

Current Trends in Surgical Airway Management of Neonates with Robin Sequence

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Background: Robin sequence is defined by the clinical triad of micrognathia, glossoptosis, and upper airway obstruction, and is frequently associated with cleft palate and failure to thrive. Though the efficacy of certain surgical interventions to relieve airway obstruction has been well established, algorithms dictating decision making and perioperative protocols are poorly defined.

Methods: A 22-question survey was sent via e-mail to members of the American Cleft Palate-Craniofacial Association and International Society of Craniofacial Surgeons. Questions were related to surgeon experience in treating neonates with Robin sequence, and specific perioperative protocols.

Results: One hundred fifty-one responses were collected. Most respondents were surgeons practicing in North America (82.8%), in a university hospital setting (81.5%) and had completed a fellowship in pediatric plastic surgery or craniofacial surgery (76.2%). Preoperative protocols varied widely by years in training and location of practice. Although 78.8% of respondents always performed direct laryngoscopy, only 49.7% of respondents routinely obtained preoperative polysomnography. Mandibular distraction osteogenesis was the most common primary surgical airway intervention reported by 74.2%, with only 12.6% primarily utilizing tongue-lip adhesion. Slightly less than half of respondents ever performed tongue-lip adhesion. Operative selection was influenced by surgeon experience, with 80% of those in practice 0–5 years primarily utilizing mandibular distraction, compared with 56% of respondents in practice >15 years.

Conclusions: This study documents wide variations in preoperative, operative, and postoperative protocols for the surgical airway management of neonates with severe Robin sequence. These results underscore the need to acquire more objective data, to compare different protocols and outcome measures. (*Plast Reconstr Surg Glob Open* 2018;6:e1973; doi: 10.1097/GOX.0000000000001973; Published online 7 November 2018.)

INTRODUCTION

The clinical triad of micrognathia, glossoptosis, and upper airway obstruction defines Robin sequence (RS) and often manifests with respiratory insufficiency, feeding difficulties, and failure to thrive. A U-shaped cleft of the

hard and soft palate is a common comorbid finding. Although neonates may present with isolated RS, additional syndromic disorders may be present, including Treacher Collins syndrome, Stickler syndrome, and velocardiofacial syndrome.¹ The diagnosis of an underlying syndrome in patients with RS is critical, as these patients tend to have poorer prognoses with structural abnormalities affecting both upper and lower airways.² Nevertheless, the overall rate of morbidity and mortality in neonates with RS has recently been reduced with improved nutritional support and airway management, with the most severe cases benefiting from early surgical intervention.^{3–5}

Surgical goals in RS include sustainable relief of airway obstruction, optimizing weight gain, and minimizing iatrogenic morbidity.^{6,7} There remains a paucity of evidence-based recommendations secondary to lack of objective and long-term comparisons of various treatment

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protocols, a challenge compounded by the wide spectrum of RS phenotypes.⁸ Neonates with mild presentations can often be managed conservatively with nonsurgical techniques including prone or lateral positioning, temporary nasopharyngeal airway, nasogastric feeding, and continuous positive airway pressure.⁶ With certain clinical findings, nonsurgical management has been demonstrated to be successful.⁹ Alternatively, those with more severe upper airway obstruction may require surgical interventions, such as floor of mouth release, tongue-lip adhesion (TLA), mandibular distraction osteogenesis (MDO), or tracheostomy.⁶ The optimal choice of operation remains poorly elucidated.

Surgical indications and preoperative evaluation also remain controversial and variable among institutions. Although polysomnography (PSG) and apnea hypoxia index (AHI) may have a role in classifying the severity of RS symptoms and guiding appropriate surgical management, they are not universally obtained and are subject to interpretation.^{2,8,10,11} Delineation of upper and lower airway anatomy can be aided by preoperative direct laryngoscopy,^{7,8} though is also not standard of care among institutions.

Two critical questions remain to be answered in managing upper airway obstruction in neonates with RS. First, which patients require surgical airway intervention? Second, which is the best surgical airway procedure for a particular patient? Although multiple institutions have documented their experiences with various procedures (ie, describing what each group believes to be the answer to the second question above), there are few studies that definitively address these questions.^{9,12–14} No prospective randomized controlled studies provide therapeutic guidelines or criteria. The lack of consensus on appropriate treatment protocols makes surgical decision-making highly variable and dependent on institutional or surgeon preference.⁶ The objective of this study was to characterize the landscape of diagnosis and treatment for this highly heterogeneous spectrum of disease.

METHODS

The study was given IRB-exempt status by the corresponding author's institution. In July 2017, an anonymous 22-question study regarding evaluation and management of neonates with RS was sent to surgeon members of the American Cleft Palate-Craniofacial Association (ACPA, *n* = 1,033) and International Society of Craniofacial Surgeons (*n* = 200; Table 1). Recipients were asked to complete the survey if they actively perform surgical airway management of RS. Designed to be completed within 5 minutes, the survey study (<http://www.surveymonkey.com>) was formed to evaluate respondent demographics (Q1–4), preoperative evaluation (Q5–8), type of surgical procedure preferences (Q9–11), and MDO preferences (Q12–22) when treating Robin sequence neonates (<6 months). Responses were required for all questions to be tabulated, except for question 7 and 11, which only populated if the respondent answered “Yes” to question 6 and 10, respectively.

Answers were tabulated into Excel (Microsoft Excel, Redmond, WA), and statistical analysis was performed. The unpaired Student's *t* test was used when comparing mean difference of numeric variables between groups. The chi-square test or the Fisher's exact test (if *n* < 5) was applied when testing the homogeneity between 2 categorical variables. Odds ratios and 95% confidence intervals were calculated to reveal trends. In an effort to simplify statistical analysis, variables, such as years in practice, were dichotomized (eg, < 10 versus ≥ 10 years in practice). Odds ratios and confidence intervals were reported.

RESULTS

Demographics and Work Up

There were a total of 151 respondents (response rate: 12.2%). Of the respondents, 33.1% of them were >15 years in practice. The next largest group (29.8%) were 0–5 years out in practice. The majority practiced in a University setting (81.5%), in North America (82.8%), and completed a pediatric plastic surgery/craniofacial fellowship (76.2%). Among all respondents, 8% were from Europe, 6% were from Asia, and 2% from South or Central America.

The majority of respondents obtained (78.8%) or sometimes obtained (11.3%) direct laryngoscopy/bronchoscopy (DLB) before surgical treatment, while 9.93% never do. Those 0–10 years in practice were 5.2 times more likely to order a DLB on initial evaluation compared with those >10 years in practice (*P* = 0.011; OR = 5.22, 1.41–19.34; Fig. 1). North American surgeons were 15 times more likely to order a DLB than international surgeons (*P* = 7.34×10^{-6} ; OR = 15, 4.55–49.48; Fig. 2). The majority of respondents always (49.67%) or sometimes (29.1%) ordered a PSG, whereas some never obtained a sleep study (21.2%). Those 0–10 in practice were 2.6 times more likely to order a PSG than those who were >10 years in practice (*P* = 0.017, OR = 2.63, 1.17–5.95; Fig. 3). North American respondents were 3.6 times more likely to order a PSG than their International colleagues (*P* = 0.00378; OR = 3.63, 1.46–9.01; Fig. 4). Fellowship-trained surgeons were no more likely to order these tests than their nonfellowship-trained counterparts (*P* > 0.05). Among those that answered “Yes” or “Sometimes” to ordering PSG, the minimum AHI for surgical intervention was 11–19 for 42.7%, 20–29 for 29.13%, >10 for 21.35%, and >30 for 5.8% (Fig. 5). The presence of syndrome changed the treatment of choice always in 11.9% of surgeons, never in 10.6% of surgeons, and depended on the syndrome in 77.5% of surgeons.

Surgical Management

The most common primary surgical management was MDO (74.2%), followed by TLA (12.6%), and tracheostomy (6.6%; Fig. 6). Responses in other surgical management included Delorme subperiosteal release of the tongue (*n* = 3). There was no correlation between years in practice and likelihood of performing tracheostomy over other procedures. International respondents were 30 times more likely to perform tracheostomy as primary surgical management versus those of other North American

Table 1. The 22-question Survey Administered**Children's National Medical Center****Mandibular Distraction Osteogenesis Survey**

All questions pertain to neonatal (i.e., <6 months of age) mandibular distraction to treat airway dysfunction.

1. Years in practice
 - a. 0–5
 - b. 6–10
 - c. 11–15
 - d. >15
2. Hospital setting (check all that apply)
 - a. University
 - b. Public
 - c. Private
 - d. Government
3. In what region do you practice medicine?
 - a. North America
 - b. South or Central America
 - c. Europe
 - d. Asia
 - e. Middle East
 - f. Other (please specify)
4. Did you complete a pediatric plastic surgery / craniofacial fellowship?
 - a. Yes
 - b. No
5. Do you obtain a direct laryngoscopy bronchoscopy to evaluate the lower airway prior to distraction?
 - a. Yes
 - b. Sometimes
 - c. No
6. Do you obtain polysomnography (sleep study)?
 - a. Yes
 - b. Sometimes
 - c. No
7. If yes (7), what is your minimum apnea hypopnea index (AHI) for surgical intervention?
 - a. <10
 - b. 11–19
 - c. 20–29
 - d. >30
8. Does the presence of a syndrome change your treatment choice?
 - a. Yes
 - b. Depends on the syndrome
 - c. No
9. What is your most common primary surgical management for patients who have failed nonoperative management of airway obstruction?
 - a. Tracheostomy
 - b. Tongue lip adhesion
 - c. Mandibular distraction
 - d. Other (please specify)
10. Do you perform tongue lip adhesion?
 - a. Yes
 - b. Sometimes
 - c. No
11. If yes to 10, what criteria are used to select TLA (select all that apply)?
 - a. Physical findings
 - b. Physiologic findings
 - c. Clinical scoring system
 - d. Other (please specify)
12. Number of neonatal mandibular distraction procedures performed annually.
 - a. 0
 - b. 1–5
 - c. 6–10
 - d. 11–15
 - e. >15
13. Type of mandibular osteotomy you typically perform.
 - a. Inverted L ramus osteotomy
 - b. Mandibular angle osteotomy
 - c. Mandibular body osteotomy
 - d. Other:
14. What types of distraction vector do you typically use?
 - a. Vertical (parallel to ramus)
 - b. Oblique
 - c. Horizontal (parallel to body)
15. What devices do you typically use? (check all that apply)
 - a. Buried
 - b. External
 - c. Absorbable
16. Do you use virtual surgical planning for mandibular distraction?
 - a. Yes
 - b. Sometimes
 - c. No
17. How long is your latency phase?
 - a. 0 days
 - b. 1–3 days
 - c. 4–6 days
 - d. 7–14 days
18. During activation phase, how often do you distract?
 - a. 1×/day
 - b. 2×/day
 - c. 3×/day
 - d. >3×/day
19. Total distraction length per day?
 - a. 0.5 mm
 - b. 1.0 mm
 - c. 1.5 mm
 - d. 2.0 mm
 - e. > 2.0 mm
20. What is your endpoint for distraction?
 - a. Cessation of apnea
 - b. Class I
 - c. Class III
 - d. As far as possible
21. How long is your typical consolidation phase?
 - a. <4 weeks
 - b. 4–6 weeks
 - c. 6–8 weeks
 - d. 8–10 weeks
 - e. >10 weeks
 - f. Comments:
22. What percentage of distracted patients required further intervention for apnea?
 - a. 0–24%
 - b. 25–49%
 - c. 50–74%
 - d. 75–100%

countries ($P = 5.64 \times 10^{-6}$; OR = 30.93, 6.00–159.50; Fig. 7). Interestingly, those who did not typically order DLB as part of preoperative work up were 8 times more likely to perform tracheostomy as their primary surgical modality ($P = 0.00923$; OR = 8.07, 1.95–33.37). North American respondents were 8 times more likely to use MDO as primary surgical management versus other surgeries than interna-

tional respondents ($P = 2.75 \times 10^{-5}$; OR = 8.29, 3.12–22.05). Respondents 0–10 years in practice were more than twice as likely to perform MDO as primary surgical management versus those >10 years in practice ($P = 0.0345$; OR = 2.44, 1.05–5.64). Fellowship-trained surgeons were not more likely to prefer a particular technique over nonfellowship-trained surgeons ($P = 0.44$).

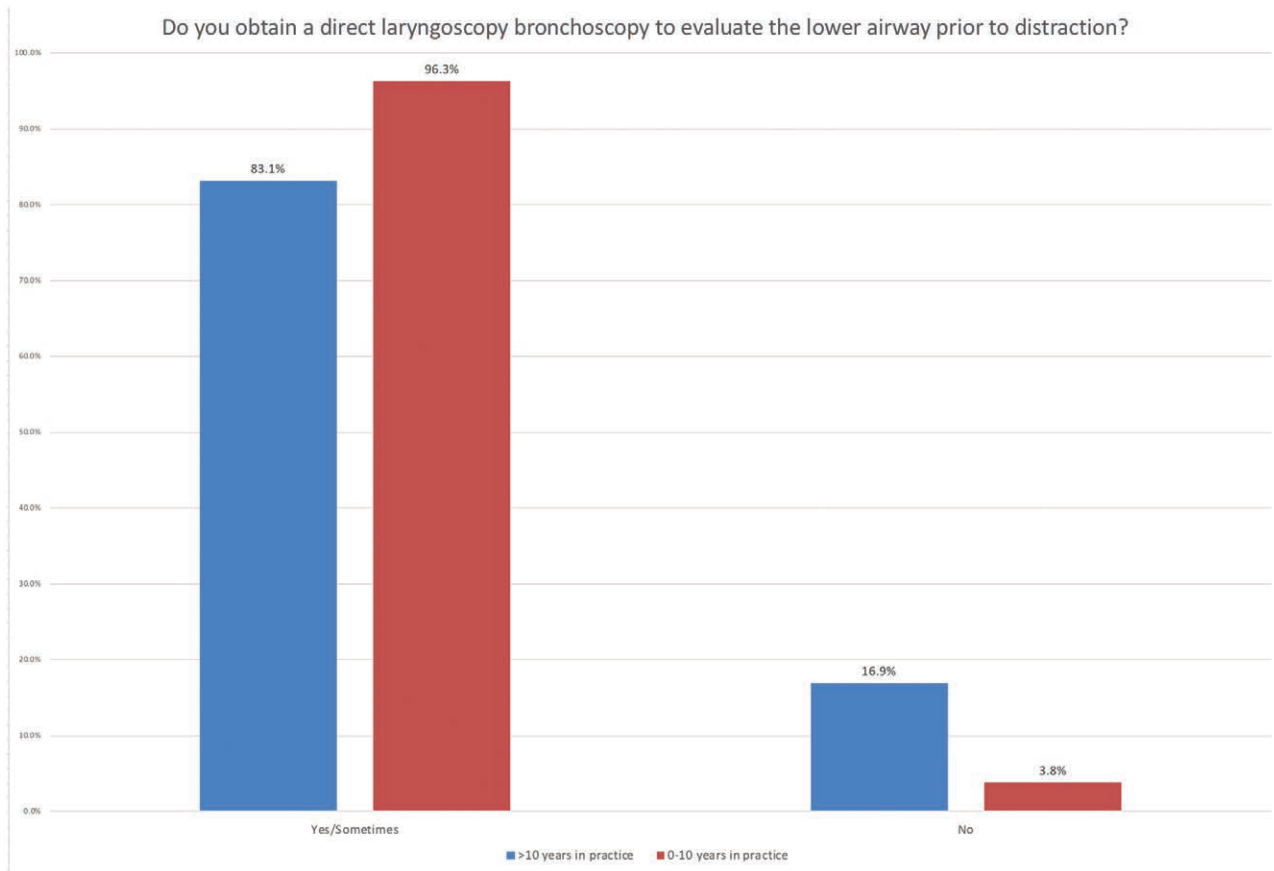


Fig. 1. Do you obtain a direct laryngoscopy to evaluate the lower airway? Surgeons 0–10 years in practice are 5.2 times more likely to order diagnostic laryngoscopy/bronchoscopy on initial evaluation compare to those >10 years in practice ($P = 0.011$; OR = 5.22, 1.41–19.34).

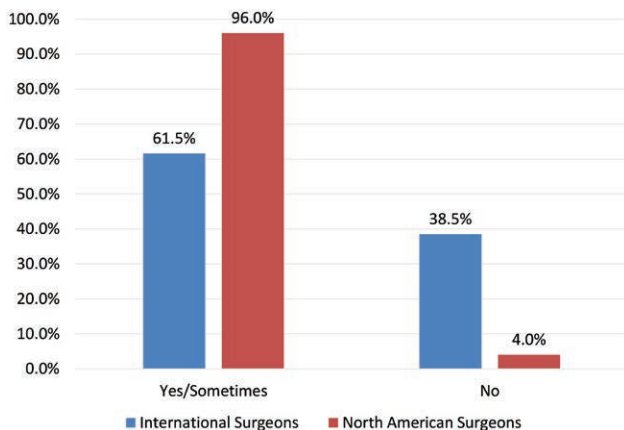


Fig. 2. Do you obtain a direct laryngoscopy to evaluate the lower airway? North American surgeons are 15 times more likely to order a diagnostic laryngoscopy/bronchoscopy than International surgeons ($P = 7.34 \times 10^{-6}$; OR = 15, 4.55–49.48).

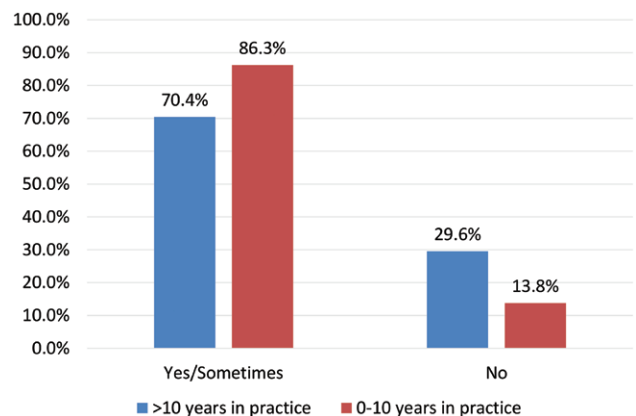


Fig. 3. Do you obtain a PSG (sleep study) as part of your preoperative work up? Those 0–10 years in practice are 2.6 times more likely to order a PSG than those who are >10 years in practice ($P = 0.017$; OR = 2.63, 1.17–5.95).

The respondents were split when queried if they performed tongue lip adhesions with 54.3% answering “No” and 23.84% and 21.85% responding “Sometimes” and “Yes”, respectively. Those 0–10 years out in practice were twice as likely to not perform TLA versus someone >10 years out in practice ($P = 0.032$; OR = 2.03, 1.06–3.89).

Country of practice or fellowship training did not influence likelihood of performing TLA. For the most part, for those who did perform TLA ($n = 69$), respondents tended to base their surgical modality on anatomical or physiological findings on clinical examination (Fig. 8). Five respondents included parental input in

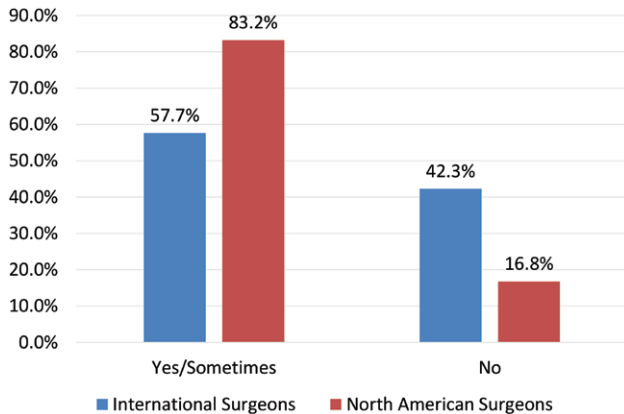


Fig. 4. Do you obtain PSG (sleep study)? North American respondents are 3.6 times more likely to order a polysomnography than their international colleagues ($P = 0.00378$; OR = 3.63, 1.46–9.01).

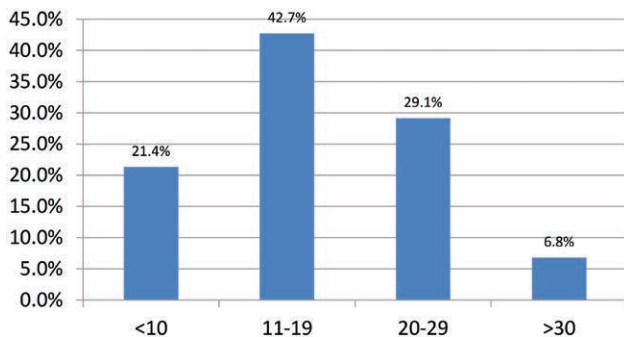


Fig. 5. If yes, what is your minimum AHI for surgical intervention?

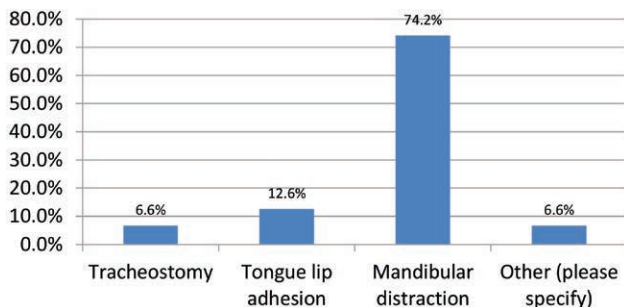


Fig. 6. What is your most common primary surgical management for patients who have failed nonoperative management of airway obstruction?

their decision for type of operative intervention, and one answered that TLA was performed when “catch-up mandibular growth” was believed to be a factor. Three respondents reported that they used to perform TLA but no longer do.

Mandibular Distraction

The majority of respondents performed 1–5 distractions annually (53.6%; Fig. 9). In total, 29.1% of respondents performed 6–10 distractions per year. There was no difference between the years in practice and the number of MDO performed ($P = 0.60$). Among those that perform MDO, 39.2% of surgeons performed an inverted

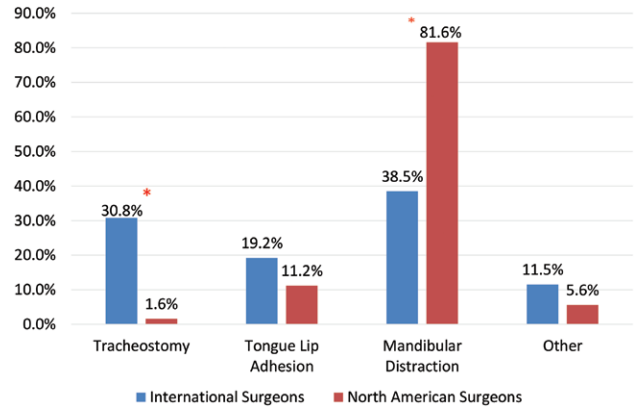


Fig. 7. What is your most common primary surgical management for patients who have failed nonoperative management of airway obstruction? North American respondents are 8 times more likely to use MDO as primary surgical management vs. other surgeries than international respondents ($P = 2.75 \times 10^{-5}$; OR = 8.29, 3.12–22.05). International respondents were 30 times more likely to perform tracheostomy as primary surgical management vs. those of other North American countries ($P = 5.64 \times 10^{-6}$; OR = 30.93, 6.00–159.50). *Indicates significance.

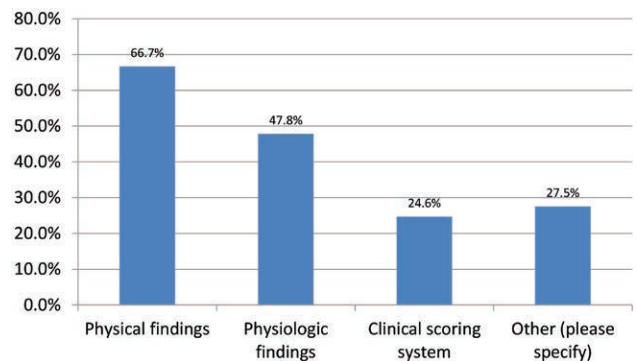


Fig. 8. If you do perform TLA, what are your surgical criteria?

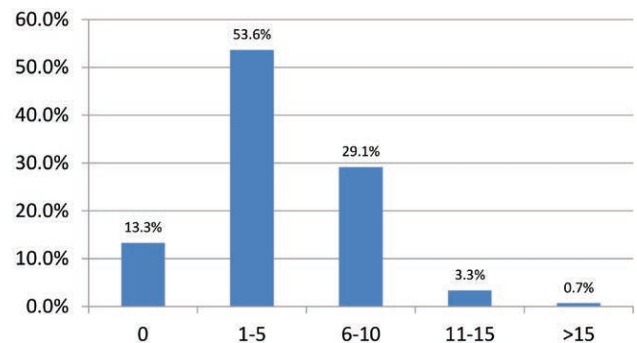


Fig. 9. Number of neonatal mandibular distraction procedures performed annually?

L ramus osteotomy, 37.7% performed a mandibular angle osteotomy, and 17.0% performed a mandibular body osteotomy. Surgeons 0–10 years in practice were twice as likely to use inverted L ramus osteotomies as opposed to other techniques compared with surgeons >10 years in practice ($P = 0.044$, OR = 2.12, 1.02–4.42; Fig. 10).

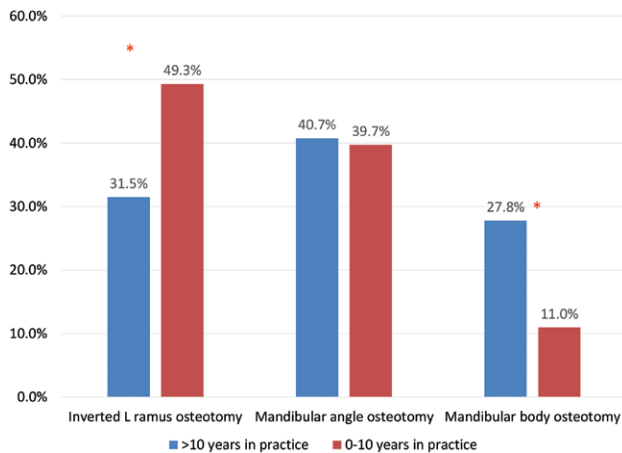


Fig. 10. What type of mandibular osteotomy do you typically perform? Surgeons 0–10 years in practice were twice as likely to use inverted L ramus osteotomies ($P = 0.044$; $OR = 2.12, 1.02–4.42$). Surgeons >10 years in practice were 3 times as likely to use mandibular body osteotomies ($P = 0.015$; $OR = 3.13, 1.21–8.04$). *Indicates significance.

Surgeons >10 years in practice were 3 times as likely to use mandibular body osteotomies instead of other techniques than surgeons 0–10 years in practice ($P = 0.015$; $OR = 3.13, 1.21–8.04$). Surgeons that performed >5 MDO a year were twice as likely to perform an inverted L ramus osteotomy as opposed to other techniques ($P = 0.045$; $OR = 2.11, 1.01–4.40$; Fig. 11). Surgeons that performed 0–5 MDO a year were 5 times more likely to perform mandibular body osteotomies as opposed to other techniques ($P = 0.0085$; $OR = 4.89, 1.37–17.48$). For the most part, most distractions underwent a horizontal vector (64%) while some were done in an oblique (22.1%) or vertical vector (6.6%). There was no correlation between distraction vector and years in practice or number performed. Surgeons generally used a buried device (79.9%). Respondents were split on whether they used virtual surgical planning: 50% answered “No”, while 26.8% and 23.2% answered “Sometimes” and “Yes.” There was no difference in years in practice, country of practice, or number of MDO performed a year in virtual surgical planning use.

Most surgeons used a 1–3 day latency phase (63%) while some used a 0 day latency phase (25.2%). During the activation phase, most surgeons distracted twice a day (72.6%), while others distracted once (16.3%) or 3 times (8.9%) a day. Half of surgeons distracted 1.0 mm (45.1%) during the activation phase, while the other half distracted 1.5 mm (15.8%) and 2.0 mm (32.3%) a day. The endpoint for distraction tended to be Class III occlusion (56%) or “As far as possible” (28.3%). A minority of respondents stopped distraction at Class I occlusion (6.7%) or cessation of apnea (9.0%). The consolidation phase was more variable (Fig. 12). Most surgeons incorporated a 6–8 week consolidation phase (31.30%), while some incorporated a >10 week distraction phase (25.2%). The majority of surgeons used a >6 week consolidation phase (75.57%). In total, 90.3% of surgeons answered that 0–24% of their patients required further

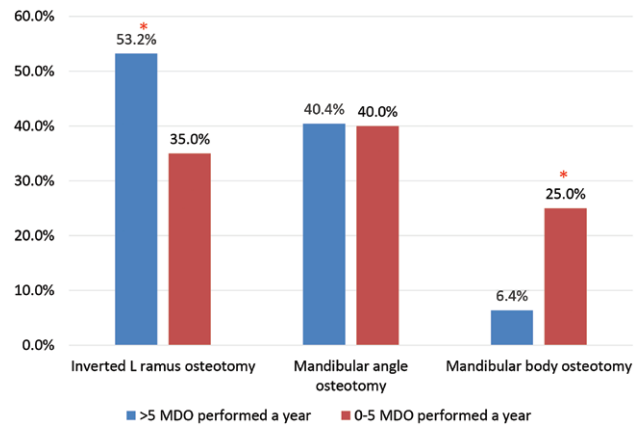


Fig. 11. What type of mandibular osteotomy do you typically perform? Surgeons that perform >5 MDO a year are twice as likely to perform an inverted L ramus osteotomy ($P = 0.045$; $OR = 2.11, 1.01–4.40$). Surgeons that perform 0–5 MDO a year are 5 times more likely to perform mandibular body osteotomies ($P = 0.0085$; $OR = 4.89, 1.37–17.48$). *Indicates significance.

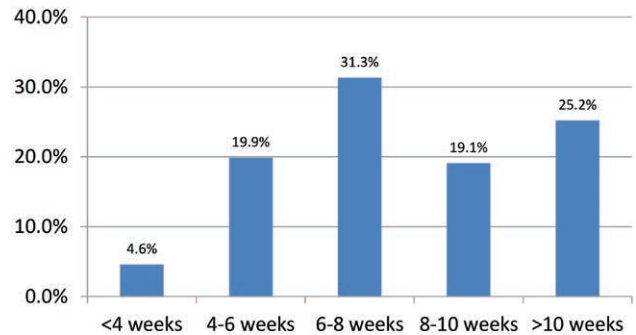


Fig. 12. How long is your typical consolidation phase?

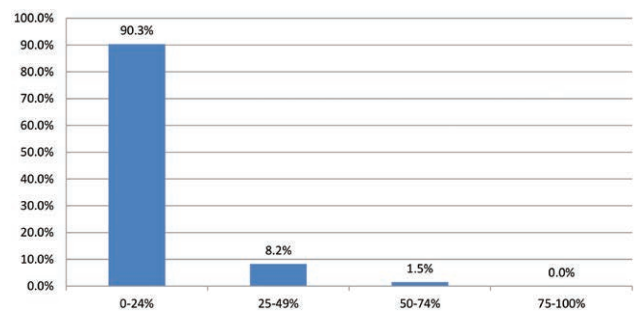


Fig. 13. What percentage of distracted patients require further intervention for apnea?

intervention for apnea after MDO, suggesting MDO in their hands is successful in treatment of airway abnormalities (Fig. 13).

DISCUSSION

The lack of consensus in the literature in RS management is evident.⁸ With over 100 syndromes with known micrognathia and wide spread variability in the very definition of RS, management of this phenotype is highly heterogenous, with several management and treatment

algorithms described with varying success.^{4,6} It has been demonstrated through examination of cephalograms that RS comprises a wide range of mandibular morphology and positions.¹⁵ Syndromic diagnosis leads to poorer outcomes and tend to fail nonsurgical intervention; only 10.6% of respondents report that a concomitant syndrome diagnosis does not affect surgical treatment choice.⁹

Many algorithms combine DLB with PSG in their initial evaluation of patients who are unresponsive to positioning.^{5,6} Although the majority of our respondents ordered DLB, not all do. Those who were >10 years in practice or practice internationally tended not to order this modality, despite the fact that up to 28% of children have concomitant lower airway abnormality beyond the base of tongue.⁵ Careful evaluation of the lower airway with laryngoscopy is necessary when considering surgical interventions that only address the upper airway.¹⁶ However, experience and regional differences in availability can preclude use. This may explain the increased likelihood of surgeons who did not order DLB resorting to tracheostomy as their initial surgical modality.

Overnight PSG is considered by some to be the gold standard investigating and quantifying obstructive sleep apnea (OSA).² The majority of respondents always or sometimes use PSG, with North American surgeons and those <10 years in practice have a higher chance of doing so. Although a number of studies have used PSG to show improvement in apnea, thresholds for intervention remain highly variable with a recent systematic review highlighting studies using an AHI of 6.4 to 52.6 as their respective indications for surgical airway intervention.² In our cohort, most surgeons selected an AHI of 11–29 as their minimum for intervention, but more than a quarter of surgeons selected either <11 and ≥ 30 as their thresholds. The wide variability may be explained by the (1) lack of normative data among different neonatal age groups and (2) lack of guidance from comparative data on severities of OSA among RS and the interventions that improve OSA at various severities.^{2,17} Furthermore, the lack of PSG in all institutions limits its use and may explain why only half of surgeons always use PSG.

Previously, our group presented results of a retrospective analysis of over 60 infants with RS, comparing characteristics of patients who did not require surgical airway interventions with those who did.¹⁸ Multivariate and ROC analysis demonstrated 4 features on PSG that distinguished the surgical group from the nonsurgical group, which we coined the MIST criteria. These characteristics were (1) Maximum $\text{CO}_2 > 62$ mm Hg; (2) AHI > 23; (3) O_2 Saturation nadir < 79.4%; and (4) Total sleep time with < 90% O_2 saturation of > 5.7%. Interestingly, Fahradyan et al.¹⁹ found higher levels of carbon dioxide (pCO_2) through a heel stick capillary blood gas to better “predict” surgical intervention than PSG, using similar ROC analyses. Both studies suffer from similar methodological problems that inherently stem from identifying predictors for surgery *ex post facto*, as these “predictors” likely influenced the clinical decision to proceed with surgical intervention. Further study is underway.

The wide spectrum of practice patterns seen in our survey may also in part be due to comfort and experiences

in training.^{20,21} In a survey of ACPA members, Collins et al.²⁰ found those trained in certain surgical techniques in RS are likely to perform those techniques at their current institution. This would explain the use of tracheostomy among international respondents, the increased use of MDO by those 0–10 years out of practice, and the preference for mandibular body osteotomies in surgeons >10 years in practice. A study examining practice patterns in Boston found that regional hospitals vary widely in initial treatment modality independent of disease severity, with some resorting to tracheostomy over other methods.²¹ Interestingly, in the same ACPA study performed by Collins in 2010, 48% of respondents perform MDO, 28% perform TLA, and 17% perform tracheostomies as their most common or preferred method.²⁰ In our study that included ACPA membership, we found that 74.2% of surgeons preferred MDO, 12.6% perform TLA, and 6.6% perform tracheostomies as their surgical intervention of choice, suggesting an interval increase of MDO usage as the primary surgical modality over the last 8 years.

Since its original description, variations in technical aspects of mandibular distraction and postoperative protocols have been successfully used. Osteotomy techniques are generally a matter of preference so long as they are able to minimize morbidity to the inferior alveolar nerve and tooth buds and adequately lengthen the mandible. The majority of surgeons performed an inverted L ramus osteotomy or a mandibular angle osteotomy. Inverted L osteotomies were more likely in younger surgeons 0–10 years out in practice and in surgeons that perform >5 MDO a year. Proponents of the inverted L cite the ability to make the vertical osteotomy away from the tooth roots and under direct visualization to avoid the inferior alveolar nerve.²² This may be a reflection of training, as younger surgeons graduating from high volume academic centers may be inclined to utilize newer techniques.

The majority of surgeons in our study utilized a 1–3 day latency phase, while a quarter of surgeons did not incorporate a latency phase. A landmark survey of 3,278 distraction cases by Mofid et al.²³ found an increase in self-reported premature consolidation among surgeons using a latency phase, on average 4.9 days. There are some clinical reports of eliminating the latency phase all together. Surgeons who eliminate the latency phase may rationalize this choice by noting that the initial distraction protocols were devised for the lower extremity in older patients, who are less likely to consolidate the distraction gap within a short period of time. Furthermore, distraction devices have inherent “play” in them by resistance of soft tissue and turns left in the distraction arm before the osteotomy sites actually distract.²⁴ In clinical series, there was no difference in clinical outcome when the latency phase was eliminated.^{25,26} With regard to the activation phase, surgeons were split, with one half distracting 1.0 mm/d (in accordance to McCarthy’s original protocol), and the other half distracting 1.5–2.0 mm/d.²⁷ Clinical study has demonstrated increasing distraction to 2.0 mm/d does not result in fibrous union or premature consolidation.²⁵ Further clarification of the age at which surgeons are distracting the majority of their patients

would help clarify discrepancies. Despite variations in protocols, most surgeons in our study had self-reported high rates of successfully relieving upper airway obstruction with mandibular distraction, similar to findings in previous studies.²⁰

There are several limitations to our study. Our relatively low response rate likely reflects the small proportion of surgeons who routinely treat RS. A selection bias must be considered, as many of the treatment modalities and diagnostic tests likely reflect the practice patterns of those who are highly specialized in treating this heterogeneous condition. Further, our study was not designed to elucidate subtleties in management among each practitioner's algorithm. This would be outside of the scope of a 5-minute survey. Further investigations may clarify the nuances, clinical characteristics, and syndromes that can affect overall management. "Catch-up" growth, which behooves consideration when considering intervention, is a well-debated phenomenon.^{15,28–32} Although current longitudinal studies have failed to clearly demonstrate its existence, it is clear syndromic mandibles are different in morphology and physiology when compared with the nonsyndromic variants.^{28,33,34} Careful consideration to the pathogenic diagnosis is paramount in successful treatment. Ultimately, this survey was able to establish clear trends in the current workup and surgical management of this heterogeneous sequence. However, the need for large scale multi-institutional trials establishing treatment guidelines is ever apparent.

CONCLUSIONS

The current study demonstrated that while the majority of respondents seem to have developed successful, institutional specific algorithms for the treatment of upper airway obstruction in infants with severe RS, specific differences are evident, particularly between North American versus international craniofacial surgeons, and between those who were 0–10 years versus greater than 10 years of practice. Nevertheless, broad trends have been documented, including the use of DLB and PSG by the majority of respondents. MDO was the most favored primary surgical intervention, a trend increasing among newer generations of craniofacial surgeons. Large multi-institutional prospective studies are likely necessary to further define the roles various treatment protocols and syndromic diagnoses may play in successfully treating these complex patients, and to develop objective data-driven indications for surgical airway management.

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