Risk of Persistent Palatal Fistula in Patients With Cleft Palate
Mairaj K. Ahmed, DDS, MS; Anthony L. Maganzini, DDS, MSD; Paul R. Marantz, MD, MPH; Joseph J. Rousso, MD

IMPORTANCE Many individuals with a cleft palate also have an associated craniofacial syndrome or anomaly.

OBJECTIVE To investigate the predictive associations of persistent palatal fistulas in patients with previously repaired cleft palate.

DESIGN, SETTING, AND PARTICIPANTS We performed a case-control study of patients with cleft palate repairs from January 1, 1986, through December 31, 2000, at a major tertiary care hospital center in the Bronx, New York. The study population consisted of patients who had their primary surgery before the age of 3 years and had all their cleft-related treatment completed at the same hospital center. Palatal fistula was defined as a breakdown of the primary surgical repair of the palate, resulting in persistent patency between the oral and nasal cavities. Data collection was conducted by using the hospital centers’ electronic medical records and patient tracking systems and confirmed by review of hard copies of patient records.

MAIN OUTCOMES AND MEASURES The Veau classification system was used to classify the preoperative cleft severity.

RESULTS A total of 130 patients were identified—23 patients with palatal fistula and 107 controls. A total of 12 girls and 11 boys were identified in the palatal fistula group and 56 girls and 51 boys in the control group. The mean patient age at the time of palatoplasty was 12.6 and 14.5 months in the palatal fistula and control groups, respectively. A statistically significant association was found between the outcome of fistula and severity of cleft, as defined by the Veau classification system ($P = .01$). Furthermore, for each Veau class increase, the odds of a palatal fistula increased by 2.64 (95% CI, 1.35-5.13; $P = .004$). No statistically significant associations were found between the outcome of fistula and the following independent variables: patient sex ($P = .98$), patient age at palatoplasty ($P = .82$), type of palatoplasty ($P = .57$), surgeon ($P = .15$), orthodontic treatment ($P = .59$), ear infection ($P = .30$), or clefts associated with syndromes ($P = .96$).

CONCLUSIONS AND RELEVANCE Palatal fistulas are reliably associated with severity of cleft, as defined by the Veau classification system. This knowledge gives the health care professional a more reliable method of preoperatively assessing the risk of postoperative palatal fistula in the cleft palate population.

LEVEL OF EVIDENCE 3.

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Cleft lip or palate is the fourth most common birth defect and the most common craniofacial anomaly, affecting 1 in 500 to 750 live births in the United States and totaling approximately 7500 cases per year.1,2 Between 5% and 15% of individuals with a cleft have an associated craniofacial syndrome or anomaly.3

Historically, the treatment of palatal clefts was through the use of obturators. In 1837, Diefenbach4 first described the use of relaxing incisions to aid in palatal closure, and several techniques have evolved since then to surgically close the palate.4,5 This method of closure has markedly improved speech, feeding, and other functional outcomes.6,7 The drawbacks of primary closure include palatal fistulas, velopharyngeal insufficiency, deficient anterior-posterior maxillary growth, and deficient vertical midfacial development.9,10

Palatal fistulas are defined as a failure of healing or a breakdown in the primary surgical repair of the palate, resulting in a patency between the oral and nasal cavities. The Pittsburgh Fistula Classification System11 includes 7 types of fistula (uvula, soft palate, junction of hard and soft palate, hard palate, junction of primary and secondary palate, lingual-alveolar, and labial-alveolar). Fistulas may be symptomatic or asymptomatic. Partially because of this, several studies11-13 have documented vague or nonexistent descriptions of fistulas in medical records. In addition, the definition of fistulas in the literature has been ambiguous and inconsistent.14,15

A symptomatic fistula can lead to several problems, such as oronasal fluid and food regurgitation, malodorous discharging, rhinitis, hearing loss, audible nasal air escape during speech, and hypernasality. The incidence of fistula diagnosis during cleft palate treatment has been reported to range from 9% to 50%.14,15 The rate of fistula recurrence ranges from 35% to 75%.12,16

The basis of this study is to determine a reliable method to help the physician make a preoperative, evidence-based assessment of the risk of fistula formation. This assessment is based on specific characteristics associated with the patient with cleft palate. Risk factors can be divided into factors determined during initial clinical presentation or factors related to treatment rendered. Risk factors based on clinical presentation include extent of clefting, presence or absence of syndrome, and medical or dental history. Extrinsic factors include type of palatoplasty, age at palatoplasty, operator skill or experience, feeding protocols after surgery, and orthodontic treatment. Although there are many possible untoward postoperative complications that can arise from palatoplasty, our study is limited to investigating postoperative palatal fistula.

### Methods

Montefiore Medical Center Internal Review Board approval for this case-control study was granted on October 9, 2008. Patients who received cleft-related surgical care at outside institutions were excluded based on the variability of protocols and techniques among different institutions. Patients with syndrome-associated and non-syndrome-associated cleft were included. Additional exclusion criteria consisted of incomplete medical records, insufficient follow-up as defined by fewer than 3 postoperative examinations, and/or patients who had palatoplasty surgery at older than 3 years. Matching based on race was not performed because of a lack of biologic plausibility and challenges in data collections. Specifically, in our hospital system, race is a self-reported entry, which has poor levels of consensus (60%-66%).17,18 Generally speaking, the patient population at the hospital center where this study was conducted is racially heterogeneous.

Data collection was conducted digitally using the electronic medical records system and data mining system. Once patient demographic information was obtained, additional clinical information was obtained and confirmed by searching hard copies of patient records. Data were collected for all male and female patients with cleft palate from January 1, 1986, through December 31, 2000.

Palatal fistula was defined as a failure of healing or a breakdown in the primary surgical repair of the palate, resulting in a patency between the oral and nasal cavities that persisted for at least 1 year after surgery. Controls were those who did not have the presence of a fistula, had all cleft-related treatment at the main hospital center, and were examined at least 3 times postoperatively. The Veau classification system was used to classify the preoperative cleft severity (Table 1).

Exploratory univariate associations were investigated using t tests and 2 × 2 and 2 × 4 contingency table analysis. Those variables with P < .20 were included in an ordinal logistic regression model. Statistical analysis was performed using STATA statistical software, version 10 (Stata Corp).

### Results

Statistical analysis was performed for 23 cases and 107 controls. As indicated in Table 2, a statistically significant association was found between the outcome of fistula and severity of cleft, as defined by the Veau classification system. No significant associations were found between the outcome of fistula and the following predictor variables: patient sex, patient age at palatoplasty, type of palatoplasty, orthodontic treatment, ear infection, surgeon, or clefts associated with syndromes.

Logistic regression analysis revealed that for each Veau class increase, the odds of a palatal fistula increased by 2.64 (95% CI, 1.35-5.13; P = .004). This association is shown in the Figure. This association remained statistically significant after including the variable “surgeon” in the model.

### Table 1. Veau Classification of Clefts

<table>
<thead>
<tr>
<th>Veau Class</th>
<th>Description</th>
<th>Cases (n = 23)</th>
<th>Controls (n = 107)</th>
</tr>
</thead>
<tbody>
<tr>
<td>I</td>
<td>Soft palate</td>
<td>0</td>
<td>15 (14.0)</td>
</tr>
<tr>
<td>II</td>
<td>Soft and hard palate</td>
<td>6 (26.1)</td>
<td>44 (41.1)</td>
</tr>
<tr>
<td>III</td>
<td>Soft and hard palate and unilateral prepalatal clefts</td>
<td>12 (52.2)</td>
<td>42 (39.3)</td>
</tr>
<tr>
<td>IV</td>
<td>Soft and hard palate and bilateral prepalatal clefts</td>
<td>5 (21.7)</td>
<td>6 (5.6)</td>
</tr>
</tbody>
</table>
Discussion

Our finding of increasing severity of cleft palate that resulted in increased postoperative complication rates is not surprising. This concept has been supported in many forms, with various outcome measures. The Veau classification system is commonly used as an objective measure of cleft severity and is outlined in Table 1. The general association between fistula rates and the Veau classification system has been recognized by several authors.12,16,19-21 Among these studies,12,16,19-21 which found an association between increased risk of fistula with higher Veau class, there was no quantified risk in terms of odds ratios. However, Jackson et al.22 found a statistically significant difference in the odds of developing postoperative fistulas when comparing preoperative Veau class IV and other classes, but this finding did not hold significance when comparing Veau class III with Veau class I and II.

To our knowledge, this is the first study to report a statistically significant quantification of the risk among individual Veau classes; for each Veau class increase, the odds of a palatal fistula increased by 2.64 times.

Because of the multifactorial nature of cleft palate care, data are contradictory regarding risk factors for fistulas. Regardless of discrepancies, the increased data from large centers continue to paint a clearer picture of optimal care of the patients with cleft palate.

Other studies21,24 have assessed similar outcomes and associations with preoperative cleft widths. In 2009, Parwaz et al.23 found that a width of 15 mm or greater was significantly associated with risk of fistula. In assessing the development of postoperative velopharyngeal insufficiency, Lam et al.24 found that a cleft width of 10 mm increased the risk by roughly 4.5 times compared with narrower clefts. Similarly, we noted corresponding physician annotation with preoperative statements that indicated wide clefts in the patients with fistula; however, no quantifiable classification system was used to describe the width in many of these records. Therefore, we suggest objective and consistent measurements of cleft width as standard documentation in all patients with cleft palate.

This study did not find an association between the use of orthodontic treatment and fistula formation. These findings support those of Muzaffar et al.20 and contrast those of other studies.36,37 However, orthodontic protocols, including technique and timing, vary widely among institutions, which may account for these discrepancies in study data. Regarding presurgical orthopedics, protocols of nasoalveolar molding are associated with low rates of postoperative fistula formation, as demonstrated by Dec et al.25 This finding may be due to its role in decreasing the preoperative cleft width; therefore, we recommend that this variable should be included in any multivariable assessment of fistulas.

Although statistical significance was not obtained in regard to surgical timing, we saw an association in palatal fistula formation in those patients whose primary surgery was performed before 6 months of age. This group was relatively small (3 fistula formations in 5 patients) because of the institutional acceptance of waiting until 10 months of age before performing most primary palatoplasty operations. In addition, no statistically significant association was found between operating surgeon and fistula outcome. This finding is

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Table 2: Summary of Univariate Analysis*

<table>
<thead>
<tr>
<th>Variable</th>
<th>Cases (n = 23)</th>
<th>Controls (n = 107)</th>
<th>P Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Patient sex</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>11 (47.8)</td>
<td>51 (47.7)</td>
<td>.98</td>
</tr>
<tr>
<td>Female</td>
<td>12 (52.5)</td>
<td>56 (52.3)</td>
<td></td>
</tr>
<tr>
<td>Age at palatoplasty, mean, mo</td>
<td>12.6</td>
<td>14.5</td>
<td>.82</td>
</tr>
<tr>
<td>Type of palatoplasty</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Furlow</td>
<td>0</td>
<td>6 (5.6)</td>
<td>.57</td>
</tr>
<tr>
<td>Vomer</td>
<td>5 (21.7)</td>
<td>16 (15.0)</td>
<td></td>
</tr>
<tr>
<td>von Langenbeck</td>
<td>1 (4.3)</td>
<td>3 (2.8)</td>
<td></td>
</tr>
<tr>
<td>Other</td>
<td>17 (73.9)</td>
<td>82 (76.6)</td>
<td></td>
</tr>
<tr>
<td>Veau class</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>I</td>
<td>0</td>
<td>15 (14.0)</td>
<td>.01</td>
</tr>
<tr>
<td>II</td>
<td>6 (26.1)</td>
<td>44 (41.1)</td>
<td></td>
</tr>
<tr>
<td>III</td>
<td>12 (52.2)</td>
<td>42 (39.3)</td>
<td></td>
</tr>
<tr>
<td>IV</td>
<td>5 (21.7)</td>
<td>6 (5.6)</td>
<td></td>
</tr>
<tr>
<td>Clefts associated with syndromes</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nonsyndromic</td>
<td>21 (91.3)</td>
<td>98 (91.6)</td>
<td>.96</td>
</tr>
<tr>
<td>Syndromic</td>
<td>2 (8.7)</td>
<td>9 (8.4)</td>
<td></td>
</tr>
<tr>
<td>Ear infection</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>16 (69.6)</td>
<td>62 (57.9)</td>
<td>.30</td>
</tr>
<tr>
<td>No</td>
<td>7 (30.4)</td>
<td>45 (42.1)</td>
<td></td>
</tr>
<tr>
<td>Orthodontic treatment</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>4 (17.4)</td>
<td>14 (13.1)</td>
<td>.59</td>
</tr>
<tr>
<td>No</td>
<td>19 (82.6)</td>
<td>93 (86.9)</td>
<td></td>
</tr>
<tr>
<td>Surgeon</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>X</td>
<td>9 (39.1)</td>
<td>61 (57.0)</td>
<td>.15</td>
</tr>
<tr>
<td>Y</td>
<td>9 (39.1)</td>
<td>36 (33.6)</td>
<td></td>
</tr>
<tr>
<td>Others</td>
<td>5 (21.7)</td>
<td>10 (9.3)</td>
<td></td>
</tr>
</tbody>
</table>

* Data are presented as number (percentage) of study participants unless otherwise indicated.
in agreement with the study by Al-Nawas et al,27 who found no significant association among patient age, experience of surgeon, and duration of surgery in early outcomes of palatoplasty. Conversely, Lu et al28 found associations between surgeon, and duration of surgery in early outcomes of palatoplasty. The lack of significance in comparing surgeons may be a result of similar surgical techniques used among surgeons at the institution. Studies that compared differing surgical techniques have produced different results. For example, in a high-volume review, Landheer et al29 found that 2-stage palate closures have higher rates of fistula formation (27%) when compared with a 1-stage repair (14%).

Admittedly, some factors have limited the power of this study, particularly incomplete records and the large number of patients who have had portions of their early treatment at other centers. However, inclusion of any of these patients in our study would have added too many confounding variables and delegitimized our statistically significant findings.

Bresnich et al30 reported that patients with Treacher Collins syndrome were more likely to develop fistulas than non-syndromic individuals. Most studies have seemingly excluded syndromic patients based on biological concerns. For example, the presence of hypotonic palatal musculature in velocardiofacial syndrome can be a confounding factor. In addition, there is some thought that syndromic cleft cases generally necessitate relatively complex care. We did not find this to be the case and found no significant association between patients with syndrome-associated clefts and those with isolated clefts. This finding is in concordance with Stransky et al,31 who found no significant association in the rates of secondary surgery for velopharyngeal insufficiency or postoperative oronasal fistula between patients with and without Pierre Robin sequence.

Individuals with cleft palate have a higher incidence of otitis media with effusion.32-34 Sheahan et al35 found that 76% of patients with cleft lip and palate, 68% of patients with cleft palate only, and 16% of patients with cleft lip had a history of an ear infection or hearing loss. The research of Bluestone et al36-37 implicated an anatomical functional obstruction of the eustachian tube at the nasopharyngeal end, where it meets 2 muscles—the levator palatini and tensor veli palatini. Because of the anatomical communication between the cavity of the middle ear and the muscles of the palate, a history of middle ear disease and subsequent surgical treatment has been postulated as a risk factor for palatal fistulas. This finding led us to research otitis media as an independent variable in palatal fistula formation, though we found no associations.

Conclusions

This study indicates that the risk of developing a persistent postoperative palatal fistula is related to the preoperative Veau class, and each increase in classification level independently increases the risk by a multiple of 2.64. Armed with this information and data, the cleft surgeon can have a reasonable understanding of the postoperative likelihood of palatal fistula in preoperatively assessing a child with a cleft palate.

Study supervision: Maganzini, Marantz.
Conflict of Interest Disclosures: None reported.
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REFERENCES