Airway Management in Pierre Robin Sequence: The Vancouver Classification

L'ouverture des voies respiratoires en cas de syndrome de Pierre-Robin : la classification de Vancouver

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Abstract

Background: Pierre Robin sequence (PRS) is a triad of micrognathia, glossoptosis, and respiratory distress. There is no standard clinical classification used in the management of neonatal airway in patients with PRS. The goal of our study was to review the presentation and management of patients with PRS and formulate a clinical grading system and treatment algorithm. **Methods:** A 10-year retrospective review of all neonates diagnosed with PRS was performed after obtaining institutional ethics approval. Patients were identified using our cleft lip and palate program database. Inclusion criteria were 2 of the following 3 clinical features—glossoptosis, retrognathia, or airway obstruction. We collected demographic data, clinical information (coexisting airway morbidity, maxillary-mandibular discrepancy, type of intervention used, complications, and outcomes (feeding, length of stay, and airway status) during the first year of life. **Results:** Sixty-three patients met our inclusion criteria. Of these, 55 (87%) had cleft palate and 17 (27%) were syndromic. Forty-eight (76%) patients were managed by prone positioning. Of the 15 surgically managed patients, the initial procedure was floor of mouth release in 7, mandibular distraction osteogenesis (MDO) in 4, and tongue-lip adhesion in 4. Five patients with coexisting airway morbidity needed a second surgery; 2 had MDO and 3 tracheostomies (one patient was later decannulated). Seven (47%) of the surgically managed patients required a gastrostomy tube. **Conclusion:** At present, there is no consensus on neonatal airway management in infants with PRS. From our review of 63 patients with PRS, we hereby propose a simple 4-point classification system and treatment algorithm, based on clinical features.

Résumé

Historique : Le syndrome de Pierre-Robin (SPR) désigne une triade de micrognathie, de glossoptose et de détresse respiratoire. Aucune classification clinique standard n'est utilisée pour assurer l'ouverture des voies respiratoires chez les nouveau-nés présentant un SPR. La présente étude visait à examiner la présentation et la prise en charge des patients ayant un SPR ainsi qu'à formuler un système de classement clinique et un algorithme de traitement. **Méthodologie :** Après avoir obtenu l'approbation du comité d'éthique de leur établissement, les chercheurs ont procédé à une analyse rétrospective sur dix ans de tous les nouveau-nés ayant reçu un diagnostic de SPR. Ils ont recensé les patients dans leur base de données de fentes labiales et palatines. Deux des trois caractéristiques cliniques suivantes constituaient les critères d'inclusion : glossoptose, rétrognathie ou obstruction

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des voies respiratoires. Les chercheurs ont recueilli les données démographiques, l'information clinique (morbidité coexistante des voies respiratoires, malocclusion maxillo-mandibulaire, type d'intervention utilisé, complications et résultats cliniques [alimentation, durée du séjour hospitalier et état des voies respiratoires]) jusqu'à l'âge d'un an. **Résultats :** Soixante-trois patients respectaient les critères d'inclusion. De ce nombre, 55 (87 %) avaient une fente palatine et 17 (27 %), un SPR. Quarante-huit patients (76 %) ont été traités par positionnement en décubitus ventral. Des 15 patients opérés, sept ont subi une libération du plancher buccal, quatre, une ostéogénèse par distraction de la mandibule (ODM) et quatre, une adhésion de la langue et de la lèvre. Cinq patients ayant une morbidité concomitante des voies respiratoires ont dû être opérés une deuxième fois. Deux avaient une ODM et trois, une trachéostomie (un a été décannulé par la suite). Sept patients opérés (47 %) ont eu besoin d'une sonde de gastrostomie. **Conclusion :** À l'heure actuelle, il n'y a pas de consensus sur la prise en charge des voies respiratoires chez les nourrissons ayant un SPR. D'après l'analyse de 63 patients ayant ce syndrome, les auteurs proposent un système de classification simple en quatre points et un algorithme de traitement reposant sur les caractéristiques cliniques.

Keywords

Pierre Robin sequence, classification, mandibular distraction osteogenesis, tongue-lip adhesion, floor of mouth release, respiratory distress, indications

Introduction

Pierre Robin reported a series of neonates with a triad of micrognathia, glossoptosis, and respiratory distress in 1923.¹ The association of cleft palate was added to the sequence in 1934.² Despite earlier descriptions by others, the eponym of Pierre Robin sequence (PRS) has stood the test of time, although confusion in nomenclature remains.³ Regardless, these infants present with respiratory distress, manifested by stridor and desaturations, as well as feeding difficulties and failure to thrive. To add to the confusion, the spectrum of symptom severity varies significantly, from problems with feeding and weight gain, to airway obstruction and sleeping difficulties. This results in a lack of consistency in the management of these patients.

At present, no classification system exists to define patients with PRS. A universal clinical classification system would facilitate better communication between individuals on the multidisciplinary team that are involved in the care of these patients. Furthermore, a classification system would allow guidance in management and comparison of clinical outcomes between institutions.

Upon review of the literature, there is 1 retrospective study that divides infants with PRS into 3 groups according to severity of respiratory symptoms and the mode of feeding. This study was published in 1994, prior to the use of mandibular distraction osteogenesis (MDO) in this population, and does not separate out the different surgical approaches used.⁴ There is a more recent study describing a grading system based on clinical nursing protocol, focused on positioning the infant, need for nasopharyngeal airway, and nasogastric feeding.⁵ Again, this article does not take into account the need for surgical intervention nor the various surgical approaches used to manage respiratory distress in these patients.

Current management of infants with PRS has been recently reviewed⁶ and can be divided into nonsurgical techniques and surgical procedures. Nonsurgical techniques include prone or lateral positioning, nasopharyngeal or endotracheal airway, and continuous positive airway pressure. Surgical interventions

currently used for the management of airway obstruction are floor of mouth release (FMR), tongue-lip adhesion (TLA), MDO, and tracheostomy.

Neonates who are managed successfully with nonsurgical methods have a milder anatomical problem, which is resolved after a period of "partial catch-up" mandibular growth. The latter was demonstrated in a retrospective longitudinal cephalometric study comparing PRS versus isolated cleft palate children versus normal controls.⁷ However, clearly only a subset of patients with PRS exhibit sufficient "catch-up" growth to avoid the need for surgery.⁸

Each surgical intervention comes with inherent risks and benefits. At present, it seems that the surgical approach used by different institutions is dependent on the individual surgeon's preference rather than a logical algorithm.⁹ Since Denny et al published the use of MDO for the management of infants with PRS,¹⁰ this has become increasingly popular in many centres. A recent survey of members of the American Cleft Palate-Craniofacial Association reported that 48% of the 87 respondents preferred the use of MDO for airway management in infants with PRS.¹¹ In another recent study, the senior surgeon switched from TLA to MDO and reported superior outcome in terms of oxygen saturation, apnea-hypopnea index, and need for tracheostomy.¹² Some authors reserve MDO for patients with failed TLA or FMR.¹³⁻¹⁵

Due to the relatively low incidence of PRS, coupled by the fact that only a minority of patients require surgical intervention, there is a lack of prospective comparison studies between MDO, TLA, and FMR. The ultimate goal in these infants is to perform a single operation that will successfully manage the airway, improve the ability to feed, and avoid the morbidity of tracheostomy. Although TLA and FMR have shown to be successful in a subset of infants, it is clear that this is not the optimal procedure in all patients and doomed to fail in some. This latter group would be better served with MDO as their initial and only procedure. It would therefore be helpful to devise an algorithm to categorize these various groups of infants to predict which are most likely to succeed with the different surgical approaches.

The goals of this study were to review our experience with infants with PRS managed over a 10-year period and devise a universal classification system, based on clinical severity. Using this classification system, we propose a management algorithm to minimize the number of procedures needed for successful airway management in patients with PRS.

Methods

Following approval from the ethics board of University of British Columbia Children's and Women's Research Ethics Board (# H14-00353), we performed a 10-year retrospective chart review of all patients with PRS, seen between 2004 and 2013. Patients were identified using the hospital discharge database, as well as the cleft lip and palate program database. All patients (syndromic or nonsyndromic) identified as having 2 of the following features were included—glossoptosis, micrognathia, or airway obstruction. Demographic data collected included gender, gestational age, associated syndromes, presence of cleft palate, symptoms of failure to thrive, airway obstruction, desaturations, feeding difficulties, nonsurgical and surgical airway approaches, feeding management (within the first year of life), age at surgical intervention, and outcomes of management (symptom recurrence, length of hospital stay, and the need for further intervention). All data were analyzed using descriptive statistics.

Patients were identified as having airway obstruction if any of the following signs of airway obstruction were mentioned in their chart—stridor, noisy breathing, tracheal tug, intercostal retractions, or cyanosis. Patients were defined as having desaturations if any single, recorded hospital oxygen saturation was less than 80% or if more than 1 hospital oxygen saturation was less than 90%. Our institution does not routinely perform polysomnography for these neonates to document apnea but rely on oximetry to document desaturations. Participants were identified as having difficulty feeding, poor weight gain/failure to thrive if feeding time exceeded 30 minutes, if patients had <30 g/24 h weight gain, or if the patient required nasogastric tube or gastrostomy tube feedings.

All patients who failed nonoperative management underwent surgical treatment, after nasendoscopy and bronchoscopy, to exclude preexisting airway pathology. Depending on each individual surgeon's preference, techniques performed were FMR, TLA, and MDO. Tracheostomy was reserved for those patients with failed FMR and MDO.

Results

A total of 63 patients were managed for PRS during our study period. Fifty-five (87%) of these patients had cleft palate (Table 1). There were 30 females and 33 males. Seventeen patients were syndromic, with the most common syndrome being Stickler.

Forty-eight (76%) patients were managed successfully by nonsurgical treatment (Figure 1). Fifteen patients required surgical management of the airway, with a mean age of 51 days at the time of surgery (ranging from 5–246 days). Ten (67%)

Table 1. Primary Diagnosis of Patients Managed Nonsurgically Compared to Surgically.

	Nonsurgical	Surgical	Total
No. of patients	48	15	63
Female	25	5	30
Male	23	10	33
Cleft palate			
Yes	41	14	55
No	7	1	8
Nonsyndromic	38	8	46
Syndromic	10	7	17
Stickler	4	5	9
Hemifacial microsomia	2	1	3
Oromandibular limb hypoplasia	1	1	2
Craniosynostosis	1	0	1
Smith-Magenis	1	0	1
Genitopatellar	1	0	1

required only a single procedure, but 5 (33%) ultimately needed a second surgery (Table 2). The initial surgical intervention was FMR in 7 patients, TLA in 4 patients, and MDO in 4. In the 5 patients who needed multiple procedures, 3 ultimately required tracheostomies and 2 were managed successfully with a subsequent MDO. Of the patients managed surgically, 7 demonstrated feeding difficulties postoperatively and required a gastrostomy tube.

Successful single-procedure airway management was achieved in all 4 patients undergoing TLA, 3 of 7 for FMR, and 3 of 4 for MDO. Of the 4 patients who had a failed FMR, 3 had coexisting airway morbidity (tracheomalacia, laryngomalacia, and bronchomalacia). Two of these patients subsequently did well with MDO, and the other patient was managed with tracheostomy. The 1 patient with failed MDO had laryngomalacia and 2 attempts at supraglottoplasty by the otolaryngology service. That patient also had congenital VI and VII cranial nerve palsies.

Discussion

Patients with PRS are a heterogeneous group of infants who present with a combination of respiratory distress and feeding difficulties. In keeping with the literature, we found that the majority (76%) of our patients could be managed successfully by nonsurgical means such as prone positioning and nasopharyngeal airway.^{15,16} We found that the majority (87%) of patients had cleft palate. Fourteen (93%) of 15 of those infants requiring surgery had cleft palate compared to 87% of those managed without surgery. Overall, 27% of patients with PRS in our series were syndromic. It has been suggested that infants with PRS as part of a syndrome had poorer outcomes than those with isolated PRS.¹⁷ This is consistent with the present series, as we found that the number of syndromic patients in the surgical group (7 of 15, 47%) was higher than those managed without surgery (10 of 48, 21%).

For those who require surgical intervention, the ultimate goal is to perform a single procedure for successful airway

Figure 1. Flowchart showing initial and secondary surgical procedures in surgically managed patients.

management and avoid tracheostomy. Procedures are broadly divided into tongue-repositioning techniques (TLA and FMR) and mandible lengthening, namely MDO. Of the 15 patients who underwent surgery, all 4 (100%) patients having TLA had successful surgery and did not require any further intervention (Figure 1 and Table 2). This is consistent with other reported success rates of 71% to 89% for TLA and is frequently considered the first-line surgical treatment of those failing nonoperative management.¹² It is important to note that none of these patients had coexisting airway pathology such as tracheomalacia or laryngomalacia.

Of our patients undergoing FMR, 3 of 7 patients were successfully managed with FMR alone. Of the 4 who needed further surgical intervention, 3 patients had associated airway pathology, namely bronchomalacia, tracheomalacia, and tracheoesophageal fistula and laryngomalacia. Of these, 2 were managed successfully by MDO and the third patient was managed by tracheostomy, at a time prior to our institution routinely performing MDO. Our findings suggest that patients with associated airway pathology are unlikely to be managed successfully with FMR alone and should undergo MDO as the initial approach.

At our institution, MDO has been used since 2008 for patients with either a coexisting airway pathology or maxillo-mandibular discrepancy (MMD) of ≥ 10 mm. Three of the patients who had MDO as the initial procedure and 2 of the patients who had MDO after failed FMR were successful (83%) and did not require any further intervention. Only 1 patient, (participant #63), who underwent MDO as the initial procedure, at the age of 9 days, failed extubation and ultimately had a tracheostomy. This patient also had congenital VI and VII cranial nerve palsies, laryngomalacia, and tracheomalacia. His postoperative course was complicated by cellulitis of the abdominal wall from the gastrostomy tube site.

There are inherent limitations to our study, due to several factors, including the retrospective design, the variability among the 4 surgeons involved, and the small sample size. The latter is partly a result of the low incidence of PRS and the high number of patients managed nonsurgically. Nonetheless, our results show that the selection of patients for surgery and the chance of surgical success can be improved by correctly identifying and addressing the etiology of the airway obstruction on an individual basis. At present, there still appears to be a culture of using the surgical technique based on surgeons' preference.^{9,11} We propose that each patient with PRS poses a specific anatomic deformity, whether it is predominantly glossoptosis, or mandibular hypoplasia, and this should be addressed surgically, instead of a "one size fits all" philosophy.

Vancouver Classification for Airway Management of PRS: When to Do Which Intervention

From our findings, we present a clinical classification system of patients with PRS (Table 3). This 4-grade system, from grade 0 to 3, is dependent on the presence and absence of key clinical features and can be used to determine methods of non-surgical and surgical management.

- Grade 0—Patients with the mildest presentation have no coexisting airway pathology, MMD < 10 mm with mild glossoptosis, and no feeding difficulties. These patients respond to nonsurgical management, such as prone positioning.
- Grade 1—These patients have MMD < 10 mm with moderate or severe glossoptosis, ongoing feeding difficulties, and/or naso-gastric (NG) tube feed dependency. They have no coexisting airway pathology and have

Table 2. Demographics of Surgically Managed Patients With PRS.

Participant Number	Gest. Age	CP	Syndrome	MMD	Ongoing O ₂ Desaturations	Ongoing Feeding Difficulties	Airway Pathology	Initial Surgery	Age at Surgery, Days	Second Surgery	LOS (Days After Surgery)	Feeding at 6 Months Postop	Complications
8	37	Yes	No	14 mm	Yes	Yes	None	TLA	60	None	106	Bottle	None
16	37	Yes	Stickler	4 mm	Yes	Yes	None	TLA	51	None	16	Bottle	None
17	40	Yes	No	8-10 mm	Yes	Yes	None	TLA	9	None	28	Bottle	None
74	36	Yes	Stickler	3 mm	Yes	Yes	None	TLA	25	None	11	Bottle	None
54	40	Yes	No	<10 mm	Yes	Yes	None	FMR	246	Tracheostomy (decannulated)	57	G tube	Failed extubation and cardiac arrest (survived)
66	38	Yes	No	5 mm	Yes	Yes	None	FMR	9	None	80	G tube	None
60	39	Yes	Stickler	<10 mm	Yes	Yes	Tracheomalacia and tracheoesophageal fistula	FMR	19	Tracheostomy (permanent)	29	G tube	Failed extubation
64	29	Yes	Stickler	<10 mm	Yes	Yes	None	FMR	109	None	13	Bottle	None
20	36	Yes	Genitopatellar	<10 mm	Yes	Yes	Laryngomalacia	FMR	43	MDO	24	G tube	None
40	38	Yes	Stickler	<10 mm	Yes	Yes	Bronchomalacia	FMR	5	MDO	52	Bottle	Right pneumothorax and failed extubation after FMR
46	37	Yes	No	>10 mm	Yes	No	None	FMR	6	None	42	Bottle	None
1	37	Yes	No	14 mm	Yes	Yes	None	MDO	54	None	15	Bottle	None
4	40	Yes	No	12 mm	Yes	Yes	None	MDO	92	None	15	G tube	Developed oral aversion
34	39	Yes	No	>10 mm	Yes	Yes	Tracheomalacia	MDO	29	None	154	G tube	Infection of device and cheek abscess and osteomyelitis
63	38	No	Oromandibular limb hypoplasia	12 mm	Yes	Yes	Laryngomalacia and tracheomalacia	MDO	9	Tracheostomy (permanent)	150	G tube	None

Abbreviations: CP, cleft palate; FMR, floor of mouth release; G tube, gastrostomy tube; LOS, length of stay; MDO, mandibular distraction osteogenesis; MMD, maxillary-mandibular discrepancy; PRS, Pierre Robin sequence; TLA, tongue-lip adhesion.

Table 3. The Vancouver Classification and Treatment Algorithm: When to Do Which Intervention.

Severity Grade	Clinical Features	Treatment
Grade 0	<ol style="list-style-type: none"> 1. MMD <10 mm 2. Mild glossoptosis 3. Maintains saturations with prone positioning 4. No feeding difficulties 5. No coexisting airway morbidity 	Prone positioning or nasopharyngeal airway
Grade 1	<ol style="list-style-type: none"> 1. MMD <10 mm 2. Moderate or severe glossoptosis 3. Ongoing desaturations with prone positioning 4. Ongoing feeding difficulties and/or NG dependent 5. No coexisting airway morbidity 	Tongue-lip adhesion or floor of mouth release
Grade 2	<ol style="list-style-type: none"> 1. MMD >10 mm 2. Moderate or severe glossoptosis 3. Ongoing desaturations with prone positioning 4. Ongoing feeding difficulties and NG dependent 5. Coexisting airway morbidity (laryngomalacia, tracheomalacia, or bronchomalacia) 	Mandibular distraction osteogenesis
Grade 3	<ol style="list-style-type: none"> 1. MMD >10 mm 2. Severe glossoptosis 3. Ongoing desaturations with prone positioning 4. Ongoing feeding difficulties and NG dependent 5. Coexisting airway morbidity (laryngomalacia, tracheomalacia, or bronchomalacia) 6. $\pm$ failed MDO 	Tracheostomy

Abbreviations: MDO, mandibular distraction osteogenesis; MMD, maxillary-mandibular discrepancy.

ongoing desaturations, despite prone positioning. In these patients, we would recommend a tongue-repositioning procedure, either TLA or FMR.

- Grade 2—These patients have MMD $\geq$ 10 mm with moderate or severe glossoptosis, ongoing desaturations with prone positioning, ongoing feeding difficulties, and/or NG dependency, coexisting airway pathology, and fail to respond to nonsurgical means. These patients are unlikely to respond to soft tissue tongue-repositioning surgery, given the severity of mandibular hypoplasia, and should undergo MDO.
- Grade 3—These patients have the severest pathology and, if after failed MDO, should undergo tracheostomy.

Conclusion

There is currently no consensus in the surgical management of respiratory distress in patients with PRS that fail to respond to nonsurgical management. Following a 10-year retrospective review of 63 patients with PRS, we hereby propose a simple 4-point classification system, based on clinical features. Using our clinical grades, we suggest a corresponding treatment algorithm to guide management.

Authors' Note

Li contributed to literature search, data collection, data interpretation, and the manuscript writing and revision. Poon contributed to literature search and data collection. Courtemanche and Verchere contributed to project idea, study design, and editing of the manuscript. Robertson contributed to project idea and study design. Bucevska contributed to supervision of the manuscript writing. Malic contributed to literature

search, project idea, and study design. Arneja contributed to project idea, study design, data interpretation, and supervision of the manuscript writing and revision.

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