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Defining failure and its predictors in mandibular distraction for Robin sequence

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ABSTRACT

Introduction: Robin sequence (RS) is defined as the triad of micrognathia, glossoptosis and airway obstruction. A popular surgical treatment is mandibular distraction osteogenesis (MDO). In this study, it is demonstrated that the associated variables change, dependent on the manner in which failure is defined. These multiple failure outcomes are used to construct a scoring system to predict MDO failure.
Methods: A retrospective database of neonatal MDO patients was constructed. Failure outcomes studied included tracheostomy; a decrease in the apnea-hypopnea index (AHI) but an AHI >20; and death. A combination of bivariate and regression analysis was used to produce significantly associated variables and a scoring system.
Results: Statistical analysis demonstrated the association of gastroesophageal reflux; age >30 days; neurologic anomaly; airway anomalies, other than laryngomalacia; an intact palate; and pre-operative intubation on the outcome variables studied. Multiple scoring systems were produced with reasonable sensitivity, specificity, and positive and negative predictive value.
Conclusions: When reporting surgical outcomes of MDO in the setting of RS, it is important to consider the AHI as well as avoidance of tracheostomy as an outcome variable. Incomplete amelioration of AHI accounts for half of the patients with a problem after MDO. The predictive scores presented will be used and validated on a larger, prospectively collected dataset.

1. Introduction

Upper airway obstruction caused by micrognathia and subsequent glossoptosis with or without cleft palate defines the triad of Robin sequence (RS) (Robin, 1929, 1934). Affected patients may present with airway obstruction causing detriment to breathing, growth, neurocognitive development and, in advanced cases, life threatening airway stenosis. Indeed mortality associated with Robin sequence is reported to be between 1.7 and 65% (Costa et al., 2014). Surgical intervention has been reserved for patients with severe airway obstruction in which conservative treatment has been unsuccessful. Mandibular distraction osteogenesis (MDO) is the first line of surgical therapy at many craniofacial centers treating Robin sequence. MDO has been demonstrated as a more functional and cost-effective alternative to tracheostomy (Kohan et al., 2010; Hong et al., 2012) and a more effective intervention compared with tongue-lip adhesion in the treatment of isolated Robin sequence (Flores et al., 2014).

Several investigators have reported on the efficacy of MDO in relieving airway obstruction in the RS population (Denny et al., 2001; Denny and Kalantarian, 2002; Monasterio et al., 2002; Denny, 2004; Mandell et al., 2004; Wittenborn et al., 2004; Burstein and Williams, 2005; Dauria and Marsh, 2008; Iatrou et al., 2010; Cascone et al., 2014). Critical appraisal of the literature demonstrates that the definition of successful distraction varies across studies (Denny et al., 2001; Denny and Kalantarian, 2002; Monasterio et al., 2002; Denny, 2004; Mandell et al., 2004; Wittenborn et al., 2004; Burstein and Williams, 2005; Dauria and

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Marsh, 2008; Paes et al., 2013; Flores et al., 2014; Lam et al., 2014; Murage et al., 2014; Rachmiel et al., 2014; Runyan et al., 2014; Tahiri et al., 2014) and can include resolution of apnea by clinical exam or polysomnogram (PSG) improvement; avoidance of tracheostomy; changes in airway obstruction patterns; or mortality. Unfortunately, this variation in definitions creates a confounder in determining patient characteristics leading to favorable or unfavorable results and is problematic to formulating definitive treatment protocols of care.

The main hypothesis of this study is that the variables associated with MDO failure depend on the definition of success for this patient population. A total institutional outcomes analysis for the treatment of MDO was performed with regard to the varying dependent definitions of success. These variables were then used to construct a tool with which failure could be predicted. This information will provide greater clarity in the analysis of surgical outcomes of MDO and draws attention to the need for standardized assessment of surgical outcomes in this challenging patient population. It also provides a set of pre-operative variables that can assist the clinician in patient counseling.

2. Material and methods

Institutional Review Board approval was obtained prior to the start of this study. A 10-year retrospective review was conducted of all patients with RS treated with MDO at a single tertiary care children's hospital between 2003 and 2012. RS was defined as micrognathia, glossoptosis, and airway obstruction with or without cleft palate. Study patients required a clinical follow-up of at least one year, pre-operative laryngoscopy/bronchoscopy, and preoperative and postoperative PSGs, unless precluded by tracheostomy, intubation, or other airway intervention. Patients were not excluded based on secondary diagnosis or age at the time of distraction.

Work up and indication for distraction was based on a previously described, institutionally-derived protocol (Murage et al., 2013, 2014; Flores et al., 2014). A multidisciplinary team with members from plastic surgery, neonatology, genetics, pulmonology, otolaryngology, and nursing participated in patient assessment and surgical indications. Briefly, patients with airway obstruction unresponsive to conservative airway interventions were assessed by PSG. Those patients with an apnea/hypopnea index (AHI) >20 or significant CO_2 retention were considered for MDO unless central sleep apnea was noted. Prior to surgery, non-contrast computed tomography (CT) of the maxillofacial skeleton was obtained to assess mandibular form, bone quality, associated malformations of the condyle, or TMJ ankylosis. A laryngoscopy and bronchoscopy was also obtained prior to distraction to rule out secondary airway anomalies which could preclude successful MDO. Laryngomalacia was not a contraindication to distraction.

Mandibular distraction was performed using a Risdon incision placed 1 cm inferior to the mandibular border. A vertical ramus osteotomy was performed with a sagittal saw in combination with a coronoidectomy. A micro Zurich mandibular distractor (KLS Martin, Jacksonville, FL, USA) was then applied using a horizontal vector. The activation arm exited anteriorly or posteriorly according to each surgeon's preference. After a latency period of 5 days, activation commenced at a rate of 1 mm/day to the maximal allowable length of the distraction device (20–30 mm). Devices were removed in a second operation after 8 weeks of consolidation.

Multiple patient variables were recorded to correlate with the surgical outcomes of MDO. These included: sex, age, low birth weight (LBW, defined as <2500 g), intrauterine growth retardation (IUGR), prematurity (defined as <37 weeks gestation), age at the time of surgery, presurgical intubation, presence of a cleft palate, syndromic or genetic anomaly, cardiac anomaly, central nervous system (CNS) anomaly, respiratory anomaly excluding laryngomalacia, gastrointestinal (GI) anomaly, gastroesophageal reflux (GER), genitourinary (GU) anomaly, or other system anomaly. Outcome variables defining failure were: a decrease in AHI but still above 20, the need for post-intervention tracheostomy, and death. Repeat distraction was considered under the same paradigm but not considered to be a failure unless it led to no further reduction in AHI, tracheostomy, or death.

Statistical analysis was performed using SAS for Windows (SAS Institute, Cary, NC, USA). Changes in AHI in response to surgery were assessed using a paired *t*-test. A chi-square test was used to analyze each dependent variable's effect on the failure of MDO as classified by: an AHI not decreasing below 20; the need for tracheostomy; or death. Statistical significance was defined as $p \leq 0.05$. Identified variables that had a statistically significant association with failure were then used to construct a scoring system that was tested for the best sensitivity, specificity, and positive and negative predictive value. The receiver operating characteristic (ROC) curves for each was then calculated in order to stratify well-performing predictive tests from poorly performing ones.

3. Results

3.1. Study demographics

Eighty-one patients met the inclusion criteria for this study. Patient characteristics included a mean age of 33.5 days at operation, a mean birth weight of 2.92 kg, and a mean operative weight of 3.36 kg. Other systemic anomaly data was collected as per previous studies. All demographic data is represented in Table 1. These variables include: male sex (58.02%); LBW (29.63%); premature (24.69%); GER (41.98%); Nissen (14.81%); gastrostomy tube (67.9%); laryngomalacia (25.93%); syndromic (30.86%); cleft palate (83.75%); isolated RS (20.99%); CNS anomaly (22.22%); cardiac anomaly (24.69%); GI anomaly (2.47%); GU anomaly (14.81%); airway anomaly (other than laryngomalacia) (34.57%); other

Table 1
Pre-operative demographics of mandibular distraction osteogenesis study patients.

	Mean, *n* (%)
Age (days)	33.49
Birth weight (kg)	2.92
Weight (kg)	3.36
Male	47 (58.02)
Female	34 (41.98)
LBW	24 (29.63)
IUGR	24 (29.63)
Premature	20 (24.69)
GER	34 (41.98)
Nissen	12 (14.81)
Gastrostomy tube	55 (67.9)
Laryngomalacia	21 (25.93)
Syndromic	25 (30.86)
Cleft palate	67 (83.75)
Isolated RS	17 (20.99)
CNS anomaly	18 (22.22)
Cardiac anomaly	20 (24.69)
GI anomaly	2 (2.47)
GU anomaly	12 (14.81)
Other airway anomaly	28 (34.57)
Other anomaly	21 (25.93)
Intubated	6 (7.41)

CNS: central nervous system; GER: gastroesophageal reflux; GI: gastrointestinal; GU: genitourinary; IUGR: intrauterine growth restriction; LBW: low birth weight; RS: Robin sequence.

congenital anomaly (25.93%); and pre-operatively intubated (7.41%).

3.2. Bivariate and regression analysis of variables associated with failure

Failure was defined as follows, with parenthesized numbers indicating the number of patients within that group: need for tracheostomy (7), death due to apneic disease (1), AHI >20 after distraction (6), failure due to tracheostomy or insufficient reduction in AHI (12), any of these failures (13), and all failures as well as all-cause mortality (16). These failures were then analyzed in a bivariate fashion to reveal variables that were specifically associated with each cluster of failure variables. Table 2 outlines all variables in this analysis. Values in bold indicate variables significantly associated with failure.

In this analysis of specific causes of failure, certain variables were important across all types of failure. These include **G**ER, **A**ge >30 days, **N**eurologic anomaly, airway anomalies **O**ther than laryngomalacia, **I**ntact palate, and pre-operative in**T**ubation. Paired *t*-test analysis for numeric variables demonstrated an age of approximately 30 days as being significant in failure by tracheostomy, AHI, and any failure (Table 3). Interestingly, there was a trend towards failure in children below 2.5 kg birth weight, but this only reached significance in the failure by tracheostomy or AHI >20 group.

3.3. Construction of a tool to predict failure in the MDO population

Elucidation of variables associated with failure provided the material with which to create a scoring system for the prediction of failure of MDO. The variables assessed were **G**ER, **A**ge >30 days, **N**eurologic anomaly, airway anomalies **O**ther than laryngomalacia, **I**ntact palate, and pre-operative in**T**ubation. Scores were created for every variation possible for these variables. A sample of the analysis is demonstrated in Table 4. The top eight scores by ROC curve analysis were listed for each mode of failure. ROC curve analysis was performed for the outcome variable denoting failures due to all causes (Fig. 1).

4. Discussion

There have been multiple publications demonstrating the effectiveness of MDO in relieving airway obstruction in patients affected by severe airway stenosis secondary to Robin sequence (Denny et al., 2001; Denny and Kalantarian, 2002; Denny, 2004). As a result MDO is increasingly used as a first line intervention for the surgical treatment of MDO. Unfortunately, standardized protocols of assessment and intervention have not yet been formulated to treat this challenging patient population. To construct these standardized care plans, a consistent means of assessing surgical outcomes needs to be defined. The current literature demonstrates varying definitions of 'failure' of MDO including: the clinical presence of apnea; an objective drop in AHI; the need for tracheostomy, redistraction, or other airway procedures; and death (Dauria and Marsh, 2008; Paes et al., 2013; Papoff et al., 2013; Flores et al., 2014; Lam et al., 2014; Tahiri et al., 2014). Agreement on the definition of failure is critical to assessing differing patient variables associated with successful and unsuccessful distraction and is ultimately required to create definitive treatment protocols.

In this study it is shown that differing definitions of successful distraction not only have an effect on the success rate of distraction but also implicate differing sets of patient variables associated with unsuccessful distraction (Table 2). An almost equal number of patients were characterized as failures by need for tracheostomy ($n = 7$) and inadequate improvement of AHI ($n = 6$). Furthermore, an additional patient died from apnea-related disease. Commonly, the success rate is defined as avoidance of tracheostomy; if this measure is used, only 50% of patients with a problem would be identified.

The variables associated with failure of distraction are also affected by the definitions of failure. This can most clearly be seen in Table 2. The table provides an easily visualized data representation of important variables of failure across differing definitions. When failure is defined by avoidance of tracheostomy, the previously described standard variables appear as important: CNS

Table 2
Bivariate analysis of pre-operative demographic variables against all causes of failure.

	% (*p* value)			
	Failure by tracheostomy	Failure by AHI	Any failure	Any failure + deceased
Total	8.64%	8.11%	16.67%	19.75%
Male	12.77 (0.229)	9.3 (1)	21.74 (0.219)	23.4 (0.405)
Female	2.94 (0.229)	6.45 (1)	9.38 (0.219)	14.71 (0.405)
LBW	16.67 (0.187)	5.26 (1)	23.81 (0.32)	33.33 (0.066)
IUGR	4.17 (0.668)	14.29 (0.343)	18.18 (1)	25 (0.543)
Premature	20 (0.059)	11.76 (0.616)	31.58 (0.073)	30 (0.206)
Isolated RS	0 (0.335)	5.88 (1)	5.88 (0.277)	5.88 (0.171)
CNS anomaly	**22.22 (0.04)**	13.33 (0.595)	29.41 (0.143)	**38.89 (0.039)**
Cardiac anomaly	10 (1)	5.26 (1)	15 (1)	20 (1)
GI anomaly	50 (0.166)	0 (1)	50 (0.307)	50 (0.358)
GU anomaly	8.33 (1)	0 (0.588)	9.09 (0.68)	8.33 (0.443)
Other anomalies	4.76 (0.67)	5.88 (1)	15.79 (1)	23.81 (0.751)
GER	**17.65 (0.038)**	13.79 (0.202)	**28.13 (0.032)**	29.41 (0.09)
NISSEN	**41.67 (<0.0001)**	22.22 (0.153)	**54.55 (0.002)**	**50 (0.011)**
Gastrostomy	12.73 (0.09)	12.24 (0.091)	**23.08 (0.05)**	25.45 (0.077)
Other airway anomalies	**17.86 (0.045)**	11.54 (0.659)	28.57 (0.055)	28.57 (0.158)
Laryngomalacia	19.05 (0.07)	10 (0.659)	28.57 (0.1)	28.57 (0.339)
Syndromic	8 (1)	4.76 (0.668)	16.67 (1)	20 (1)
Intact palate	**30.77 (0.012)**	16.67 (0.249)	**41.67 (0.024)**	38.46 (0.122)
Age >30 days	**19.23 (0.031)**	12.50 (0.38)	26.92 (0.11)	**34.62 (0.034)**
Intubated	**50 (0.007)**	50 (0.15)	**80 (0.002)**	**66.67 (0.012)**

AHI: apnea-hypopnea index; CNS: central nervous system; GER: gastroesophageal reflux; GI: gastrointestinal; GU: genitourinary; IUGR: intrauterine growth restriction; LBW: low birth weight; RS: Robin sequence.
Significant values ($p < 0.05$) are listed in bold.

Table 3
Paired *t*-test of numeric variables between mandibular distraction osteogenesis successes and failures.

	Failure by tracheostomy			Failure by AHI			Any failure			Any failure + deceases		
	No	Yes	*p* value	No	Yes	*p* value	No	Yes	*p* value	No	Yes	*p* value
n	74	7	–	68	6	–	65	13	-	64	16	–
Birth weight (kg)	2.97	2.46	0.067	3.00	2.69	0.307	3.02	2.61	0.053	**3.03**	**2.50**	**0.006**
Age (days)	**29.9**	**70.5**	**0.001**	32.2	43.0	0.423	**30.8**	**51.7**	**0.037**	**29.6**	**49.3**	**0.031**
Weight (kg)	3.33	3.75	0.417	3.43	3.16	0.419	3.39	3.44	0.898	3.38	3.28	0.707

AHI: apnea-hypopnea index.
Significant values ($p < 0.05$) are listed in bold.

anomalies, GER, intact palate, airway anomalies, and pre-operative intubation. However, when failure is defined as limited improvement in AHI, there are no variables statistically associated with failure. This suggests that multifactorial or unanalyzed variables are influencing failure in this unanalyzed and previously unreported sub-population.

As reported previously, laryngomalacia is not associated with failure of MDO across any of the analyzed variables ($p < 0.05$) (Tholpady et al., 2015). When this supraglottic disease is separated from other airway anomalies, a clear difference can be seen between the two variables. Non-laryngomalacia airway anomalies are associated with failure by tracheostomy and so should still be approached with the knowledge that MDO will not be successful at a higher rate.

The analysis of this patient population provides the basis for score creation, much like the GILLS score (Rogers et al., 2011; Abramowicz et al., 2012). The score is a well-known predictor of success of tongue-lip adhesion (TLA) in the RS population. It has identified **G**ER, **I**ntubation pre-operatively, **L**ate operation, **L**ow birth weight, and **S**yndromic diagnosis as important predictors of success; fewer than three of these predicts a 100% successful TLA. Of these variables GER and intubation pre-operatively were identified as being important in this mandibular distraction study. Low birth weight was not shown to be significant, but approached significance in the deceased population ($p < 0.06$). Syndromic status was not significant.

Interestingly, using paired *t*-test analysis, a breakpoint was identified between successful and unsuccessful MDO with regard to the age at performance of distraction. RS patients below 30 days of age at the time of distraction were more likely to be successful than children older than 2 months. The reasons for this age difference could be many, but this is similar to the GILLS score in that

Table 4
Sensitivity, specificity, positive and negative predictive value for the combinations of the six variables with corresponding receiver operating characteristic curve scores.

Combination of variables analyzed[a]	GAITO	GIANTO	GIT	OAINT	OTIG	TINGO	TING	TONI
Failure by tracheostomy								
Specificity	100	100	85.7	100	100	100	85.7	100
Negative predictive value	22.6	19.4	54.5	28	31.8	25	31.6	43.8
Positive predictive value	100	100	98.6	100	100	100	98.4	100
Sensitivity	67.6	60.8	93.2	75.7	79.7	71.6	82.4	87.8
Area under ROC curve	0.93 (0.86–1)	0.94 (0.87–1)	0.93 (0.86–1)	0.94 (0.88–1)	0.95 (0.9–1)	0.96 (0.9–1)	0.92 (0.82–1)	0.96 (0.92–0.99)
Failure by AHI								
Specificity	83.3	83.3	33.3	66.7	83.3	83.3	50	50
Negative predictive value	18.5	16.1	28.6	18.2	27.8	21.7	21.4	23.1
Positive predictive value	97.9	97.7	94	96.2	98.2	98	95	95.1
Sensitivity	67.6	61.8	92.6	73.5	80.9	73.5	83.8	85.3
Area under ROC curve	0.75 (0.5–1)	0.75 (0.49–1)	0.73 (0.53–0.94)	0.71 (0.47–0.96)	0.77 (0.53–1)	0.75 (0.51–1)	0.72 (0.5–0.95)	0.72 (0.5–0.94)
Any failure								
Specificity	84.6	84.6	53.8	76.9	84.6	84.6	61.5	69.2
Negative predictive value	36.7	32.4	70	40	52.4	42.3	47.1	56.3
Positive predictive value	95.8	95.5	91.2	94.3	96.5	96.2	91.8	93.5
Sensitivity	70.8	64.6	95.4	76.9	84.6	76.9	86.2	89.2
Area under ROC curve	0.8 (0.64–0.97)	0.8 (0.63–0.97)	0.8 (0.66–0.95)	0.79 (0.63–0.95)	0.83 (0.67–0.99)	0.81 (0.65–0.98)	0.78 (0.63–0.94)	0.81 (0.66–0.96)
Any failure including all deaths								
Specificity	75	81.3	43.8	68.8	68.8	68.8	50	56.3
Negative predictive value	38.7	36.1	63.6	44	50	39.3	42.1	56.3
Positive predictive value	92	93.3	87.1	91.1	91.5	90.6	87.1	89.2
Sensitivity	70.8	64.6	93.8	78.5	83.1	73.8	83.1	89.2
Area under ROC curve	0.74 (0.58–0.91)	0.77 (0.63–0.92)	0.72 (0.57–0.87)	0.77 (0.63–0.91)	0.73 (0.56–0.9)	0.76 (0.61–0.9)	0.75 (0.62–0.88)	0.75 (0.61–0.9)

AHI: apnea-hypopnea index; ROC: receiver operating characteristic.
[a] The combinations of variables analyzed are drawn from the following: **G**astroesophageal reflux; **A**ge >30 days; **N**eurologic anomaly; airway anomalies **O**ther than laryngomalacia; **I**ntact palate; and pre-operative intubation.

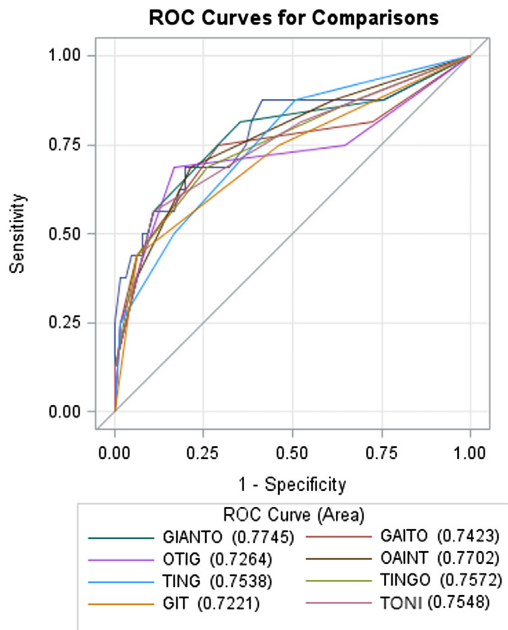

Fig. 1. Receiver operating characteristic (ROC) curve analysis for the top eight predictor variable groupings for outcome variable denoting all failures.

there is a critical period during which an operation has a better chance of success. In the GILLS score, late operation is defined as two weeks; in this study it is 30 days. This indicates there may be physiological changes that become ingrained and are irreversible past a certain age even with intervention.

This report does identify the following patient variables associated with failure of distraction as defined by post-procedure tracheostomy, limited improvement in AHI, or death by apnea: **G**ER, **A**ge >30 days, **N**eurologic anomaly, airway anomalies **O**ther than laryngomalacia, **I**ntact palate, and pre-operative in**T**ubation. Gastrostomy tube and fundoplication were eliminated from the score construction because they are usually procedures that occur after distraction, and so are not useful for prospectively identifying failures of distraction.

These scores were most simply chosen by evaluation of the ROC curves. These are graphical plots that illustrate the performance of a binary classifier system (will MDO work or not) as the discrimination values are varied. It is created by plotting the true positive rate against the false positive rate over differing threshold values. In this case, 0.5 indicates what would be seen when guessing, 1 indicates a perfectly discriminative test, and 0 is completely incorrect. Although no score is perfect, ROC values ranged between 0.7 and 0.9 for all failure variables (Fig. 1).

Depending on the question asked, each score has a differing value. A comparison of the GIT and GIANTO scores illustrate this point. If a score with a high sensitivity is required, the GIT score demonstrates a higher sensitivity and positive predictive value than the GIANTO score. As more variables are added to the score, the specificity increases, at the expense of sensitivity. No score has a 100% specificity and sensitivity, and the needs of the patient and provider determine which test will be useful (Loong, 2013).

These data suggest that a unified definition of successful distraction should be established for consistent assessment of surgical outcomes of MDO as applied to RS patients. The authors suggest this definition would be the tripartite avoidance of tracheostomy, improvement in AHI, and avoidance of mortality associated with airway obstruction. The avoidance of tracheostomy and mortality are commonly used measures; however, these outcomes fail to identify patients who avoid tracheostomy and are still affected by severe airway obstruction.

Decrease in AHI is a metric that requires further attention. AHI measurement requires the use of polysomnography as an assessment tool for quantifying surgical outcomes, and is not consistently used across and within all studies (Denny, 2004; Schaefer et al., 2004; Dauria and Marsh, 2008; Cicchetti et al., 2012). Furthermore, the requirement of an improvement in AHI may identify patients who have avoided tracheostomy but still have a high degree of airway obstruction (AHI >5–10) and as a result, require major supplemental airway support such as continuous positive airway pressure (CPAP) or home oxygen. This scenario demonstrates an incomplete beneficial effect of MDO and therefore cannot be considered a completely successful distraction. The specific definition of 'improvement in AHI' is subject to debate and limited clinical data exists suggesting a normal acceptable range.

An AHI below 5 would be considered acceptable by most specialists treating airway obstruction. The authors, however, would urge caution in following this definition based on their clinical experience in treating Robin sequence. Certainly patients with RS that is isolated or lacks an additional craniofacial anomaly can be expected to respond favorably to mandibular distraction, when indicated. However, patients with severe Treacher Collins or Nager syndrome uncommonly achieve AHI below 5, even in the best of circumstances, precluding them from any possibility of achieving a successful distraction as defined by a 'normal' AHI. Further complicating the issue of a 'normal' AHI is the recent report of isolated cleft lip/palate patients having AHIs above 20 in infancy that then rapidly ameliorate without any intervention except growth (Smith et al., 2014). Defining the normal/acceptable range of airway obstruction in the RS population after MDO is beyond the scope of this report. Based on the clinical data presented, and to improve the classification of successful and unsuccessful operations, the authors suggest an AHI below a certain threshold should be considered as an indicator of successful MDO for the relief of airway obstruction in the RS population.

This study is limited by the retrospective design and single-institutional experience which includes the collective surgical outcomes of multiple surgeons over many years. The patients in this report were treated following a previously published, institutionally derived, treatment protocol (Flores et al., 2014; Murage et al., 2014). It is certainly possible that other surgeons, following different indications for intervention would produce different surgical outcomes.

5. Conclusion

In conclusion, variables significantly associated with failure of distraction are shown to be **G**ER, **A**ge >30 days, **N**eurologic anomaly, airway anomalies **O**ther than laryngomalacia, **I**ntact palate, and pre-operative in**T**ubation. Failure can be defined as: the need for tracheostomy, an incomplete amelioration in AHI, or any cause of death; these dependent outcome variables have different contributing independent variables, with no variable appearing to significantly contribute in patients where there is incomplete amelioration of AHI. The variables allow score construction with varying levels of specificity and sensitivity, depending on the needs of the treating physician and the question asked. These variables will be studied along with others in a larger prospective study on this patient population.

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