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Review

The management of upper airway obstruction in Pierre Robin Sequence

Katrina Zaballa [a,b,*], Jagdev Singh [a,c], Karen Waters [a,c]

[a] Department of Sleep Medicine, The Children's Hospital at Westmead, Sydney, NSW, Australia
[b] Department of General Medicine, The Children's Hospital at Westmead, Sydney, NSW, Australia
[c] Department of Child and Adolescent Health, Faculty of Medicine, University of Sydney, NSW, Australia

Educational aims

The reader will come to appreciate:

- Definition and pathophysiology of syndromal and isolated Pierre Robin Sequence [PRS].
- The different management options available for patients with PRS.
- Evidence for the effectiveness of mandibular distraction osteogenesis, non-invasive ventilation and a nasopharyngeal airway as management options for PRS.

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ABSTRACT

Pierre Robin Sequence (PRS) is defined by a constellation of characteristics including micrognathia, glossoptosis and airway obstruction. PRS can occur in isolation or can be associated with syndromes and another anomalies. Airway obstruction and feeding difficulties are the major presenting issues, and the severity of the condition ranges from mild, with minimal to no symptoms, to severe, with overt obstruction resulting in apnoeas, severe respiratory distress and cyanosis. The presence of airway obstruction can result in obstructive sleep apnoea and abnormalities in gas exchange, as well as exacerbation of already present feeding difficulties and failure to thrive, secondary to mismatch of caloric intake to energy usage associated with increased effort of breathing. Management of airway obstruction for infants with PRS varies between centres. This paper explores the surgical and non-surgical management options available, their effectiveness and pitfalls in children with PRS. Despite the pros and cons of each management option, it is evident that resource availability and multidisciplinary clinical support are key factors to successful management.

INTRODUCTION

In 1934, French dentist Pierre Robin first described a condition involving a constellation of features including micrognathia, glossoptosis, and upper airway obstruction, known today as Pierre Robin Sequence (PRS) [1–3]. Of those with PRS,up to 90% have an associated cleft palate [3–5]. Over the years there has been discrepancy in the literature regarding which features define PRS. In 2014, an international consensus meeting was held in the Netherlands, where an expert panel was convened to develop a consensus report on PRS. The report outlined the diagnostic characteristics of PRS which included micrognathia, glossoptosis and airway obstruction; cleft palate was not deemed to be a mandatory characteristic of the condition [6]. PRS occurs in 1:3000–1:30,000 live births [1–8], the variation in rate is related in part to the inconsistent definition of PRS used previously.

Although PRS can occur as an isolated condition, more than 50% will have an associated syndromic condition or chromosomal abnormality [3]. Over 50 different syndromes have been identified to be associated with PRS [3–4,9]. The most commonly associated syndrome is Stickler syndrome,which represents 11–18% of the PRS population,other commonly associated syndromes include 22q11.2 deletion and Treacher Collins [1,4]. Mortality rates for PRS of up to 21% have been reported, with the mortality rate for infants with syndromes or other anomalies being significantly higher than those with isolated PRS [3]. A study by Costa *et al.* [10] evaluated 181 infants with PRS, and found an overall mortality

* Corresponding author.

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rate of 16%; with no deaths observed in infants with isolated PRS. This study also found that major risk factors for mortality in this population were cardiac anomalies, central nervous system anomalies and having 2 or more organ system abnormalities. The severity of OSA was not found to be an independent risk factor for mortality. Antenatal diagnosis of PRS is technically difficult and dependent on subjective ultrasound findings, hence PRS is often diagnosed postnatally [4].

The most common presenting issues for infants with PRS are airway obstruction and feeding difficulties. In PRS, the most common area of airway obstruction is at the level of the tongue base [3]. This is largely attributed to glossoptosis (posterior displacement of the tongue base) [6],resulting in the narrowing of the oropharynx and/or hypopharynx. Additionally, there is a dynamic component of the airway obstruction, in which the increased resistance to inspiratory airflow in the already narrow airway results in negative pressure within the pharynx and further posterior pull of the tongue, leading to collapse of the pharyngeal muscles [7]. Further airway narrowing progresses and becomes evident by the second month of life [3]. Staudt et al. demonstrated that despite the increase in airway size with growth over the first two years of life, children with PRS achieve lower limits of normal pharyngeal airway size in comparison to individuals without PRS. Clinically, obstruction in PRS presents as a spectrum from infants with no symptoms of airway obstruction; airway obstruction that only becomes evident with feeding or crying in the form of increased respiratory effort or stridor; sleep-related breathing disorder or obstructive sleep apnoea (OSA). Airway obstruction is most notable in the supine position. Some infants present with overt obstruction, cyanosis and apnoeas, requiring immediate intervention [6]. van Lieshout *et al.* found that the prevalence of OSA in infants with PRS can be 46–100%, regardless of the presence or absence of symptoms of a sleep-related breathing disorder. It is well known that within the first two years of life, the mandible and upper airway increase with size, resulting in natural, gradual improvement in airway obstruction with age [3,11].

From birth, airway narrowing progresses and becomes more pronounced by the second month of life [3]. However, Staudt et al. demonstrated that despite the increase in airway size with growth, the pharyngeal airway size of children with PRS that they achieve is at the lower limits of normal in comparison to individuals without PRS. Therefore, not only is the early identification and management of airway obstruction important, especially in the context of potential neurocognitive, metabolic and cardiovascular complications associated with untreated OSA, but serial monitoring and review for persistent or re-emergence of symptoms of airway obstruction throughout childhood [eg with enlargement of tonsils and adenoids] and into adulthood is prudent.

Associated problems

Feeding difficulties are a common presenting issue for infants with PRS, and similar to airway obstruction, severity is variable among infants. Feeding difficulties may manifest as prolonged feeding time, reduced oral intake, unsafe swallow and subsequent failure to thrive [6]. The upper airway obstruction may also contribute to feeding difficulties due to the effort of breathing and associated energy expenditure exceeding that of the caloric intake [3,7–8]. In addition, if a cleft palate is present,there may be ineffective and insufficient intraoral pressure to allow for extraction of milk from the breast or bottle [3]. Up to 75% of infants with PRS have severe feeding difficulties and require assisted feeding via a nasogastric tube,with some requiring gastrostomy feeding [1,3–4]. In infants with isolated PRS, feeding difficulties often resolve after the first 12 months of life, in comparison to those infants with associated syndromic conditions who tend to continue to require ongoing feeding support [3–4]. Given the high prevalence of feeding issues seen within the PRS population, early recognition, assessment and proactive management of feeding should be considered.

Additional issues faced by infants and children with PRS include gastro-oesophageal reflux, with a prevalence of up to 80% of infants, which not only exacerbates feeding difficulties but can also impact upper airway obstruction through the development of reflux laryngitis [3]. Speech and language skill development should be monitored with time, where velopharyngeal insufficiency can develop in up to 20% of individuals post cleft palate repair [12] and speech and language delay have been reported in up to 46% of patients with PRS [13].

Pierre Robin Sequence, with or without an associated syndrome, is a complex condition involving significant health sequelae, with airway obstruction being the greatest contributor to morbidity. Review of available literature has brought to light the differences in management of airway obstruction between different centres around the world. Given the significant impact of airway obstruction on growth and development of infants with PRS, the aim of this paper is to discuss the available treatment strategies to manage airway obstruction in this unique population.

AIRWAY INTERVENTIONS UTILISED IN THE MANAGEMENT OF PRS

Non-surgical approaches

Prone positioning

The simplest and often the first option in the management of infants with PRS is to nurse the infant in the prone position where the tongue and mandible fall forward, thus improving the upper airway obstruction [3]. Prone positioning is reportedly successful in 40–70% of patients with PRS [3–4,14] and has been an effective management strategy for infants with mild to moderate OSA [9]. Van Leishout *et al.* demonstrated that patients who only required prone positioning to overcome their airway obstruction were unlikely to develop significant airway obstruction later in life. Although airway obstruction in PRS is known to improve in the first 2 years of life [4–5], there is currently no consensus on the duration of the management of PRS using this method and there is paucity of polysomnography data on the extent of relief airway obstruction achieved through positioning infants with PRS [2]. The major risk to be considered in association with prone positioning is the increased risk for SIDS, necessitating consideration of apnoea monitoring in the first six months of life [2,16].

Nasopharyngeal airway

The nasopharyngeal airway (NPA) creates a patent airway by bypassing the obstruction due to the glossoptosis, with the tip of the NPA ideally positioned at the top of the larynx [4,15]. Successful resolution of obstruction with the use of an NPA varies between studies from 67 to 100% [15]. However, an NPA is considered a temporising measure whilst awaiting mandibular growth [3,16]. Similar to prone positioning; no consensus has been established regarding the recommended duration that the NPA remains *in situ*, with studies reporting a duration of 2–8 months [3,14–15,17]. Complications associated with the use of NPAs include exacerbation of gastroesophageal reflux; intranasal stenosis and blockage of the NPA with secretions [17]. Additionally, the feasibility and safety of an NPA as a management in the home setting needs to be considered, especially concerning parental education (care, accidental dislodgement) and access to services equipped to manage an NPA.

Orthodontic appliance

The premice of an orthodontic appliance is to shift the tongue anteriorly and therefore increase the hypopharyngeal space [15]. Orthodontic appliances have been utilised for several decades and are primarily used to aid with feeding; particularly in those with an associated cleft palate [3,18]. However, it is not often used as a management strategy in many centres [19]. A randomised clinical trial conducted by Buchenau et al., using customised orthodontic plates via casting, demonstrated a reduction in mixed and obstructive events from a median of 13.8 events/hour to 3.9 events/hour, as well as a reduction in the oxygen desaturation index. While initially each plate had to be crafted by a specialist orthodontist, the availability of digital oral scanning and 3-D printing technologies mean that this may become more widely available for use in other centres. Complications associated with the use of oral appliances include local irritation and tenderness [3].

Continuous positive airway pressure

Continuous positive airway pressure (CPAP) therapy has become more easily accessible, particularly over the past 10 years. However, few centres have adopted CPAP as part of their management strategy for infants with PRS [3]. CPAP provides positive pressure to maintain airway patency; resulting in a reduction in the effort of breathing and improvements in gas exchange [20]. Leboulanger *et al.* demonstrated effective treatment of OSA in patients with PRS using CPAP; none of whom progressed to tracheostomy within their cohort; whilst Van Leishout *et al.* demonstrated 100% relief in airway obstruction with CPAP as a sole therapy. The mean duration of therapy ranged from 4 to 16 months [15,20–21]; and cessation of therapy was based on the number of obstructive events and gas exchange profile identified on polysomnography. Complications associated with CPAP include local skin irritation or breakdown, conjunctivitis and midface hypoplasia [15,21]. Additionally, adherence to CPAP therapy can be challenging in children [22].

SURGICAL APPROACHES

Tongue lip adhesion

Tongue lip adhesion (TLA) involves the tongue being tethered to the lower lip, resulting in the advancement of tongue positioning and relief of the airway obstruction caused by glossoptosis [11,23]. There has been large variability in reported success rates with TLA; ranging from 33 to 100% [15,23–26]. However, it is widely recognised that TLA does not result in complete resolution of OSA [24]. Camacho *et al.* demonstrated; that in patients with severe OSA, there was a mean reduction in apnoea-hypopnoea-index (AHI) by 50% post-TLA, although they remained within the severe OSA range postoperatively (Table 1). Similarly, Mermans *et al.* showed within their cohort, the mean pre-operative AHI was 26 events/hour and post-operatively the mean AHI was 15 events/hour. TLA is often conducted within the first 2 months of life [28]; with the release of the TLA ranging from 9 to 15 months [15,27]. The most common complication associated with TLA was dehiscence. Zhang et al. reported up to 45% of patients required revision of TLA secondary to dehiscence. Other complications reported with TLA include infection/abscess formation; granuloma formation and requirement for assisted feeding support [17,20]. Overall, although TLA is more cost-effective than other surgical options [29], infants with moderate to severe OSA were found to be more likely to require subsequent procedures to alleviate the airway obstruction [3].

Mandibular distraction osteogenesis

Mandibular distraction osteogenesis (MDO) involves the gradual lengthening of the mandible which allows the tongue base to move forward, thereby directly targeting and relieving the airway obstruction [11,30–31]. The stages involved in MDO include: initial surgery; osteotomy and device insertion; the latency period to allow for callus formation; the distraction period; where the mandible is gradually lengthened by 1–2 mm per day; and the consolidation period, the time in which the stretched callus can mineralize [32]. After a 6–12 week consolidation period; the internal devices will be removed [24]. MDO has been utilised as an initial surgical management option or as a method to facilitate decannulation in infants with severe OSA who required tracheostomy [29,31]. The timing of MDO varies between centres with data suggesting that those who underwent earlier MDO within the first 30 days of life; had more successful outcomes [33]. As such; for a number of centres; this is the first line therapy for infants who require active intervention. The overall reported success rate of MDO has been as high as 75–95% [29,31,34]; with the most common cause of failure being undiagnosed lower airway abnormalities [31,35]. Several studies have demonstrated a significant reduction in AHI following MDO and improvements in oxygen nadir [26,28,31,36–37] (Table 2). Additionally, Zhang *et al.* found that in patients with tracheostomy, 80% were able to achieve successful decannulation post-MDO. Reported complication rates associated with MDO range from 22 to 40% [30,32–34]. These include dental complications, premature consolidation, nerve damage, device malfunction, scarring, infection, non-union and temporomandibular joint ankylosis as the most common complications [24,30,32]. Hamilton *et al.* demonstrated that surgical options; including MDO, were associated with ICU admissions, a longer length of stay, delay in the establishment of oral feeding and increased tympanostomy tube insertion. However, studies have found that MDO has a lower cost burden compared to tracheostomy [29,38].

Tracheostomy

Historically, a tracheostomy was the only definitive option for stabilisation of the airway [3,29]; with up to 2–25% of patients with PRS requiring tracheostomy [14,20]. Tracheostomy was a very successful method of securing the airway; whilst awaiting mandibular growth [3,15]. The mean duration of stay in the hospital; post tracheostomy, is 1 year [21] and decannulation was considered anywhere from 1 to 4 years following the initial procedure [11,15,29]. However; tracheostomy is associated with significant

Table 1
Studies on the effectiveness of tongue lip adhesion surgery. *AHI: apnea-hypopnea index.*

	Pre-operative AHI (events/hour)	Post operative AHI (events/hour)	Pre-op O_2 Nadir (%)	Post-op O_2 Nadir (%)
Khansa (2017)	15.2	2.8	–	–
Mermans (2018)	26.8	15.4	–	–
Zhang (2018)	–	–	63–73	81–88
Viezel-Mathieu (2016)	30.8	15.4	75.8	84.4

Table 2
Summary of effectiveness of mandibular distraction osteogenesis (MDO), AHI – apnoea-hypopnoea index, events/hour, iPRS – isolated Pierre Robin Sequence, sPRS – syndromic Pierre Robin Sequence.

	Mean Pre-op AHI	Mean Post-op AHI	Pre-op O_2 Nadir	Post-op O_2 nadir	
Ehsan et al (2020)	43.8	14.2	74	83	
Soto et al (2021)	iPRS	23.08	5.64	–	–
	sPRS	21.02	4.28	–	–
Khansa (2017)	27.7	1.5	–	–	
Breik (2016)	31.2	4.34	–	–	
Ren (2017)	–	–	49	91	
Zhang (2018)	–	–	72–76	91–95	

morbidity and mortality; with up to 43% rate of complications and a mortality rate of up to 7% [3,29,21]. Complications included accidental decannulation; haemorrhage, tracheitis, recurrent chest infections, formation of granulation tissue, subglottic stenosis, tracheomalacia and delayed speech and language skills [11,5]. Other considerations associated with tracheostomy include the financial cost; which is considerable, and the subsequent significant carer burden [3,11,29]. Tracheostomy is now considered in many centres as a last resort when other previously described treatments have failed [28,39].

FUTURE RESEARCH

The availability of objective data to define "success" of treatment options were limited or ambiguous, in particular for prone positioning, NPA and CPAP. To further evaluate and improve comparability of these management strategies, future research should consider the use of polysomnography pre- and post- intervention to obtain objective data of their effectiveness.

CONCLUSION

Airway obstruction is one of the defining characteristics for a diagnosis of PRS. While the severity of airway obstruction varies, infants with PRS are at significant risk of upper airway obstruction and feeding difficulties, along with other significant comorbities in those with syndromic assocaitions. Given the multiple treatment options and variable effectiveness, the evaluation and initial management of infants with PRS would be most ideal in paediatric centres with access to multidisciplinary allied health and subspecialty teams. There continues to be variation in management approaches to infants with PRS. This variation, is in part attributeable to available resources, clinical support and expertise of different centres. Non-surgical treatment options have been proven to be effective, though clinical support and parental education are key to their success. Surgical treatment options have also been demonstrated to be effective, however, are associated with longer hospital admissions, significant costs and carer burden. In addition, should surgical pathways be considered, it is evident that a full evaluation of lower airway anatomy should be conducted, as failure is often associated with undiagnosed lower airway anomalies. Although this paper discusses the individual treatment options available, it is possible to combine multiple interventions to achieve optimum results for the patient.

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