INTRODUCTION

Velopharyngeal dysfunction describes any condition in which the velopharyngeal valve does not properly close during the production of oral sounds. Velopharyngeal dysfunction has multiple causes, including velopharyngeal insufficiency (VPI) due to abnormal anatomy, velopharyngeal incompetence due to neurophysiologic dysfunction, and velopharyngeal mislearning due to incorrectly learned speech sounds. Palatal shortening, scarring, insufficient palatal muscle function, and palatal fistulas may lead to VPI, which manifests as hypernasality, nasal emissions, and weak pressure consonants. Furthermore, children with cleft palate (CP) and VPI may learn to use abnormal speech sounds due to abnormal anatomy. This leads to compensatory articulation including glottal stops and glottal, nasal, or pharyngeal fricatives. The overall result is decreased intelligibility of speech and social impairment.

How Early Can We Predict the Need for VPI Surgery?

Background: Velopharyngeal dimensions change as a child with cleft palate (CP) grows. The aim of this study was to assess if the decision for velopharyngeal insufficiency (VPI) surgery can be made by the age of 3 years among CP children with moderate-to-severe VPI. In addition, we sought to clarify if speech therapy before VPI surgery is beneficial for VPI speech characteristics.

Methods: This retrospective study reviewed documentation of children with moderate-to-severe VPI at age 3 years who did not undergo VPI surgery until age 5 years. Based on the national cleft register, 959 patients with syndromic and nonsyndromic CP were treated by the craniofacial team at Helsinki University Hospital, Finland between 2000 and 2014. Eighty-six patients fulfilled the study inclusion criteria. The speech pathologist evaluated velopharyngeal function at age 3, 5, and 8 years.

Results: Of the 86 children presenting with moderate-to-severe VPI at age 3 years, 94% still had moderate-to-severe VPI at age 5 years, even though speech therapy was offered to 77%. Of those whose velopharyngeal function improved by age 5 years, function regressed to incompetent over time. Overall, 93% underwent VPI surgery and 82% underwent VPI surgery between ages 5 and 8 years. Only 23% at age 8 years still had moderate-to-severe VPI. Speech therapy alone did not improve VPI speech characteristics.

Conclusions: Moderate-to-severe VPI did not improve from 3 to 5 years or improved but subsequently relapsed. This suggests that the decision for VPI surgery can be made for children aged 3 years with moderate-to-severe VPI. (Plast Reconstr Surg Glob Open 2022;10:e4678; doi: 10.1097/GOX.0000000000004678; Published online 21 November 2022.)

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nasal emissions, or weak pressure consonants even with only a small velopharyngeal gap. However, compensatory articulation can be treated with speech therapy. Some cleft teams start speech therapy for compensatory articulation before VPI surgery. Some recommend correcting the anatomy first, as speech therapy may be more effective and less time-consuming if performed after anatomical correction. The frequency of VPI surgery after primary cleft palatoplasty appears to vary with cleft anatomy, palatoplasty method, surgeon experience, and the presence of 22q11.2 deletion syndrome or Robin sequence (RS); the frequency range is 3%–56%. The surgical methods and timing of VPI surgery vary among cleft teams. Careful monitoring of speech remains paramount in early identification of VPI. Surgeons should not plan VPI surgery before a reliable assessment of velopharyngeal function (VPF). A normally developing child is usually talking and sufficiently cooperative for a speech assessment by the age of 3 years. Traditionally, VPI surgery is performed during preschool years to achieve the best possible speech development by utilizing this critical period of brain development. Furthermore, some patients with CP require VPI surgery at an older age if VPI characteristics continue to improve with growth or after osteotomy. The mean age of VPI surgery is between 5 and 10 years. Many children with CP and VPI receive extensive speech therapy, either alone or with VPI surgery. Although speech therapy can address to compensatory articulation and not VPI, a common misconception about the role of speech therapy in the management of “cleft palate speech” still exists. Although conservative management with speech therapy is attempted to treat VPI before surgical intervention, such management has not been shown to be effective. Traditionally at our cleft center, children with CP and VPI at age 3 years have been followed and usually referred to speech therapy; these children have been referred to VPI surgery if VPI is still evident at age 5 years. In recent years, we have performed VPI surgery earlier if moderate-to-severe VPI is evident. Such a decision is based on gained clinical experience and the results presented in this study. However, as the child grows, the nasopharyngeal dimensions can change as craniofacial structures grow and adenoids involute, causing changes in VPF.

The purpose of this study was to assess if the decision to perform VPI surgery can be made already at age 3 years among children with speech characteristics related to moderate-to-severe VPI. Accordingly, we assessed if VPF improved by age 5 or 8 years without VPI surgery. We also evaluated if syndrome or RS presence influence change in VPF. In addition, we sought to clarify if speech therapy before VPI surgery is beneficial for VPI speech characteristics.

MATERIAL AND METHODS

This was a retrospective single-center study approved by Helsinki University Hospital, Finland. We followed the principles of the Declaration of Helsinki. The study had approval to use patient data from the patient database of the Cleft and Craniofacial Center of Helsinki University Hospital.

Participants

Based on the national cleft register, 959 patients with CP were treated by the Craniofacial Team at Helsinki University Hospital, Finland, between 2000 and 2014. Of these 959 patients, we included only those who had speech characteristics related to moderate-to-severe VPI at the age of 3 years and who did not undergo VPI surgery before the 5-year follow-up visit (Fig. 1). One hundred children met the inclusion criteria. These patients were identified by the cleft center’s speech pathologist after primary palatoplasty at 3 years of age. Fourteen children were excluded; three were lost from follow-up visits, four missed the 8-year follow-up visit, and seven had a severe speech disorder that prevented reliable assessment of VPF at age 3 years. Four of these seven excluded children were syndromic, including one child with Kabuki syndrome, one with Goldenhar, one with 22q11.2 deletion syndrome, and one with an unknown syndrome. The studied group consisted of 86 children with all cleft types; those with syndromes and

Takeaways

**Question:** Can we assume that moderate-to-severe VPI does not improve without surgery and decide to re-operate at as early as 3 years of age?

**Findings:** Although most 3-year-old children with moderate-to-severe VPI received speech therapy from 3 to 5 years of age, their VPF did not change by age 5 or improved but later relapsed back to incompetent.

**Meaning:** Moderate-to-severe VPI in 3-year-old children with cleft palate does not resolve without surgery and the decision to re-operate can be made already at 3 years of age.

![Fig. 1. Study participants.](image-url)
Table 1. Overall VPF Graded by Speech Pathologist

| Degree          | Scale |
|-----------------|-------|
| Competent       | A     |
| Borderline competent | B     |
| Mild-to-moderate VPI | C     |
| Moderate-to-severe VPI | D     |

Speech was clear without any nasality, nasal emissions, or difficulties producing pressure consonants or high vowels. In Finnish, a nasometry score greater than 29% refers to hypernasality. The speech pathologist graded overall VPF into the following categories: competent (A), borderline competent (B), mild-to-moderate VPI (C), and moderate-to-severe VPI (D) (Table 1). Nasoendoscopy and videofluoroscopy were performed in the case of an uncertain role of VPI in perceptual speech evaluation and, if needed, in preoperative planning: 37 children underwent videofluoroscopy and two nasoendoscopy at age 5.

We retrospectively evaluated speech data from hospital records at ages 3, 5, and 8 years. We rated the speech variables and intelligibility on four-point ordinal scales (0–3) at age 3, 5, and 8 years (Table 2) and overall VPF on a four-point ordinal scale (A–D) (Table 1). We pooled the four possible ratings into three scale values corresponding to the traffic light color coding system, where green indicates normal/no speech deviances and slight deviances/single occurrences within the normal limit (value 0/value A and B); yellow indicates mild deviation/some occurrences (value 1/value C); and red indicates moderate deviation/frequently occurring and severe/occurs indicates always or almost always (values 2 and 3/value D) (Tables 1 and 2). A two-point scale was used (occurrence or no occurrence) for compensatory articulation and weak pressure consonants.

Speech Therapy

Speech therapy was recorded if the child received speech therapy by the 5-year follow-up visit before undergoing VPI surgery. The type of speech therapy was designed to target speech problems concerning the cleft diagnosis, such as compensatory articulation, phonological delay, difficulties with articulation, and speech characteristics related to VPI. In addition, some children also received speech therapy for other speech deficiencies. Complete documentation of the speech therapy targets and amount were unavailable, as speech therapy may have been provided by local hospitals or schools.

Table 2. Assessment of Speech Variables Related to VPI and Intelligibility

| Scale  | Degree                  | Frequency of Occurrence               |
|--------|-------------------------|---------------------------------------|
| 0      | Normal                  | No occurrence                         |
| 1      | Mildly incompetent/deviant | Completely intelligible               |
| 2      | Moderately incompetent/deviant | Single to some occurrences Mostly intelligible |
| 3      | Severely incompetent/deviant | Frequently occurring Mostly unintelligible |
| 4      | Completely unintelligible | Occurs always or almost always Completely unintelligible |
Statistical Analysis

SPSS version 27 (IBM Corp., Armonk, N.Y.) was used for data analysis. We performed descriptive analyses and counted the frequency of occurrence for the different speech variables in the three age groups. Data distribution was verified by the Kolmogorov–Smirnov test. Differences between age groups (related samples) were compared with two-tailed Wilcoxon signed-rank test because of the skewed data. Differences in the same age groups (independent samples) were tested with two-tailed \( \chi^2 \) and Fisher exact tests. Differences in mean age (independent samples) were tested with Mann–Whitney \( U \) test. We considered a probability less than 0.05 as significant.

RESULTS

Speech Characteristics Related to VPI

Overall VPF, audible nasal emissions, weak pressure consonants, compensatory articulation, and overall intelligibility were recorded in all 86 children at age 3, 5, and 8 years. Hypernasality was recorded in 84 children at age 3 years (two were not reliably assessed; data not shown) and in all children at age 5 and 8 years. Results of the assessment are shown in Figure 2 for overall VPF, hypernasality, nasal emissions, and intelligibility. A nasometer was not used at age 3 years. At age 5 years, a nasometer was used on 61 children; the mean value was 45 (SD 12.4, range 18–72). At age 8 years, a nasometer was used on 85 children; the mean value was 25 (SD 14.4, range 6–65). The difference between nasalance scores at age 5 and 8 years was statistically significant \( (P < 0.001) \). The prevalence of weak pressure consonants and compensatory articulation in the age groups is shown in Figure 3.

The changes between age 3 and 5 years in overall VPF \( (P = 0.083) \), hypernasality \( (P = 0.929) \), weak pressure consonant \( (P = 0.237) \), compensatory articulation \( (P = 0.108) \), nasal emissions \( (P = 0.506) \), or intelligibility \( (P = 0.489) \) were not statistically significant; none received VPI surgery before age 5 years. As most children had VPI surgery between age 5 and 8 years, the speech outcome improvement from age 3 to 8 years was statistically significant in overall VPF \( (P < 0.001) \), hypernasality \( (P < 0.001) \), weak pressure consonant \( (P < 0.001) \), compensatory articulation \( (P < 0.001) \), nasal emissions \( (P = 0.007) \), and intelligibility \( (P < 0.001) \).

Secondary Surgery

Secondary procedures for VPI and fistula repair are presented in Table 3. Mean age of VPI surgery was significantly higher in children with a syndrome or RS (6.3 years, SD 1.5, range 4.8–10.6) than in nonsyndromic children (5.7 years, SD 1.3, range 4.7–12.1) \( (P = 0.030) \). Speech outcomes of children who underwent VPI surgery before age 8 years and those who did not are presented in Table 3. Speech outcomes of children who underwent VPI surgery between ages 5 and 8 years had significantly worse VPF at age 5 years than children who did not \( (P = 0.002) \). Children who underwent VPI surgery between ages 5 and 8 years had significantly better VPF at age 8 years than children who did not \( (P < 0.001) \). Thirteen children did not have VPI surgery until age 8 years. Twelve of these children \( (92\%) \) still had VPI at age 8 years; seven of these children had VPI surgery after age 8 years. The one patient who achieved competent VPF by age 8 years had speech characteristics related to moderate-to-severe VPI at age 5 years which improved after tonsillectomy.

Speech Therapy before VPI Surgery

Between age 3 and 5 years, 66 \( (77\%) \) children underwent speech therapy prior to VPI surgery. Speech outcomes of children who received speech therapy and those who did not are presented in Supplemental Digital Content 2 (http://links.lww.com/PRSGO/C283). Groups of children who received speech therapy before age 5 years and children who did not were not significantly different in the primary surgery method \( (P = 0.770) \), cleft type \( (P = 0.111) \), or the presence of syndrome or RS \( (P = 0.218) \). There was no significant correlation between administered speech therapy and overall VPF or speech characteristics related to VPI at age 5 or 8 years. Speech intelligibility was significantly better in the 8-year-old children who received speech therapy compared to children who did not \( (P = 0.012) \), but not in the 5-year-old children \( (P = 0.289) \).

Syndrome or RS

Speech outcomes of children with a syndrome or RS and nonsyndromic children are shown in Supplemental Digital Content 2 (http://links.lww.com/PRSGO/C283). No significant effect on overall VPF or speech characteristics related to VPI was observed at age 5 years before reoperations. Children with a syndrome or RS had significantly worse overall VPF \( (P = 0.013) \) and more hypernasality \( (P = 0.021) \) at age 8 years than nonsyndromic children after 74% and 88% of them had undergone VPI surgery, respectively.

Longitudinal Data of Children with Improved VPF

VPF improved by age 5 years in five \( (6\%) \) children. In two of these children, overall VPF improved to mildly incompetent by age 5 years. In one child, overall VPF relapsed back to moderately to severely incompetent by age 8 years. However, VPI surgery was not performed as intelligibility was good. In the other child, overall VPF remained mildly incompetent by age 8 years but later relapsed to moderate-to-severe incompetent; VPI surgery was performed at age 12 years. Three of the studied children reached normal overall VPF by age 5 years. However, two developed moderate-to-severe incompetent VPF and both received VPI surgery (age 7 and 8 years). The remaining child’s VPF relapsed to mildly incompetent by age 8 years and VPI surgery was considered.
Fig. 2. Overall VPF, hypernasality, nasal emissions, and intelligibility at age 3, 5, and 8 years. No children had VPI surgery before age 5, and 73 (85%) underwent VPI surgery between ages 5 and 8 years. Green indicates 0 (no occurrence or normal); yellow, 1 (mild deviances/occurrences); red, 2 (moderate-to-severe degree).
This study sought to determine if a decision for VPI surgery can be made at age 3 years for children with moderate-to-severe VPI. This assumes that 3-year-old children with CP and moderate-to-severe VPI still remain symptomatic despite speech therapy and growth. The perceptual assessment of speech to identify children with moderate-to-severe VPI at age 3 years by a cleft speech pathologist was very reliable, and speech characteristics related to VPI appeared to be very persistent. Interestingly, although 77% received speech therapy, 94% of VPI speech characteristics did not change by age 5 years. Particularly interesting was that even though VPI speech characteristics of five (6%) children improved by age 5 years, VPF relapsed back to incompetent with time and growth in all five.

Early management of VPI may lead to better outcomes. Riski et al. observed greater success in children aged younger than 6 years in two studies of 48 and 139 patients undergoing VPI surgery. However, better outcomes in younger patients have not been observed in all studies. Nevertheless, VPI results in poor quality-of-life for patients and their parents, and children with severe hypernasality have improved quality-of-life after VPI surgery. Moreover, speech disorders in patients with CP, such as compensatory articulation, may become incorporated into the child’s developing phonological rule system and produce a phonological disorder. Treatment of such disorders at an early age may be beneficial for normal speech development. Furthermore, children with CP and persistent speech problems are at greater risk of delayed acquisition of reading skills.

However, the lack of objective measurements is often present at a young age: not all children at age 3 years are cooperative for instrumental assessment of speech. Instrumental assessment with a nasometer provides an objective evaluation of nasalance. Furthermore, nasoendoscopy and videofluoroscopy provide important functional and anatomic findings to facilitate planning of VPI surgery. The role of preoperative dynamic examination using such instrumental methods is especially important in more complex cases, such as in residual VPI and syndromic patients. Interestingly, a recent study revealed that routine preoperative nasoendoscopy may not provide new information or a better outcome in patients undergoing double-opposing Z-plasty for VPI but leads to increased time to surgery after diagnosis of VPI. In this material, some patients underwent mostly videofluoroscopy at age 5.
years for preoperative planning if needed. More recently, we have been developing our preoperative examination protocol by performing nasoendoscopy more routinely.

Parents of children with only mild VPI report better quality-of-life than parents of children with severe VPI. Furthermore, the speech pathologist may have difficulties in identifying speech characteristics related to VPI in very young children if the condition is mild, especially if the child has other speech deviations in addition to VPI. For these reasons, we did not include children with mild VPI in our study; we believe these children should undergo a longer follow-up period before deciding on VPI surgery. We included children with a diagnosis of a syndrome or RS (19 of 86) whose VPF the speech pathologist could assess. Nevertheless, if moderate-to-severe VPI was noted at age 3 years, the condition was also very persistent in these children before reoperations. However, the assessment of VPF is not always easy in these children, and a few syndromic children with some speech characteristics of VPI were excluded because the VPF assessment was not reliable. At age 8 years, when most children had received VPI surgery, children with a syndrome or RS had more residual VPI. Most of the children received double-opposing Z-plasty for VPI surgery, and we previously noted that the method might lead to more residual VPI in syndromic patients.

If the speech pathologist has difficulties in drawing definite conclusions about VPF in young children, the speech pathologist and cleft surgeon might traditionally have chosen speech therapy as the primary course of rehabilitation, even though no evidence supports this approach. Ruscello addressed the issue of speech therapy and especially oral motor exercises (such as blowing and sucking) without surgery in treatment of VPI and concluded that such treatment is ineffective and would benefit only a small proportion of patients. In the current study, 77% of the children received speech therapy prior to VPI surgery between age 3 and 5 years; although intelligibility was better in the 8-year-olds who received speech therapy, these children had no significant improvement in VPF or in the presence of compensatory articulation errors. Although speech therapy was targeted for VPI, the specific content and amount of speech therapy was not recorded because it was given in local hospitals and schools; thus, our results are only preliminary. Based on these preliminary findings and the current literature, we agree with Kummer that in the case of VPI, a rational approach is to correct the anatomy first; once the anatomy is corrected, speech therapy is not as time-consuming and is effective in correcting the remaining articulation placement errors.

With a tradition of centralized cleft care in Northern Europe, we treat a relatively large number of cleft patients and have a comprehensive follow-up protocol, which enables thorough clinical assessment of the conditions that the cleft team encounters. Certain limitations of the retrospective design of this study exist. We were not able to use a validated scale for speech evaluation, as the speech evaluations were only driven by patient records. We did not have specific information about the given speech therapy. The lack of consistent record keeping and the lack of detailed information on the kind of interventions used in speech therapy are major limitations; thus, the study’s results on the efficacy of speech therapy for VPI treatment are only preliminary. Further studies with exact methods of speech therapy are needed to determine the effectiveness of speech therapy in VPI and compensatory articulation treatment.

CONCLUSIONS

The speech characteristics related to moderate-to-severe VPI in this study did not improve from age 3 to 5 years without VPI surgery or improved, but later relapsed to become incompetent even though speech therapy was offered to most children. We conclude that the decision to perform VPI surgery can be made already at age 3 years.
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