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Management of Airway Obstruction in Infants With Pierre Robin Sequence

Christopher M. Runyan, MD, PhD*
Armando Uribe-Rivera, DDS†
Shahryar Tork, MD‡
Tasneem A. Shikary, MD§
Zarmina Ehsan, MD¶
K. Nicole Weaver, MD‖
Md Monir Hossain, PhD, MSc**
Christopher B. Gordon, MD††
Brian S. Pan, MD‡‡

Background: Pierre Robin sequence (PRS)-related airway obstruction is often treated surgically; however, objective measures predicting the need for surgery are poorly defined.

Methods: A retrospective chart review was performed on 171 neonates with PRS. Infants were grouped based upon intervention modality: nonsurgical (conservative) or surgical [mandibular distraction osteogenesis (MDO) or tracheostomy]. Demographic data, physical examination findings, and study results were compared between groups to determine risk factors for surgical intervention, and to predict long-term success or failure of those interventions.

Results: The most significant, objective risk factor among those receiving surgery was a poor preintervention sleep study [obstructive index (OI): 42.4 versus 12.9 for the conservative treatment group; $P < 0.001$]. Only 11% of those treated conservatively had an OI >20, whereas 67.5% of those treated surgically met this severity measure. Of those receiving surgery, tracheostomy was associated with neurologic impairment ($P = 0.030$) and low birth weight ($P = 0.046$) compared with the MDO group. Together with syndromic status, these risk factors were useful for predicting failure of MDO to avoid subsequent tracheostomy (test sensitivity and specificity were 64.2% and 100.0%, respectively). No long-term differences in speech or micrognathia were detected between the 3 groups; however, those treated conservatively or with MDO had improved long-term feeding and airway obstruction outcomes compared with the tracheostomy group.

Conclusions: Surgical intervention for PRS-related tongue-based airway obstruction should be strongly considered with an OI >20. Tracheostomy should be reserved for complex patients with concomitant syndromic diagnosis, neurologic impairment, and low birth weight. *(Plast Reconstr Surg Glob Open 2018;6:e1688; doi: 10.1097/GOX.0000000000001688; Published online 10 May 2018.)*

INTRODUCTION

Pierre Robin sequence (PRS) is defined by 3 related findings: micrognathia, glossoptosis, and tongue-based airway obstruction (TBAO).[1] Although not required for the diagnosis,[2] cleft palate is commonly found among those with PRS because the hypoplastic mandible causes superior- and retropositioning of the tongue, which may interfere with palatal shelf fusion. PRS may occur more frequently than previously appreciated,[3,4] with a recent demographic study identifying 1:3,128 with PRS among

*From the *Department of Plastic Surgery, Wake Forest University, Winston-Salem, N.C.; †Department of General Dentistry, Eastman Institute for Oral Health, University of Rochester, Rochester, N.Y.; ‡Department of Plastic Surgery, Ohio State University, Columbus, Ohio; §Otolaryngology, Trihealth, Cincinnati, Ohio; ¶Department of Pulmonary and Sleep Medicine, Children's Mercy Hospital and University of Missouri-Kansas City, Kansas City, Mo.; ‖Division of Genetics, Cincinnati Children's Hospital Medical Center, Cincinnati, Ohio; **Division of Biostatistics and Epidemiology, Cincinnati Children's Hospital Medical Center, Cincinnati, Ohio; ††Department of Plastic Surgery, Dayton Children's Hospital, Dayton, Ohio; and ‡‡Division of Plastic Surgery, Cincinnati Children's Hospital Medical Center, Cincinnati, Ohio.*

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all infants born during 2006 and 2009 within the United States.[5] The presence of a concomitant syndrome or other dysmorphology occurs in 50%–70% of infants with PRS,[5–9] most commonly stickler, velocardiofacial, teratogenic-related, and Treacher Collins syndromes.[6,10]

Treatment modalities vary widely based upon center preference and severity of associated TBAO. In most cases, affected neonates may be treated without surgery, using prone positioning or a palatal obturator to clear the obstructed airway.[11,12] Others report good results bypassing the TBAO using long-term nasopharyngeal tubes or continuous positive airway pressure (CPAP) mask therapy.[13,14] Surgical interventions are generally reserved for those determined to have failed conservative measures, either with severe, persistent TBAO or inadequate oral feeding. The gold standard for correction of severe TBAO is tracheostomy; however, tracheostomy does not correct the underlying anatomical problem and relies upon mandibular growth to adequately clear the tongue base to allow decannulation. Tracheostomy is also associated with significant mortality[15] and increased costs[16,17] compared with mandibular distraction osteogenesis (MDO) for the treatment of PRS-related TBAO. MDO corrects micrognathia by gradually lengthening the mandible and can safely be performed in infancy.[18–21] Tongue–lip adhesion helps to clear TBAO by temporarily connecting the tongue to the lower lip. This helps correct glossoptosis in the short term, but must be reversed as the lower teeth erupt and, like tracheostomy, also relies on mandibular "catch-up" growth.[22,23] Mandibular growth in infants with PRS may be deficient compared with control infants,[24,25] and some initially treated with tongue–lip adhesion or tracheostomy benefit from subsequent MDO.[26]

There are few published and no widely accepted algorithms for deciding which interventions to offer infants with PRS-related TBAO.[18,27] Workup of infants with PRS may include numerous specialties, including neonatology, genetics, pulmonology, otolaryngology, plastic surgery, feeding, and speech/swallow specialists.[28] Monitoring/tests commonly administered include pulse oximetry, polysomnography, nasopharyngoscopy, direct laryngoscopy, computed tomography, cephalometry, and esophageal pH monitoring. While these all may have a role in PRS-related workup, there are no accepted objective measures to define PRS or PRS-related TBAO as severe. The important question remains, "When is surgery indicated for PRS-related airway obstruction?" Other important considerations that follow this question are which surgical intervention to offer when surgery is indicated, and how should successful treatment be defined?

To attempt to answer these questions, we conducted a review of all infants evaluated for PRS-related TBAO at a large, tertiary airway referral center over a 20-year period. To our knowledge, this study represents the largest reported population of infants with PRS, which includes both those treated conservatively (nonsurgically) and with surgery.

METHODS

Under institutional review board approval (#2009-0162), a retrospective chart review was performed for neonates (infants younger than 1 year old) with PRS treated at Cincinnati Children's Hospital Medical Center between 1994 and 2014. Patients were identified from databases maintained within the Genetics and Plastic Surgery divisions at Cincinnati Children's Hospital Medical Center. Patients identified as having PRS were evaluated within the neonatal intensive care unit by a multidisciplinary airway team including neonatologists, geneticists, otolaryngologists, pulmonologists, speech and swallow therapists, and plastic surgeons. The PRS diagnosis was assigned to those with micrognathia on physical examination, glossoptosis on nasopharyngoscopy, and signs of airway obstruction including observed apneic events with oxygen desaturation. Infants with PRS were subsequently evaluated based upon team recommendations; this workup varied but typically included lateral cephalogram or computed tomography, pulse oximetry monitoring, feeding assessment, and a polysomnogram. Treatment modalities included conservative treatment (prone repositioning), tracheostomy, or mandibular distraction. These were selected based upon team recommendation reflecting severity of airway obstruction and concomitant comorbidities.

Demographic data collected included gestational age, gender, age at the time of intervention, syndromic diagnosis, presence of cleft palate, neurologic impairment, and mortality. Prematurity was defined as gestational age less than 37 weeks. All patients were evaluated by a clinical geneticist, and were classified as syndromic or nonsyndromic based on the presence or absence of additional physical examination findings, medical or developmental concerns, or positive genetic testing results. Airway-related data collected included the total apnea–hypopnea index (AHI, which includes central events), obstructive AHI (OI) from pre- and postintervention polysomnograms, the presence of multilevel obstruction from microlaryngoscopic/bronchoscopic assessment, and whether there was a long-term requirement for supplemental oxygen, continuous positive airway pressure (CPAP), or bilevel-PAP. OI was calculated by dividing the total number of obstructive apneas and hypopneas by the total sleep time. Nutrition-related data collected included birth weight (with associated Z scores), weight at 12 months, weight at most recent examination, diagnosis of gastroesophageal reflux (GER), need for supplemental feeding tube (nasogastric, gastrostomy, or jejunostomy), and the age at exclusive mouth feeding. Need for subsequent airway intervention, including repeat mandibular distraction or tracheostomy, was recorded, as was the age of tracheostomy decannulation. Long-term data collected also included the presence of velopharyngeal insufficiency, persistent/recurrent micrognathia, or operative speech intervention.

Demographic and clinical characteristics of infants with PRS were compared between treatment groups. For continuous variables, mean and SD are reported. Either 1-way analysis of variance or the nonparametric Kruskal–Wallis test was used depending on the validity of normality assumption for comparing the means or medians among the groups. For categorical variables, frequencies and percentages were reported. Chi-square or Fisher exact test was used to determine if there was any difference in the

Table 1. Demographics and Clinical Findings of Infants With Pierre Robin Sequence-related Airway Obstruction Treated Conservatively or With Surgical Intervention

	Conservative (Nonsurgical)	Surgical (MDO/Tracheostomy)	*P*
Total no. patients treated	49	122	
Gender (female)	59.2%	49.2%	0.236
Age at intervention (d)	93.2±172.6	43.5±57.4[3]	0.009*
Age at the time of study (y)	7.5±1.5	6.6±0.6	0.181
Premature (<37 wk)	19.1%	25.4%[4]	0.404
Cleft palate	77.6%	68.9%	0.255
Birth weight (kg)	3.1±0.7[4]	2.8±0.7[4]	0.046*
Birth weight Z score	−0.4±1.0	−0.4±1.2	0.959
Change in weight Z score from birth to 1 y	−0.4±1.4	−0.3±1.5	0.677
Syndromic diagnosis	51.0%	54.1%	0.715
Neurologic impairment	26.2%[4]	35.2%[4]	0.291
Gastroesophageal reflux	44.9%	42.6%	0.786
Surgical (G- or J-) feeding tube requirement	23.7%[4]	46.6%[4]	<0.001*
Now able to feed exclusively by mouth	81.1%[4]	70.6%[4]	0.215
Age at feeding exclusively by mouth (d)	53.2±154.3	326.2±479.7	0.003*
Multilevel airway obstruction	50.0%[2]	36.1%[4]	0.283
Preintervention obstructive index (mean)	12.9±9.9[2]	42.4±29.8[3]	<0.001*
Postintervention obstructive index (mean)	3.1±4.4[2]	6.8±9.1[2]	0.090
Change in obstructive index (mean)	−9.7±8.1[2]	−35.4±27.4[2]	<0.001*

All data were available for each patient with the exception of those marked, for whom only the following percentages of patients had that data point available: [1]<25%, [2]2%–49%, [3]50%–75%, [4]>75%.
*Statistically significant for the error rate 5%.

distribution between treatment groups for discrete variables. In the 3 treatment groups (conservative, tracheostomy, and MDO) comparison, pairwise comparisons were performed and Tukey–Kramer multiplicity adjustments were applied for multiple comparisons. A multivariable regression model was used in examining the effects of syndromic diagnosis, neurologic impairment, low birth weight (≤2.5 kg), and the presence of cleft palate on the success of tracheostomy avoidance for those receiving primary MDO. The odds ratio estimates with 95% confidence intervals (CIs) and the values for sensitivity and specificity from the logistic model are reported. In all analyses, the statistical significance was set a priori at $\alpha = 0.05$. All analyses were performed using SAS software, version 9.3 (SAS Institute, Cary, NC).

RESULTS

When Is Surgery Indicated for PRS-related Airway Obstruction?

To investigate this question, demographics and clinical findings were compared between those patients treated conservatively and those receiving surgery (MDO and/or tracheostomy). A high percentage (71.3%) of patients were treated with surgery (**Table 1**). These patients presented to the hospital at an earlier age and had a lower birth weight, which was not significant when adjusting for gestational age (Z score). No differences were observed between the groups for other demographic variables, including gender, prematurity status, the presence of a cleft palate, syndromic status, neurologic impairment, GER, or incidence of multilevel obstruction. However, patients treated with surgery had a higher incidence of requiring a surgical feeding tube, a significant delay in the time required to feed exclusively by mouth, and an objectively worse sleep study. Although no differences in mean oxygen nadir or mean end tidal carbon dioxide were observed (not shown), patients receiving surgery had significantly higher obstructive AHIs (OI) compared to those treated conservatively (OI: 42.4±29.8 versus 12.9±9.9). Similar differences were observed in the total AHI, which includes central events (not shown).

We then asked whether the degree of polysomnogram obstructive severity may be used to distinguish those treated conservatively or surgically. We found that the greatest discrepancy between the 2 groups occurred with an OI >20 (Fig. 1). Over two thirds of those treated surgically (67.5%) had an OI >20, compared to only 11.5% of those treated conservatively.

When Surgery Is Performed, Which One Should Be Used?

Patients with surgically treated PRS-related airway obstruction were compared based upon surgical treatment modalities, tracheostomy versus MDO (**Table 2**). Several factors were significantly increased among those receiving tracheostomy, including syndromic status, neurologic impairment, GER, and surgical feeding intervention. Nine of the neonates receiving tracheostomy (16.7%) were treated via ex utero intrapartum procedure, which contributed to the significant increase of early (<5 days of life) intervention in this group compared to MDO.

To identify predictors for failure of MDO, failure was first defined as the requirement of secondary tracheostomy following MDO. A comparison was then made between those with failed or successful outcomes following MDO [**see Table**, **Supplemental Digital Content 2**, which displays demographic and clinical findings of infants with PRS-related airway obstruction treated with mandibular distraction, based upon treatment outcome (success defined as tracheostomy avoidance), ***http://links.lww.com/PRSGO/A710***]. Of 68 patients in the MDO group, 6 required subsequent tracheostomy (8.8%). Risk factors for MDO failure were limited to syndromic status and neu-

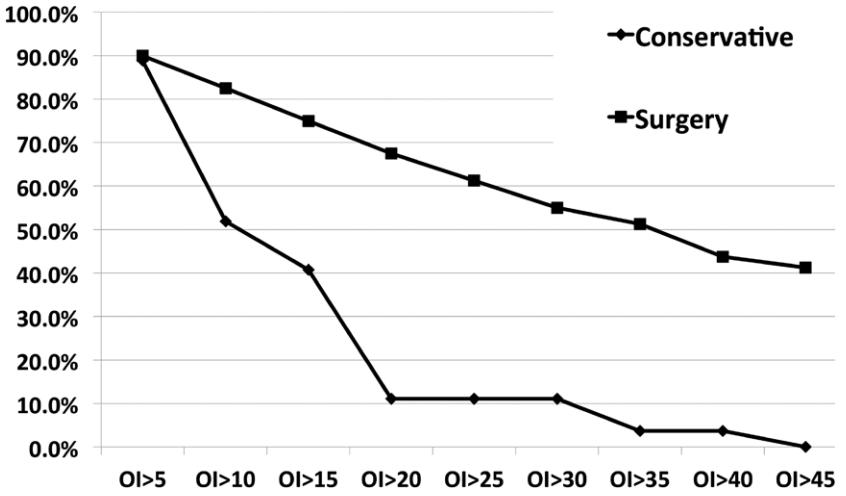

Fig. 1. Percentage of sleep studies with an OI > listed value, based upon treatment modality.

Table 2. Demographic and Clinical Findings of Infants With Pierre Robin Sequence-related Airway Obstruction Treated Surgically

	Tracheostomy	Mandibular Distraction	*P*
Total no. patients treated	54	68	
Gender (female)	53.7%	45.6%	0.373
Age at intervention (d)	37.3±54.1	48.3±59.9	0.295
Age at the time of study (y)	8.0±1.0	5.4±0.7	<0.001*
Premature (<37 wk)	29.6%	22.1%	0.340
Early intervention (<5 d of life)	25.9%	0.0%	<0.001*
Cleft palate	64.8%	72.1%	0.391
Birth weight (kg)	2.6±0.7[3]	2.9±0.7[4]	0.084
Birth weight Z score	−0.7±1.5	−0.3±1.1	0.250
Change in weight Z score from birth to 1 y	−0.4±1.8	−0.3±1.4	0.807
Syndromic diagnosis	66.7%	44.1%	0.013*
Neurologic impairment	48.8%[4]	25.8%[4]	0.015*
Gastroesophageal reflux	55.6%	32.4%	0.010*
Surgical (G or J) feeding tube requirement	80%[4]	22.1%	<0.001*
Now able to feed exclusively by mouth	47.8%[4]	87.3%[4]	<0.001*
Age at feeding exclusively by mouth (d)	763.2±677.3[2]	167.3±239.8[4]	<0.001*
Multilevel airway obstruction	44.2%[4]	29.9%[4]	0.105
Preintervention obstructive index (mean)	39.2±25.4[2]	43.3±31.1[4]	0.633
Postintervention obstructive index (mean)	6.0±9.1[1]	7.0±9.2[2]	0.742
Change in obstructive index (mean)	−49.5±1.7[1]	−33.9±28.4[2]	0.359

All data were available for each patient with the exception of those marked, for whom only the following percentages of patients had that data point available: [1]<25%, [2]25%–49%, [3]50%–75%, [4]>75%.
*Statistically significant for the error rate 5%.
G, gastrostomy; J, jejunostomy.

rologic impairment, with a trend toward significance for low birth weight. Not surprisingly, those that failed also had significantly worse post-MDO obstructive indices on polysomnography. To assess these factors for their value in generating a test for predicting MDO failure, a multivariable logistic regression analysis was performed examining syndromic diagnosis, neurologic impairment, low birth weight (≤2.5 kg), and the presence of cleft palate (due to a possible interaction between syndromic diagnosis and absence of cleft palate). When considering intervariable interactions, syndromic status was no longer predictive of MDO failure, whereas low birth weight and neurologic impairment were [**see Table**, **Supplemental Digital Content 3**, which displays odds ratio estimate with 95% CI from logistic regression model where success is tracheostomy avoidance, ***http://links.lww.com/PRSGO/A711***]. Using these, a pretest was created to assess the ability to predict failure among those treated with MDO and observed that the model predicted area under the receiver operating characteristic (ROC) curve was 0.90 (95% CI, 0.78–1.00) [**see Fig.**, **Supplemental Digital Content 1**, which displays ROC curve from the logistic regression model for the success of tracheostomy avoidance, ***http://links.lww.com/PRSGO/A709***]. The values for pretest sensitivity and specificity were 77.6% and 60.0%, respectively, when including the neurologic impairment and low birth weight variables. Although syndromic status was not independently a significant predictor of MDO failure, when included in the logistic regression model together with neurologic impairment and low birth weight, the values for test sensitivity

and specificity are 64.2% and 100.0%, respectively; this model predicted that area under the ROC curve was 0.93 (95% CI, 0.84–1.00).

Of 54 patients treated primarily with tracheostomy, 33 received secondary MDO both to correct micrognathia and to help achieve decannulation. Of these, 57.5% were successfully decannulated at the time of our study. We then identified risk factors for failure to be decannulated following MDO, which included low birth weight, syndromic status, and absence of cleft palate [**see Table**, **Supplemental Digital Content 4**, which displays demographic and clinical findings of infants with PRS-related airway obstruction treated with tracheostomy and subsequent mandibular distraction, based upon treatment outcome (success defined as tracheostomy decannulation), ***http://links.lww.com/PRSGO/A712***].

How Should Successful Treatment Be Defined?

The 3 treatment groups (conservative, tracheostomy, and MDO) were then compared with each other individually to assess long-term feeding and growth, speech, and airway outcomes (**Table 3**). With regard to feeding, patients treated conservatively or with MDO both had a significantly lower requirement for surgical feeding assistance, and earlier onset of feeding exclusively by mouth compared with the tracheostomy group. Patients receiving tracheostomy had a significantly lower birth weight than those in the conservative group; however, by 1 year of life, there were no differences in Z scores between all 3 groups.

To assess speech outcomes, patients with cleft palate underwent formal speech evaluations once they reached 3 years of age. All patients had previously undergone palatoplasty usually between 10 and 18 months of age. The incidence of velopharyngeal incompetence was not significantly different between the 3 treatment groups, nor were there differences in the incidence of requiring subsequent speech surgery.

To assess long-term airway obstruction, the most recent clinical notes available for all patients were assessed for recommendation of CPAP ventilation, continued tracheostomy, or oxygen supplementation. Excluding patients developing obstructive sleep apnea secondary to speech surgery, we found that patients treated conservatively or with MDO had similar rates of long-term airway obstruction (near 16%), which was significantly lower compared with those receiving tracheostomy.

DISCUSSION

The determination of when to treat PRS-related TBAO conservatively versus surgically is poorly defined due to the wide range of severity, the low incidence of this patient population, and the high number of clinical variables that

Table 3. Demographic and Clinical Findings of Infants With Pierre Robin Sequence-related Airway Obstruction Treated Conservatively, With Tracheostomy or With Mandibular Distraction, Based on Feeding, and Long-term Speech and Airway Outcomes

	Conservative	Tracheostomy	MDO	*P*
Total no. patients treated	49	54	68	
Age at study (y)	7.5±1.5	8.0±1.0	5.4±0.7	0.611 (C versus T) 0.008* (C versus M) <0.001* (T versus M)
Birth weight (kg)	3.1±0.7[3]	2.6±0.7[3]	2.9±0.7[4]	0.026* (C versus T) 0.493 (C versus M) 0.245 (T versus M)
Birth weight Z score	−0.4±1.0	−0.7±1.5	−0.3±1.1	1.000 (C versus T) 1.000 (C versus M) 0.554 (T versus M)
Change in weight Z score from birth to 1 y	−0.2±1.7[2]	0.6±1.8[2]	0.5±1.7[3]	0.387 (C versus T) 0.592 (C versus M) 1.000 (T versus M)
G-tube/J-tube requirement	23.7%[4]	80%[4]	22.1%	0.843 (C versus T) 0.505 (C versus M) 0.033* (T versus M)
Age at feeding exclusively by mouth (d)	53.2±154.3[3]	763.2±677.3[2]	167.3±239.8[4]	<0.001* (C versus T) 0.462 (C versus M) <0.001* (T versus M)
Velopharyngeal insufficiency	60.7%[3]	44.0%[2]	47.1%[3]	0.659 (C versus T) 0.839 (C versus M) 1.000 (T versus M)
Received speech surgery	53.6%[3]	36.0%[2]	27.3%[2]	0.532 (C versus T) 0.092 (C versus M) 1.000 (T versus M)
Persistent or recurrent micrognathia	16.3%[4]	18.9%[3]	15.8%[4]	1.000 (C versus T) 1.000 (C versus M) 1.000 (T versus M)
Obstructive sleep apnea requiring home oxygen, CPAP or, tracheostomy	15.6%[4]	44.4%[3]	15.8%[4]	0.001* (C versus T) 0.977 (C versus M) <0.001 * (T versus M)

All data were available for each patient with the exception of those marked, for whom only the following percentages of patients had that data point available: [1]<25%, [2]25%–49%, [3]50%–75%, [4]>75%.

*Statistically significant for the error rate 5%.

C, conservative; M, mandibular distraction osteogenesis; T, tracheostomy.

must be factored into the decision of when to intervene. In their decision tree model, Bradley assigns those with "mild" obstruction to prone positioning and home monitoring; however, the criteria separating mild from "moderate/severe" obstruction are not defined.[18,29] Another review recommended that those treated conservatively should include those without reflux, central apnea, or neurologic comorbidities, with an AHI 0–5, normal endoscopic examination, and with positive response to prone positioning.[30] While these criteria seem reasonable, if applied together, they would divert most patients with PRS toward surgery (indeed over 60% of our conservative cohort did not meet these criteria). Handley et al[9] compared micrognathic infants treated surgically or conservatively and identified 3 independent risk factors for needing surgery: the absence of cleft palate, neurologic impairment, and the need for intervention within the first day of life. We found that the majority of cited factors, including the presence or absence of cleft palate, neurologic impairment, presence of multilevel obstruction, syndromic status, and GER, did not predict the need for surgery in our cohort. This is not because these factors are absent among those receiving surgery, but because many of those treated conservatively had these risk factors present.

Two important distinguishing criteria identified between those treated conservatively and surgically are the need for a surgical feeding tube and a poor PSG. The ability to feed is an indirect measure of severity of airway obstruction. Infants cannot be easily fed in the prone position, so those that cannot safely or adequately swallow when supine require tube feeding assistance.[30] Esophageal dysmotility and increased rates of GER are common features of those with PRS, due in part to altered intrathoracic pressures due to airway obstruction.[31,32] Consistent with others,[14,31] we observed a high incidence of feeding tube assistance; 50% of our conservative group and 100% of those receiving surgery required temporary nasogastric (NG) or surgical feeding tubes. Surgical (gastrostomy or jejunostomy) feeding tubes were generally reserved for those who are subjectively determined to require long-term support. The incidence of surgical feeding tube is roughly doubled in those receiving surgery. Interestingly, the ability to feed exclusively by mouth occurred at much greater frequency in the MDO compared with the tracheostomy group, likely reflecting improved feeding potential with correction of micrognathia and glossoptosis.

A poor polysomnogram is identified as a distinguishing, objective measure among those treated surgically in our study. We selected the obstructive AHI (OI) as our primary outcome measure to maintain focus on anatomic causes of obstruction. Those with a severe central component typically receive tracheostomy. PSG interpretation is not standardized for neonates, and there is no accepted scale of obstructive severity.[33,34] A severe obstructive index in neonates has been interpreted as >6,[30] >10,[14] and >24.[12] Our study is not designed to help stratify obstructive severity; however, we observed a significant threshold between the conservative and surgical groups at an OI >20. Combined with other examination findings, this threshold may help guide surgical decision making; surgery should be considered at an OI >20. At an OI <20, one should rely more heavily upon the presence of multiple concomitant comorbidities including persistent feeding difficulties, syndromic status, and neurologic impairment.

When surgery has been deemed necessary, the specific modality must be considered. In our cohort, high-risk patients more commonly received tracheostomy, including those with syndromic status, neurologic impairment, low birth weight, GER, and need for surgical feeding intervention. However, the mean age at the time of the study was significantly older in the tracheostomy group compared to MDO group given the later implementation of the latter surgical modality in our institution. As reported by others,[21,35,36] we found MDO highly effective for correction of TBAO (91% in our study). As we experienced success utilizing MDO, we began offering this option as the first-line surgical modality. Tracheostomy bypasses but does not correct the tongue-based anatomic site of obstruction, and over half of those treated primarily with tracheostomy received secondary MDO. Given the mortality risk associated with tracheostomy,[15] and its comparatively poor feeding and airway outcomes in our study, we recommend considering MDO as the first-line therapy for those with severe TBAO due to PRS. Failure of MDO to avoid tracheostomy can be predicted with excellent specificity using the combined high-risk factors identified in this study (syndromic status, neurologic impairment, and low birth weight) or those reported by Flores et al.[8]

This study has a number of limitations. PRS is a relatively uncommon condition making it difficult to collect large number of patients at a single center. A high percentage (71.3%) of those in our study received surgery, reflecting our referral base, which is skewed toward severe airway disease, as well as a potential selection bias toward surgery in this cohort given assembly of the database in large part from surgical services' records. This may limit the general applicability of our findings to PRS patients in other centers, particularly those that have a more normal distribution of TBAO severity, and those offering other interventions such as long-term nasopharyngeal airways, palatal obturators, and tongue–lip adhesion. We also were limited by incomplete data sets for all patients, with more complete records available on younger patients (more often treated with MDO). Follow-up of older patients was likely biased toward those with ongoing cleft-related concomitant problems, for example, velopharyngeal insufficiency (VPI). This likely accounts for the trend toward higher VPI and speech surgery in those within the conservative treatment group.

In conclusion, our series supports the use of MDO as an effective, first-line surgical intervention in the treatment of neonates with PRS-related TBAO. By directly addressing the anatomic source of airway obstruction, the resultant physiologic consequences of micrognathia may largely be avoided, even in patients possessing comorbidities independently deemed to be contraindications to MDO. This is evidenced by the similarities between the clinical characteristics of the conservatively managed and surgically managed cohorts. Not surprisingly, OIs >20 coupled with the need for nutritional support were found to be strong predictors for determining surgical interven-

tion; however, when combined, the presence of low birth weight, neurologic impairment, and syndromic status are highly predictive of failure of MDO and conversion to tracheostomy. Considering its effectiveness, the long-term cost savings compared to tracheostomy, and the improved potential to feed independently following surgery, surgical intervention with MDO should be strongly considered when conservative measures have failed.

Christopher M. Runyan, MD, PhD
Department of Plastic Surgery
Wake Forest Baptist Medical Center
Medical Center Boulevard
Winston-Salem, NC 27157
E-mail: crunyan@wakehealth.edu

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