Comparative Effectiveness of Preepiglottic Baton Plates and Mandibular Distraction in Infants with Robin Sequence

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Background: Mandibular distraction osteogenesis (MDO) and preepiglottic baton plates (PEBP) are both effective for early management of upper-airway obstruction (UAO) in infants with Robin sequence (RS), but have not been directly compared. The purpose of this study was to compare early airway, feeding, and growth outcomes between these treatments.

Methods: This is a bicentric retrospective cohort study from 2015 through 2021 including infants with RS treated with MDO or PEBP before 6 months of age with pretreatment and posttreatment sleep studies and follow-up at least through age 1 year. The primary outcome was immediate postintervention UAO, measured as obstructive apnea-hypopnea index or obstructive apnea index, as available. Latest follow-up sleep studies, feeding, and growth characteristics were also assessed.

Results: A total of 114 participants were included (MDO, $n = 31$; PEBP, $n = 83$). Pretreatment UAO was similar between groups ($P = 0.61$). PEBP was initiated at a younger age (median (interquartile range) 31 (14, 53) versus 41 (28, 84) days of life; $P < 0.05$). Significant reduction in obstructive apnea-hypopnea index or obstructive apnea index was achieved in both groups, greater with MDO (98%) compared with PEBP (94%) ($P < 0.05$). PEBP demonstrated better early feeding and growth outcomes compared with MDO, with fewer surgical feeding tubes needed ($P < 0.001$), and more rapid early growth ($P = 0.038$). When stratified by preintervention UAO, infants starting with moderate UAO experienced similar airway outcomes with both treatments ($P = 0.11$), whereas those with severe UAO had greater resolution with MDO ($P < 0.001$).

Conclusion: Both treatments effectively relieved moderate UAO in infants with RS, but MDO was more effective for infants with severe respiratory compromise. *(Plast. Reconstr. Surg.* 156: 267e, 2025.)

CLINICAL QUESTION/LEVEL OF EVIDENCE: Therapeutic, III.

THERAPEUTIC

Mandibular distraction osteogenesis (MDO) is commonly used in North America to treat upper-airway obstruction (UAO) in infants with Robin sequence (RS).[1,2] MDO ameliorates UAO in more than 94% of infants with nonsyndromic RS.[3–6] MDO is also associated with improved oral feeding,[7–10] reduced need for surgical feeding tubes,[11,12] better weight gain,[13] improved laryngoscopy view, ease of intubation,[14] and improved health-related quality of life.[15,16] Despite these benefits, MDO imparts considerable treatment burden. Two operations are required (distractor placement and removal), prolonged hospitalization and intensive care

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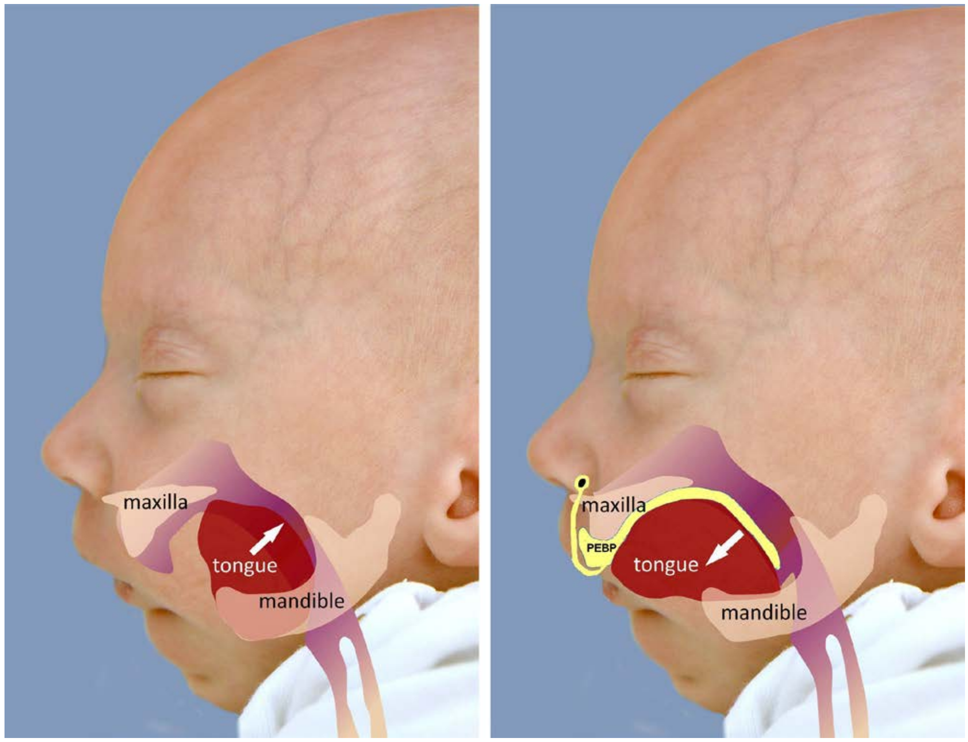

Fig. 1. The PEBP effect.

needs are common,[17] minor complications are frequent,[18,19] and implications to dental development and growth remain unclear.[20–24]

An expanded repertoire of nonoperative treatments is used in select European centers, where MDO is less commonly performed.[25] An oral appliance with a preepiglottic baton to mitigate tongue-base prolapse into the oropharynx (termed the preepiglottic baton plate [PEBP], Tübingen palatal plate, or orthodontic airway plate) is the first-line treatment in Tübingen, Germany.[26] Similar appliances are used in other German institutions,[27] and, recently, in the United States.[28,29]

The PEBP is a patient-specific device designed from an alginate or digital impression of the palate and maxillary alveolus with a velar extension that promotes a nonobstructing forward position of the tongue base (Fig. 1). It consists of a palatal plate that covers the alveolar ridges and hard palate and that obturates the cleft, when present, and a customized velar extension (approximately 3 cm) that is dorsally attached to the plate and terminates just above the epiglottis. First, a prototype is fabricated and evaluated in situ by unsedated fiberoptic nasopharyngoscopy, during which the position, length, and angle of the velar extension are adjusted so that it sufficiently pushes the tongue base forward to erect the epiglottis and widen the pharyngeal airway space. When the protoype is deemed satisfactory, a final device is created based on the protoype characteristics and with a strengthening wire incorporated into the velar extension to safeguard against mechanical failure (Fig. 2). In recent years, physical oral impressions have been replaced by digital scanning (Trios 3 Intraoral Scanner; 3Shape) and 3-dimensional (3D) printing has been used (3Shape Ortho Appliance Designer) to produce the PEBP, which is less traumatic to the infant and less costly.[30,31] The PEBP is retained by mucosal contact with the palate and alveolar ridges, enhanced by fixative cream (Corega Super-Haftcreme; Procter &

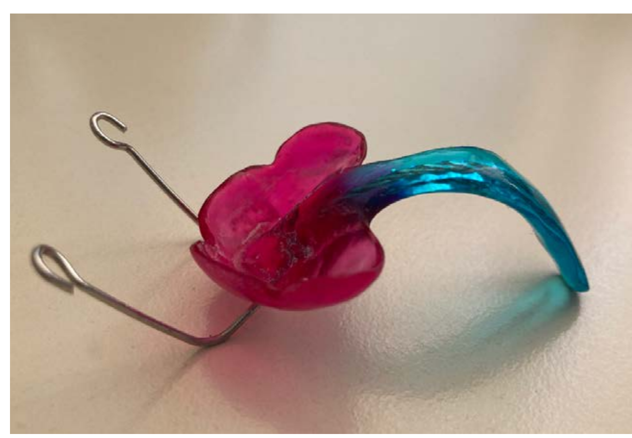

Fig. 2. The PEBP.

Element	0 (Normal)	1 (Minor)	2 (Moderate)	3 (Severe)
Micrognathia	Normal (overjet <3mm)	Mild (overjet 3-<6mm)	Moderate (overjet 6- <10mm)	Severe (overjet ≥10mm)
Nutrition	Full PO, meeting caloric and weight goals	Full PO, not fully meeting caloric and/or weight goals	Feeds partially PO and partially enteric	Full enteric feeds
Airway	oAHI < 5/hr AND SpO2 nadir >85%	oAHI 5 – 10/hr OR SpO2 nadir <85%	oAHI 11 – 20/hr	oAHI >20/hr OR $P_{ET}CO_2$ > 50 torr for >50% TST OR Tracheostomy OR Intubated (may be downgraded after extubation)
Palate	Intact/Submucous	Veau I cleft	Veau II cleft	Veau ≥ III cleft
Syndrome/ Comorbidities	No impactful syndrome or comorbidities	MINOR impact to neonatal management	MODERATE impact to neonatal management	SEVERE impact to neonatal management

Stage	Micro	N	A	P	S
TBAO	0	1-3	1-3	Any	0-3
R0	1-3	Any	0	Any	0-1
R1	1-3	1	1	Any	0-2
R2	1-3	2	2	Any	0-2
R3	1-3	3	3	Any	0-2
R4	1-3	1-3	1-3	Any	3

Fig. 3. The MicroNAPS grading system for RS. OAHI, obstructive apnea-hypopnea index; TST, total sleep time.

Gamble), and secured by extraoral wire bows to the forehead skin using adhesive tape (Steri-Strip and Cavilon No Sting Barrier Film or Steri-Strip Compound Benzoin Tincture; 3M Health Care). The plate is briefly removed once daily for cleaning and to inspect the oral mucosa for pressure marks or decubitus, which are relieved by abrasion of the offending areas when present. Oral feeding is initiated before PEBP insertion and continues uninterrupted with the plate. PEBP effectiveness is confirmed by a repeat sleep study before hospital discharge. The PEBP is worn continuously for approximately 6 months[26] while the infant is at home, where daily maintenance is managed by the family. PEBP use is typically discontinued before cleft repair.

The literature suggests that PEBP is effective in alleviating UAO[32–36] and improving oral feeding by obturating the cleft palate, if present. Additional benefits, including promotion of mandibular growth and improved weight gain, have also been reported.[33,35] These studies suggest that, in patients with adequate mandibular catch-up growth, a childhood operation for UAO may be avoided by using a PEBP.[34,37] Despite promising outcomes, this technique is rarely used outside of select centers. Existing literature suffers from selection bias, lack of a control population, and use of outcome measures not comparable with those reported for MDO.[38] The 2 techniques have never been directly compared.

The purpose of this study was to compare airway, feeding, and growth outcomes in similar series of infants treated with MDO or PEBP over the first year of life. We hypothesized that airway and growth outcomes would be similar, but that oral feeding would be achieved more quickly in the PEBP group due to lack of operative recovery and the benefit of cleft obturation.

PATIENTS AND METHODS

Study Design

This bicentric retrospective cohort study included infants with RS treated with MDO (Boston) or PEBP (Tübingen) from January of 2015 to December of 2021. To be included, patients had to have a clinical diagnosis of micrognathia; to have been treated by MDO or PEBP in their first 6 months of life; to have completed both preintervention and postintervention sleep studies; to have a MicroNAPS[39] grade of R2 or R3 (Fig. 3) and airway score of A_2 or A_3; and to have been followed up at least until their first birthday.

Infants with previous surgical procedures for airway management (eg, tongue–lip adhesion, tracheostomy) or insufficient records were excluded. Patients with MicroNAPS[39,40] grades of stage R1 or R4 or with starting airway scores other than A_2 or A_3 were also excluded. This stratification allowed inclusion of infants with syndromes and comorbidities with minor to moderate impact on neonatal airway management (eg, Stickler syndrome), while excluding those with major impact (eg, Treacher Collins syndrome). This deviates from the more traditional stratification of isolated versus syndromic, which artificially narrows the sample.

Participants were grouped into MDO and PEBP cohorts. Pretreatment management occurred according to standardized clinical pathways for each institution (Fig. 4). The processes for MDO and PEBP are shown in Figures 5 and 6, respectively. This study was approved by the institutional review boards of both participating institutions.

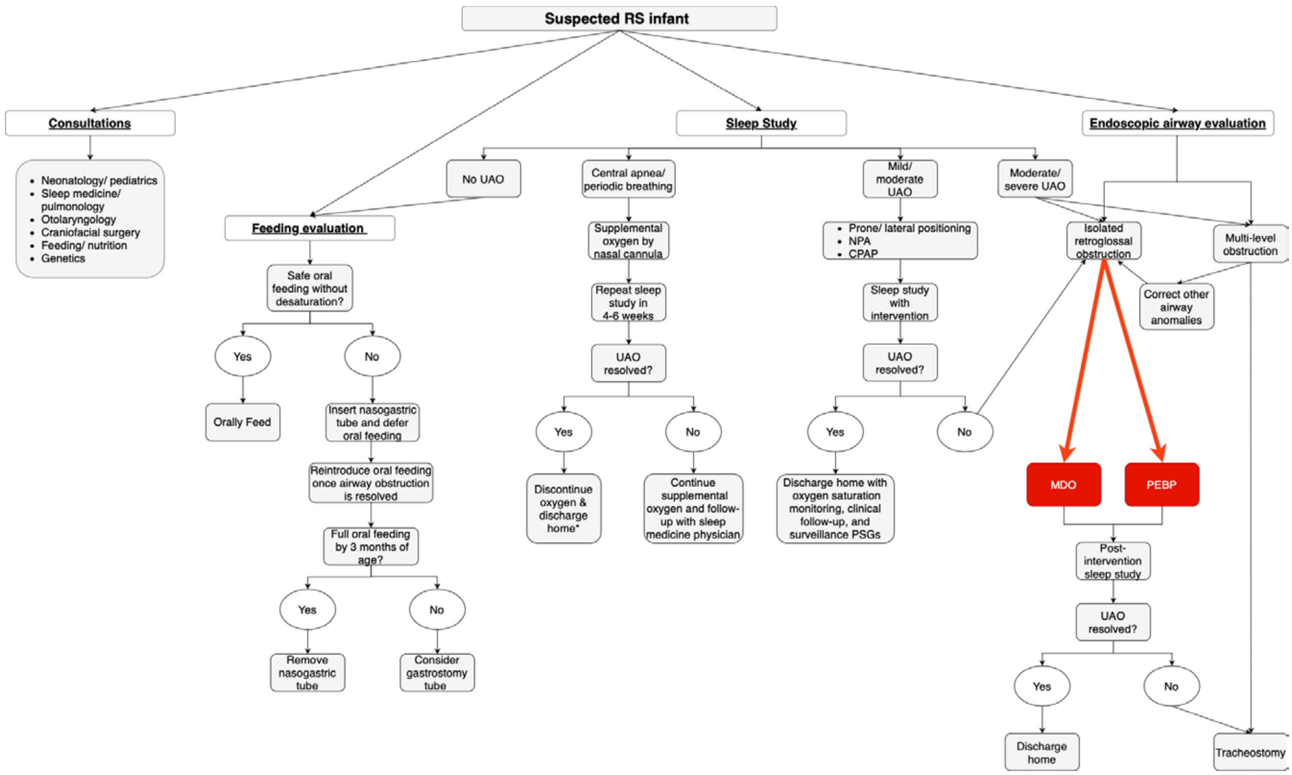

Fig. 4. Clinical pathway for workup and management of infants with suspected RS. The presented pathway represents an amalgamation of the pathways used at each participating institution. CPAP, continuous positive airway pressure; NPA, nasopharyngeal airway; PSG, polysomnography.

Neonatal MDO Protocol

- **Internal device with activation arm posterior**
- **Horizontal vector**
- **2 mm intraoperative distraction**
- **No latency**
- **2 mm distraction per day, split into 2 sessions**
- **Overcorrect 2-4 mm**
- **Intubation for 3-4 post-operative days**
- **Oral feeding as early as possible**
- **Polysomnogram at end of MDO**
- **Remove turning arms at bedside when MDO completed**
- **Consolidation for 6-8 weeks**

Fig. 5. Process for MDO.

Variables

The primary predictor variable was treatment type (MDO or PEBP). Secondary predictors included sex; race; gestational age at birth; birthweight; age at intervention; preintervention severity of UAO; MicroNAPS scores for nutrition, palate, and syndrome/comorbidities[39]; occurrence of and age at palatoplasty; comorbidities; use of airway adjuncts (eg, supplemental oxygen, nasopharyngeal tubes, continuous positive airway pressure [CPAP]); need for intubation; use of nasogastric tube (NGT) or surgical feeding tube; weight-for-age *z* scores at birth and immediately preintervention; and length of stay. The time point "intervention" was defined as initiation of MDO (osteotomies and placement of devices) or insertion of the definitive PEBP.

Sleep studies in the MDO group were performed using comprehensive polysomnography (PSG).[41] In the PEBP group, sleep studies were performed using limited respiratory polygraphy in a sleep laboratory.[42] Collected PSG or respiratory polygraphy parameters included obstructive apnea-hypopnea index (OAHI) from PSG, obstructive apnea index (OAI) from respiratory polygraphy, and oxygen saturation (Spo_2) nadir. All studies were scored using the same standardized criteria.[43] Only supine data were analyzed.

Normalization of OAHI and OAI values was achieved using the conversion factor established by Lim et al.[42] UAO was defined as OAHI of 5 or more events/hour, OAI of 1 or more events/hour, or Spo_2 nadir less than 85%.[39] UAO was further categorized as moderate (A_2 = OAHI 11 to 20 events/hour or OAI 5 to 10 events/hour)

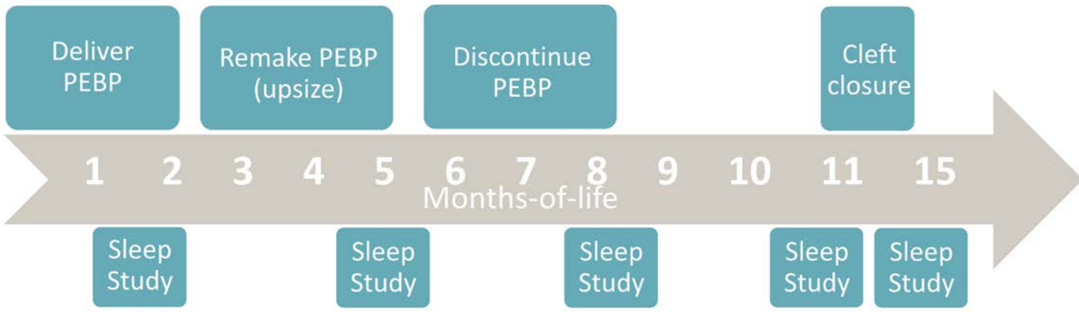

Fig. 6. Process for PEBP.

or severe (A_3 = OAHI more than 20 events/hour or OAI more than 10 events/hour). Participants with minor UAO (A_1, stage R1[39]) were excluded, as these infants are not typically considered for either intervention.

To validate the relationship between OAHI and OAI within this sample, an age- and sex-matched subgroup from the PEBP cohort underwent prospective reevaluation of their original respiratory polygraph results by a sleep medicine physician blinded to the original results to allow calculation of OAHIs. This was done by rescoring obstructive apneas and obstructive hypopneas per American Academy of Sleep Medicine guidelines,[43] but without EEG arousals, because EEG results were not available.

The primary outcome variable was immediate postintervention residual UAO, defined as none (A_0 = OAHI less than 5 events/hour or OAI less than 1 event/hour and Spo_2 nadir greater than 85%) or minor (A_1 = OAHI 5 to 10 events/hour or OAI 1 to 5 events/hour or Spo_2 nadir less than 85%).[39] However, threshold values for UAO remain debated, and there is a trend toward accepting higher OAHIs as normal based on recently published normative infant data,[44–46] and European centers typically use a target of OAHI less than 10 or OAI less than 3 events/hour as "success" with treatment.[47] As such, a secondary analysis was performed using this more liberal threshold.

Secondary outcome variables included (1) change in OAHI/OAI from before to after intervention; (2) residual UAO at latest follow-up; (3) feeding at discharge (eg, use of nasogastric or surgical feeding tubes, time to recommencing oral feeding after intervention); and (4) growth velocity (weight-for-age *z* scores immediately after intervention; at ages 3, 6, 9, and 12 months; and at latest follow-up, calculated using World Health Organization Child Growth Standards[48]).

Data Analysis

Distributions of continuous data were evaluated by Kolmogorov-Smirnov tests, box plots, and histograms. Normally distributed data were analyzed by unpaired *t* tests and are presented as mean ± SD. Data with skewed distributions were assessed by Kruskal-Wallis rank sum tests and presented as median (interquartile range) to diminish the impact of outliers on the data. Categorical data were analyzed using chi-square tests, except when frequency was less than 5, in which case Fisher exact tests were used. An adjusted median regression model was created for the continuous variable pre to post ΔOAHI/OAI, including predictors with clinical or statistical significance. A *P* value less than 0.05 was considered statistically significant. Analyses were performed with R statistical software (version 4.2.2) and RStudio interface (version 2022.12).

RESULTS

Sample

During the study period, 59 infants underwent MDO and 111 had PEBP at the corresponding centers. Some patients were excluded because of previous operation (MDO, *n* = 5; PEBP, *n* = 2), inadequate follow-up (MDO, *n* = 6), missing PSG/respiratory polygraphy data (MDO, *n* = 2; PEBP, *n* = 7), or excluded MicroNAPS scores (MDO, *n* = 15; PEBP, *n* = 19). The final sample included 31 patients in the MDO cohort and 83 in the PEBP group. Dropout analysis revealed no significant differences between included and excluded patients. (**See Table, Supplemental Digital Content 1**, which shows dropout analysis of included versus excluded patients in the MDO group. Results presented as number [%], median [IQR], *http://links.lww.com/PRS/H853*. **See Table, Supplemental Digital Content 2**, which shows dropout analysis of included versus excluded

Table 1. Preintervention Sample Characteristics[a]

Variables	MDO	PEBP	*P*
No.	31	83	
Female	15 (48.4)	42 (50.6)	1.00
Age at intervention, days	41.00 (28.00, 84.00)	31.00 (13.50, 52.50)	0.01[b]
Race			<0.001[b]
White	22 (71.0)	83 (100.0)	
Black/African American	4 (12.9)	0 (0.0)	
Other	1 (3.2)	0 (0.0)	
Unknown	4 (12.9)	0 (0.0)	
Gestational age at birth, wk	38.93 (36.00, 39.57)	39.14 (38.14, 40.14)	0.15
Birthweight, kg	3.13 (2.39, 3.40)	3.30 (2.84, 3.55)	0.11
Cleft palate	26 (83.9)	79 (95.2)	0.06
Age at palatoplasty, mo	10.00 (10.00, 11.00)	11.00 (10.00, 12.00)	0.06
Comorbidities			
GERD	12 (38.7)	5 (6.0)	<0.001[b]
Tracheo- or laryngomalacia	8 (25.8)	2 (2.4)	<0.001[b]
Length of stay, days	32.00 (18.50, 46.00)	17.00 (14.00, 24.00)	<0.001[b]
Intubation before intervention	3 (9.7)	4 (4.8)	0.34
Preintervention PSG/respiratory polygraphy			
OAHI	26.10 (18.95, 37.70)	—	—
OAI	—	19.90 (9.91, 40.05)	
Spo_2 nadir	76.27 ± 10.24	76.80 ± 10.26	0.511
UAO severity			0.61
Moderate (A_2)	10 (32.3)	21 (25.3)	
Severe (A_3)	21 (67.7)	62 (74.4)	
Use of supplemental oxygen	6 (19.6)	2 (2.4)	0.005[b]
Use of NPA	0 (0.0)	3 (3.6)	0.56
Use of CPAP	5 (16.1)	17 (20.5)	0.80
NGT	27 (87.1)	83 (100.0)	0.005[b]
Surgical feeding tube	9 (29.0)	0 (0.0)	<0.001[b]
z Score at birth	−0.43 (−2.12, 0.34)	0.00 (−0.94, 0.55)	0.13
z Score before intervention	−1.36 (−2.52, −0.59)	−1.36 (−2.04, −0.36)	0.53

GERD, gastroesophageal reflux disease; NPA, nasopharyngeal airway.
[a]Results presented as number (%), median (interquartile range), or mean ± SD.
[b]Significant.

patients in the PEBP group. Results presented as number [%], median [IQR], ***http://links.lww.com/PRS/H854***. **See Table, Supplemental Digital Content 3**, which shows dropout analysis between groups. Results presented as number [%], median [IQR], ***http://links.lww.com/PRS/H855***.)

There was no difference in sex ($P = 1.000$), but race was more diverse in the MDO group ($P < 0.001$). Pretreatment MicroNAPS stages were similar ($P = 0.667$). Gastroesophageal reflux disease and tracheolaryngomalacia were diagnosed more frequently in the MDO group ($P < 0.001$) (Table 1).

Before intervention, median OAHI was 26.1 (19.0, 37.7) events/hour for the MDO group and OAI was 19.9 (9.9, 40.1) events/hour for the PEBP group. Spo_2 nadirs were 76.3±10.2% and 76.8±10.3% for the MDO and PEBP groups, respectively ($P = 0.51$). These values correspond to moderate UAO (A_2) for 10 patients (32.3%) in the MDO group and 21 patients (25.3%) in the PEBP group, and severe UAO (A_3) for 21 patients (67.7%) in the MDO group and 62 patients (74.7%) in the PEBP group ($P = 0.61$). Supplemental oxygen was used for 6 patients (19.4%) in the MDO group compared with 2 patients (2.4%) in the PEBP group ($P = 0.005$; Table 1).

Median ages at initiation of interventions were 41 (28, 84) days of life for the MDO group and 31 (14, 50) days of life for the PEBP group ($P = 0.01$). Distractors were removed at 9 (7, 10) weeks after placement, and PEBP was discontinued after 25 (21, 29) weeks. Length of stay was 32 days (19, 46) for MDO and 17 days (14, 24) for PEBP ($P < 0.001$). Age at palatoplasty was not significantly different between groups ($P = 0.62$ and $P = 0.06$, respectively).

Immediate UAO Outcomes

First postintervention PSG/respiratory polygraphy was obtained at 12 (9, 15) and 10 (7, 17) days

Table 2. Immediate Postintervention Airway Outcomes[a]

Variables	MDO	PEBP	P
UAO severity			<0.001[b]
Normal (A_0)	29 (93.5)	39 (47.0)	
Minor (A_1)	2 (6.5)	44 (53.0)	
OAHI	0.40 (0.00, 1.85)		—
OAI		1.02 (0.40, 2.10)	
Δ OAHI/OAI from before intervention	−98 (−1.00, −0.94)	−94 (−0.99, −0.89)	0.006[b]
Spo_2 nadir	92.00 (87.90, 93.95)	87.00 (80.50, 90.00)	<0.001[b]
Use of supplemental oxygen	1 (3.2)	2 (2.4)	1.00
Use of CPAP	0 (0.0)	3 (3.6)	0.56

[a]Results presented as number (%) or median (interquartile range).
[b]Significant.

Table 3. Adjusted Logistic Regression Model for Preintervention to Postintervention Change in OAHI/OAI

Predictors	Coefficient	95% CI	P
Group	0.017	−0.020, 0055	0.361
Age, days	0.000	−0.001, 0.00	0.069
GERD	−0.037	−0.081, 0.008	0.104
Tracheo- or laryngomalacia	0.003	−0.024, 0.090	0.254

GERD, gastroesophageal reflux disease.

after intervention for MDO and PEBP, respectively (P = 0.144). Median OAHI for the MDO group was 0.4 (0, 1.9) events/hour (decrease of 98% [1, 0.9]) and median OAI for the PEBP cohort was 1 (0.4, 2.1) events/hour (decrease 94% [1, 0.9]; P = 0.006). Spo_2 nadirs were 92.0% (87.9, 94.0) and 87.0% (80.5, 90.0) for the MDO and PEBP groups, respectively (P < 0.001). These values correspond to no UAO (A_0) for 29 patients (93.5%) in the MDO group and 39 (47%) in the PEBP group, and minor UAO (A_1) for all remaining patients (P < 0.001). (Table 2). Secondary analysis using the more liberal OAHI/OAI targets found complete resolution of UAO in all patients in the MDO group and 73 patients (88%) in the PEBP group (P = 0.060). No predictors were significant in the adjusted regression model (Table 3).

Respiratory Polygraphy Rescoring Subanalysis

Rescoring of the matched series of respiratory polygraphs found immediate postintervention OAHI of 7.6 (4.35, 11.10) events/hour for the PEBP group (n = 30). Reanalysis using this outcome did not alter results of the original analysis (Table 4).

UAO Outcomes Stratified by Starting Severity

For patients with preintervention moderate UAO (A_2), median immediate postintervention OAHI/OAI and Spo_2 nadirs were 0.45 (0.05, 0.78) events/hour and 91.5% (88.6, 94.8) for MDO, and 0.86 (0.58, 1.50) events/hour and 89.0% (84.0, 90.0) for PEBP. These values correspond to no UAO (A_0) for 9 patients (90.0%) in the MDO group and 12 (57.1%) in the PEBP group, and minor UAO (A_1) for the remaining patient (10.0%) in the MDO group and 9 patients (42.9%) from the PEBP group (P = 0.106) (Table 5).

For patients with severe UAO (A_3) before intervention, median immediate postintervention OAHI/OAI and Spo_2 nadirs were 0.3 (0.0, 2.0) events/hour and 92.1% (88.0, 93.7) for the MDO group and 1.2 (0.33, 2.25) events/hour and 85.0% (80.0, 90.0) for the PEBP group. These values correspond to no UAO (A_0) for 20 patients (95.2%) in the MDO group compared with 27 (43.5%) in the PEBP group, and minor UAO (A_1) for 1 patient (4.8%) in the MDO group and 35 (56.5%) in the PEBP group (P < 0.001) (Table 5).

Latest Follow-up Respiratory Outcomes

The latest follow-up PSG/respiratory polygraphs were obtained at a median of 7.0 (5.8, 8.5) months after MDO and 8.1 (6.2, 10.9) months after PEBP initiation (P = 0.43), often corresponding to shortly before palatoplasty. By latest follow-up, median OAHI for the MDO group was 0 (0, 0.8) events/hour and median OAI for the PEBP group was 0.1 (0, 0.6) events/hour. Median Spo_2 nadirs were 88.0% (81.0, 92.0) and 90.0% (84.5, 94.0) for the MDO and PEBP groups, respectively (P = 0.223). These values correspond to no or minor UAO for 12 patients (92.3%) in the MDO group and 75 (98.7%) from the PEBP cohort (MDO, 84.6% A_0, 7.7% A_1; PEBP, 84.2% A_0, 14.5% A_1), but return of moderate or severe UAO for 1 patient from each group (MDO, 0% A_2, 7.7% A_3; PEBP, 1.3% A_2, 0% A_3) (P = 0.28). From before intervention to latest follow-up, OAHI/OAI decreased by a median of 100% (100, 96) with MDO and 99% (100, 97) with PEBP (P = 0.388).

Table 4. Rescored Airway Outcomes of Matched Patients[a]

Variables	MDO	PEBP	P
No.	31	31	
Before intervention			
OAI	13.00 (5.70, 18.55)	14.6 (9.50, 22.77)	0.29
OAHI	29.50 (10.30, 49.15)	36.00 (22.55, 49.95)	0.17
Postintervention OAHI			
All patients	0.00 (0.00, 1.55)	7.60 (4.35, 11.10)	<0.001[b]
Moderate (A_2)	0.00 (0.00, 0.45)	9.40 (4.78, 11.75)	0.005[b]
Severe (A_3)	0.00 (0.00, 2.60)	7.20 (4.35, 9.80)	<0.001[b]

[a]Results presented as median (interquartile range).
[b]Significant.

Table 5. Immediate Postintervention Airway Outcomes Stratified by Severity of Preintervention UAO[a]

Preintervention UAO and Outcome	MDO	PEBP	P
Moderate (A_2)			
UAO severity			0.11
Normal (A_0)	9 (90.0)	12 (57.1)	
Minor (A_1)	1 (10.0)	9 (42.9)	
Use of supplemental O_2	1 (10.0)	1 (4.8)	1.00
Use of CPAP	0 (0.0)	1 (4.8)	1.00
NGT use at discharge	3 (30.0)	3 (14.3)	0.36
Surgical feeding tube	6 (60.0)	0 (0.0)	<0.001[b]
Severe (A_3)			
UAO severity			<0.001[b]
Normal (A_0)	20 (95.2)	27 (43.5)	
Minor (A_1)	1 (4.8)	35 (56.5)	
Use of supplemental O_2	0 (0.0)	1 (1.6)	1.00
Use of CPAP	0 (0.0)	2 (3.2)	1.00
NGT use at discharge	14 (66.7)	12 (19.4)	<0.001[b]
Surgical feeding tube	3 (14.3)	0 (0)	0.01[b]

[a]Results presented as number (%).
[b]Significant.

Three patients (8.6%) in the MDO group used CPAP at latest follow-up, compared with none in the PEBP group ($P = 0.02$).

Feeding Outcomes

Application of PEBP did not interrupt oral feeding, but, for the MDO group, oral feeding was discontinued for a median of 9.5 (6.25, 13) days after the initial operation ($P < 0.001$). Patients in the MDO group had higher rates of both NGT use at discharge (n = 17 [54.8%] versus n = 15 [18.1%]; $P < 0.001$) and insertion of surgical feeding tubes ($n = 9$ [29.0%] versus 0) compared with the PEBP group ($P < 0.001$) (Table 6).

Infants starting with moderate UAO (A_2) showed similar rates of postintervention NGT use between treatments but a higher rate of surgical feeding tube insertion in the MDO group ($P <$ 0.001). For those starting with severe UAO (A_3), MDO was associated with higher rates of both NGT and surgical feeding tube use ($P < 0.001$, $P =$ 0.014, respectively) (Table 5).

Weight Outcomes

Weight-for-age z scores were similar between groups before intervention and at immediate and 3-, 6-, 9-, and 12-month postintervention timepoints. However, at latest follow-up (MDO, n = 18; PEBP, $n = 26$), z scores were significantly lower (indicating poorer growth velocity) for the MDO group compared with the PEBP group ($P = 0.038$) (Table 6 and Fig. 7).

DISCUSSION

MDO and PEBP have both been shown to improve upper-airway obstruction in infants with RS.[6,32,49] Both treatments have also demonstrated additional benefits regarding feeding,[10,11,26,50] weight gain,[13,35,51] quality of life, and neurocognitive development.[15,16,52–54] Existing data for each treatment, however, are not directly comparable, and no single study has evaluated both modalities. To determine the role for each treatment in the algorithm of early RS care, a direct comparison is

Table 6. Feeding and Growth Outcomes[a]

Variables	MDO	PEBP	*P*
Days to restart PO feeds	9.5 (6.3, 13.0)	0.0 (0.0, 0.0)	<0.001[b]
NGT use at discharge	17 (54.8)	15 (18.1)	<0.001[b]
Surgical tube insertion	9 (29.0)	0 (0.0)	<0.001[b]
z Score			
Immediately after intervention	−1.70 (−2.42, −0.28)	−1.33 (−2.04, −0.55)	0.91
3 mo	−1.83 (−2.12, −0.86)	−1.29 (−1.85, −0.60)	0.15
6 mo	−1.29 (−1.94, −0.45)	−0.99 (−1.40, −0.44)	0.22
9 mo	−0.76 (−1.38, −0.16)	−0.31 (−0.98, 0.40)	0.05
1 yr	−0.25 (−1.31, 0.29)	−0.17 (−0.89, 0.22)	0.67
Latest follow-up	−0.64 (−1.19, 0.29)	0.24 (−0.06, 1.01)	0.04[b]

[a]Results presented as number (%) or median (interquartile range).
[b]Significant.

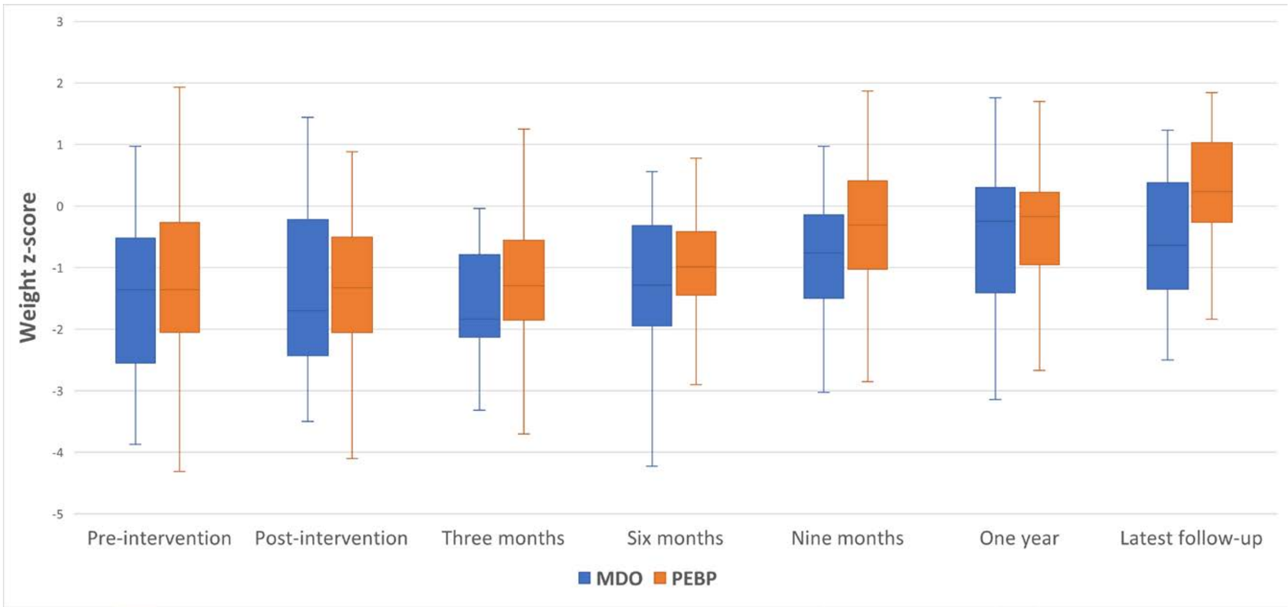

Fig. 7. Weight-for-age *z* scores.

necessary. The objective of this study was to compare early airway, feeding, and growth outcomes in similar series of infants with RS who were managed with either MDO or PEBP.

This study demonstrated substantial improvement in all 3 domains with both treatments. Overall, MDO was more effective than PEBP in providing complete or near-complete resolution of UAO when strict UAO outcome criteria were applied. A secondary analysis using more liberal outcome targets that are commonly applied in European centers[47] closed the UAO outcomes gap between treatments, eliminating the statistically significant difference between them. Furthermore, when stratified by severity of preintervention airway dysfunction, the superiority of MDO for resolution of UAO was limited to infants starting with severe obstruction (A_3). Those with moderate preintervention UAO (A_2) had statistically similar early airway outcomes with both treatments.

Airway outcomes 7 to 8 months after MDO or PEBP were similar between groups. Neither group had a significant requirement for posttreatment airway adjuncts, such as supplemental O_2 or CPAP, although CPAP use was more frequent in the MDO group at latest follow-up. These findings suggest a natural progression toward airway improvement over time that is independent of the early intervention, further emphasizing that the goal of treatment in early infancy should be to support breathing, feeding, and growth during the first few months of life with the least possible treatment burden.

Early feeding outcomes were inverse to the airway results in this study. Infants treated with PEBP required fewer feeding tubes and had superior growth velocities than those managed by MDO. One factor that likely contributed to this effect is the mean 9-day interruption of oral feeding by initiation of MDO. The PEBP group continued feeding orally without interruption after appliance delivery. In addition, obturation of the palatal cleft with PEBP, when present, likely enhanced the suck-swallow-breathe reflex, thereby accelerating oral feeding competence.[35]

Our results agree with previous literature. MDO has demonstrated a reduction in OAHI by 79% to 94%[55–57] and adequate resolution of UAO in more than 94% of infants with RS.[6,49,58] A similar decrease of 88.9% in OAI has been reported at 3-month follow-up after treatment with PEBP.[33] Improvement in feeding and growth trajectories have also been observed with both treatments.[10,13,35,51,59] MDO is associated with a lower rate of surgical feeding tube insertion compared with other operations used to treat UAO, such as tongue–lip adhesion and tracheostomy.[11,12] In addition, despite impairment of pretreatment growth velocity, infants demonstrate faster weight gain than age-matched peers after MDO.[10,13] PEBP typically facilitates hospital discharge without feeding tubes.[35,60]

This study has several limitations that must be considered. Due to its retrospective nature, patient characteristics and outcome assessments could not be completely standardized between groups. Some differences exist between groups that may represent different populations (eg, racial diversity), may reflect variation in diagnostic approaches (eg, gastroesophageal reflux disease, tracheo/laryngomalacia), or may be related to the primary treatment (eg, length of stay, rates of NGTs or gastrostomy tubes) that could not be completely reconciled. Most notably, this includes differences in sleep study parameters. The primary difference between study types is that electroencephalography (EEG) was used with PSG but not for respiratory polygraphy. Lack of EEG complicates scoring of hypopneas, because obstructive and central hypopneas may be indistinguishable due to physiologic thoracoabdominal asynchrony in infants. In addition, EEG is used to measure total sleep time for PSG, whereas total sleep time is calculated using behavioral observation for respiratory polygraphy, likely a small difference. Another minor variation between study types was data averaging times, which were 2 to 4 seconds for PSG and 2 seconds for respiratory polygraphy studies in this investigation. Because there is a linear relationship between averaging times and desaturation rates, the benefit of PEBP as measured by with shorter averaging times may be even more profound than this study suggests. Considerable effort was made to harmonize these metrics, including use of the same standardized definitions and scoring techniques and by performing a prospective reanalysis of a matched group of patients to test direct assessment of the same measures. To further mitigate the impact of these differences, we relied heavily on stratification of UAO variables using the MicroNAPS classification, which is a new system that has not been fully validated. In addition, no consensus exists for the OAHI/OAI threshold that constitutes UAO in infants.[61] Recent data indicate that occasional obstructive apneas occur in healthy infants.[44–46] We therefore analyzed 2 cutoffs. Threshold values for best practice must be interpreted by each institution and will continue to evolve.

CONCLUSIONS

Both MDO and PEBP demonstrated significant relief of airway obstruction and improvement in feeding and growth for infants with RS. Overall, MDO outmatched PEBP in resolution of UAO, whereas PEBP portended better early feeding and growth outcomes. When stratified by preintervention severity, however, the airway superiority of MDO was realized primarily for infants starting with severe UAO. Following the concept that no operation is as good as operation (when outcomes are similar), we suggest that PEBP be considered an alternative to MDO for infants starting with moderate UAO (A_2), and that MDO remain the treatment of choice for those with severe obstruction (A_3). As a result of these findings, a PEBP program has been initiated at Boston Children's Hospital to allow both treatments to be offered.

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DISCLOSURE

Dr. Resnick is a consultant for AbbVie Pharmaceuticals. The other authors have no financial relationships or conflicts of interest to disclose.

PATIENT CONSENT

Parents or guardians provided written informed consent for use of the patient's images.

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