



## Long term speech outcomes following late cleft palate repair using the modified Furlow technique



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### ABSTRACT

**Objectives:** Published reports and previous studies from our institution have reported worse overall speech results, including significantly higher rates of persistent articulation errors, in patients undergoing palatoplasty at age >18 months. This study further investigates the effects of late repair on long term speech outcomes.

**Methods:** A retrospective review was performed of non-syndromic patients undergoing primary palatoplasty at age >18 months between 1980 and 2006 at our institution. Longitudinal speech results were compared based on reason for late repair and age at repair.

**Results:** Forty-one patients were greater than 18 months of age at the time of palatoplasty, and 24 fit criteria for longitudinal data analysis. There was a statistically significant improvement in nasality scores at Time Point 1 for international adoptees compared to the non-adopted population ( $p = 0.04$ ). Patients with submucosal clefts were found to have significantly less severe nasal emission scores at Time Point 1 compared to those with overt clefts ( $p = 0.04$ ), but not at Time Point 2. There were no significant differences between scores if repair was performed between 18 and 36 months or >36 months, nor any difference in incidence of articulation errors between subgroups of patients with late repair at either Time Point.

**Conclusion:** Our experience demonstrates that cleft palate repair after 18 months of age is associated with a significantly increased incidence of articulation errors associated with VPI, irrespective of reason for late repair, highlighting the persistence of learned compensatory behaviors in speech and the importance of proceeding with early repair.

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## 1. Introduction

The major functional goal of cleft palate repair is to allow normal speech production. The ideal timing and technique of cleft palate repair is still debated, however, in order to optimize both speech outcomes and maxillary bony development [1]. Controversy regarding the timing of palatoplasty exists between the need for early palate repair to improve speech vs. delaying cleft palate repair to allow for undisturbed maxillary growth [2].

The recommendation for early palatal repair to improve speech production stems from the theory that the palate must be

functional when palate-related sounds are first learned to avoid maladaptive speech development and integration [2,3]. Compensatory strategies may develop in the presence of a cleft because of the need to regulate pressures and flows in the vocal tract, even at the expense of undermining speech performance [4,5]. Kaplan suggested the ideal age for palate closure is 3–6 months [3]. This allows for an additional 3–6 months for post-operative edema to resolve before the palate can be fully functional at 9–12 months. If the cleft palate closure is delayed past 2 years and the compensatory mechanisms for speech have already been developed, the chances for normal speech are significantly diminished [6]. Articulation errors are more likely to develop with late palatal repair. As speech develops, compensatory and obligatory articulation patterns occur in the setting of an abnormal palate and velopharyngeal insufficiency (VPI) [7]. VPI is defined as insufficient seal between the soft palate and the posterior pharynx during speech, resulting in air leak up into the nasopharynx [8,9]. Previous studies have found a significantly

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increased incidence of articulation errors, including non-natural productions not seen in noncleft speakers, some of which include glottal stops, pharyngeal fricatives, and reduced pressure nasalized consonants [2,10,11]. Once learned, compensatory speech patterns are more difficult to correct compared with naturally occurring developmental errors [11].

Early cleft palate repair has been complicated, however, by evidence that it increases the incidence of maxillary growth deformities. Gillies and Fry [12] noted that earlier closure of the hard palate leads to increased deformity such as a narrow arch and posteriorly positioned maxilla. For this reason, Ortiz-Monasterio et al. [13] have previously advocated delaying cleft repair until maxillary growth is complete. Aiming to balance the need for early repair to improve speech and delay of repair to achieve normal maxillary growth, Schweckendiek advocated a staged hard palate closure: early closure of the mucosa first for prevention of VPI allowing normal speech development, but delay of hard palate closure with the goal of avoiding scarring and growth hindrance of the bony palate [14]. This technique has been in use in several institutions [15–17]. However, it has been suggested that this approach results in both poor speech production with compensatory articulation and increased maxillary hypoplasia (likely secondary to scarring from prosthesis use or increased number of secondary procedures for speech correction) in comparison to single stage palate repair [1].

In fact, it is palatoplasty itself, performed at any point before facial growth is complete, which results in maxillofacial hypoplasia in the absence of orthognathic intervention [18]. Ten to 40% of cleft lip and palate patients who undergo surgical repair develop maxillary hypoplasia requiring orthognathic correction, while patients with un-operated cleft palate have been observed to have normal maxillary growth [2,12,13,19]. However, with orthodontic treatments such as alveolar collapse correction, midfacial retrusion correction, and dental alignment, cleft palate patients operated on as early as 4 months of age can achieve normal cephalometric parameters during adolescence [20]. In light of these findings, cleft palate repair timing has been largely driven by the need for normal speech development.

It is well known that late cleft repair leads to documented worse speech outcomes and higher rates of secondary surgery for VPI [1,2,4,21]. A study from our institution reported significantly worse overall speech scores using the Pittsburgh Weighted Values for Speech Symptoms Associated with VPI in patients who underwent palatoplasty at greater than 18 months of age compared to those who underwent repair at earlier ages ( $p = 0.025$ ) [21]. There was a significantly higher incidence of velopharyngeal insufficiency in the late repair population when compared to those repaired at earlier ages ( $p = 0.024$ ). Both nasality and nasal emission did not significantly differ between late repair and repair at 18 months or younger ( $p = 0.13$  and  $p = 0.51$ , respectively); however, this late repair population did have a significantly higher rate of persistent articulation errors associated with VPI ( $p = 0.0001$ ). There was no significant difference in rates of secondary surgery for VPI between the two groups ( $p = 0.57$ ).

The purpose of this study is to further investigate nasality, nasal emission, and articulation after late cleft palate repair at our institution in non-syndromic patients. In addition, by evaluating longitudinal assessments of speech in this population, this study aims to further describe speech patterns over time after late repair to determine which patients are at highest risk for persistent compensatory articulation errors.

## 2. Materials and methods

A retrospective review was performed of all non-syndromic patients undergoing primary cleft palate repair by the modified

Furlow technique between 1980 and 2006 at The Children's Hospital of Philadelphia. Patients presenting late for repair (>18 months of age) were included in this study and the reason for late repair was investigated through patient records. If patients presented with multiple congenital anomalies, genetic assessments were performed and patients were only included if they were found to have no identified syndrome. Patients were also excluded for the following reasons: patients less than 5 years of age at their last speech examination or with incomplete speech records, evaluation by outside speech pathologists, large unrepaired oronasal or nasoalveolar fistulas that would affect resonance, and persistent hearing impairment of significant magnitude to influence speech development. This study was approved by The Children's Hospital of Philadelphia's Institutional Review Board.

### 2.1. Surgical technique

Cleft palate repair was performed on all patients using the modified Furlow technique as described by Randall et al. [22] in 1986. This included use of opposing mirror image z-plasties of the oral and nasal mucosa, with tension on the closure decreased by lateral relaxing incisions on the hard palate. In cases of both cleft lip and cleft palate, the alveolar cleft was left for closure at the time of bone grafting.

### 2.2. Speech evaluation

Speech outcomes were assessed at a minimum of 5 years of age by the Pittsburgh Weighted Values for Speech Symptoms Associated with VPI [23]. Speech evaluations were performed by two senior licensed speech pathologists with documented inter-rater reliability [24]. Speech symptoms during clinic evaluation were scored using this standard format which assesses nasal emission, facial grimace, nasality, phonation, and articulation. Based on the total score, velopharyngeal function was characterized as competent to borderline competent (composite score  $\leq 2$ ), borderline competent to borderline incompetent (composite score = 3–6), or incompetent (composite score  $\geq 7$ ). Longitudinal speech evaluations after late repair (greater than 18 months old) were collected and compared in patients fitting inclusion criteria with two documented Pittsburgh Weighted Values for Speech Symptoms Associated with VPI at least one year apart. Speech evaluations were collected initially following cleft palate repair and at the most recent recorded examination, and were titled Time Points 1 and 2, respectively.

The reason for late repair fell into four categories: children who were internationally adopted, patients with submucosal cleft palate presenting with hypernasal speech, late presentation, and children whose medical needs precluded earlier surgery. The speech results were compared based on two categories: the international adoptee population was compared to the non-adopted population, and the submucosal cleft palate population compared to those with overt cleft palate. Additionally, these late repair patients were compared according to age of repair in subcategories of repair at >18 months but <36 months and repair at 36 months or greater.

### 2.3. Statistical analysis

Using Stata statistical software, data analysis included ordered logistical regression test, chi square test, and Fisher's exact test.

## 3. Results

### 3.1. Patient characteristics

Forty-one patients were greater than 18 months of age at the time of palatoplasty. Average age at surgery in this late repair

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