Optimizing surgical treatment of internationally adopted children with cleft lip and/or palate: Understanding the family experience

Maren Shipe

A thesis
submitted in partial fulfillment of the
requirements of the degree of

Master of Public Health

University of Washington
2014

Committee:
Todd C. Edwards, Chair
Raymond Tse

Program Authorized to Offer Degree:
School of Public Health –Department of Health Services

University of Washington

Abstract

Optimizing surgical treatment of internationally adopted children with cleft lip and/or palate: Understanding the family experience

Maren Shipe

Chair of the Supervisory Committee:
Dr. Todd C. Edwards
Department of Health Service

Background: Seattle Children's Hospital has seen a 5-fold increase in the number of internationally adopted children who present with cleft lip and/or palate over the past 15 years. Many of these children have unrepaired clefts and would benefit from surgery as soon as feasible after arrival. Little is known about the surgical experience following adoption however, and the ideal timing for proceeding with surgery is unclear. The benefits for speech outcome from an early repair must be weighed against the evolving family relationships, communication challenges and culture adjustments experienced by newly adopted children and their adoptive families. This study was conducted to investigate the experience of families with children undergoing cleft surgery soon after adopting the child from another country.

Methods: Semi-structured qualitative interviews were conducted with parents of internationally adopted children post-primary repair of cleft lip and/or palate. Interviews were audio-recorded and transcribed and transcripts coded by pairs of investigators from a multidisciplinary study team. Mixed methods using both qualitative and quantitative data sources were used to further contextualize themes relating to timing of surgery that were derived from the parent interviews.

Results: Twenty parent interviews were conducted and four core themes were identified: 1) parent sources of anxieties prior to surgery, 2) considerations for the timing of surgery, 3) impact of surgical experience on child and family, and 4) modifiable socio-contextual factors. Parents experienced numerous anxieties prior to cleft surgery such as their ability to communicate with their child and their child feeling like he/she may be abandoned. Parents considered a strong child bond with at least one parent and the ability of the child to communicate basic needs to be important before undergoing surgery. In retrospect, parents generally felt the surgical experience did not negatively impact their child or their families, and the majority of parents felt that the surgical experience facilitated bonding and attachment with their child. No difference was found in perceived bonding before or after surgery in families who had initiated surgery sooner after adoption compared to families that waited longer, but surgery was found to facilitate perceived bonding more often in families with children older than 2 years old.

Conclusion: In our study, parents reported that cleft surgery soon after international adoption did not appear to impair child bonding or adjustment. Specific family and provider factors that could optimize the experience for families were identified.

ACKNOWLEDGMENTS

The lead researcher would like to thank thesis committee members Dr. Todd C. Edwards and Dr. Raymond Tse for their support and guidance throughout this process. Additionally, the other members of the study team Kelly Evans, Carolyn Schook, Dawn Leavitt, Ashley Peter, Babette Saltzman, and Julian Davies contributed substantially to data collection and analysis and provided invaluable input. Thank you to Laura Stueckle for assembling the IRB application, Karina Martinex-Lopez for administrative support, and to Jerrie Bishop for the management of funding. Finally, we are grateful to the CCTR of Seattle Children's Hospital for funding this project via the Faculty Research Support Fund.

TABLE OF CONTENTS

INTRODUCTION

In the United States craniofacial centers have seen a significant increase in the number of children presenting with unrepaired cleft lip and/or cleft palate who have been recently adopted from other countries (Swanson 2104; Hansson & Becket 2012; Goldstein 2012; Haynes 2010; Bellucci 2014; Morgan 2014). Seattle Children's Hospital (SCH) has experienced a 5-fold increase over the past 15 years, with 216 international adoptees with oral clefts presenting between 1997-2013 (Swanson 2014).

These children differ from American-born children with cleft lip and/or palate in several important ways. They are on average older at presentation (median age of 2.6 years rather than at or before birth) (Swanson 2014). While many adoptees have had repairs prior to adoption, 46% of adoptees present with unrepaired cleft lips and 70% with unrepaired cleft palates (Swanson 2014). In contrast, American-born children with cleft lip and/or palate typically undergo cleft lip and cleft palate repairs before one year of age. As a result of these presenting characteristics, there are fewer options in preoperative care (including non-surgical management) and for the timing of surgical treatment (Hansson & Becker 2012).

Adoptees also differ psychosocially from American-born children with clefts. In addition to being older and more aware, adoptees typically present to clinic a few weeks following adoption, and often have significant communication and cultural challenges that can affect the child's understanding of the situation and their ability to cope with a major surgical procedure (Eigsti 2011; Hansson 2013; Van Dyke & Canady 1995). Additionally, unlike American-born children with clefts, the adoptees are still in the process of forming new social bonds and relationships with their adoptive family and the family unit may be especially vulnerable to significant stress (Niemann & Weiss 2012).

While many craniofacial centers in the US choose to repair clefts as soon as possible in adoptees in order to optimize speech outcomes, concern has been raised over whether surgery should be delayed until the bonding and attachment of the new family is more secure (Hansson & Becker 2012).

Only one study has assessed the impact of cleft surgery on families with new adoptees. In Sweden, where their policy is to operate on adoptees with clefts as soon as possible after arrival, Hansson et al (2013) surveyed 30 parents of adoptees with cleft lip and/or palate to gather information on the families' experiences. They found that the majority of parents (85%) felt early hospitalization and surgery did not negatively affect their attachment with their child. In the discussion the authors suggest that that early surgery may in fact be beneficial to the new parent-child relationship via increased parental response to social cues and by improving the child's speech development. This was a small, community-based study however, in which parents agreed, or disagreed, with survey questions rather than being asked to identify issues important to their families.

Timing of Cleft Lip and Palate Repairs

The main impetus toward quick repair of adoptees' clefts is to improve speech outcomes. Research consistently demonstrates that an earlier repair of the hard palate, before 12-18 months of age, improves speech outcomes compared to children who had their palates repaired at an older age (Rohrich 2004). Specifically, an earlier repair improves articulation, phonation, nasal resonance, and intelligibility of speech and likely influences the child's propensity to vocalize and practice articulations (Willadsen 2012).

As the international adoptees with cleft palate are presenting at older ages, they are often past the ideal age for palate repair when they are adopted. Delaying cleft repair further increases the risk of impairment of speech outcomes.

Internationally Adopted Children

The adoption of children from foreign countries to the United States has increased in recent decades, with over 150,000 children adopted from outside of the US in the past 10 years (US Department of State 2014). The number of annual adoptions has decreased over the past few years, largely due to restrictions on the number of adoptions allowed by some countries. However, few restrictions have been placed on the adoption of children with special needs, including children with cleft lip and/or palate, and may explain the increased number of adopted children with clefts seen by SCH's Craniofacial Center. In 2013 there were 7,094 international adoptions in the US, of which 33% were from China (US Department of State 2014). At SCH, Chinese adoptions composed 81% of the adopted population presenting to the Craniofacial Center (Swanson 2014).

Health & Development

Children who have been adopted have an increased incidence of physical, developmental, and mental health concerns (Jones 2012). Health problems common to adoptees include nutritional and vitamin deficiencies, intestinal parasites, infectious diseases, congenital abnormalities, undiagnosed vision and hearing impairments, and chronic Hepatitis B (Jenista & Chapman 1987). Risk factors for health problems in adoptees include poverty, little or no prenatal care, malnutrition, exposure to blood-borne pathogens and environmental toxins, and inadequate developmental stimulation and emotional support (Jones 2012).

Attachment & Bonding

International adoptees often spend their early years in institutions such as orphanages where they may experience multiple caregivers, lack of stimulation, neglect, or abuse (Murphy 2009). This time period often coincides with the developmental stage when infants should be forming secure attachments, which may lead to a delay or variation in the type of attachments the children are able to form later in life (van Londen 2007). Adoptees often develop indiscriminate friendliness or disorganized attachment and in severe cases may develop reactive attachment disorder (van den Dries 2012). In contrast, an increasing number of international adoptees are living in foster homes prior to adoption where they are able to develop more stable attachments, resulting in attachment patterns similar to non-adopted children of similar ages (van den Dries 2012). While adoptees may have poor attachment immediately after adoption the majority of children have formed secure attachments with their adopted parents by 12 months after adoption, and a significant increase in child responsiveness occurs within the first 6 months following adoption (van den Dries 2010).

Communication & Language

Children who are adopted typically experience an abrupt change in language exposure after adoption, which results in a phenomenon termed “second-first language acquisition” where the child ceases exposure to their birth language and begins to learn a new first language (Gauthier & Genese 2011). While children may have no understanding of their new language at the time of adoption they typically acquire their adoptive language quickly over a short period of time (Eigsti 2011). However, when compared to monolingual children, the adopted children display weaker expressive and receptive language skills for several years following adoption (Gauthier & Genese 2011). These delays are directly correlated to the amount of time spent in institutions pre-adoption. Delays in language and an inability to communicate often affect cognitive function and educational achievement as well as short-term psycho-emotional function (Eigsti 2011). Adopted children with cleft lip and/or palate face the challenges not only of communicating in a new language, but communicating despite the presence of a cleft lip and/or palate that significantly impairs their ability to articulate in any language.

In summary, internationally adopted children with cleft lip and/or palate often present with health, attachment, and communication challenges as well as issues related to their clefts.

Purpose of Study

The purpose of this study was to identify psychosocial and other factors that may justify delay of cleft surgery for internationally adopted children, such as family bonding status and child communication barriers. The study aims were to

- Characterize the experience of families going through post-adoption cleft surgery.
- Determine factors related to the optimal timing for cleft surgery following adoption.
- Identify potentially modifiable factors that can be used to improve the experience of families undergoing post-adoption cleft surgery.

METHODS

Study Design

This was an observational exploratory mixed-methods study combining statistical analysis of patient data with a qualitative analysis of semi-structured interviews to achieve the study objectives. Given the paucity of information available and the nature of this clinical dilemma, qualitative methods were chosen to inductively generate hypotheses about relatively unknown topics and guide future research to refine or test hypotheses. The qualitative methodology used in this study was based on the grounded theory approach, which is “a qualitative research design in which the inquirer generates a general explanation (a theory) of a process, an action, or an interaction shaped by the views of a large number of participants” (Creswell 2012). This study design was chosen due to the lack of knowledge about the experience of families of international adoptees undergoing cleft surgery and the factors that may inform timing of surgery or improve the experience.

Study Setting & Team

The study took place at Seattle Children’s Hospital (SCH) in Seattle, WA, a referral center for craniofacial patients that covers a large geographic area including Washington, Alaska, Montana, and Idaho. In-person interviews were conducted in the Craniofacial Center’s library. A multidisciplinary study team was composed of a plastic/cleft surgeon, a craniofacial pediatrician, a pediatric resident, a nurse, a social worker, an adoption medicine pediatrician, an epidemiologist, an outcomes researcher, and a graduate student.

The research protocol was approved by Seattle Children’s Institutional Review Board, which has a cooperative review agreement with the University of Washington.

Participant Recruitment & Enrollment

The participants in this study were caregivers of children with cleft lip and/or palate who had been adopted internationally; this included the parents, with whom interviews were conducted, and the children, who were not direct participants but on whom data was collected via chart review. Potential study participants were identified from the SCH Craniofacial Center

Database or by craniofacial staff during clinic visits. Parents were invited to participate via letter with a follow-up telephone call to answer questions and determine if they would like to participate. If so, an interview time and date were scheduled.

Criteria for inclusion/exclusion were: 1) parents of children with cleft lip and/or palate who were adopted from outside of the United States, and 2) planned to undergo or had undergone surgery at SCH. Participants were excluded if their children were adopted from within the United States or had not planned to undergo primary or secondary cleft surgery at SCH.

Written informed consent was obtained at the time of the interview by the researcher conducting the interview. For in-person interviews, it was collected at the time of the interview. For telephone interviews, consent was obtained over the phone and the consent form was mailed back to the study team after the interview.

Data Collection

Chart Review

A chart review on the index child using SCH's electronic medical record was completed prior to each interview to establish inclusion criteria and to collect information on child demographics, cleft type, surgical history, medical history, and adoption history (Appendix 1:Data Collection Sheet). These data were made available to the interviewer prior to the interview to familiarize her with the study participant. Sections of the Data Collection Sheet that could not be completed via chart review, including parent income and education level, were completed during the parent interview.

Semi-Structured Parent Interviews

Parent interviews were conducted by pediatricians not directly involved in the patients' care who were trained in qualitative interviewing methods by a member of the research team experienced with the methods. Parent interviews occurred either in person or via telephone for participants who lived a significant distance from SCH and could not travel for the interview. The interview questions were derived from the research aims, but were semi-structured in that they permitted discovery of relevant new ideas and themes (Appendix 2: Interview Guide). The interviewer recorded field notes during each interview to assist in later analysis. The interviews were also audio-recorded and transcribed by a professional transcription service. Each transcript was checked for accuracy, edited to remove participant identifiers, and uploaded into Dedoose, an online qualitative analysis program (Dedoose.com).

Adequate sample size for a qualitative study based in grounded theory is determined by thematic saturation, defined as the point in data collection and analysis when new information produces little or no change to the existing themes (Creswell 2012). Participants were enrolled continually during the study period, with interview transcripts analyzed following every 5 interviews, until no new themes emerged from the analysis of the interview transcripts.

Data Analysis

Descriptive Statistics

Basic descriptive statistics (means, standard deviations) were performed using Microsoft Excel on the information gathered from the Data Collection Sheet (Appendix 1). The length of time between key events (adoption, first appointment, first surgery, etc.) was calculated.

Coding & Identification of Interview Themes

The first five parent interviews were open coded (researchers formed broad categories (codes) for ideas represented by small excerpts of text within the interviews) by all study team members to develop the codebook. All interviews were analyzed by at least two team members independently during axial coding (refining open codes and sorting into relevant themes) who selected relevant excerpts and assigned axial codes. Meetings were held periodically throughout this process to identify additional themes, reconcile any coding discrepancies, and monitor progress towards thematic saturation. Once all interviews were complete, the lead researcher performed selective coding (consolidating themes into core domains (broad categories) that reflect the relationships between themes discovered through the coding process). The core domains were presented to the entire study team who refined the phrasing of the domains and themes and selected excerpts to illustrate them.

Mixed Methods Analysis

Additional analysis was completed using mixed methods to explore which themes differed when sorted by two descriptors: 1) length of time from adoption to the first surgery, and 2) age of child at adoption. These descriptors were chosen based on clinically-driven hypotheses that age at adoption and length of time before surgery may affect the sense of urgency for surgery and the bonding experience of the family, especially in adoptees with unrepaired clefts. Therefore for this portion of the analysis only interviews with families of adoptees whose first surgery at SCH was a primary repair for cleft lip or palate were included. Interviews with parents of adoptees whose children had previously repaired clefts and only underwent revisions or secondary cleft surgeries at SCH were excluded from this analysis. These participants were *not* excluded from the thematic analysis as families of adoptees with prior repairs make up a significant portion of the population of adoptees presenting to SCH.

Parent interviews were sorted by descriptor and arranged numerically from smallest to greatest, then divided into groups that were clinically relevant and approximately equal in number. The thematic analysis was then compared to determine differences in the occurrence of themes between these groups for each descriptor.

RESULTS

Patient Characteristics

Participant characteristics are presented in Table 1. There were 9 male adoptees and 11 female adoptees included who were a mean of 2.3 years old at adoption. Adoptees were a mean of 3.1 years at the time of their first surgery at SCH. Adoptees undergoing primary cleft repairs were a mean of 2.3 years old, and adoptees undergoing secondary repairs or revisions were a mean of 6.2 years old. All but one adoptee was from China. The majority had cleft lip repairs prior to adoption (70%). All of the adoptees in our sample had spent time in an orphanage prior to adoption, and only 3 adoptees lived in both an orphanage and with a foster family prior to adoption. Several of the adoptees had growth delays (15%) or speech delays (30%), and one adoptee had an associated syndrome (Van der Woude).

The characteristics of the parents and families of the adoptees are presented in Table 1A (Appendix 3: Supplementary Tables). All of the families had two parents living at home, and had high levels of education and income compared to national averages (US Census Bureau 2013). Almost half of the adoptees had adoptive family siblings, and 3 families had other adopted children. All but one parent interviewee had previous surgical experience (either themselves or with one of their children), but only 25% of parents had any experience with oral clefts (through the experiences of friends or relatives with clefts).

TABLE 1: Adoptee Characteristics[1]

	N (%)
Sex	
Male	9 (45)
Female	11 (55)
Mean age at adoption (years)	2.3 ± 1.8
Mean age at first surgery @ SCH (years)	
Primary cleft repairs	2.3 ± 0.9
Secondary repairs or revisions	6.2 ± 3.1
Country of Adoption	
China	19 (95)
Kazakhstan	1 (5)
Pre-Adoption Housing	
Orphanage only	17 (85)
Foster care only	0 (0)
Orphanage & foster care	3 (15)
Cleft type	
Unilateral Cleft Lip (UCL)	1 (5)
Bilateral Cleft Lip (BCL)	0 (0)
Unilateral Cleft Lip and Palate (UCLP)	12 (60)
Bilateral Cleft Lip and Palate (BCLP)	7 (35)
Surgical repair prior to adoption	
None	4 (20)
Cleft Lip Repair	14 (70)
Cleft Lip & Cleft Palate Repair	2 (10)
Developmental Delays	
None	10 (50)
Growth	3 (15)
Speech	6 (30)
Global	1 (5)
Associated Anomalies/Syndromes	1 (5)

[1] Plus-minus values are mean ± SD

Eleven interviews were conducted with the mother only, 3 with the father only, and 6 with mother and father together (Table 2A, Appendix 3: Supplementary Tables). Twelve interviews were conducted in person and 8 were conducted via telephone.

Average length of time between adoption and the first appointment, first surgery, and parent interview, and average length of time between first surgery and parent interview are presented in Table 2.

TABLE 2: Lengths of Time[2]

Events	**Mean time (months)**
Adoption and 1st appointment	
Primary cleft repairs	1.6 ± 1.2
Secondary repair or revision	1.7 ± 0.1[3]
1st appointment and 1st surgery	
Primary cleft repairs	3.2 ± 3.3
Secondary repair or revision	6.9 ± 2.6[3]
1st surgery and interview	19.9 ± 12.9

[2] Plus-minus values are mean ± SD

[3] *Excludes one interview with adoptee who presented to other US institution before SCH*

Mixed Methods Demographics

Four parent interviews were excluded from the mixed methods analysis. The characteristics of the adoptees excluded and included in the mixed methods analysis are presented in Table 3A (Appendix 3: Supplementary Tables). By excluding adoptees who did not have their primary cleft repairs at SCH, the average age at adoption, at the time of the first surgery, and the length of time between adoption and the first surgery were all decreased.

Thematic Analysis

Open coding of the initial parent interviews led to 11 themes and 25 sub-categories within those themes. Throughout the coding process themes were refined and further themes emerged, eventually leading to 15 themes with 31 sub-categories (Appendix 4: Codebook). After thematic saturation was reached, the themes were combined into four major thematic categories: parental anxieties surrounding surgery, considerations for the timing of surgery, impact of the surgical experience, and modifiable socio-contextual factors.

Parental Anxieties Surrounding Surgery

When parents discussed the process of adopting a child with a cleft, coming to the Craniofacial Center for their initial appointment, and going through the first surgery and recovery they reflected on numerous anxieties they experienced and that they perceived in their children. Common themes related to parental anxieties included not being able to communicate with their child before and after surgery (present in 50% of the interviews), worrying their child may feel like he/she may be abandoned (50%), not being able to comfort and care for their child after surgery (35%), and the potential vulnerability of the family bond (35%). The following quotes portray how parents described several of these anxieties:

> "We weren't that attached yet, and how we were gonna comfort this child we couldn't communicate with and how we would begin to explain to him that he was in pain but we didn't 'cause it and that it would get better when he didn't understand us."
>
> --Mother of 2yo boy w/BCLP, no prior repairs[4]

---

[4] *Quotes used to illustrate themes are de-identified, attributed to the parent(s) interviewed, the age of the child at his/her first surgery, cleft presentation (unilateral cleft lip (UCL), unilateral cleft lip and palate (UCLP), bilateral cleft lip and palate (BCLP), and any prior cleft surgeries (cleft lip repair (CL), cleft palate repair (CP)).*

> "Given the trauma that she went through with this huge life change, and she's, you know, living with strangers; she's in a completely different country; she's having to learn a whole new language that she doesn't understand what people are saying right now and people can't understand her; and just thinking about how, you know, how that just creates stress and how that effect her."
>
> --Mother of 5.5yo girl w/BCLP, prior CL & CP repairs

Considerations for the Timing of Surgery

Parents were asked about how the timing of the first surgery was determined and how parents knew when they felt "ready for surgery". Parents often listed the following factors when they discussed what determines readiness for surgery or the optimal timing for surgery: at least one parent with an established bond or a feeling of trust (mentioned in 75% of interviews); the ability to communicate basic needs with their child (via spoken or sign language) such as food, drink, and pain (65%); a sense of normalcy or routine within the family (50%); medical advice to hasten or delay the surgery (50%); and the age of the child (30%). The parents who mentioned the child's age as a factor felt that younger children would be ready to undergo surgery sooner than older children who may take longer to establish a bond. Parents did not discuss a specific length of time they felt was necessary before proceeding with surgery and instead felt the timeline should be based off of the above factors rather than a proscribed length of time. The following quotes from parents discuss the need for communication and an established bond before proceeding with surgery:

> "Given that [our son] wasn't talking and we had to interpret his needs, if you're not able to figure out a need in three tries, then you might think about whether it's the right time for surgery."
>
> --Mother & father of 2yo boy w/UCLP, prior CL repair

> "I think that [our timeline] worked really well. It gave us enough time that she had realized, 'Okay. This is mom and dad. This is my family.' She trusted us."
>
> --Mother of 2yo girl w/UCLP, no prior repairs

Some parents discussed their sense of urgency for surgery, citing goals of improving speech and communication. Other parents discussed their motivations to delay surgery due to concerns about bonding and communication, as well as medical reasons to delay surgery such as the child's nutrition and ability to gain weight. While parents had numerous motivations to hasten or delay surgery, ultimately they said they relied on medical advice in determining the optimal timing for surgery.

Impact of the Surgical Experience

When reflecting back on the surgical experience, the parents felt that surgery had no negative impact on the relationship between them and their child. Many parents felt that the surgery and recovery period accelerated bonding (mentioned in 70% of interviews) and attributed this to realized dependence of the child on the parent (55%); a heightened sense of feeling like a mom or dad rather than a caretaker (30%); and an intense period of round-the-clock care (25%). The following quotes relate three parents' perceptions of the impact of surgery on the parent-child relationship:

> "Being the mom when she recovered, and helping her through the recovery and seeing her heal and rebound, and expand from the new function that she has, and whatnot, really built the bond with my wife and her... I think that [our daughter] really learned to trust us. She learned to depend that we would be there. "
>
> -- Father of 1.5yo girl w/BCLP, no prior repairs

> "And to have that [couch time] for 30 days straight you're gonna fast track a bonding process that she didn't have in the previous three and a half years 'cause no one ever held her."
>
> --Mother & father of 4yo girl w/UCLP, prior CL repair

> "It probably helped it a little you know just because she did need us and we were meeting a need and we were there when she hurt…You know you think well you're putting her in pain but she didn't blame us, I didn't sense that from her."
>
> --Mother of 1.5yo girl w/BCLP, prior CL repair

Additionally, many parents mentioned that subsequent cleft-related surgeries were far less stressful than the initial operative experience (50% of interviews with families that had two or more surgeries by the time of the interview) and attributed this to a more established bond and a better ability to communicate with their child.

Modifiable Socio-contextual Factors

Parents identified certain aspects of the surgical experience as going particularly well, or notably poorly, and gave advice for future parents of adoptees with cleft lip and/or palate. The most common potentially modifiable socio-contextual factors (excluding logistical factors) were combined into the following list:

- Minimizing situations of separation from their child and the resulting anxiety (present in 65% of the interviews), by utilizing sedation pre-operatively, allowing parents to be present with the child at anesthesia induction, and allowing early reunion with the child post-operatively
- Improving communication with the child (60%), by using interpreters for older children while in the clinic or hospital and using sign language with younger children
- Identifying specific sources of comfort prior to the day of surgery (60%), such as bringing familiar items from home (favorite foods, blankets, toys) and maintaining close physical contact (including co-sleeping with their child while hospitalized)
- Avoiding reminders of previous institutions (25%), by avoiding crib confinement and visible straps on beds
- Connecting with other parents via social media throughout the process of adoption and cleft surgery (25%).

The following quotes from parent interviews illustrate two of these recommendations:

> "She was a little bit apprehensive, I think, about the straps that were on the bed…So, I think that was also a little bit disconcerting to her to see those and not knowing how they would be applied or, you know, how long she might be strapped to the bed or what might happen to her while she was strapped in."
>
> --Mother of 5.5yo girl w/BCLP, prior CL & CP repair

> "I mean she didn't speak. She couldn't really. So, her cleft was really big, and she never – we've been told since then by speech people that she probably tried to talk at the natural time to talk, and when it didn't work, she just stopped doing. [Sign language] was so helpful. Because she just – there – you know, she couldn't speak, but she could sign. And it's – they pick it up so quickly that it was a help for her to be able to communicate . So, it gives her some control. So, that was awesome."
>
> --Mother of 2.5yo girl w/UCLP, prior CL repair

Mixed Methods Analysis

Sixteen parent interviews were included in the mixed methods analysis. The descriptors used for the comparative analysis were length of time between adoption and first cleft surgery, and age at adoption. The number of parent interviews falling into each category is displayed in Table 3.

TABLE 3: Mixed Methods Analysis Descriptor Groups

	Time: adoption to 1st surgery			
		≤4 months	>4 months	*Total*
Age @ Adoption	≤18 months	5	0	*5*
	19-24 months	3	4	*7*
	≥25 months	1	3	*4*
	Total	*9*	*7*	*16*

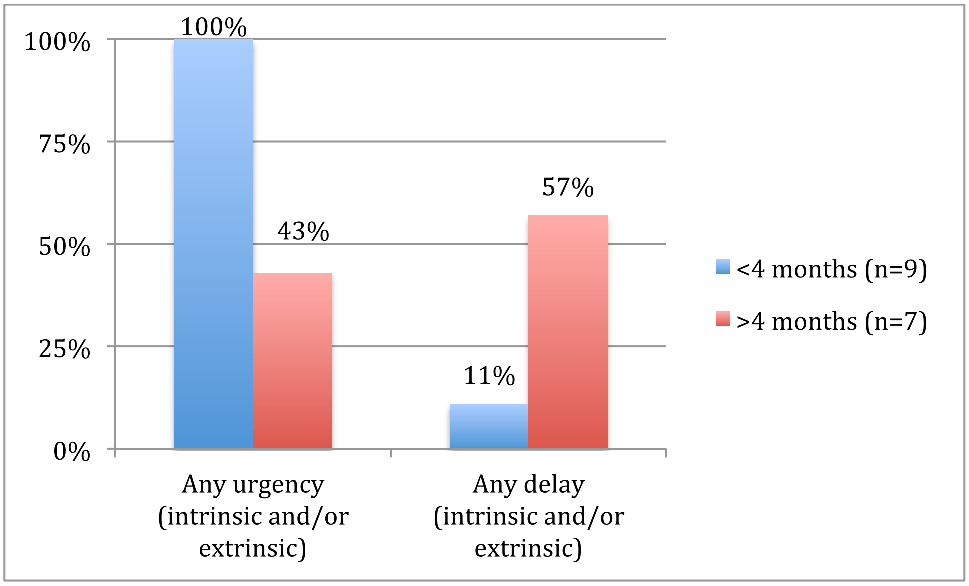
Themes analyzed included those relating to the timing of surgery and whether the parents perceived reasons to hasten or delay surgery, and whether those reasons were perceived to be intrinsic (stemming from the parents) or extrinsic (stemming from health professionals). Other themes analyzed related to bonding and attachment: whether parents felt that the bond was well established (with at least one parent) before the first surgery, and whether the parents felt that surgery facilitated or improved the bonding process.

Length of Time: Adoption to First Surgery

The parent interviews were divided into two groups: adoptees undergoing their first surgery in 4 months or less (9 interviews), and adoptees undergoing their first surgery in over 4 months (7 interviews).

Themes relating to the timing of surgery were analyzed within each group and the frequencies of interviews containing the themes were compared. The families that had surgery within the first 4 months following adoption all perceived a greater urgency for surgery compared to those that waited longer for surgery (themes present in 100% of interviews vs. 43% of interviews), and cited both intrinsic reasons for the urgency (89%) and extrinsic reasons (33%). The families that waited longer than 4 months for the first surgery following adoption perceived more reasons to delay surgery than the families that had surgery sooner (57% vs. 11%), and cited both intrinsic (29%) and extrinsic (43%) reasons for delaying the first surgery. These values are reflected in Table 4A (Appendix 3: Supplementary Tables) and Figure 1.

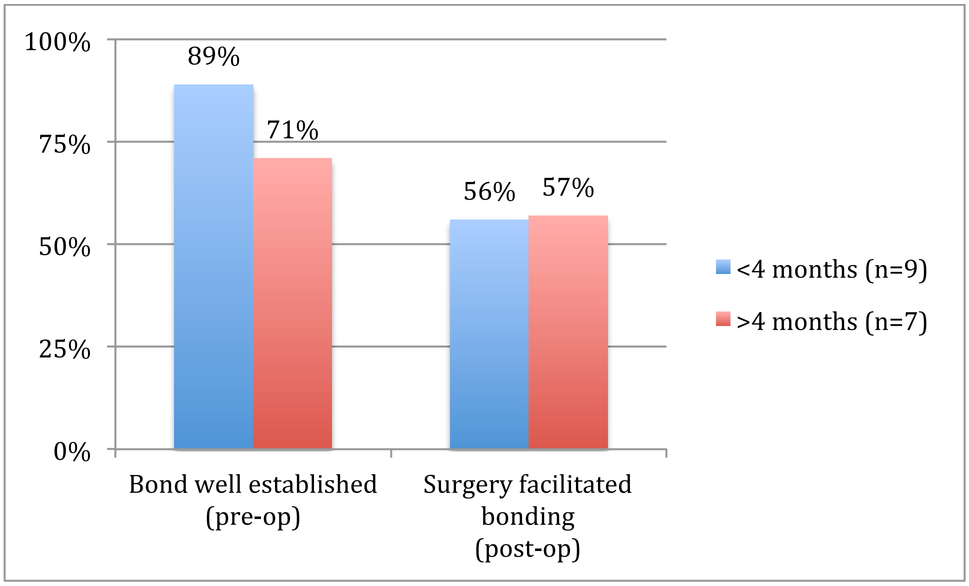
FIGURE 1: Frequency of interviews with themes related to timing by length of time from adoption to first surgery[5]



[5]Extrinsic refers to healthcare providers affecting timing.
Intrinsic refers to parents affecting timing.
Any refers to an interview with intrinsic and/or extrinsic themes related to timing.

Themes relating to bonding and attachment were analyzed by the length of time to adoption and the frequencies of interviews containing the themes were compared. More parents of adoptees who had surgery sooner felt that their bond was well established with at least one parent by the time of surgery compared to those that waited longer for surgery (89% vs. 71%). The same proportion of parents in both groups felt that surgery facilitated or improved the bonding process. These values are reflected in Figure 2.

FIGURE 2: Frequency of interviews with themes related to bonding/attachment by length of time from adoption to first surgery

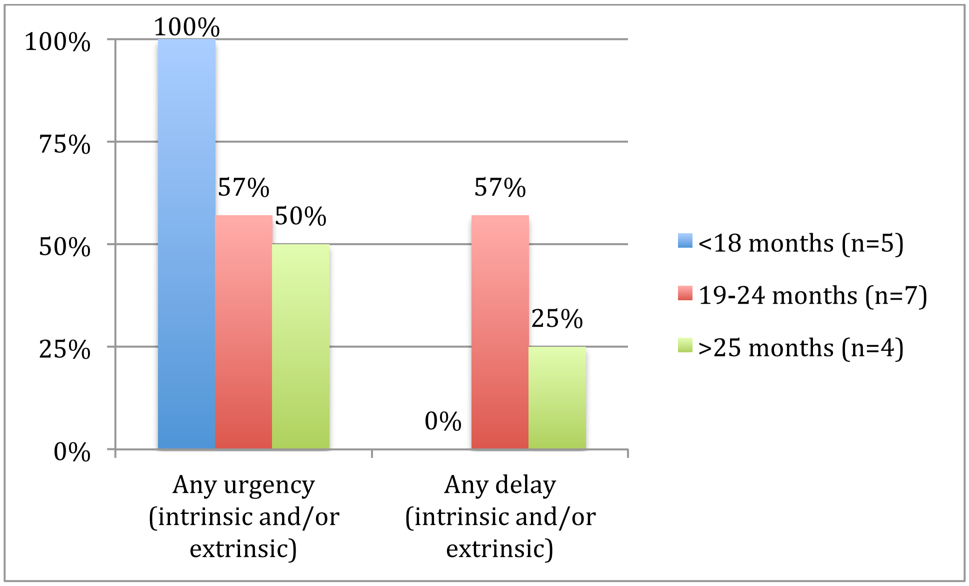


Age at Adoption

The parent interviews were sorted into three categories: adoptees 18 months or younger (5 interviews), those 19-24 months old (7 interviews), and those 25 months old or older (4 interviews) at the time of adoption.

Themes relating to the timing of surgery were analyzed by the age of the child at adoption and the frequencies of interviews containing themes were compared. Parents of the youngest children perceived the most urgency for surgery (intrinsic and/or extrinsic urgency, present in100% of interviews), and the fewest reasons to delay surgery (intrinsic and/or extrinsic delay, 0% of interviews). The parents of children aged 19-24 months at adoption perceived equal numbers of reasons for urgency and for delay (57%). The parents of the oldest children perceived the fewest reasons for urgency (50%) compared to younger children. These values are reflected in Table 5A (Appendix 3: Supplementary Tables) and Figure 3.

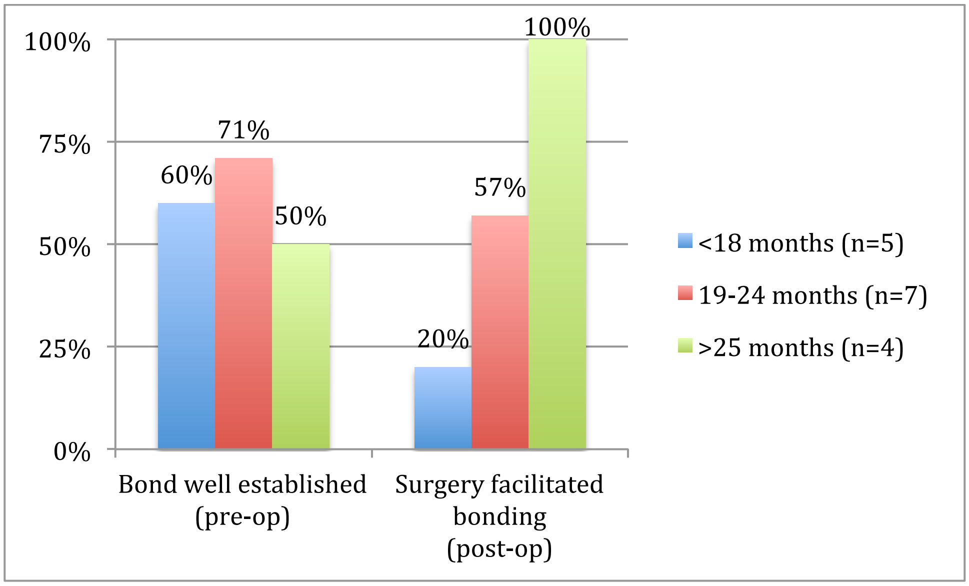
FIGURE 3: Frequency of interviews with themes related to timing by age at adoption[6]



[6] Extrinsic refers to healthcare providers affecting timing.
Intrinsic refers to parents affecting timing.
Any refers to an interview with intrinsic and/or extrinsic themes related to timing.

Themes relating to bonding and attachment were analyzed by the age of the child at adoption and the frequencies of interviews containing themes were compared. All three age groups had similar frequencies of parents who felt that the bond with their child was established with at least one parent prior to the first surgery, though the oldest children had the lowest number of parents who expressed this theme (50%). The parents of the oldest children had the highest frequency of parents who felt that surgery facilitated or improved the bonding process (100%), followed by the parents of children aged 19-24 months (57%), followed by the parents of the youngest children (20%). These values are reflected in Figure 4.

FIGURE 4: Frequency of interviews with themes related to bonding/attachment by age at adoption



DISCUSSION

Key Findings

In this study sample, cleft surgery soon after international adoption was not perceived to negatively impact the parent-child relationship, and was felt to facilitate bonding and attachment by the majority of parents (70%). This is consistent with the findings by Hansson et al (2013) that Swedish parents of international adoptees did not perceive a negative impact of early hospitalization and surgery on bonding or attachment and their postulation that surgery may promote bonding within the new family. In this study the positive effect of surgery on bonding/attachment was perceived by parents to be present regardless of the length of time between adoption and the first surgery, but was perceived more often by parents of children older than 2 years. Some parent interviews explored reasons behind why this potentially stressful surgical experience may have accelerated bonding, and the most frequent response related to a realized dependence of the child on the parent. The adoptee was put into a situation in which they had to rely on and trust their parents to care for them, and this experience was perceived to benefit bonding and attachment. One can see how this may especially affect older adoptees who may be more independent in day-to-day life and are thus more impacted by having to fully rely on their parents. Future studies may reveal if realized dependence is a significant factor in the acceleration of bonding or if other factors have a greater effect on bonding. Regardless, the finding that surgery does not negatively impact the parent-child relationship should provide reassurance to parents; families can proceed with cleft surgery soon after adoption to optimize surgical outcomes with decreased anxiety over the effect on the new family relationships.

Additionally, this study identifies factors that parents of international adoptees considered before undergoing cleft surgery, including having an established bond with at least one parent, their child being able to communicate basic needs, and creating a sense of normalcy or routine within the family. Notably, the parent interviews did not identify a specific period of time they should wait before the first cleft surgery but instead discussed signs of 'readiness' that helped families in this study identify when their child was ready for surgery. As there have been no studies on the impact of delaying cleft palate repair on speech outcomes in international adoptees, it is difficult to impose time-based deadlines for achieving palate repair in this population, especially in light of these new findings. Clinically, these factors can be used in the future to help inform and guide decision-making conversations with families.

While this study characterized the numerous anxieties parents of international adoptees with cleft lip and/or palate experienced during adoption, clinic appointments, and cleft surgery, it also elicited modifiable factors that can improve the experience for other families. Parents in this study recommended minimizing situations of separation and improving communication with the child, identifying specific sources of comfort prior to the day of surgery, and avoiding reminders of previous institutions. Some of these recommendations have already been put into practice at SCH, such as using anti-anxiety medications pre-operatively, allowing for early reunion with the child post-operatively, and allowing co-sleeping in hospital beds during recovery. The findings from this study can be further used to provide anticipatory guidance to parents of adoptees with clefts about commonly experienced anxieties and potential ways to avoid stressful situations during the operative experience.

The mixed methods analysis explored the differences in themes related to bonding and to the timing of surgery when compared across ages at adoption and lengths of time between surgery and adoption. An interesting correlation was present between descriptor groups; the youngest children (≤18 months) had surgery sooner (≤4 months) and the majority of the oldest children (≥25 months) waited longer for the first surgery (>4 months). These results may appear counterintuitive, as medically one would expect a greater urgency for surgery in the older children with unrepaired clefts. However, the parents of the youngest children felt more urgency than other parents, and those who perceived more urgency had surgery sooner. Several parents of younger children cited the child's age as the reason to proceed quickly with surgery, as they wanted to have the surgery while their child was less likely to remember the experience. Parents who felt the bond with their child was well established before surgery also tended to have surgery sooner. Conversely, parents of the oldest children perceived the least urgency for surgery, and had the lowest proportion of parents who felt well bonded prior to surgery. This may be due to the nature of bonding with older adopted children; developing secure attachments can be more of a challenge for late-adopted children (van Londen 2007), and may take longer to bond with their adoptive parents than younger children. Parents of older adoptees may have waited longer for surgery to allow more time for bonding. Additionally, providers may have felt that as these children were already far outside of the usual time period for cleft palate repair, delaying a few months more would unlikely make a difference in surgical outcomes. Further studies are required to explore this relationship between older adoptees with unrepaired clefts, longer times to surgery, and the possible impact of delays on speech outcomes.

Limitations and Strengths

The qualitative nature of this study has some inherent limitations. The parents interviewed were from a relatively small, non-random convenience sample, which may affect the external validity of the findings. The characteristics of the adoptees in our study were consistent with the population of international adoptees at SCH in terms of country of adoption (majority from China), but had a higher proportion of adoptees with previous repairs and a lower proportion of adoptees with isolated cleft lip (UCL or BCL) (Swanson 2014). As a result this study may not reflect the perceptions of the larger populations of parents with internationally adopted children with clefts. Additionally, this study was retrospective as parent interviews were conducted a mean of 20 months following surgery, and recall bias may influence parents' perceptions on the impact of the surgical experience. Finally, the themes and domains have not yet been member-checked by the parents interviewed and have not yet been replicated in other populations.

Strengths of the study are that it addresses a vulnerable population of patients that has been increasing over the past two decades with a paucity of published studies describing this population; only one other study has sought to characterize the experiences of families of international adoptees with clefts. Another strength of this study is the methodology: the paired coding of semi-structured interviews allowed themes to emerge organically rather than gathering answers from a predetermined set of questions. Finally, the multidisciplinary study team (surgeon, pediatricians, nurse, social worker, researchers) provided unique and complementary perspectives during the analysis of data that are rarely obtained in similar study designs.

Future Directions

Many of the findings from this study have already begun to be incorporated into clinical practice at SCH, such as minimizing separation between adoptees and their parents, decreasing anxiety by removing nasal stents in clinic rather than the operating room, and counseling parents on the positive effects surgery may have on bonding and attachment. In the future, clinical practice can continue to change with increased anticipatory guidance for parents of international adoptees, and the creation of a 'surgical tool kit' to help prepare families for surgery utilizing the identified modifiable factors to reduce stress.

The findings of this study can also be used to re-examine policies at hospitals that disproportionately affect families with international adoptees. At SCH, policy was amended to allow parents of adoptees into the recovery room post-operatively to minimize separation. Additionally, nursing policy at SCH was examined to empower families to request a bed for their child post-operatively rather than confining the child to a barred crib, which decreases reminders of previous institutions and allows for co-sleeping with parents. Future policy considerations involve establishing a timeline for cleft surgery following adoption that is not based on calendar time but rather fulfillment of milestones (bonding, communication, etc.) related to surgical readiness.

Additional research is needed to further characterize the experiences of this population and to optimize outcomes surgically and emotionally. The findings of this study should be validated though surveys of these 20 families, and the generalizability of the findings should be established through surveys in a broader population of parents of international adoptees with clefts. Ideally, to study how different lengths of time between adoption and surgery affect speech and emotional outcomes in adoptees of differing ages, a prospective randomized study should be undertaken. However, a retrospective review of speech outcomes in adoptees with unrepaired cleft palates would also provide missing information on the effect of delayed repairs in this population.

CONCLUSION

In this study, parents reported that cleft surgery soon after international adoption did not appear to impair family relationships and the majority of parents felt that the surgical experience facilitate bonding and attachment with their child. Parents identified a number of factors that they felt were important to consider before undergoing surgery as well as a number of modifiable socio-contextual factors. The results of this study can be used to develop tools to guide decision-making conversations between providers and parents of internationally adopted children with cleft lip and/or palate, and to adapt clinical practice to optimize the surgical experience in this vulnerable population.

REFERENCES

Bellucci, C., Collett, B., Morgan, A., Curtis, A., Kowalewicz, E., Coppersmith, J., & Kapp-Simon, K. (2014). Language development in children with orofacial clefts adopted from non-English speaking countries [Abstract]. *Proceedings of the American Cleft Palate-Craniofacial Association 71st Annual Meeting & Pre-Conference Symposia March 24-29, 2010*. Indianapolis, IN.

Chapman, K. L., Hardin-Jones, M. A., Goldstein, J. A., Halter, K. A., Havlik, R. J., & Schulte, J. (2008). Timing of palatal surgery and speech outcome. *Cleft Palate Craniofac J, 45*(3), 297-308. doi: 10.1597/06-244

Cohen, N. J., Lojkasek, M., Zadeh, Z. Y., Pugliese, M., & Kiefer, H. (2008). Children adopted from China: a prospective study of their growth and development. *J Child Psychol Psychiatry, 49*(4), 458-468. doi: 10.1111/j.1469-7610.2007.01853.x

Creswell, JW (2012). Qualitative Inquiry and Research Design: Choosing Among Five Approaches (Third Edition). Thousand Oaks, CA: SAGE Publications, Inc.

Eigsti, I. M., Weitzman, C., Schuh, J., de Marchena, A., & Casey, B. J. (2011). Language and cognitive outcomes in internationally adopted children. *Dev Psychopathol, 23*(2), 629-646. doi: 10.1017/S0954579411000204

Gauthier, K., & Genesee, F. (2011). Language development in internationally adopted children: a special case of early second language learning. *Child Dev, 82*(3), 887-901. doi: 10.1111/j.1467-8624.2011.01578.x

Goldstein, J. et al (2012). Cleft Care in International Adoption [Abstract]. *Proceedings of the American Cleft Palate-Craniofacial Association 69th Annual Meeting & Pre-Conference Symposia March 15-20, 2010*

Hansson, E., Ostman, J., & Becker, M. (2013). Adopting a child with cleft lip and palate: a study of parents' experiences. *J Plast Surg Hand Surg, 47*(1), 30-35. doi: 10.3109/2000656X.2012.736866

Hansson, E., Svensson, H., & Becker, M. (2012). Adopted children with cleft lip or palate, or both, require special needs cleft surgery. *J Plast Surg Hand Surg, 46*(2), 75-79. doi: 10.3109/2000656X.2012.668774

Haynes, K (2010). Health maintenance for the Internationally Adopted Child with Cleft [Abstract]. *Proceedings of the American Cleft Palate-Craniofacial Association 67th Annual Meeting & Pre-Conference Symposia March 15-20, 2010*. Fort Worth, TX.

Jenista, J. A., & Chapman, D. (1987). Medical problems of foreign-born adopted children. *Am J Dis Child, 141*(3), 298-302.

Jones, V. F., & Committee On Early Childhood, A., A.d Dependent Care. (2012). Comprehensive health evaluation of the newly adopted child. *Pediatrics, 129*(1), e214-223. doi: 10.1542/peds.2011-2381

Jones, V. F., Schulte, E. E., Childhood, C. o. E., & Council on Foster Care, A. o., and Kinship Care. (2012). The pediatrician's role in supporting adoptive families. *Pediatrics, 130*(4), e1040-1049. doi: 10.1542/peds.2012-2261

Krakow, R. A., Tao, S., & Roberts, J. (2005). Adoption age effects on english language acquisition: infants and toddlers from China. *Semin Speech Lang, 26*(1), 33-43. doi: 10.1055/s-2005-864214

Miller, L., Chan, W., Comfort, K., & Tirella, L. (2005). Health of children adopted from Guatemala: comparison of orphanage and foster care. *Pediatrics, 115*(6), e710-717. doi: 10.1542/peds.2004-2359

Miller, L. C. (1999). Caring for internationally adopted children. *N Engl J Med, 341*(20), 1539-1540. doi: 10.1056/NEJM199911113412009

Miller, L. C. (2000). Initial assessment of growth, development, and the effects of institutionalization in internationally adopted children.*Pediatr Ann, 29*(4), 224-232.

Miller, L. C. (2005a). Immediate behavioral and developmental considerations for internationally adopted children transitioning to families.*Pediatr Clin North Am, 52*(5), 1311-1330, vi-vii. doi: 10.1016/j.pcl.2005.06.011

Miller, L. C. (2005b). Immediate behavioral and developmental considerations for internationally adopted children transitioning to families.*Pediatr Clin North Am, 52*(5), 1311-1330, vi-vii. doi: 10.1016/j.pcl.2005.06.011

Miller, L. C. (2005c). International adoption, behavior, and mental health. *JAMA, 293*(20), 2533-2535. doi: 10.1001/jama.293.20.2533

Miller, L. C., & Hendrie, N. W. (2000). Health of children adopted from China. *Pediatrics, 105*(6), E76.

Morgan, A., Bellucci, C., O'Gara, M., Collett, B., Patel, P., Kowalewicz, W., & Kapp-Simon, K., (2014). Articulation outcomes in children who are internationally adopted [Abstract]. *Proceedings of the American Cleft Palate-Craniofacial Association 71st Annual Meeting & Pre-Conference Symposia March 24-29, 2010*. Indianapolis, IN.

Murphy, N. L. (2009). Facilitating attachment after international adoption. *MCN Am J Matern Child Nurs, 34*(4), 210-215; quiz 216-217. doi: 10.1097/01.NMC.0000357911.10606.63

Nelson, P. A., Kirk, S. A., Caress, A. L., & Glenny, A. M. (2012). Parents' emotional and social experiences of caring for a child through cleft treatment. *Qual Health Res, 22*(3), 346-359. doi: 10.1177/1049732311421178

Niemann, S., & Weiss, S. (2012). Factors Affecting Attachment in International Adoptees at 6 Months Post Adoption. *Child Youth Serv Rev, 34*(1), 205-212. doi: 10.1016/j.childyouth.2011.10.001

Pace, C. S., & Zavattini, G. C. (2011). 'Adoption and attachment theory' the attachment models of adoptive mothers and the revision of attachment patterns of their late-adopted children. *Child Care Health Dev, 37*(1), 82-88. doi: 10.1111/j.1365-2214.2010.01135.x

Pace, C. S., Zavattini, G. C., & D'Alessio, M. (2012). Continuity and discontinuity of attachment patterns: a short-term longitudinal pilot study using a sample of late-adopted children and their adoptive mothers. *Attach Hum Dev, 14*(1), 45-61. doi: 10.1080/14616734.2012.636658

Rettig, M. A., & McCarthy-Rettig, K. (2006). A survey of the health, sleep, and development of children adopted from China. *Health Soc Work, 31*(3), 201-207.

Rohrich, R. J., & Gosman, A. A. (2004). An update on the timing of hard palate closure: a critical long-term analysis. *Plast Reconstr Surg, 113*(1), 350-352. doi: 10.1097/01.PRS.0000097286.00667.4F

Rushton, A., Grant, M., Feast, J., & Simmonds, J. (2013). The British Chinese Adoption Study: orphanage care, adoption and mid-life outcomes. *J Child Psychol Psychiatry, 54*(11), 1215-1222. doi: 10.1111/jcpp.12088

Swanson JW, Smartt JM, Saltzman BS, Birgfeld CB, Hopper RA, Gruss JS, Tse R. Adopted Children with Cleft Lip and/or Palate: A Unique and Growing Population. Plastic and Reconstructive Surgery (accepted for publication).

Tan, T. X., Loker, T., Dedrick, R. F., & Marfo, K. (2012). Second-first language acquisition: analysis of expressive language skills in a sample of girls adopted from China. *J Child Lang, 39*(2), 365-382. doi: 10.1017/S0305000911000109

United States Department of State (2013). Intercountry Adoption Statistics [Data file]. Retrieved on March 15, 2014 from http://adoption.state.gov/about_us/statistics.php

United States Census Bureau (2013). State & County QuickFacts [Data file]. Retrieved on March 15, 2014 from http://quickfacts.census.gov/qfd/states/00000.html van den Dries, L., Juffer, F., van Ijzendoorn, M. H., & Bakermans-Kranenburg, M. J. (2010). Infants' physical and cognitive development after international adoption from foster care or institutions in China. *J Dev Behav Pediatr, 31*(2), 144-150. doi: 10.1097/DBP.0b013e3181cdaa3a

van den Dries, L., Juffer, F., Van Ijzendoorn, M. H., Bakermans-Kranenburg, M. J., & Alink, L. R. (2012). Infants' responsiveness, attachment, and indiscriminate friendliness after international adoption from institutions or foster care in China: application of Emotional Availability Scales to adoptive families. *Dev Psychopathol, 24*(1), 49-64. doi: 10.1017/S0954579411000654

Van Dyke, D. C., & Canady, J. W. (1995). Management of the adopted child in the craniofacial clinic. *J Craniofac Surg, 6*(2), 143-146.

van Londen, W. M., Juffer, F., & van Ijzendoorn, M. H. (2007). Attachment, cognitive, and motor development in adopted children: short-term outcomes after international adoption. *J Pediatr Psychol, 32*(10), 1249-1258. doi: 10.1093/jpepsy/jsm062

Weitzman, C., & Albers, L. (2005). Long-term developmental, behavioral, and attachment outcomes after international adoption. *Pediatr Clin North Am, 52*(5), 1395-1419, viii. doi: 10.1016/j.pcl.2005.06.009

Willadsen, E. (2012). Influence of timing of hard palate repair in a two-stage procedure on early speech development in Danish children with cleft palate. *Cleft Palate Craniofac J, 49*(5), 574-595. doi: 10.1597/09-120

APPENDICES

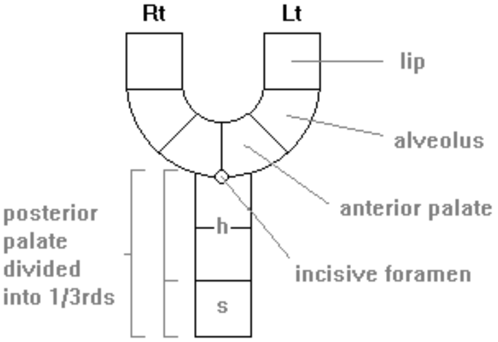
APPENDIX 1: Data Collection Sheet

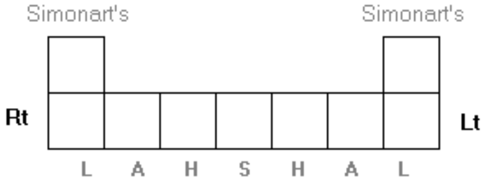
All shaded columns must be selected to be eligible		
Inclusion Criteria	***Yes***	***No***
1.) Child with cleft who has or will be undergoing surgery at SCH	▨	
2.) Child must be adopted from outside of the USA	▨	
Exclusion Criteria	***Yes***	***No***
1.) Child adopted within the USA or through foster care		▨

Subject Number: ______

Information about the child:
Chart Review:
Date of Birth (MM/DD/YY): ______________
Gender: ☐ M ☐ F
Place/Country of Birth: ___________________________________
Child adopted from: ☐ Orphanage ☐ Another family
Date of adoption(MM/DD/YY): ______________
Date of arrival in USA (MM/DD/YY): ______________

Type of cleft:





Associated anomalies/syndromes: ☐ No ☐ Yes, details: ______________________________
Developmental Delay: ☐ No ☐ Yes, Date of diagnosis (MM/DD/YY): ______________
Type: _______________________________________
Previous surgery: ☐ No ☐ Yes, Date of surgery (MM/DD/YY): __________________
Procedure: ____________________________________

Information about the family:

☐ Single parent ☐ Two parents

Other siblings: ☐ No ☐ Yes: ___________________________ (relative ages and gender)

Other adopted children: ☐ No ☐ Yes: ___________________________ (relative ages and gender)

Parents have surgical experience (as patient or as parent of a child?) specifics?

____

____

Parents have cleft surgery experience (as patient or as parent of a child?) specifics?

____

____

Socioeconomic status: Household income the year before adoption of participant including income such as Medicaid, Social security, and Unemployment payments

☐ < $15 000 ☐ $15 000 - $19 999 ☐ $20 000 - $24 999 ☐ $25 000 - $34 999

☐ $35 000 - $49 999 ☐ $50 000 - $69 999 ☐ $70 000 - $89 999

☐ $90 000 or more

☐ Unkonwn ☐ Question deferred

Number of people, including parents, supported by total household income for the year prior to adoption: __________

Parental education level: Highest grade of year of school/college completed prior to adoption

☐ No formal schooling ☐ 1-6 years ☐ 7-8 years ☐ 9-11 years

☐ 12 years, completed high school or equivalent ☐ 1-3 years of College

☐ Completed Technical College ☐ 4 years College or Bachelor's degree

☐ Master's degree ☐ Advanced degree (MD, PhD, JD)

☐ Unkonwn ☐ Question deferred

The SCH Craniofacial team:

Surgeon _________________

Pediatrician ______________

Nurse ___________________

Social worker______________

The intervention:

Primary vs Secondary (lip, palate, alveolus, fistula, rhinoplasty, orthognathic)

____

____

Date of Procedure (MM/DD/YY): ______________

Number of days of admission: __________________

Surgical complications: : ☐ No ☐ Yes, specify ___________________________

Date complication was noted (MM/DD/YY): __________

APPENDIX 2: Interview Guide

Questions	Probes
1. What was your child's language capacity at the time of adoption? At the time of the first clinic visit? At surgery?	*Ask about expressive and receptive forms*
2. Can you describe from the time of adoption to surgery, what level of bonding your child had with you and with your spouse?	*Inquire regarding insecure and/or preferential attachment*
3. What kinds of thoughts and feelings did you experience when you brought your child to the Craniofacial Clinic for her/his first visit?	*Indications of fears/hopes/expectations*
4. What kinds of things did you observe about your child when you brought her/him to the clinic for her/his first visit?	Anything out of the ordinary? *Indications of fears/hopes/expectations*
5. What kinds of thoughts and feelings did you experience on the day of your child's surgery?	*Indications of fears/hopes/expectations*
6. What kinds of things did you observe about your child on the day of her/his surgery?	Anything out of the ordinary? *Indications of fears/hopes/expectations*
7. What kinds of thoughts and feelings did you experience during your child's hospital stay after surgery?	*Indications of fears/hopes/expectations*
8. What kinds of things did you observe about your child during her/his hospital stay after surgery?	Anything out of the ordinary? *Indications of fears/hopes/expectations*
9. Are there any specific tools or techniques you found helpful to sooth your child through challenge experiences/interactions?	*e.g. objects, behavior, etc.*
10. What are the ways in which you felt you had to advocate for your child throughout this process?	
11. How would you say your relationship with your (adopted) child was affected by her/him having surgery?	How was it different before compared with after surgery?
12. What do you think about the timing of the surgical procedure your child had in relation to the amount of time since you adopted her/him?	Was the timing too fast? Too slow? Why would you say that?
13. What do you think is important to consider when deciding upon the timing of surgery for a newly adopted child? Is there an ideal time after adoption to proceed with the surgical procedure you have gone through?	What factored into your own decision regarding timing?
14. Do you have any recommendations for helping families to decide upon the timing of surgery for their newly adopted child?	
15. Is there anything about the experience either before, during, or after the surgery you wished you had known about ahead of time?	

APPENDIX 3: Supplementary Tables

TABLE 1A: Parent & Family Characteristics

	N (%)
Parents in family	
1	0
2	20
Other children in family	11 (55)
Other adopted children in family	4 (20)
Highest Education in Family	
4 yrs college or Bachelor's degree	6 (30)
Master's degree	9 (45)
Advanced degree	4 (20)
Deferred	1 (5)
Family Income	
<$25,000	0
$25,000-$49,999	3 (15)
$50,000-$89,999	6 (30)
>$90,000	8 (40)
Unknown	1 (5)
Deferred	2 (10)
Family Prior Experience	
Surgical experiences	19 (95)
Cleft lip and/or palate experience	5 (25)

TABLE 2A: Interview Characteristics

	N (%)
Conducted with	
Mother only	11 (55)
Father only	3 (15)
Mother & Father	6 (30)
Conducted via	
In-person interview	12 (60)
Telephone interview	8 (40)
Number of cleft surgeries between adoption and interview	
1	10 (50)
2 or more	10 (50)

TABLE 3A: Abbreviated Characteristics of Adoptees Excluded and Included in Mixed Methods Analysis[7]

	EXLCUDED (N=4)	**INCLUDED (N=16)**
Gender		
Male	1	8
Female	3	8
Cleft presentation		
UCL	1	0
UCLP	2	10
BCLP	1	6
Mean Age (years)		
At adoption	4.1 ± 3.3	1.9 ± 0.9
At 1st surgery @ SCH	6.2	2.3
Mean time from adoption to 1st surgery (months)	25.4 ± 33.7	4.8 ± 3.9

[7] Plus-minus values are mean ± SD

TABLE 4A: Frequency of interviews with themes related to timing by length of time from adoption to first surgery[8]

	Time from adoption to first surgery	
Theme	**≤4 months (n=9)**	**>4 months (n=7)**
Any delay	**11%**	**57%**
Extrinsic delay	*0%*	*43%*
Intrinsic delay	*11%*	*29%*
Any urgency	**100%**	**43%**
Extrinsic urgency	*33%*	*14%*
Intrinsic urgency	*89%*	*43%*

[8] Extrinsic refers to healthcare providers affecting timing.
Intrinsic refers to parents affecting timing.
Any refers to an interview with intrinsic and/or extrinsic themes related to timing.

TABLE 5A: Frequency of interviews with themes related to timing by age at adoption[9]

	Age at Adoption		
Themes	**≤18 months (n=5)**	**19-24 months (n=7)**	**≥25 months (n=4)**
Any delay	**0%**	**57%**	**25%**
Extrinsic delay	*0%*	*29%*	*25%*
Intrinsic delay	*0%*	*29%*	*25%*
Any urgency	**100%**	**57%**	**50%**
Extrinsic urgency	*0%*	*43%*	*25%*
Intrinsic urgency	*100%*	*43%*	*50%*

[9] Extrinsic refers to healthcare providers affecting timing.
Intrinsic refers to parents affecting timing.
Any refers to an interview with intrinsic and/or extrinsic themes related to timing.

APPENDIX 4: Codebook

Code	Sub-code	Definition	Code Count
Anxiety	Not otherwise specified (NOS)	Anxiety in parent or child, not covered by one of below sub-codes	25
	Breakdown of established trust	Parent anxiety about 'betraying' level of trust that has been established with their child	14
	Fear of institution	Similarity to orphanage setting causing fear/anxiety in child	11
	Lack of anxiety	Parents reporting being comfortable with upcoming surgery, or specific elements of surgery.	7
	Separation anxiety/ fear of abandonment	Child's anxiety of separation and/or fear of abandonment	27
	Stranger anxiety	Child's anxiety (or lack thereof) around strangers	9
Bonding/ attachment	NOS	Bonding/attachment not covered by one of below sub-codes	105
	Bond established	Parents report that a good bond was established. Status of bond, can provide comfort to child, etc.	51
	Independence/ stage of development	Bonding affected by child's desire for independence (may be at appropriate stage of dev for child)	2
	Potentially poor bond, indiscriminate friendliness	Child climbs into everyone's lap, doesn't differentiate family vs. strangers in behaviors	5
	Refusal to attach/bond	Child doesn't let anyone close, lack of bond with parents	9
Bucket for Modifiable		Potentially modifiable aspects of the experience, logistical, preparation/expectations, social support, etc.	155
Communication	NOS	Not covered by one of below sub-codes	61
	Expressive verbal	Child speaking in any language, intelligible or not	61
	Expressive nonverbal	Child nonverbal communication, especially sign language	54
	Receptive	Child understanding language, signing	48
Coping/ resilience	NOS	Not covered by one of below sub-codes	30
	Current emotional status	How the child is now (at time of interview)	34
	Physical comfort	Physical methods that the child/family used to cope (close contact, toys, etc.) Were they successful?	59
Establishing new normal	NOS	Family feeling established in day to day routines, general level of comfort with each other, not covered by one of below sub-codes	1
	Child adaptation	Child changing into new routine	5
	Parent adaptation	Parents changing into new routine	2
General experience		Relevant ideas that do not fall in one of established coding areas	35

Code	**Sub-code**	**Definition**	**Code Count**
General tools for preparing for surgery	NOS	Advice for other parents, lessons learned, not covered by one of below sub-codes	78
	Anxiety about regression and unlearning skills	Parents' worries that their child would be negatively affected by surgery	11
	Resilience	Parents observations that their child exceeded their expectations, were extremely resilient in the setting of the surgery and recovery	9
Food issues	NOS	Child seems especially fixated on food, hoarding behavior, etc.	28
Parental personality	NOS	Not covered by one of below sub-codes	6
	Implied through text of interview	Certain personality traits affect the surgical experience	11
	Self report	Parent talks about their personality traits, how they affected process	12
Preparation/ expectations		Family's level of prep and expectations for cleft, surgery, medical system, etc. Research, education, etc.	117
Previous experience	NOS	Not covered by one of below sub-codes.	110
	Child	Child's pre-adoption experience: orphanage? Foster family? Medical care?	11
	Parents	Parents' previous experience with clefts, surgery, adoption, etc.	26
Recognition of parent-child relationship	NOS	Feeling like they fit into parent-child roles within their family. Not covered by one of below sub-codes	5
	By child	Child recognizing this dynamic (parent vs. caregiver)	4
	By extrinsic group	Hospital staff, insurance company, etc.	1
	By parents	Parents recognizing this dynamic (their child vs. a child)	1
Timing of surgery	NOS	Parental concerns, and how they factored into their decision about the timing of their child's surgery. Not covered by one of below sub-codes.	86
	Emotional trauma	Anxiety about emotional trauma to child delaying surgery.	9
	Fear	Fear of parents or child for surgical process leading to delay in surgery.	6
Urgency	NOS	Not covered by one of below sub-codes.	24
	Extrinsic	Providers urging parents for earlier surgery (speech concerns, etc.)	20
	Extrinsic desire to delay	Providers recommending delay (nutrition concerns, etc.)	4
	Intrinsic	Parents' 'get it done' attitude. Want to put the experience behind them. Motivations (or lack thereof) for getting the surgery done earlier.	40
	Intrinsic desire to delay	Parents' motivations to delay surgery, or desire for delay.	7