)	(0.24)	(0.15)	(0.11)
<b>Operation Smile Surgery</b>	0.14	-0.09	-0.16	0.28	0.27	0.21
	(0.29)	(0.34)	(0.31)	(0.36)	(0.27)	(0.19)
<b>Birth Order</b>	0.08	0.13	0.02	0.02	0.002	0.02
	(0.09)	(0.12)	(0.10)	(0.11)	(0.09)	(0.07)
<b>Age</b>	0.006	0.04	0.006	-0.01	-0.03	-0.01
	(0.03)	(0.03)	(0.03)	(0.03)	(0.03)	(0.02)
<b>Male</b>	0.04	0.01	0.26	0.037	-0.16	-0.068
	(0.11)	(0.14)	(0.16)	(0.13)	(0.12)	(0.10)
<b>Constant</b>	-0.19	1.26*	3.55***	4.90***	5.45***	4.82***
	(0.66)	(0.74)	(0.59)	(0.71)	(0.61)	(0.50)
<i>N</i>	363	363	363	363	363	363

Standard errors in parentheses

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

NOTE: This regression output generated using parental data to capture the parent's perspective on overall health for the participants. Column (1) represents an overall health index on being born with cleft and receiving cleft surgery. Cleft severity has significant negative impact on overall health, but cleft surgery has no significant impact on the dependent variable. Columns (4), (5), and (6) represents counter intuitive results at statistically significant level. Parents may have misinterpreted the questions. N=363 because parents were asked to complete the survey on all of their children in the household.

TABLE 6: IMPACT OF CLEFT SEVERTY AND CLEFT SURGERIES RECEIVED  
USING PARENTAL DATA

--Dependent Variable: Overall Health--

	(1)	(2)	(3)	(4)	(5)	(6)
	Overall Health	Good Health	Fitness	Trouble Eating	Trouble Hearing	Trouble completing task
<b>Require 2 Surgeries Incomplete CLO</b>	0.26	0.57	0.21	-0.04	-0.14	-0.15
	(0.38)	(0.40)	(0.34)	(0.29)	(0.39)	(0.38)
<b>Require 3 Surgeries Incomplete CPO</b>	-0.13	0.74	-0.06	-1.289**	-0.01	-0.21
	(0.32)	(0.49)	(0.35)	(0.51)	(0.27)	(0.26)
<b>Require 4 Surgeries Complete CLO</b>	0.28	0.63	-0.16	-0.06	0.18	-0.05
	(0.38)	(0.48)	(0.46)	(0.43)	(0.33)	(0.29)
<b>Require 5 Surgeries Incomplete CLP</b>	0.15	1.175*	-0.46	-0.43	0.01	-0.46
	(0.59)	(0.65)	(0.67)	(0.96)	(0.42)	(0.45)
<b>Require 6 Surgeries Complete CLP</b>	-0.79	0.44	-0.23	-1.08	-1.01	-0.79
	(0.72)	(0.69)	(0.53)	(0.73)	(0.68)	(0.66)
<b>Require 7+ Surgeries Bilateral CLP</b>	-0.819*	0.77	-0.85	-1.260*	-0.81	-0.69
	(0.49)	(0.63)	(0.69)	(0.75)	(0.52)	(0.51)
<b>Received 1 Surgery</b>	-0.13	-0.08	0.06	-0.16	-0.26	0.11
	(0.36)	(0.44)	(0.40)	(0.43)	(0.35)	(0.32)
<b>Received 2 Surgeries</b>	-0.29	-0.24	-0.14	-0.44	-0.14	0.27
	(0.69)	(0.68)	(0.60)	(0.79)	(0.56)	(0.60)
<b>Received 3 Surgeries</b>	0.24	0.57	0.47	-0.15	-0.90	0.60
	(0.86)	(0.85)	(1.03)	(0.91)	(0.67)	(0.44)
<b>Received 4+ Surgeries</b>	0.41	0.43	-0.40	-0.18	0.59	0.76
	(0.55)	(0.95)	(1.00)	(1.16)	(0.55)	(0.51)
<b>Birth Order</b>	0.06	0.13	0.01	-0.01	-0.01	0.01
	(0.09)	(0.12)	(0.10)	(0.11)	(0.09)	(0.07)
<b>Age</b>	0.00	0.05	0.01	-0.02	-0.03	-0.02
	(0.03)	(0.03)	(0.03)	(0.03)	(0.02)	(0.02)
<b>Male</b>	0.09	0.06	0.312**	0.04	-0.15	-0.06
	(0.11)	(0.14)	(0.15)	(0.13)	(0.12)	(0.11)
<b>Constant</b>	-0.12	1.20	3.484***	5.133***	5.533***	4.926***
	(0.62)	(0.73)	(0.60)	(0.69)	(0.54)	(0.47)
<b>N</b>	363	363	363	363	363	363

Standard errors in parentheses

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

NOTE: This regression output generated using parental data to capture the parent's perspective on overall health for the participants. Column (1) represents an overall health index on different cleft severity and cleft surgeries received. Using parental data, bilateral CPL has significant negative impact on overall health, but cleft surgery has no significant impact on the dependent variable. Parents appear to think that first two surgeries have negative impact on health, but the results are not statistically significant. Columns (4), (5), and (6) represents counter intuitive results at statistically significant level. Parents may have misinterpreted the questions.

TABLE 7. IMPACT OF BEING BORN WITH CLEFT AND CLEFT SURGERY ON OVERALL SPEECH OUTCOME

--Dependent Variable: Speech Acceptability (Overall Speech)--

	(1)	(2)	(3)	(4)	(5)
	Speech Acceptability	Hyper-nasality	Hypo-nasality	Audible Air Emission	Speech Understand-ability
<b>Cleft Severity</b>	-0.327***	0.282***	0.262***	0.190**	-0.347***
	(0.07)	(0.06)	(0.10)	(0.08)	(0.06)
<b>Surgery Received</b>	0.12	0.17	-0.30	0.10	0.17
	(0.21)	(0.13)	(0.28)	(0.20)	(0.22)
<b>Operation Smile Surgery</b>	0.10	0.11	-0.13	0.14	-0.06
	(0.29)	(0.25)	(0.36)	(0.32)	(0.30)
<b>Birth Order</b>	0.13	0.12	0.00	-0.10	0.16
	(0.10)	(0.10)	(0.15)	(0.13)	(0.11)
<b>Age</b>	0.01	0.00	-0.0265***	0.00	0.00
	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)
<b>Male</b>	-0.17	-0.12	0.00	0.11	-0.30
	(0.25)	(0.22)	(0.31)	(0.29)	(0.25)
<b>Constant</b>	0.24	-0.892***	0.11	-0.31	0.39
	(0.36)	(0.24)	(0.44)	(0.40)	(0.36)
<b>N</b>	186	186	186	186	186

Standard errors in parentheses

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

NOTE: Overall speech outcome is measured using Universal Parameters of Speech. Speech Acceptability parameter acts as overall speech outcome. Overall speech outcome significantly decreases with increasing cleft severity and restorative surgery improves speech acceptability. The surgery is not statistically significant.

TABLE 8. IMPACT OF CLEFT SEVERITY AND SURGERIES RECEIVED ON  
OVERALL SPEECH OUTCOME

--Dependent Variable: Speech Acceptability (Overall Speech)--

	(1)	(2)	(3)	(4)	(5)
	Speech Acceptability	Hyper- -nasality	Hypo- -nasality	Audible Air Emission	Speech Understand- ability
<b>Require 2 Surgeries Incomplete CLO</b>	-0.21	0.55	0.57	0.48	-0.31
	(0.27)	(0.60)	(0.64)	(0.43)	(0.29)
<b>Require 3 Surgeries Incomplete CPO</b>	-2.058***	1.405***	0.49	1.530***	-2.085***
	(0.25)	(0.40)	(0.50)	(0.47)	(0.25)
<b>Require 4 Surgeries Complete CLO</b>	-0.29	0.884*	1.01	0.69	-0.885**
	(0.38)	(0.53)	(0.78)	(0.57)	(0.34)
<b>Require 5 Surgeries Incomplete CLP</b>	-1.330***	1.468***	1.846*	1.04	-1.640***
	(0.48)	(0.46)	(1.07)	(0.85)	(0.48)
<b>Require 6 Surgeries Complete CLP</b>	-1.241***	1.551***	1.997*	0.79	-1.497***
	(0.38)	(0.44)	(1.01)	(0.67)	(0.45)
<b>Require 7+ Surgeries Bilateral CLP</b>	-2.097***	1.235**	2.347**	1.339*	-2.098***
	(0.33)	(0.57)	(1.02)	(0.78)	(0.43)
<b>Received 1 Surgery</b>	-0.29	0.50	-0.19	-0.18	-0.07
	(0.27)	(0.48)	(0.65)	(0.49)	(0.29)
<b>Received 2 Surgeries</b>	-0.13	0.53	-1.650*	0.57	-0.03
	(0.37)	(0.48)	(0.99)	(0.70)	(0.43)
<b>Received 3 Surgeries</b>	-0.48	0.79	-1.52	0.14	-0.38
	(0.52)	(0.50)	(0.92)	(1.06)	(0.49)
<b>Received 4+ Surgeries</b>	0.97	0.77	-1.16	1.16	0.98
	(0.65)	(0.56)	(1.59)	(0.73)	(0.89)
<b>Birth Order</b>	0.09	0.10	-0.03	-0.08	0.12
	(0.07)	(0.09)	(0.15)	(0.14)	(0.09)
<b>Age</b>	0.00	0.00	-0.0248**	-0.01	0.00
	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)
<b>Male</b>	-0.32	-0.01	0.07	0.09	-0.422*
	(0.20)	(0.25)	(0.40)	(0.32)	(0.23)
<b>Constant</b>	0.591**	-1.007***	0.09	-0.31	0.708**
	(0.26)	(0.29)	(0.54)	(0.51)	(0.33)
<b>N</b>	186	186	186	186	186

Standard errors in parentheses

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

TABLE 9: OVERALL HEALTH WITH AGE RESTRICTION BETWEEN SIBLING PAIRS

--Dependent Variable: Overall Health--

	(1)	(2)	(3)	(4)	(5)
	<b>Overall Health</b>	<b>Weight</b>	<b>Height</b>	<b>Grip Strength</b>	<b>BMI</b>
<b>Cleft Surgery</b>	0.02	-0.01	0.05	-0.01	-0.05
	(0.08)	(0.03)	(0.05)	(0.06)	(0.04)
<b>Surgery Received</b>	0.09	0.00	-0.09	-0.08	0.11
	(0.21)	(0.11)	(0.13)	(0.14)	(0.19)
<b>Operation Smile Surgery</b>	0.01	0.06	0.03	0.29	0.04
	(0.33)	(0.21)	(0.26)	(0.30)	(0.33)
<b>Birth Order</b>	-0.10	0.14	0.16	0.01	0.04
	(0.22)	(0.13)	(0.16)	(0.17)	(0.21)
<b>Age</b>	0.09	0.22***	0.24***	0.12**	0.123*
	(0.08)	(0.04)	(0.05)	(0.05)	(0.07)
<b>Male</b>	-0.06	0.15	0.38**	0.60***	-0.20
	(0.23)	(0.11)	(0.16)	(0.21)	(0.17)
<b>Constant</b>	-1.23	-3.59***	-4.11***	-2.16**	-1.85
	(1.46)	(0.85)	(0.97)	(1.04)	(1.41)
<b>N</b>	168	168	168	168	168

Standard errors in parentheses

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

NOTE: The regression output is generated by imposing age restriction between patient and unaffected sibling within 5 years of difference. The main coefficients, cleft severity and surgery received, are statistically not significant. The impact of surgery received on overall health shows a positive impact compared to Table 3.

TABLE 10: OVERALL HEALTH WITH AGE RESTRICTION BETWEEN SIBLING PAIRS

	(1)	(2)	(3)	(4)	(5)
	<b>Overall Health</b>	<b>Weight</b>	<b>Height</b>	<b>Grip Strength</b>	<b>BMI</b>
<b>Require 2 Surgeries Incomplete CLO</b>	-0.17	0.08	0.04	-0.10	0.05
	(0.53)	(0.21)	(0.31)	(0.30)	(0.27)
<b>Require 3 Surgeries Incomplete CPO</b>	0.02	-0.21	-0.01	-0.17	-0.30
	(0.40)	(0.19)	(0.29)	(0.39)	(0.29)
<b>Require 4 Surgeries Complete CLO</b>	-0.97	-0.11	0.15	-0.18	-0.52
	(0.69)	(0.38)	(0.52)	(0.56)	(0.51)
<b>Require 5 Surgeries Incomplete CLP</b>	0.12	0.23	0.20	0.14	0.18
	(0.81)	(0.46)	(0.46)	(0.61)	(0.50)
<b>Require 6 Surgeries Complete CLP</b>	0.01	0.17	0.22	-0.34	0.09
	(0.76)	(0.35)	(0.44)	(0.53)	(0.45)
<b>Require 7+ Surgeries Bilateral CLP</b>	-0.06	-0.12	0.26	0.05	-0.43
	(0.97)	(0.28)	(0.46)	(0.68)	(0.35)
<b>Received 1 Surgery</b>	0.69	0.06	0.03	0.14	0.22
	(0.61)	(0.21)	(0.28)	(0.40)	(0.28)
<b>Received 2 Surgeries</b>	0.14	-0.09	-0.04	0.04	-0.08
	(0.77)	(0.39)	(0.47)	(0.61)	(0.50)
<b>Received 3 Surgeries</b>	0.49	-0.09	-1.118***	-0.24	1.02
	(0.96)	(0.63)	(0.39)	(0.81)	(0.94)
<b>Received 4+ Surgeries</b>	0.39	-0.12	0.08	0.04	-0.18
	(1.16)	(0.50)	(0.61)	(0.65)	(0.55)
<b>Birth Order</b>	-0.12	0.12	0.17	0.08	-0.01
	(0.22)	(0.13)	(0.16)	(0.18)	(0.22)
<b>Age</b>	0.09	0.216***	0.245***	0.144**	0.12
	(0.08)	(0.05)	(0.05)	(0.06)	(0.08)
<b>Male</b>	0.02	0.14	0.373**	0.588**	-0.18
	(0.27)	(0.13)	(0.18)	(0.23)	(0.20)
<b>Constant</b>	-1.20	-3.484***	-4.120***	-2.556**	-1.63
	(1.42)	(0.92)	(1.02)	(1.11)	(1.49)



<b>N</b>	168	168	168	168	168
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Standard errors in parentheses

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

NOTE: The regression result is generated using patient and sibling data restricted to age differences within 5 years. The coefficients do not return significant results on cleft severity and number of surgeries received.

TABLE 11: IMPACT OF DIFFERENT CLEFT CHARACTERISTICS AND CLEFT MANAGEMENT

	(1)	(2)	(3)	(4)	(5)
	<b>Overall Health</b>	<b>Weight</b>	<b>Height</b>	<b>Grip Strength</b>	<b>BMI</b>
<b>Cleft Lip Only</b>	0.07	0.12	0.37	-0.14	-0.11
	(0.22)	(0.18)	(0.26)	(0.30)	(0.28)
<b>Cleft Palate Only</b>	-0.14	-0.12	0.07	-0.24	-0.05
	(0.25)	(0.22)	(0.26)	(0.32)	(0.34)
<b>Cleft Lip and Palate</b>	-0.09	-0.03	0.36	-0.35	-0.26
	(0.27)	(0.22)	(0.29)	(0.39)	(0.32)
<b>Primary Surgery</b>	0.15	0.02	-0.12	0.35	0.19
	(0.24)	(0.20)	(0.27)	(0.33)	(0.32)
<b>Secondary Surgery</b>	-0.09	-0.21	-0.32	0.14	-0.08
	(0.33)	(0.32)	(0.45)	(0.44)	(0.53)
<b>Birth Order</b>	-0.02	-0.01	-0.04	-0.02	0.00
	(0.10)	(0.09)	(0.10)	(0.12)	(0.14)
<b>Age</b>	0.167***	0.179***	0.143***	0.132***	0.159***
	(0.02)	(0.02)	(0.02)	(0.03)	(0.04)
<b>Male</b>	0.461***	0.15	0.328**	0.662***	-0.13
	(0.13)	(0.11)	(0.15)	(0.19)	(0.19)
<b>Constant</b>	-2.696***	-2.728***	-2.305***	-2.265***	-2.262***
	(0.45)	(0.46)	(0.51)	(0.58)	(0.76)
<b>N</b>	228	228	228	228	228

Standard errors in parentheses  
 \*  $p < 0.10$ , \*\*  $p < 0.05$ , \*\*\*  $p < 0.01$

NOTE: This is a regression output based on different cleft characteristics (CLO, CPO, CLP) and different cleft management (primary care and secondary care) without incorporating BMI in the overall health index. The results are not significant.

TABLE 12: IMPACT OF DIFFERENT CLEFT CHARACTERISTIC AND CLEFT MANAGEMENT ON OVERALL HEALTH USING PARENTAL DATA

	(1)	(2)	(3)	(4)	(5)	(6)
	<b>Overall Health</b>	<b>Good Health</b>	<b>Fitness</b>	<b>Trouble Feeding</b>	<b>Trouble Hearing</b>	<b>Trouble Completing Task</b>
<b>Cleft Lip Only</b>	0.25	0.59	0.18	-0.04	-0.13	-0.18
	(0.35)	(0.38)	(0.33)	(0.31)	(0.34)	(0.34)
<b>Cleft Palate Only</b>	-0.16	0.71	-0.07	-1.306**	-0.04	-0.21
	(0.33)	(0.49)	(0.35)	(0.51)	(0.27)	(0.26)
<b>Cleft Lip and Palate</b>	-0.669*	0.64	-0.61	-1.119**	-0.649*	-0.610*
	(0.38)	(0.48)	(0.48)	(0.52)	(0.36)	(0.34)
<b>Primary Surgery</b>	-0.07	0.00	0.23	-0.20	-0.32	0.06
	(0.37)	(0.47)	(0.43)	(0.51)	(0.36)	(0.32)
<b>Secondary Surgery</b>	0.57	0.83	0.46	0.01	-0.46	0.54
	(0.65)	(0.74)	(0.87)	(0.78)	(0.54)	(0.35)
<b>Operation Smile Surgery</b>	-0.08	-0.14	-0.47	0.04	0.30	0.19
	(0.34)	(0.38)	(0.35)	(0.43)	(0.30)	(0.23)
<b>Birth Order</b>	0.05	0.12	-0.01	-0.02	-0.01	0.01
	(0.09)	(0.11)	(0.09)	(0.11)	(0.09)	(0.07)
<b>Age</b>	0.00	0.04	0.00	-0.03	-0.04	-0.02
	(0.03)	(0.03)	(0.02)	(0.03)	(0.03)	(0.02)
<b>Male</b>	0.06	0.04	0.298*	0.01	-0.17	-0.07
	(0.11)	(0.14)	(0.16)	(0.13)	(0.12)	(0.11)
<b>Constant</b>	0.02	1.257*	3.686***	5.234***	5.537***	4.939***
	(0.60)	(0.70)	(0.55)	(0.69)	(0.59)	(0.48)
<b>N</b>	361	361	361	361	361	361

Standard errors in parentheses  
\*  $p < 0.10$ , \*\*  $p < 0.05$ , \*\*\*  $p < 0.01$

NOTE: The regression results show how parents perceive different types of cleft and cleft management on overall health. CLP has the worst effect on overall health at 10% significance. Parents think that secondary care has positive impact compared to primary. However, cleft management does not return statistically significant results.

TABLE 13. IMPACT OF DIFFERENT CLEFT CHARACTERISTIC AND CLEFT MANAGEMENT ON OVERALL SPEECH

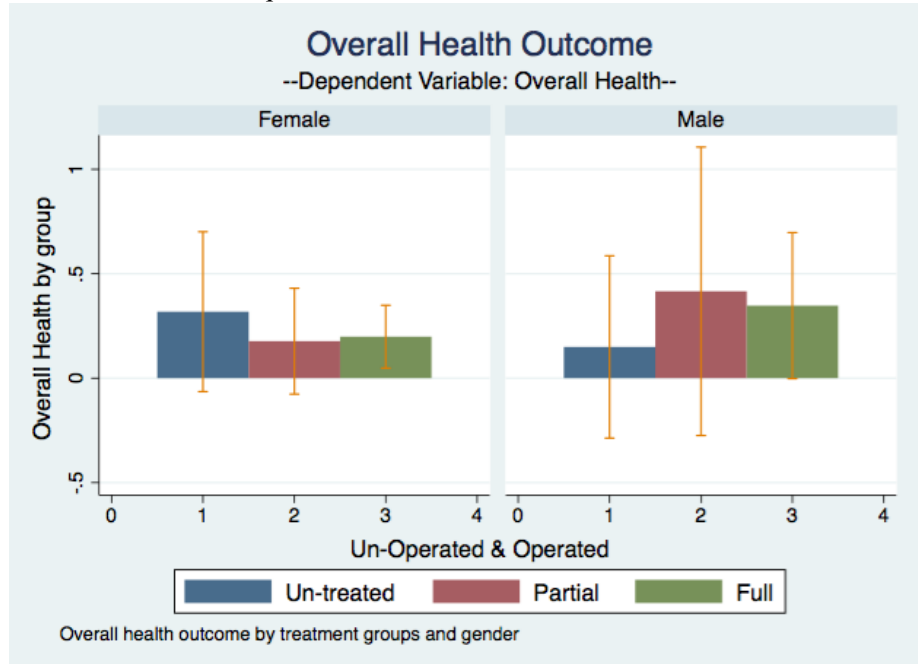
	(1)	(2)	(3)	(4)	(5)
	Speech Acceptability	Hyper-nasality	Hypo-nasality	Audible Air Emission	Speech Understand-ability
<b>Cleft Lip Only</b>	-0.28	0.67	0.94	0.46	-0.533**
	(0.24)	(0.51)	(0.62)	(0.45)	(0.27)
<b>Cleft Palate Only</b>	-2.072***	1.392***	0.44	1.516***	-2.075***
	(0.24)	(0.39)	(0.52)	(0.48)	(0.24)
<b>Cleft Lip and Palate</b>	-1.437***	1.397***	1.424**	1.374**	-1.622***
	(0.29)	(0.44)	(0.71)	(0.58)	(0.32)
<b>Primary Care</b>	-0.24	0.55	-0.46	-0.04	-0.10
	(0.24)	(0.46)	(0.64)	(0.49)	(0.28)
<b>Secondary Care</b>	-0.10	0.813*	-0.89	0.31	-0.08
	(0.53)	(0.49)	(0.81)	(0.84)	(0.60)
<b>Birth Order</b>	0.12	0.11	-0.02	-0.09	0.15
	(0.08)	(0.09)	(0.15)	(0.13)	(0.10)
<b>Age</b>	0.01	0.00	-0.0242**	0.00	0.00
	(0.01)	(0.01)	(0.01)	(0.01)	(0.01)
<b>Male</b>	-0.24	0.01	-0.01	0.14	-0.36
	(0.21)	(0.22)	(0.35)	(0.27)	(0.23)
<b>Constant</b>	0.40	-1.049***	0.12	-0.41	0.55
	(0.30)	(0.26)	(0.48)	(0.42)	(0.34)
<b>N</b>	186	186	186	186	186

Standard errors in parentheses

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

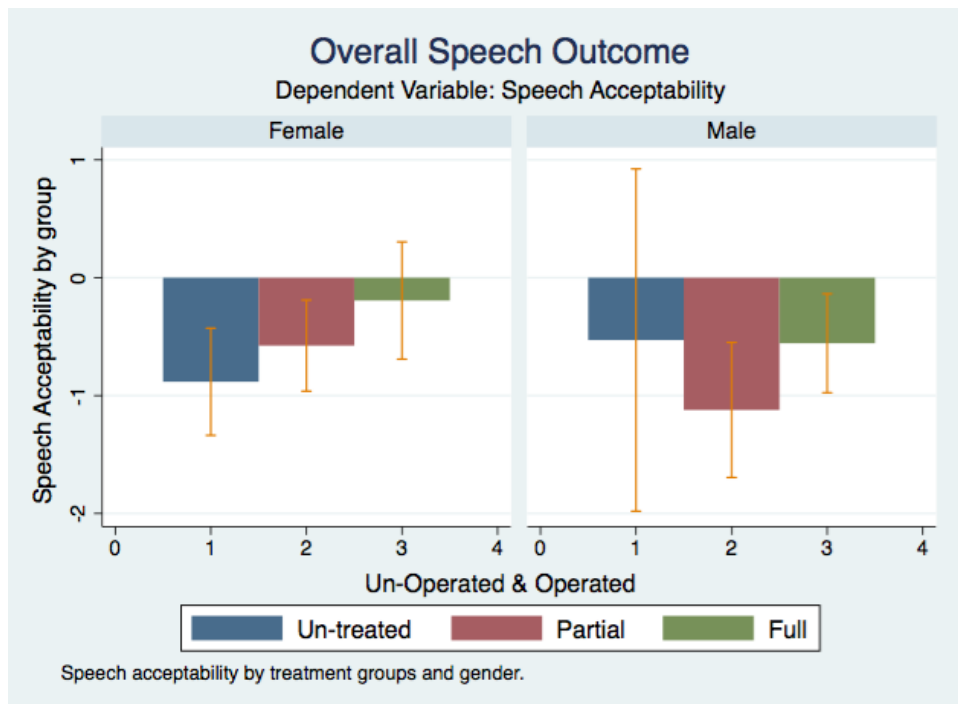
NOTE: Speech acceptability measures overall speech and it significantly decreases for individuals with CPO and CLP. Column (2) shows that hypernasality increases with secondary care, but this is driven by the property of the data.

OVERALL HEALTH BY TREATMENT GROUP AND GENDER  
 --Dependent Variable: Overall Health--



NOTE: Bar graphs showing overall health by gender and treatment group with error bars.

OVERALL HEALTH BY TREATMENT GROUP AND GENDER  
 --Dependent Variable: Speech Acceptability--



NOTE: Bar graphs showing speech acceptability by group and gender with error bars

## Appendix I

### Parental Survey Questionnaire

- 5 – Strongly Agree
- 4 – Somewhat Agree
- 3 – Neither Agree or Disagree
- 2 – Somewhat Disagree
- 1 – Strongly Disagree

Name of each child	/					
Physical Health	Cleft child	Nearest age sibling	2nd nearest age sibling	3rd nearest age sibling	4th nearest age sibling	5th nearest age sibling
B41. Relative to others his/her age, this child rarely suffers from illness or health challenges						
B42. Relative to others his/her age, this child is physically fit/strong						
B43. This child often has trouble eating or drinking without assistance						
B44. This child often has trouble hearing						

B45. This child often has trouble completing daily tasks and activities without assistance						
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**PART C Medical Information**

***For patients who plan to have surgery***

C1. When is the respondent scheduled for surgery? Date (DD/MM/YYYY)

\_\_\_\_\_/\_\_\_\_\_/\_\_\_\_\_ *if specific date is not available, circle one of the options below:*

1. August mission      2. November mission      3. Unsure      4. The respondent is not qualified for surgery, explain why not:

\_\_\_\_\_

\_\_\_\_\_

\_\_\_\_\_

***For patients who had cleft surgery in the past***

C2. When did the respondent have cleft ***lip*** surgery? Date (DD/MM/YYYY)

\_\_\_\_\_/\_\_\_\_\_/\_\_\_\_\_

C3a. How old was the respondent at the time of surgery? Age: \_\_\_\_\_

C3b. What was the location of surgery? (town and hospital name):

\_\_\_\_\_

C3c. Was this surgery done by Operation Smile? If not, who did it?:

YES Operation Smile / Other: \_\_\_\_\_

C4. When did the respondent have cleft ***palate*** surgery? Date (DD/MM/YYYY)

\_\_\_\_\_/\_\_\_\_\_/\_\_\_\_\_

C5. How old was the respondent at the time of surgery? Age: \_\_\_\_\_

C5b. What was the location of surgery? (town and hospital name):

\_\_\_\_\_

C5c. Was this surgery done by Operation Smile? If not, who did it?:

YES Operation Smile / Other: \_\_\_\_\_

**C6. [INTERVIEWER NOTE: If the patient has had surgery in the past, ask the parent to describe the patient's previous condition before they received any surgery. If the patient has not had surgery in the past, ask the parent to describe the patient's current condition.]**

C6a. If patient has or had cleft lip:

- A. Small Indentation on a side of lip (Forme Fruste Unilateral Cleft Lip)
- B. Cut on a side of upper lip but does not connect to nose (Incomplete Unilateral Cleft Lip)
- C. Cut on a side of upper lip and connected to nose (Complete Unilateral Cleft Lip)
- D. Cleft on both sides but does not connect to nose (Incomplete Bilateral Cleft)
- E. Cleft on both sides and connected to nose (Complete Bilateral Cleft)
- F. None of the above

C6b. If patient has or had cleft palate:

- A. Cleft in the back of the mouth on soft palate (Incomplete Cleft Palate)
- B. Cleft on both hard and soft palate and mouth & nose cavities are exposed (Complete Cleft Palate)
- C. Hard and soft palate are affected but covered by thin membrane on roof of mouth (Submucous Cleft Palate)
- D. None of the above



## Appendix II

### CONSENT FORMS

#### **INFORMED CONSENT FORM UNIVERSITY OF SAN FRANCISCO CONSENT TO BE A RESEARCH SUBJECT -For adults 18 and older-**

##### **Purpose and Background**

My name is \_\_\_\_\_ and I am a research assistant working on behalf of researchers at the University of San Francisco in the USA. I am asking you to participate in a project that examines the impact of receiving cleft lip and cleft palate surgery. Our study aims to measure the impact on a range of outcomes that may be affected by access to reparative cleft surgery and taking part in this survey will help us know the true value of receiving surgery as a young child.

##### **Procedures**

If I agree to allow my child to participate in this study, the following will happen:

1. I will complete a half hour survey conducted by the researchers and their assistants. I will be asked information about my age, gender, education and information about each of my children.

##### **Duration and Location of the Study**

My family's participation in this study will involve one session that lasts up to 2 hours. The study will take place in West Bengal, India. My child may quit this study at any time by simply saying "Stop" or "I do not wish to participate."

##### **Confidentiality**

Any data my child provides in this study will be kept confidential unless disclosure is required by law. In any report we publish, we will not include information that will make it possible to identify you or any individual participant. Specifically, we will transfer survey information onto a password-protected computer and remove all identifying information. Although the researchers may ask for my child's name during the interview, all identifying information will be coded as numbers in a way that does not allow researchers to distinguish one participant from another.

##### **Risks and Discomforts**

There are no expected risks or comforts associated with taking part in this study.

##### **Questions**

I have talked to one of the research assistants about this study and have had my questions answered. If I have any question about this study, please contact any of the following researchers: Kira Evsanaa (khatansuudal@yahoo.com), Jeremiah Maller (rjmaller@gmail.com), or Sam Manning (sam.j.manning@gmail.com)

##### **PARTICIPATION IN RESEARCH IS VOLUNTARY**

I am free to decline to have my child be in this study, or to withdraw my child from it at any point. My decision as to whether or not to have my child participate in this study will have no influence on the surgery or medical services my child receives. My signature below indicates that I agree to allow my

child to participate in this study. In addition, the researcher has the right to withdraw my child from participation in the study at any time.

**I HAVE READ THE ABOVE INFORMATION. ANY QUESTIONS I HAVE ASKED HAVE BEEN ANSWERED. I AGREE TO PARTICIPATE IN THIS RESEARCH PROJECT AND I WILL RECEIVE A COPY OF THIS CONSENT FORM.**

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*Child's Name* (print clearly)

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*Signature of Subject's Parent/Guardian*

*Date*

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*Signature of Person Obtaining Consent*

*Date*

**PARENTAL CONSENT FORM**  
**UNIVERSITY OF SAN FRANCISCO**  
**PARENTAL CONSENT FOR RESEARCH PARTICIPATION**  
**-For parents of survey respondents under the age of 18-**

**Purpose and Background 3**

My name is \_\_\_\_\_ and I am a research assistant working on behalf of researchers at the University of San Francisco in the USA. I am asking you to participate in a project that examines the impact of receiving cleft lip and cleft palate surgery. Our study aims to measure the impact on a range of outcomes that may be affected by access to reparative cleft surgery and taking part in this survey will help us know the true value of receiving surgery as a young child.

**Procedures**

If I agree to allow my child to participate in this study, the following will happen:

1. I will complete a half hour survey conducted by the researchers and their assistants. I will be asked information about my age, gender, education and information about each of my children.
2. The researchers will review my child's medical records to obtain information about the nature and extent of my child's cleft lip or palate.
3. The researchers will ask my child to complete 45-minute survey to answer questions about age, gender, school, their social life, and their health.

**Duration and Location of the Study**

My family's participation in this study will involve one session that lasts up to 2 hours. The study will take place in West Bengal, India. My child or I may quit this study at any time by simply saying "Stop" or "I do not wish to participate."

**Confidentiality**

Any data my child provides in this study will be kept confidential unless disclosure is required by law. In any report we publish, we will not include information that will make it possible to identify you or any individual participant. Specifically, we will transfer survey information onto a password-protected computer and remove all identifying information. Although the researchers may ask for my child's name during the interview, all identifying information will be coded as numbers in a way that does not allow researchers to distinguish one participant from another.

**Risks and Discomforts**

There are no expected risks or discomforts associated with taking part in this study.

**Questions**

I have talked to one of the research assistants about this study and have had my questions answered. If I have any question about this study, please contact any of the following researchers: Kira Evsanaa ([khatansuudal@yahoo.com](mailto:khatansuudal@yahoo.com)), Jeremiah Maller ([rjmaller@gmail.com](mailto:rjmaller@gmail.com)), or Sam Manning ([sam.j.manning@gmail.com](mailto:sam.j.manning@gmail.com))

**PARTICIPATION IN RESEARCH IS VOLUNTARY**

I am free to decline to have my child or myself to be in this study, or to withdraw my child and myself from it at any point. My decision as to whether or not to have my child participate in this study will have no influence on the surgery or medical services my child receives. My signature below indicates

that I agree to allow my child to participate in this study. In addition, the researcher has the right to withdraw me and my child from participation in the study at any time.

**I HAVE READ THE ABOVE INFORMATION. ANY QUESTIONS I HAVE ASKED HAVE BEEN ANSWERED. MY SIGNATURE BELOW INDICATES THAT I AGREE TO ALLOW MY CHILD TO PARTICIPATE IN THIS STUDY**

---

*Child's Name* (print clearly)

---

*Signature of Subject's Parent/Guardian*

*Date*

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*Signature of Person Obtaining Consent*

*Date*

**REQUESTING ASSENT FOR AN OLDER CHILD**  
**UNIVERSITY OF SAN FRANCISCO**  
**-For respondents under the age of 18-**

Dear Sir/Mam,

My name is \_\_\_\_\_ and I am a research assistant working on behalf of researchers at the University of San Francisco in the USA. I am asking you to participate in a project that examines the impact of receiving cleft lip and cleft palate surgery. Our study aims to measure the impact on a range of outcomes that may be affected by access to reparative cleft surgery and taking part in this survey will help us know the true value of receiving surgery as a young child.

I am asking you to complete a questionnaire that may take about 45 minutes. Your parents or legal guardians have already given a permission for you to participate in this study, but you do not have to participate if you choose not to. You may quit this study at any time by simply telling us that you do not want to continue. You can skip any questions or tasks that you do not want to complete. There are no known risks involved in this study.

To protect your confidentiality, your responses will not be shared with anyone unless required by law. The responses you give will be kept by the research team on a password-protected computer. Aside from the research team, nobody will know if you choose to participate in this project, nor will anyone know the answers you provide.

If you have any question about this study, please contact any of the following researchers: Kira Evsanaa ([khatansuudal@yahoo.com](mailto:khatansuudal@yahoo.com)), Jeremiah Maller ([rjmaller@gmail.com](mailto:rjmaller@gmail.com)), or Sam Manning ([sam.j.manning@gmail.com](mailto:sam.j.manning@gmail.com))

**Agreement**

I agree to participate in this research project and I have received a copy of this form.

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*Participant's Name (Please Print)*

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*Participant's Signature*

*Date*

I have explained to the above-named individual the nature and purpose, benefits and possible risks associated with participation in this research. I have answered all questions that have been raised and I have provided the participant with a copy of this form.

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*Signature of Person Obtaining Consent*

*Date*