

Airway Events and Critical Care Requirements in Patients with Robin Sequence after Palatoplasty

Stephanie M. Cohen, MD*

Melissa Kanack, MD†

Lisa Nussbaum, MS, MBA‡

Tyler T. Nguyen, BA‡

Cory M. Resnick, DMD, MD‡

Raymond Park, MD§

Faye Evans, MD§

Carolyn R. Rogers-Vizena, MD‡

Ingrid M. Ganske, MD, MPA‡

Background: Patients with Robin sequence (RS) are often thought to be at high-risk for airway complications after cleft palate repair, and may be routinely admitted to the intensive care unit after surgery. This study compares frequency of postoperative airway events in patients with and without RS undergoing palatoplasty, and assesses potential risk factors for needing intensive care.

Methods: A matched cohort study of patients with and without RS undergoing palatoplasty from February 2014 to February 2022 was conducted. Variables of interest included prior management of micrognathia, comorbidities, polysomnography, age and weight at the time of palatoplasty, operative techniques, intubation difficulty, anesthesia duration, and postoperative airway management. Airway events were defined as airway edema, secretions, stridor, laryngospasm, obstruction, and/or desaturation requiring intervention. Logistic regression was performed to identify factors predictive of airway events.

Results: Thirty-three patients with RS and 33 controls were included. There were no statistically significant differences in airway events between groups (eight RS, four controls, $P = 0.30$). Anesthetic duration over 318 minutes was associated with increased risk of postoperative airway events [(OR) 1.02 (1.00–1.04) ($P = 0.04$)] for patients with RS, but not for patients in the control cohort.

Conclusions: Postoperative intensive care unit admission is not universally necessary for patients with RS after palatoplasty if intubation was straightforward and there were no concomitant procedures being performed. Patients with longer anesthesia durations were more likely to have postoperative airway events and may need a higher level of care postoperatively. (*Plast Reconstr Surg Glob Open* 2024; 12:e6080; doi: 10.1097/GOX.0000000000006080; Published online 22 August 2024.)

INTRODUCTION

Robin sequence (RS) is characterized by micrognathia and glossoptosis causing episodes of upper airway obstruction (UAO) during infancy.^{1,2} Patients with RS and cleft palate are often considered to be at high risk of having airway issues following cleft palate repair and, as

a result, may be scheduled to be admitted to the intensive care unit (ICU) postoperatively for airway monitoring. A subset of patients with RS undergo mandibular distraction osteogenesis (MDO) before palate repair to improve UAO, and previous studies have shown that ICU admission is rarely required for these patients after distractor removal.^{3–5}

Mandatory postoperative ICU management for patients with RS may not always be necessary, and requiring every patient with RS to be admitted to the ICU after palate repair could potentially strain acute care resources. The purpose of this study was to review the postoperative respiratory outcomes of patients with RS who underwent cleft palate repair in order to (1) quantify and characterize postoperative respiratory events in patients with RS after palatoplasty, (2) compare respiratory outcomes between patients with RS and healthy controls with isolated cleft palate, and (3) identify risk factors for postoperative respiratory events to be used as criteria for planning postoperative ICU admission.

From the *Department of Surgery, Beth Israel Deaconess Medical Center, Boston, Mass.; †Division of Pediatric Plastic Surgery, Rady Children's Hospital, University of California, San Diego, Calif.; ‡Department of Plastic and Oral Surgery, Boston Children's Hospital, Boston, Mass.; and §Department of Anesthesiology, Boston Children's Hospital, Boston, Mass.

Received for publication March 28, 2024; accepted June 26, 2024.

Presented at the American Cleft Palate Association Annual Meeting, March 2023, Raleigh, North Carolina.

Copyright © 2024 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the [Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 \(CCBY-NC-ND\)](https://creativecommons.org/licenses/by-nc-nd/4.0/), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.

DOI: 10.1097/GOX.0000000000006080

Disclosure statements are at the end of this article, following the correspondence information.

METHODS

A retrospective review of the electronic medical record was performed to identify all patients with RS who underwent cleft palate repair at our tertiary care center from February 2014 to February 2022. Patients were identified by ICD-9 and ICD-10 codes. Patients undergoing primary repair before 14 months of life were included. All charts were reviewed, including operative notes from the palate repair and previous procedures, anesthesia records, polysomnography results, preoperative and postoperative clinic notes, and inpatient progress notes. Anesthesia records were reviewed by a surgeon and an anesthesiologist.

Surgical Techniques

All patients who underwent pre-palatoplasty mandibular distraction underwent distraction to overcorrect the mandibular position by 2–4mm compared with the maxillary alveolar position, which requires advancement of 20–24mm.

Patients with Veau II cleft palate were repaired with a two-flap palatoplasty with vomerine flaps and intravelar veloplasty. Patients with Veau I cleft palate were repaired either with two-flap palatoplasty with intravelar veloplasty or Furlow double-opposing Z-plasty, as per the surgeon's preference.

Case-control Study Design

A case-control study was conducted to determine if there were differences in postoperative respiratory events between patients with RS compared with an otherwise healthy cohort of patients with isolated cleft palate undergoing palatoplasty.

The control cohort consisted of otherwise healthy patients with isolated Veau I and Veau II cleft anatomy undergoing initial cleft palate repair at the same center during the inclusion period. Patients were excluded from the control cohort if they had a diagnosis of RS, obstructive sleep apnea, known severe manifestations of another syndromic diagnosis (ie, 22q11, Apert, other chromosomal abnormalities); were undergoing a secondary cleft palate procedure; or had severe cardiopulmonary disease, neurologic disorders, recent respiratory illness within one month of palate repair, or other major anomalies that would independently necessitate a postoperative admission to the ICU.

Patients with a formal diagnosis of RS were included, as long as they did not have significant medical comorbidities that would have independently necessitated a postoperative ICU admission. Patients were excluded from the RS cohort if they had a history of tracheostomy, hemodynamically significant cardiac disease, respiratory infections requiring steroids within one month of palate repair, known seizure disorders, or significant pulmonary diseases.

Variables

Data collected for both controls and patients with RS included sex, a history of prematurity, birthweight, age and weight at the time of palate repair, a history of gastrostomy

Takeaways

Question: Do all patients with a diagnosis of Robin Sequence universally require intensive care after cleft palate repair?

Findings: Postoperative ICU admission is not always required for patients with RS after palatoplasty if intubation was straightforward, and there were no concomitant procedures being performed. Patients with longer anesthesia durations were more likely to have postoperative airway events and may need a higher level of care in the postoperative period.

Meaning: The decision to admit a patient with Robin Sequence to the ICU postoperatively should be made on a case-by-case basis. Factors such as difficult intubation and longer anesthetic duration should raise heightened awareness for potential need for a higher level of care in the postoperative period.

tube placement, documentation of difficult feeding, concomitant operations performed under the same anesthetic, preoperative polysomnography results, palate repair technique, documentation of difficult intubation at the time of palate repair, number of intubation attempts at time of palate repair, airway events at any point during admission after palate repair, ICU admission (planned and unplanned), postoperative medications administered, and anesthesia time. For all patients who were admitted to the ICU postoperatively, charts were reviewed to determine if the decision to admit each patient to the ICU was determined preoperatively (planned), intraoperatively, or postoperatively. For patients with RS, difficult intubation at the time of mandibular distractor removal as well as all procedures that were performed prior to cleft palate repair were reviewed. Advanced airway techniques were defined as any technique other than conventional direct laryngoscopy. If direct laryngoscopy was successful or if a modified Cormack-Lehane⁶ grade 2 view or better direct view (a partial view of the glottis, or only the arytenoids could be seen) was obtained while using a video laryngoscope during the palate repair or the prior anesthetic encounter, the patient was not considered to have a difficult airway.

Any airway event requiring intervention was documented; these specifically included obstructive events, stridor, desaturation requiring intervention, postoperative use of airway adjuncts (oropharyngeal airway/nasopharyngeal airway), reintubation, inability to extubate in the operating room after cleft repair, and requirement of supplemental oxygen after initial blow-by oxygen in the recovery room.

Statistical Analyses

Propensity-score matching was performed for variables that demonstrated significant baseline differences between the two groups. Differences in categorical or dichotomous variables between patients with RS and controls were determined with Fisher exact tests, and continuous variables were assessed with Wilcoxon rank sum test.

Table 1. Matched Results Comparing Demographic Characteristics and Airway Outcomes after Primary Palatoplasty in Patients with RS Compared with Patients with Isolated Cleft Palate

	RS (n = 33)	Controls (n = 33)	P
Sex			0.62
Female	18	21	
Male	15	12	
Median weight at palate repair	8.49 (7.80, 8.73)	8.32 (7.46, 9.01)	0.62
Median birthweight	3.38 (2.83, 3.57)	3.14 (2.80, 3.50)	0.42
History of prematurity	4	6	0.73
Median age at repair	10.00 (10.00, 11.00)	10.00 (9.00, 10.00)	0.018
Veau anatomy			0.08
Type 1	13	21	
Type 2	20	12	
Repair type			>0.99
Furlow	3	3	
Two-flap	30	30	
Concomitant procedures*	8	2	0.08
Median # intubation attempts	1.00 (1.00, 2.00)	1.00 (1.00, 2.00)	0.19
Difficult intubation	4	0	0.11
Total length of stay	3.00 (2.00, 3.00)	2.00 (2.00, 3.00)	0.008
Airway events	8	4	0.34
Postoperative racemic Epi	3	0	0.24
Postoperative decadron	26	18	0.07
Anesthesia time (min)	231 (212, 274)	186 (139, 251)	<0.001

The reported values are averages unless the median is specified, and interquartile ranges (25th percentile to 75th percentile) are given. Wilcoxon rank sum test was used to compare all continuous variables and Fisher exact test was used to compare all dichotomous variables.

*Procedures other than tympanostomy tube placement.

Subgroup analyses were performed for patients with RS who underwent MDO. In order to identify potential predictors of postoperative airway events, all patients (control and patients with RS) were re-grouped according to whether they had an airway event and then analyzed by Wilcoxon rank sum test for continuous variables and by Fisher exact test for dichotomous variables. Variables that were different between the group that had an airway event and the group that did not have an airway event were then analyzed by both unadjusted and adjusted logistic regression models. All calculations were performed with power of 80% and alpha level of 0.05. Statistical software used was SAS Enterprise Guide Version 8.3 (SAS Institute Inc. Cary, NC) and R Version 4.2.2 (2022-10-31 ucrt).

RESULTS

During the study period, 48 infants with RS had primary cleft palate repairs. After exclusion criteria were applied, 33 patients with RS were included in the study group. Patients with RS were excluded from this study for one or more of the following reasons: seven patients had significant neurologic disease (uncontrolled seizure disorders, history of neonatal strokes/intraventricular hemorrhage, severe hypotonia, severe developmental delay); five patients had hemodynamically significant cardiac disease (truncus arteriosus, coarctation of the aorta, hemodynamically significant atrial or ventricular septal defects); four patients had a history of tracheostomy; one had restrictive lung disease; and two had pulmonary infections within 1 month of surgery. Reasons for exclusion were not mutually exclusive (ie, one patient may have had both a history

of a seizure disorder and a hemodynamically significant cardiac comorbidity).

Of the patients with RS, early management of the airway included nonoperative treatment with side or prone positioning (n = 8), MDO (n = 23), or tongue-lip adhesion (n = 2). All patients underwent palate repair by the time they were 14 months old. Because of institutional practices, 24 (81%) patients with RS were preoperatively planned for postoperative admission to the ICU.

During the same inclusion period, 251 patients without RS underwent primary palatoplasty by 14 months old. After exclusion criteria were applied, 161 patients remained. Patients with submucous cleft palate, Veau III, and Veau IV anatomy were additionally excluded from the cohort. Patients who underwent cleft palate repair at our institution's community hospital satellite locations were also excluded, leaving 44 otherwise healthy patients with isolated Veau I and Veau II cleft palates in the control group.

There were no significant differences in sex, weight, or median age at the time of palate repair between the RS and control groups prior to matching. There were significant differences in palate repair technique (two-flap or Furlow palatoplasty) between groups. Thus, 1:1 propensity-score matching was performed for palate repair technique, after which each group consisted of 33 subjects (Table 1).

The median number of intubation attempts was one for both patients with and without RS, with no significant differences between groups ($P = 0.19$). Four patients in the RS cohort were difficult to intubate compared with zero in the control group ($P = 0.11$). Eight patients with RS had postoperative airway events compared with four patients in the control group ($P = 0.34$). There were significant

Table 2. Qualitative Description of all Postoperative Airway Events

Control (n = 4)	Robin with MDO (n = 6)	Robin without MDO (n = 2)
1) "Failure to move air" after extubation requiring epinephrine and dexamethasone	1) Concern for laryngomalacia and an erythematous/inflamed airway requiring racemic epinephrine and nasal trumpet placement	1) Concern for airway edema requiring nasal trumpet placement, desaturation to 70% requiring additional blow-by-oxygen for 24h postoperatively
2) Postoperative stridor requiring dexamethasone	2) Desaturation to 60% postoperatively requiring racemic epinephrine and blow-by oxygen until postoperative day 1	2) Difficult intubation in the operating room (5 attempts) requiring intubation until postoperative day 1
3) Audible stridor and desaturation on postoperative day 1 to 80% requiring blow-by-oxygen and dexamethasone	3) Desaturation requiring CPAP as well as blow-by oxygen, furosemide for pulmonary edema, and conservative management of a small pneumothorax from barotrauma	
4) Palate bleeding requiring oxymetazoline and tranexamic acid	4) Copious secretions requiring frequent suctioning, concern for upper airway obstruction requiring racemic epinephrine	
	5) Intermittent desaturation due to breath holding requiring blow-by-oxygen	
	6) Stridor and retractions postoperatively requiring reintubation and extubation in the OR on POD 1	

differences in total anesthesia time [RS = 231 minutes (212–274), control = 186 minutes (139–251) ($P < 0.001$)] and length of stay [RS = 3.00 hospital days (2.00–3.00), control = 2.00 hospital days (2.00–3.00) ($P = 0.008$)], both longer in patients with RS (Note: length of stay is reported as hospital days, meaning 2 hospital days indicates that the patients in the control cohort, on average, were discharged on postoperative day 1 after a one night stay). Three patients in the RS cohort required postoperative racemic epinephrine compared with none in the control cohort ($P = 0.24$), and 26 patients with RS received postoperative dexamethasone compared with 18 in the control cohort ($P = 0.07$). Two patients in the RS cohort had postoperative nasopharyngeal airways (NPA) placed, and both were admitted to the ICU. One of these two patients who had a postoperative NPA placed had an airway event. Neither had prepalate repair mandibular distraction.

Of the eight patients with RS who had airway events, five had already been prebooked for the ICU, but three had unplanned ICU admissions. For these three patients with RS who had airway events that prompted unplanned ICU admissions, a higher level of care was required due to (1) multiple intubation attempts with resultant airway edema, necessitating nasal trumpet placement, blow-by oxygen and a 72-hour dexamethasone taper; (2) sustained oxygen desaturations requiring CPAP in the postanesthesia care unit (PACU); and (3) copious secretions requiring frequent suctioning in the PACU and concern for stridor, treated with racemic epinephrine and dexamethasone (Table 1). Four different surgeons performed palate repairs for the 33 patients with RS, and there were no differences in the frequency of postoperative airway events ($P = 0.078$) or postoperative ICU admissions ($P = 0.667$) when stratified by surgeon.

Of the four control patients who had postoperative airway events, three were managed in a standard surgical postoperative unit with supplemental oxygen, suctioning, dexamethasone and/or racemic epinephrine, with only one required unanticipated admission to the ICU. None of the patients in the control cohort had been prebooked for admission to the ICU before the palate repair (Table 2).

Before palate repair, all 23 patients who underwent MDO underwent both pre- and postdistraction polysomnography showed improvement in apnea-hypopnea index

score, with a mean improvement of 26.1 ± 16.8 . However, despite showing improvement, three patients still had more than five apnea-hypopnea index events per hour before palate repair due to mixed central and obstructive events. None of these patients had postoperative respiratory events after palate repair. Patients who did not undergo MDO had heterogeneous usage of polysomnography. Two had studies early in infancy demonstrating obstructive sleep apnea without subsequent follow-up sleep studies.

Nearly all patients in both groups (87.9% RS and 93.9% controls) underwent bilateral myringotomy tube placement at the time of palate repair. In the control cohort, two patients additionally underwent frenulotomy during the same anesthetic. Eight patients with RS underwent additional procedures, including frenulotomy (1), distractor removals (2), epiblepharon repair (1), tongue-lip adhesion takedowns (2), nevus removal (1), and circumcision (1). Although there can be concern about whether frenulotomy at the time of palate repair may increase risk of tongue-based obstruction postoperatively, no patient who had a frenulotomy in either cohort experienced postoperative airway events. In the RS cohort, one of the two patients who underwent tongue-lip adhesion takedown and one of the two patients who underwent distractor removal had an airway event.

Eleven patients undergoing distraction had documentation of difficult intubation during distractor placement. None of these patients were considered to have a difficult intubation at the time of distractor removal. However, three of the patients who had not previously been described as having a difficult airway were deemed to have a difficult airway at time of palate repair and required an advanced airway technique for intubation. None of these patients, however, had a postoperative airway event. None of the patients who underwent mandibular distraction before cleft palate repair had postoperative complications after distraction, and the full length of planned distraction had been achieved prior to cleft palate repair.

In order to identify potential predictors of postoperative airway events, all 33 study subjects were re-grouped by whether they had an airway event (Table 3).

Children who had airway events weighed less at the time of palate repair (7.81 kg versus 8.55 kg, $P = 0.05$), were considered difficult to intubate (three patients versus one

Table 3. Factors Associated with Postoperative Airway Events in Patients with RS

Characteristic	No Airway Event (n = 25)	Postoperative Airway Event (n = 8)	P
Sex			0.42
Female	60% (15/25)	38% (3/8)	
Male	40% (10/25)	62% (5/8)	
History of prematurity	3 (12%)	2 (25%)	0.57
History of feeding difficulty	9 (36%)	6 (75%)	0.10
Veau anatomy			0.11
1	48% (12/25)	12% (1/8)	
2	52% (13/25)	88% (7/8)	
Weight at repair	8.55 (7.95–8.80)	7.81 (7.14–8.26)	0.046
Birthweight	3.41 (3.10–3.60)	2.83 (2.40–3.30)	0.08
Gastrostomy tube dependence	7 (29%)	5 (62%)	0.11
Age	10.00 (10.00–11.00)	10.00 (9.00–11.00)	0.36
Repair type			>0.99
Furlow	8% (2/25)	12% (1/8)	
Two-flap	92% (23/25)	88% (7/8)	
Difficult intubation during palate repair			0.036
N	96% (24/25)	62% (5/8)	
Y	4% (1/25)	38% (3/8)	
Concern apnea preoperatively			>0.99
N	92% (23/25)	88% (7/8)	
Y	8% (2/25)	12% (1/8)	
Mandibular distraction osteogenesis			>0.99
N	32% (8/25)	25% (2/8)	
Y	68% (17/25)	75% (6/8)	
Median intubation attempts	1.00 (1.00–2.00)	2.00 (1.50–4.50)	0.09
Intraoperative morphine (mg/kg)	0.33 (0.19–0.41)	0.29 (0.24–0.36)	0.75
ICU LOS	1.00 (1.00–2.00)	3.50 (2.00–4.50)	0.004
Total LOS	3.00 (2.00–3.00)	4.50 (3.00–5.00)	0.014
Dexamethasone			0.15
N	28% (7/25)	0% (0/8)	
Y	72% (18/25)	100% (8/8)	
Anesthesia time (min)	223 (207–266)	317 (260–339)	0.016

P values from Wilcoxon rank sum test and Fisher exact tests.

Table 4. Logistic Regression for Variables Predictive of Postoperative Airway Events after Palatoplasty in Patients with RS

Predictor	Odds Ratio (95% CI)	P
Unadjusted models		
Anesthesia time (min)	1.02 (1.00–1.04)	0.02
Weight at the time of palate repair (kg)	0.36 (0.12–1.05)	0.06
Difficult intubation	14.40 (1.23–168.50)	0.03
Adjusted model*		
Anesthesia time (min)	1.02 (1.00–1.04)	0.04
Weight at the time of palate repair (kg)	0.48 (0.14–1.70)	0.26
Difficult intubation	9.93 (0.53–186.85)	0.13

*Model includes all three covariates.

patient, $P = 0.04$), and had longer anesthesia times (317 minutes compared with 223 minutes, $P = 0.016$). When unadjusted logistic regression was performed for the RS group for these variables, both anesthesia time [OR 1.02 (1.00–1.04), ($P = 0.02$)] and difficult intubation [OR 14.40 (1.23–168.50), ($P = 0.03$)] were significant, whereas weight at the time of palate repair was not significant [OR 0.36 (0.12–1.05), ($P = 0.06$)]. However, when all three variables were included in the same adjusted model (Table 4), for the RS cohort, only anesthesia time remained significant [OR 1.02 (1.00–1.04), ($P = 0.04$)]. When combining both

the RS and control cohorts, anesthesia time remained a significant predictor of postoperative airway events [OR = 1.013 (95% CI (1.003–1.024), ($P = 0.015$)]]. For controls alone, anesthesia time is not a significant predictor of having a postoperative airway event [OR 1.002 (95% CI (0.984–1.020), ($P = 0.852$)]].

DISCUSSION

The aim of this investigation was to understand postoperative airway outcomes and critical care requirements

of patients with RS after cleft palate repair. In our study sample, we did not find a statistically significant difference in the number of postoperative airway events after cleft palate repair in patients with RS compared with otherwise healthy patients undergoing palatoplasty. Institutional norms dictated that 27 patients with RS had planned ICU admissions following palatoplasty, of which only eight actually had postoperative airway events necessitating that level of care. In comparison, four patients without RS had postoperative airway events following palatoplasty, and only one patient in this group required ICU admission. There were likely a variety of reasons as to why certain patients were not prearranged for admission to the ICU after palatoplasty, including minor Robin phenotype, lack of comorbidities, provider variation in tolerance for trial-and-error admission to the floor, and resource availability.

The only factor we identified that was associated with an increased likelihood of postoperative airway events was anesthesia time. However, increased anesthesia time could be related to difficult intubation because it could take longer to intubate these patients. Additionally, increased anesthesia time could also have resulted from performing concomitant procedures under the same anesthetic. Or, the increased time could have been related to the additional challenge of completing the operation in a child with difficult intubation likely related to abnormal oropharyngeal anatomy. Based on our data, the point at which anesthesia time became predictive of having a postoperative airway event was at 318 minutes, and thus, this time point should be considered when deciding to admit a patient to the ICU postoperatively.

In the RS cohort, there were eight additional procedures performed under the same anesthetic, whereas there were only two additional procedures performed in the control cohort, although the specific types of procedures varied. Frenulotomy in particular may result in tongue edema and UAO⁷; however, the only patient who underwent frenulotomy did not have any postoperative airway problems. One of two patients who underwent distractor removal and one of two patients who underwent tongue-lip adhesion take-down concomitant to palate repair had airway events. Although our sample size is too small to specifically assess differences in outcomes for coordinating other associated procedures, this study found that half of patients undergoing such simultaneous operations had airway events. There should be heightened awareness and further investigation into the potential need for postoperative ICU admission in these patients. Longer overall anesthesia time could also result from longer palatoplasty surgical time. It is also possible that patients with more challenging cleft anatomy could have had longer operative time. Longer palatoplasty time would require additional duration of Dingman retractor placement and potentially increase tongue edema.⁸ Unfortunately, it is not possible to discern how much time was spent performing each procedure because procedure start time is recorded for the overall surgical experience and not by procedure.

Patients who did not have difficult airways during the most recent previous anesthetic exposure at our hospital

(distractor removal prior to palate repair, gastrostomy tube placement, hernia repair, etc.), also did not have postoperative airway issues after palate repair. In fact, Other studies have shown that patients with RS typically experience resolution of airway obstruction after MDO, that they rarely require an advanced airway team for future intubations, and they do not have a higher rate of postoperative respiratory events.^{4,9} In a recent study analyzing postoperative airway events and critical care requirements of 25 patients with RS after mandibular distractor removal, five patients (20%) had a postoperative airway event.⁴ Of these, only two required intervention, and the others resolved spontaneously. Both had undergone recent major cardiac surgery, which would have excluded them from our study, because they would have likely needed ICU admission due to preexisting conditions. In contrast to our study, the only predictive variable of postoperative airway events was congenital cardiac disease.

Costa and colleagues investigated airway compromise following palatoplasty in patients with RS.¹⁰ The authors found that patients with RS with comorbid histories of cardiac, lower respiratory, and/or gastrointestinal anomalies were more likely to have airway complications. Like our present study, the authors concluded that infants with RS in general, had no greater risk of airway compromise than patients without. Our study is unique in that we intentionally selected an otherwise healthy cohort of patients with isolated RS to compare with the nonsyndromic control cohort. Neither of our cohorts have significant comorbidities, which allowed us to understand the risk that RS poses alone.

In a case series that looked at postoperative respiratory complications after two-staged Furlow palatoplasty in patients with RS,¹¹ four of 92 (4.35%) patients experienced significant respiratory distress postoperatively, and only two of them required ICU admission. However, unlike in our RS cohort in which the majority underwent MDO to manage airway obstruction, the patients in their study were managed conservatively with oropharyngeal airways. Furthermore, they had a two-staged palatoplasty, and it is possible that leaving the hard palate open affected the postoperative airway dynamics. However, consistent with our unadjusted model, the authors found that difficult intubation at the time of the palate repair was associated with postoperative respiratory issues.

Like all investigations, there are limitations to this present study. The incidence of RS ranges from one of 8500 to one of 14,000 live births¹²; thus, the rarity of this condition as well as exclusion of patients with other significant medical comorbidities narrowed our sample size. And, although we performed propensity-score matching for cleft palate repair type, matching was not perfect because age at the time of repair became significant after matching (although age was not clinically meaningful because the median remained 10 months in both groups). Although our findings of 24% of patients with RS having a postoperative respiratory event are consistent with the literature,¹³ the sample size was likely too small to determine if this is different from patients who do not have RS. Based on our study size of 33 patients in each cohort, an

airway event would have had to occur 3.9 times more frequently in one group compared with the other to be statistically significant. Although our study found anesthesia time correlated with postoperative airway events, we were unable to accurately discern how much time was spent on each procedure or how long it took to intubate each patient in order to more fully understand the contributors to anesthesia time. Additionally, there is variation in nursing documentation, and thus, frequency of suctioning and administration of additional oxygen after the immediate postoperative period may not always have been accurately documented.

Although diagnostic and treatment algorithms exist with regard to choice of positioning, nasogastric tube, tongue-lip adhesion, and MDO to manage obstruction,¹⁴ there is currently no algorithm that suggests how patients should be managed after cleft palate repair. Although there is no one-size-fits-all pathway for patients with RS, our study found risk factors for adverse airway events, namely difficult intubation and prolonged anesthetic time. Additionally, we found that most patients with RS do well after palatoplasty and do not require the resources of an ICU setting. Thus, we recommend a patient-specific approach to ICU admission rather than obligatory admission based on this diagnosis. Although not all cases of difficult airway or prolonged anesthetic time can be anticipated in advance, factors such as a particularly challenging cleft palate or need for multiple procedures under the same anesthetic should alert the provider to the value of postoperative ICU monitoring in those individuals.

CONCLUSIONS

Based on this study, not all patients with RS require ICU level of care after palate repair. Decision to admit these patients to the ICU postoperatively should be made on a case-by-case basis. There should be heightened awareness that those with prolonged anesthetic times may be more likely to have postoperative respiratory events.

Ingrid M. Ganske, MD, MPA

Department of Plastic and Oral Surgery

Boston Children's Hospital

300 Longwood Avenue; Enders 123

Boston, MA 02115

E-mail: ingrid.ganske@childrens.harvard.edu

DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

REFERENCES

1. Mackay DR. Controversies in the diagnosis and management of the Robin sequence. *J Craniofac Surg.* 2011;22:415–420.
2. Wang C, Shi B, Li J. Management of cleft palate among patients with Pierre Robin sequence. *Br J Oral Maxillofac Surg.* 2023;61:475–481.
3. Chocron Y, Barone N, Zammit D, et al. Efficacy and complications of mandibular distraction osteogenesis for airway obstruction in the Robin sequence population: a comprehensive literature review. *J Craniofac Surg.* 2022;33:1739–1744.
4. Resnick CM, Caprio R, Evans F, et al. Is intensive care unit admission necessary after removal of mandibular distraction devices in infants with robin sequence? *Cleft Palate Craniofac J.* 2021;58:306–312.
5. Breugem CC, Logjes RJH, Nolte JW, et al. Advantages and disadvantages of mandibular distraction in Robin sequence. *Semin Fetal Neonatal Med.* 2021;26:101283.
6. Yentis SM, Lee DJ. Evaluation of an improved scoring system for the grading of direct laryngoscopy. *Anaesthesia.* 1998;53:1041–1044.
7. Genther DJ, Skinner ML, Bailey PJ, et al. Airway obstruction after lingual frenulectomy in two infants with Pierre-Robin sequence. *Int J Pediatr Otorhinolaryngol.* 2015;79:1592–1594.
8. Sherif RD, Sanati-Mehrizy P, Taub PJ. Lingual pressure during Dingman-assisted cleft palate repair: an investigatory case series. *Cleft Palate Craniofac J.* 2018;55:312–315.
9. Pius L, Jindal S, Resnick CM. Is a difficult airway team needed for intubation at removal of mandibular distraction devices for infants with Robin sequence? *J Oral Maxillofac Surg.* 2023;81:716–720.
10. Costa MA, Murage KP, Tholpady SS, et al. Airway compromise following palatoplasty in Robin sequence: improving safety and predictability. *Plast Reconstr Surg.* 2014;134:937e–945e.
11. Opdenakker Y, Swennen G, Pottel L, et al. Postoperative respiratory complications after cleft palate closure in patients with Pierre Robin sequence: operative considerations. *J Craniofac Surg.* 2017;28:1950–1954.
12. Gangopadhyay N, Mendonca DA, Woo AS. Pierre robin sequence. *Semin Plast Surg.* 2012;26:76–82.
13. van Lieshout MJS, Voshol IE, Joosten KFM, et al. Respiratory distress following cleft palate repair in children with Robin sequence. *Cleft Palate Craniofac J.* 2016;53:203–209.
14. Gómez OJ, Barón OI, Peñarredonda ML. Pierre Robin sequence: an evidence-based treatment proposal. *J Craniofac Surg.* 2018;29:332–338.