Outcomes of Cleft Palatal Repair for Internationally Adopted Children

Stephen R. Sullivan, M.D., M.P.H.
Young-Soo Jung, D.M.D., Ph.D.
John B. Mulliken, M.D.

Providence, R.I.; Boston, Mass.; and Seoul, Republic of Korea

Background: Families in the United States adopted approximately 230,000 foreign-born children over the past two decades. Age at adoption and the presence of a cleft palate impact speech and language development. The authors' purpose is to document speech outcome after palatal closure in internationally adopted children.

Methods: The authors reviewed internationally adopted children with cleft lip–cleft palate or cleft palate who had two-flap palatoplasty from 1987 to 2010. Data collected included date of birth, cleft palatal type, age at palatoplasty, palatal fistula, postoperative speech assessment, and need for secondary surgery.

Results: The authors identified 55 children adopted with unrepaired cleft palate. Palatal types were Veau I ($n = 1$), II ($n = 1$), III ($n = 37$), or IV ($n = 16$). Median age at palatoplasty was 25.6 ± 11.8 months; palatal fistula occurred in five patients (9 percent). Speech outcome was successful in 28 patients (51 percent), whereas a secondary operation was recommended for 27 patients (49 percent). Need for a secondary operation was independent of palatal type ($p = 0.6$). Children who required a pharyngeal flap were significantly older at the time of palatoplasty compared with those who did not ($p = 0.009$). There was a significant association between increasing age at palatoplasty and need for a secondary operation (OR, 1.07; 95 percent CI, 1.01 to 1.13; $p = 0.01$). Pharyngeal flap significantly improved speech ($p < 0.001$).

Conclusions: International adoption with late palatoplasty can result in disordered speech. Velopharyngeal insufficiency is associated with increasing age at palatoplasty. The authors recommend palatoplasty and speech therapy soon after adoption. (*Plast. Reconstr. Surg.* 133: 1445, 2014.)

CLINICAL QUESTION/LEVEL OF EVIDENCE: Risk, III.

Families in the United States adopted approximately 230,000 foreign-born children over the past two decades.[1] For many of these children, speech and language development can be complicated by their age at adoption,[2,3] the switch in language and culture if a native language has developed,[3] and environmental deprivation in some orphanages.[4,5] Although many internationally adopted children make rapid speech and language gains in the preschool years, some do not.[6]

Cleft lip–cleft palate or cleft palate is an additional obstacle to normal speech and language development. An increasing number of internationally adopted children are presenting to cleft and craniofacial centers with unrepaired cleft lip–cleft palate or cleft palate.[7] For these children, velopharyngeal competence is the most important outcome following palatal repair. Velopharyngeal insufficiency, characterized by hypernasal resonance and

From the Division of Plastic and Reconstructive Surgery, Warren Alpert Medical School of Brown University and the Departments of Plastic Surgery and Pediatrics, Rhode Island Hospital and Hasbro Children's Hospital; the Department of Plastic and Oral Surgery, Boston Children's Hospital, Harvard Medical School; and the Department of Oral and Maxillofacial Surgery, Oral Science Research Center, College of Dentistry, Yonsei University.
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decreased intraoral pressure for pressure-dependent consonants, is the audible hallmark of failed palatal repair.[8] The reported frequency of velopharyngeal insufficiency following palatoplasty is 5 to 30 percent.[9–18] Velopharyngeal competence is attained more often in infants who undergo repair between 7 and 11 months of age[17,19–23]; however, more than half of children adopted to the United States are older than 12 months.[1]

Our purpose is to discuss the variables that must be considered when caring for a child adopted from another country with an unrepaired cleft palate and to describe the treatment and outcomes of cleft palatal repair in a cohort of internationally adopted children. The outcome measures were perceptual speech results and complications.

PATIENTS AND METHODS

After approval by the committee on clinical investigation, we identified and reviewed the charts of a consecutive series of foreign-born patients with cleft lip–cleft palate or cleft palate who were adopted by families in the United States between 1984 and 2012. Our inclusion criteria were the presence of an unrepaired cleft palate at the time of adoption and an age of 4 years at the time of chart review because children are old enough to cooperate with speech assessment and are unlikely to subsequently develop velopharyngeal insufficiency. Data collected included date of birth, sex, syndrome/association, cleft palate type (Veau type I, II, III or IV), age at palatoplasty (two-flap palatoplasty with muscular retropositioning), incidence of palatal fistula, postoperative speech assessment, need for secondary surgery (pharyngeal flap), and duration of follow-up. Nasoalveolar and anterior palatal fistulas intentionally not repaired (Pittsburgh types VI and VII) were not included in the postoperative fistula rate.[24] As described previously, operative techniques consisted of a two-flap palatoplasty with muscular retropositioning and a tailored superiorly based pharyngeal flap when necessary.[17,25–27] A tailored superiorly based pharyngeal flap is our preferred secondary operation because we have previously demonstrated that this operation is highly effective in correcting velopharyngeal insufficiency (97 percent success), with a low incidence of obstructive sleep apnea (2.5 percent).[25] Before pharyngeal flap surgery, all patients underwent preoperative multiview videofluoroscopy. The lateral pharyngeal wall motion, symmetry of lateral wall motion, palatal length, velopharyngeal gap size, and defect pattern were considered in planning the operation.[28–30]

Speech Assessment

Patients were followed annually in a cleft lip–cleft palate clinic. A speech pathologist, specializing in cleft palate, performed postpalatoplasty and post–pharyngeal flap (at least 3 to 6 months postoperatively) perceptual assessments and scored the results using the Pittsburgh Weighted Values for Speech Symptoms Associated with Velopharyngeal Incompetence instrument.[31,32] Overall assessment of speech was graded as follows: 0, competent velopharyngeal mechanism; 1 to 2, competent to borderline competent; 3 to 6, borderline to borderline incompetent; and greater than or equal to 7, incompetent velopharyngeal mechanism. Competent and competent to borderline competent were categorized as a success, whereas borderline to borderline incompetent and incompetent were categorized as a failure. Patients for whom a secondary operation was recommended but not yet performed were recorded as equivalent to having had the procedure. Hyponasal resonance, obstructive sleep apnea, and need for a revision operation (e.g., postoperative tonsillectomy, adenoidectomy, flap division, or dilation of pharyngeal ports) were recorded. Polysomnography was conducted if a child evidenced symptoms suggestive of obstructive sleep apnea.

Statistical Analyses

Patient characteristics and descriptive statistics were summarized. Continuous data were compared using the Wilcoxon signed rank test for paired samples or the Wilcoxon rank sum test for independent samples, and proportions were analyzed using Fisher's exact test. To evaluate for a possible association between the age at primary palatoplasty and need for a secondary operation, we performed a logistic regression and presented the odds ratio and 95 percent confidence interval. Continuous data are presented as median ± SD and range. All calculated p values are two-tailed and considered significant for values of $p < 0.05$. Statistical analyses were performed using Stata SE version 12.1 (StataCorp, College Station, Texas).

RESULTS

We identified 55 consecutive foreign-born patients with cleft lip–cleft palate or cleft palate who were adopted by families in the United States and had a primary palatoplasty performed by the senior surgeon. Patient characteristics and Veau classification are presented in Table 1. The median age at palatoplasty was 25.6 ± 11.8 months; only seven children (13 percent) had palatoplasty

Table 1. Patient Characteristics

	Value (%)
No. of patients	55
Age at palatoplasty, mo	
Median ± SD	25.6 ± 11.8
Range	10.1–60.2
Male-to-female ratio	30:15
Country of adoption	
China	36 (65)
Korea	9 (16)
Russia	5 (9)
Kazakhstan	2 (4)
Japan	1 (2)
India	1 (2)
Bulgaria	1 (2)
Cleft palate type	
Veau I	1 (2)
Veau II	1 (2)
Veau III	37 (67)
Veau IV	16 (29)
Cleft lip	53 (96)
Repaired in native country	35 (66)
Repaired after adoption	18 (33)
Interval to most recent follow-up after palatoplasty, mo	
Median ± SD	59.0 ± 61.3
Range	13.0–220.5

before the age of 12 months. Most children had their cleft lip repaired in their native country before adoption, whereas fewer patients presented with both an unrepaired cleft lip and cleft palate (Fig. 1). We noted a trend toward an association between patients with bilateral cleft lip being unrepaired ($p = 0.062$). However, this relationship between the cleft lip being unilateral or bilateral and whether or not it was repaired before adoption did not reach significance.

Postoperative speech evaluations are listed in Table 2. Palatoplasty was successful, with speech outcome characterized as competent or competent to borderline competent in 28 patients (51 percent), but a nearly equal number of children [$n = 27$ (49 percent)] were categorized as palatoplasty failure because of a borderline to borderline incompetent or incompetent velopharyngeal mechanism. The need for a secondary operation was independent of Veau palatal type ($p = 0.6$) (Fig. 2).

We compared patients who did/did not require a secondary operation for velopharyngeal insufficiency (Table 3). Patients who required a secondary operation were significantly older at the time of primary palatoplasty compared with those who did not (29.7 months versus 21.6 months, respectively; $p = 0.009$) (Fig. 3). Similarly, we found a significant association between increasing age at palatoplasty and need for a secondary operation (OR, 1.07; 95 percent CI, 1.01 to 1.13; $p = 0.01$) (Fig. 4). Of the seven children who had palatoplasty before age 12 months, three required a pharyngeal flap. Children managed with a pharyngeal flap had a significant improvement in speech when median preoperative and postoperative weighted speech values were compared (10.3 ± 2.8 versus 1.4 ± 1.1, respectively; $p < 0.001$) (Fig. 5). Two patients had hyponasal resonance following pharyngeal flap, but there were no cases of sleep apnea, flap dehiscence, or other complications.

DISCUSSION

We evaluated a series of consecutive internationally adopted children with unrepaired cleft palate and found that palatal repair was usually performed at an advanced age and often resulted in poor speech outcome. In one study, parents of

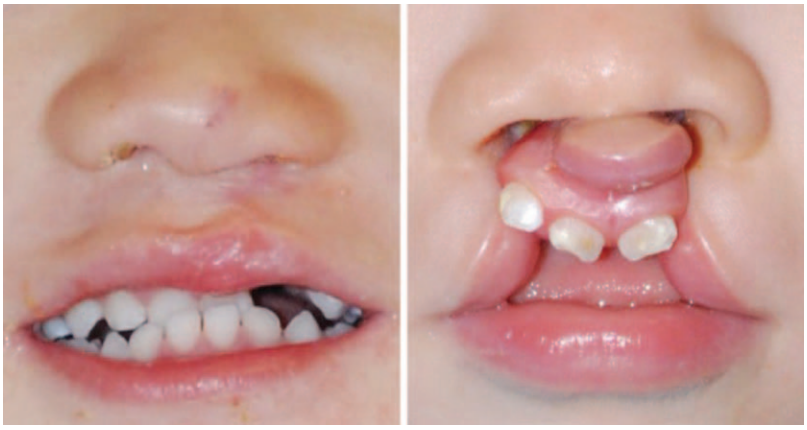

Fig. 1. (*Left*) Chinese child with a left complete cleft lip and palate, with labial repair before adoption; the palate is unrepaired. (*Right*) Chinese child with a bilateral complete cleft lip and palate, unrepaired at the time of adoption.

Table 2. Postoperative Results of Perceptual Speech Evaluation, Need for a Secondary Operation, and Fistula Incidence

	Value (%)
No. of patients	55
Velopharyngeal function	
Competent or competent to borderline competent	28 (51)
Borderline to borderline incompetent or incompetent	27 (49)
Secondary operation recommended	27 (49)
Fistula	5 (9)

children adopted with cleft palate identified speech as the most troubling issue.[33] Factors contributing to poor speech outcome include palatal type, age at adoption and palatoplasty, fistula occurrence, infectious diseases, correcting compensatory articulatory errors, learning a new language, and deprivation as a result of living in an orphanage.

Most children in our series were adopted from China, Korea, and Russia. These countries represent some of the most common countries from which children are adopted to the United States.[1]

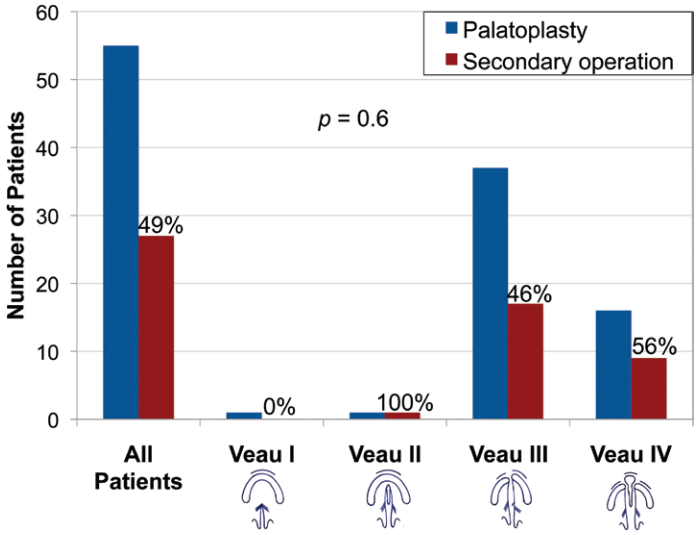

Fig. 2. Veau cleft palatal type compared with need for a secondary operation. Percentage indicates number of patients requiring secondary operation per total number of patients categorized by Veau type. Need for a secondary operation is independent of palatal type ($p = 0.6$, Fisher's exact test).

Table 3. Characteristics of Patients Who Did/Did Not Require a Secondary Operation following Palatoplasty Based on Speech Outcome

Patient Characteristic	Secondary Operation (%)	No Secondary Operation (%)	*p**
No. of patients	27	28	
Age at palatoplasty, mo			
Median ± SD	29.9 ± 12.8	21.7 ± 9.2	0.009
Range	11.0–60.2	10.1–45.9	
Male-to-female ratio	16:11	14:14	0.6
Country of adoption			1.0
China	18 (67)	18 (64)	
Korea	4 (15)	5 (18)	
Russia	3 (11)	2 (7)	
Kazakhstan	1 (4)	1 (4)	
Japan	0 (0)	1 (4)	
India	0 (0)	1 (4)	
Bulgaria	1 (1)	0 (0)	
Cleft palatal type			0.6
Veau I	0 (0)	1 (4)	
Veau II	1 (4)	0 (0)	
Veau III	7 (63)	20 (71)	
Veau IV	9 (33)	7 (25)	

*The *p* values were calculated using the Wilcoxon rank sum test to compare median age and Fisher's exact test to compare all other proportions.

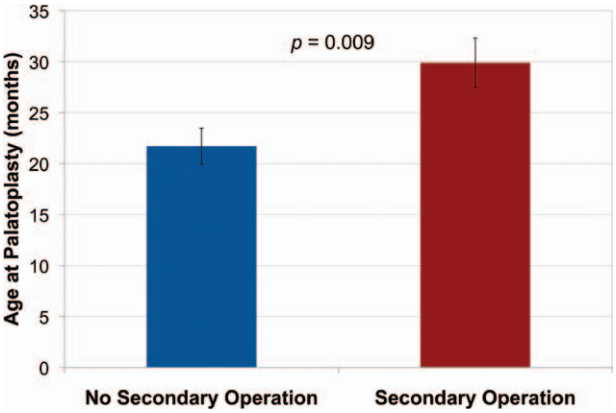

Fig. 3. Comparison of median age at primary palatoplasty between patients who did/did not require a secondary operation ($p = 0.009$, Wilcoxon rank sum test). *Error bars* = standard error.

We found a 2:1 male-to-female ratio in our cohort. Although this ratio is consistent with epidemiologic studies of children with a cleft lip and palate,[34] it is the inverse of the sex ratio of internationally adopted children.[1] Our finding of a 2:1 ratio of Veau III to Veau IV palatal clefts was similar to our previous reports of nonadopted children[17,21]; however, the frequencies of Veau I and II palatal clefts in adopted children were disproportionately lower than expected. Epidemiologic studies, including those from Korea, describe cleft lip and palate (Veau III and IV) to be twice as common as isolated cleft palate (Veau I and II),[34] but our series of adopted children had a Veau III/IV–to–Veau I/II palatal cleft ratio of more than 25:1. Our findings may represent selection bias for the children who sought care in our clinic. Our data indicated that the likelihood of an adoptee having a palatoplasty in their native country follows the Veau hierarchy, namely, that less severe palatal clefts (Veau I and II) are more likely to be repaired before adoption. Furthermore, we found that children with a bilateral cleft lip and palate (Veau IV) were more likely to have had neither the cleft lip nor the cleft palate repaired before adoption. Hansson and colleagues[35] also noted a greater percentage of patients with a Veau IV palatal cleft among adopted patients compared with those not adopted.

We did not find an association between the Veau hierarchy and speech outcomes, as we and others have previously reported.[17,21,36,37] The lack of a relationship in this series may be a type II statistical error resulting from the small sample of patients with a Veau I or II palatal cleft. It may also be that older children, regardless of their cleft palatal type, are more likely to develop velopharyngeal insufficiency such that any potential relationship with palatal type was not detected.

The overall incidence of velopharyngeal insufficiency in these adopted patients (49 percent) is much higher than our previously reported series of 449 consecutive nonsyndromic patients who underwent palatoplasty at a mean age of 11.6 ± 4.9 months and had an overall incidence of velopharyngeal insufficiency of 14.9 percent, which decreased to less than 12.5 percent when the operation was performed before 11 months of age.[17] As before, we found a significant association between increasing age at the time of palatoplasty and risk of velopharyngeal insufficiency. This finding provides further evidence of the

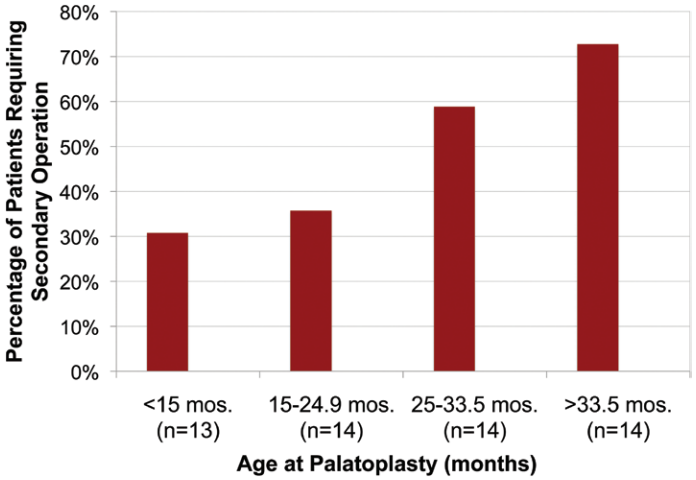

Fig. 4. Age at palatoplasty compared with need for a secondary operation. There is a significant association between increasing age at palatoplasty and need for a secondary operation ($p = 0.01$, logistic regression).

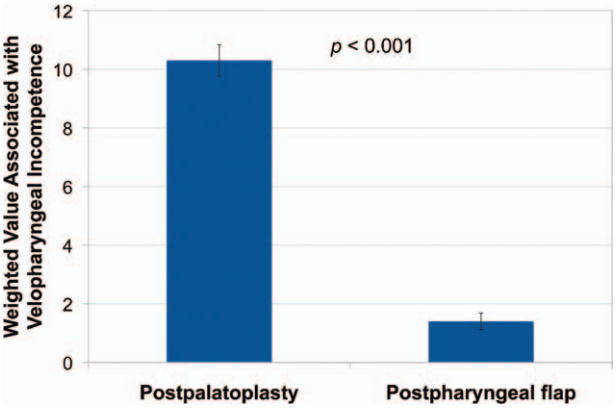

Fig. 5. Comparison of average pre–pharyngeal flap and post–pharyngeal flap Pittsburgh Weighted Values for speech symptoms associated with velopharyngeal incompetence scores. Pharyngeal flap surgery resulted in significant improvement in speech ($p < 0.001$, Wilcoxon signed rank test). *Error bars* = standard error.

relationship between increasing age at palatoplasty and decreased likelihood of achieving velopharyngeal competence.[17,19–23,38,39] Spoken words typically begin at approximately 12 months of age, and few children in our series underwent repair before this critical age. Even when treated before 12 months of age, a higher than expected number of children required a secondary operation, which suggests that additional variables contribute to speech development in these adopted children. When necessary, we treated velopharyngeal insufficiency in this group of adopted children with a pharyngeal flap, which was highly successful, with low risk. These results are corroborated by our previous findings[25–27,38] and by others.[28,29,37,40–49]

We agree with Hansson and colleagues,[33,35] who advocated palatal repair and not cleft lip repair as the priority following adoption. When patients present at an advanced age with unrepaired unilateral cleft lip and palate, we recommend altering the usual order of operations to first schedule simultaneous palatoplasty and labial adhesion followed by delayed cleft lip repair. Children with unrepaired bilateral cleft lip and palate should also undergo prompt palatoplasty; if the premaxilla is protuberant, palatal closure should be combined with premaxillary setback and bilateral alveolar gingivoperiosteoplasty followed by delayed synchronous bilateral cleft lip repair.[50] Audiologic evaluation was not studied in our audit but deserves future review.

The frequency of palatal fistulae in our series (9 percent) was higher than our previously reported incidence of 2.9 percent[17] but within the range of other reports (4.7 to 30 percent).[10,11,13,14,16,22,51–56] The higher fistula rate in adopted children may be related to infection. Hansson and colleagues[35] also found a considerably higher incidence of fistula (14 percent) in adopted Chinese patients compared with their native patients with cleft palate in Sweden (5 percent). They also found that approximately half of adopted children were carriers of methicillin-resistant *Staphylococcus aureus*, and some were carriers of extended-spectrum beta-lactamase–producing bacteria or penicillin-resistant pneumococci. Preoperative screening and treatment for methicillin-resistant *Staphylococcus aureus* or other resistant organisms may be indicated, and proper perioperative antibiotics should be selected.

Another factor contributing to the high fistula rate may be the advanced age at palatoplasty. Emory and colleagues[52] found a significantly higher fistular incidence following palatal closure between the ages of 12 and 25 months (19.4 percent) compared with palatoplasty before 12 months of age (7.8 percent). Rohrich and colleagues[55] also reported that late palatal repair (average, 48.6 months) was associated with a significantly increased occurrence of fistula compared with staged early palatal repair (average, 10.8 months).

In addition to having delayed primary palatal repair, internationally adopted children face many potential barriers to speech and language development following palatal repair. Many children are reluctant to speak their birth language and learn new articulatory skills, and a new language may slow development of velopharyngeal function.[42,57] Age at adoption alone is also likely critical. Even in the absence of a cleft palate, foreign-born children adopted at an advanced age are at increased risk for delayed or disordered speech and language development.[2,3,58,59] If a native language has developed, the switch in language and culture compounds potential speech and language problems.[3] The environmental deprivation suffered in some orphanages contributes to this delayed development.[4,5] Adoption into an enriched environment does not always fully erase the effects of early deprivation on development for children adopted after 9 to 12 months of age.[4,5] Nevertheless, internationally adopted children can make rapid speech and language gains in the preschool years and can catch up to their native-born peers,[6,42,57] even following late palatal repair.[42]

Our study has some limitations. Speech outcomes were assessed by more than one speech pathologist, and interrater reliability was not evaluated. Although the speech pathologists who

evaluated children in this study specialize in cleft and craniofacial anomalies, they may vary in their description of speech characteristics. The threshold for recommending a secondary operation may vary between surgeons and institutions, which makes it difficult to generalize our findings. We did not study the potentially confounding relationship between velopharyngeal insufficiency and change of language if a child was adopted after learning a native language. Polysomnography was not routinely conducted on all children after pharyngeal flap surgery, but only on those who presented with complaints of obstructive symptoms or sleep disturbance.

CONCLUSIONS

We audited a consecutive series of foreign-born adopted children with unrepaired cleft palate. Our results confirm that an advanced age and delayed primary palatoplasty were associated with poorer speech outcomes and an increased likelihood for requiring a secondary operation. Palatal closure should be performed in the first year of life if possible; if children are adopted beyond this age, palatoplasty should be prioritized over lip repair. Craniofacial centers and plastic surgeons should work with adoption agencies and families planning adoption to promote early palatal closure.

Stephen R. Sullivan, M.D., M.P.H.
Division of Plastic and Reconstructive Surgery
Department of Pediatrics
Warren Alpert Medical School of Brown University
Department of Plastic and Reconstructive Surgery
Rhode Island Hospital and Hasbro Children's Hospital
2 Dudley Street, MOC 180
Providence, R.I. 02905
stephen.sullivan@brown.edu

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