

Evaluating the efficacy of the Tübingen Palatal Plate in managing upper airway obstruction in Pierre Robin Sequence: Experience from a tertiary hospital in Sydney, Australia

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Abstract

Study objective:

To evaluate the efficacy of the Tübingen Palatal Plate in managing upper airway obstruction in Pierre Robin Sequence during its introduction into a tertiary hospital in Sydney, Australia.

Methods

We evaluated polysomnography and treatment outcomes in children diagnosed with PRS and managed with TPP from October 2023 - March 2025 at the Children's Hospital in Westmead. A level 1 polysomnography was performed at baseline, prior to discharge from hospital and at the end of treatment. Sleep study parameters were compared between initial and follow-up studies.

Results

From 14 admissions of infants with PRS, ten infants completed TPP treatment. At baseline all ten had severe obstructive sleep apnea (OSA) with a median apnea-hypopnea index (AHI) of 53.9 events/hr, and mixed and obstructive apnea index (MOAI) of 37.8 events/hr. There was evidence of impaired gas exchange with an oxygen nadir of 88% (71–91%) and peak carbon dioxide (CO₂) level of 51.7mmHg (46.1–65.8). The final median AHI of 8.1 (2.9–13.3) events/hr, and MOAI of 2.9 (1.3–8.8) events/hr. When compared to baseline there were statistically significant improvements in AHI, OAH, MOAI and peak CO₂ levels. Excluded Infants included one patient with an intact soft palate, and three others with clinical and/or practical issues preventing implementation of the TPP.

Conclusions

This study demonstrates that when first introduced in a new health setting, TPP therapy is effective for treating airway obstruction in infants with PRS.

Brief Summary

Current Knowledge/Study Rationale: PRS is a congenital triad of micrognathia, glossoptosis and upper airway obstruction with a range of management options. This study assesses the effectiveness of TPP for treating airway obstruction in neonates with PRS during the establishment of the method in a new center.

Study Impact: As the first Australian center to introduce the TPP, the Children's Hospital at Westmead has demonstrated that the TPP is an effective non-invasive treatment modality for managing OSA in infants with PRS while introducing the treatment.

Introduction

Pierre Robin sequence (PRS) is a congenital triad of micrognathia, glossoptosis and airway obstruction[1]. Up to 50% of PRS have an underlying genetic component, of which there are currently 34 known genetic syndromes, including Stickler, Treacher Collins and Velocardiofacial syndrome[2, 3]. Cleft palate is also present in up to 90% of cases[3]. The incidence of PRS ranges from 1/8500 to 1/14,000 births[4], with a higher incidence of 1/5500 reported in Australia[5].

First line management of airway obstruction in PRS varies worldwide and according to the severity of disease. Graded approaches for managing airway problems usually start with non-invasive approaches that include prone positioning, nasopharyngeal airway (NPA), continuous positive airway pressure (CPAP) and orthodontic devices such as palatal plates. Prone positioning has been shown to be an effective method of relieving obstruction by allowing the mandible and tongue to fall forwards[6]. However prone positioning alongside prematurity and smoking are well recognized risk factors for sudden infant death syndrome (SIDS). Centers that use positioning treatment may recommend home monitoring such as apnea devices, and polysomnogram studies to confirm the effect of sleep position on airway obstruction[7, 8]. This treatment is used infrequently at the Children's Hospital at Westmead (CHW), where infants with PRS are generally referred for treatment because of significant airway and feeding issues, and the severity of their airway obstruction requires more aggressive intervention. Surgical treatment options and tracheostomy are reserved for infants with the most severe airway disturbances.

Orthodontic appliances have been in use to treat PRS for the last 15 years but have gained increasing popularity as evidence for their effectiveness as a non-invasive approach increases and technological developments make their implementation easier. The group in Tübingen Germany have published broadly on the use of the Tübingen Palate Plate (TPP), which features a palatal base plate covering the hard palate and cleft, and a velopharyngeal spur which works by displacing the tongue anteriorly to widen the hypopharynx and thereby relieve upper airway obstruction[9, 10]. They have documented positive outcomes with improvement of obstructive events on PSG within 2 weeks of treatment, with normalization of PSG scores at 6–8 months when treatment is discontinued [11].

Technological advances allowing use of 3D scanning and 3D printing have facilitated the wider adoption of this therapy. At CHW, we were able to introduce the TPP because, in addition to the technologies, we had access to dental specialists and a specialist dental center with a well-appointed dental laboratory (Westmead Centre for Oral Health). This study was undertaken to review the clinical experiences and outcomes in our initial cohort of patients as we introduced the therapy, being the first center in Australia to implement the TPP as an alternate non-surgical intervention for PRS.

Methods

Electronic medical records for all infants under 6 months of age who were diagnosed with PRS from June 2023 to March 2025 were reviewed retrospectively. Specific analysis was undertaken for infants

who underwent TPP treatment at the Children's Hospital at Westmead (CHW). Ethics approval was provided by the Sydney Children's Hospital Network (SCHN) Human Research Ethics Committee (ETH01484), which operates in accordance with the National Health and Medical Research Council's National Statement on Ethical Conduct in Human Research and CPMP/ICH Note for Guidance on Good Clinical Practice.

For each infant, we collated the following information: demographic data, comorbid conditions, hospital length of stay, interventions undertaken, any recorded adverse events, and outcomes of sleep studies for the duration of their TPP therapy. One clinician (KW) scored and reported all PSGs to maintain consistency.

Our clinical team developed a standardized workflow plan for patients undergoing TPP treatment including the following eligibility and exclusion criteria:

Inclusion criteria

- Pierre Robin Sequence
- Under 6 months of age
- Cleft palate
- Obstructive sleep apnea (OSA)

Exclusion criteria

- Multi-level airway obstruction
- Multi-organ/system involvement
- Bulbar dysfunction, impaired swallow
- Presence of teeth

Our primary outcome was the efficacy of TPP in managing severe obstructive sleep apnea in this cohort of patients. This was documented by performing polysomnography at baseline, prior to initial hospital discharge, following placement of a new TPP when infants outgrew their first plate, and at the conclusion of treatment when they outgrew their second plate and polysomnography confirmed resolution of their OSA. Respiratory parameters from the sleep study include AHI, obstructive apnea-hypopnea index (OAHI), obstructive apnea index (OAI), mixed and obstructive apnea index (MOAI), mean oxygen saturations and peak carbon dioxide levels. The OAHI includes hypopneic events and indices which are less consistent among analysts and potentially attributable to central arousals, rather than airway events, so the MOAI is the recommended marker to compare assessments of obstructive events as infants are known to have a higher number of central events at baseline [12–15].

Outcomes are presented as descriptive statistics, or for statistical analyses, the Wilcoxon signed-ranked test. A statistically significant difference was defined by a p-value of < 0.05.

Results

Over the study period, fourteen infants were admitted to CHW with PRS for airway management. Three were excluded from the TPP pathway, one each due to the following: complex multi-level airway obstruction, previous MDO complicated by infection and unexpected staff absences precluding use of the TPP. One patient, with an intact soft palate, did not tolerate the plate and developed worse clinical symptoms so the TPP was ceased after a 1-week trial. Ten infants completed TPP treatment, including 3 referred from interstate centers.

Results are presented for the ten infants who completed their therapy. Table 1 summarizes the demographic data. The majority were male (6/10; 60%), had an antenatal diagnosis of PRS (6/10; 60%) and were born at term (10/11; 91%; 32–40 + 2 weeks). All infants required respiratory support therapies to manage airway obstruction while the TPP was manufactured and fitted and included CPAP (7/10; 70%) and nasopharyngeal airway (NPA) (5/10; 50%; some infants had both). After the TPP was established, these interventions were able to be ceased within 0.5 (0–5, mean range) days. The median length of stay (LOS) in hospital was 36 days with the bulk of the patient management occurring in the outpatient setting. The therapy finished when the infant outgrew their second plate (around 6 months of age) and polysomnography confirmed that OSA had improved/resolved. The median duration of treatment for our cohort was 131.5 (58–208) days (approximately 4–5 months) which is comparably shorter than published data (6–8 months) in Tübingen [12].

Table 1
Demographic data of patients who underwent TPP (n = 10)

Male	6/10 (60%)
Female	4/10 (40%)
Antenatal diagnosis	6/10 (60%)
Gestation	38 + 5 (32–40 + 2; 1 + 2) weeks
Birth weight	3030g (2144-3400g; 636g)
Comorbidities	
Cleft palate	10/10 (100%)
VOUS	2/10 (20%)
Other symptoms/events	4/10 (40%)
Intervention	
CPAP	7/10 (70%)
NPA	5/10 (50%)
TPP	
Age at intervention (n = 10)	23 (6–63; 39) days
Length of stay (n = 10)	36 (8–45; 17) days
Adverse events	8/10 (80%)
Duration of treatment (n = 10)	131.5 (58–208; 55.75) days

Data expressed in median, range and interquartile range where possible

VOUS = genetic variant of unknown significance

NPA = nasopharyngeal airway

Other symptoms/events included: congenital heart defect, aspiration, scaphocephaly, hip dysplasia.

Table 2 shows the polysomnography results, pre and post TPP treatment. At baseline all patients had severe OSA with a median OAHl of 46.7 events/hr and MOAI of 37.8 events/hr. There was evidence of impaired gas exchange with an oxygen nadir of 88% (71–91%) and peak carbon dioxide (CO₂) level of 51.7mmHg (46.1–65.8). PSG with the plate in-situ prior to the first hospital discharge demonstrated statistically significant improvement to AHI and OAHl with a median difference of 25.1 and 24.6 events/hr respectively. At the conclusion of TPP treatment, these 10 patients had a median AHI of 8.1 events/hr, OAHl 3.2 events/hr, and MOAI 2.5 events/hr and improvements other PSG parameters

included a median drop in AHI (45.2 events/hr), OAHl (43.4 events/hr), OAI (33.8 events/hr), MOAI (37.2 events/hr) and in peak CO₂ levels (10.9mmHg). Small numbers did not allow statistical comparison of oxygen nadir levels.

Table 2
Summary of polysomnography data at baseline and post intervention (n = 10)

AHI (events/hr)	Baseline (n = 10)	TPP in (n = 10)	Difference	Post TPP treatment (n = 10)	Difference
	53.9 (40-88.5; 21.8)	23.5 (4.6-58.1; 25.9)	25.1 (W = 2; p < 0.05)	8.1 (1.3-13.3; 7.2)	45.2 [W = 0; p < 0.05]
OAHl (events/hr)	46.7 (15.5-88.5; 25.2)	21.8 (25.9-76.1; 26.5)	24.6 [W = 6; p < 0.05]	3.2 (0.1-12.4; 5.2)	43.4 [W = 0; p < 0.05]
OAI (events/hr)	35 (10.1-79; 35.9)	19.3 (2.1-37.2; 18.2)	20.9 [W = 10; p = 0.08]	2.5 (0-8.3; 3.5)	33.8 [W = 0; p < 0.05]
MOAI (events/hr)	37.8 (11.4-86; 32.8)	23.4 (3.2-41.1; 29.5)	19.9 [W = 11; p > 0.09]	2.9 (0-8.8; 5.1)	37.2 [W = 0; p < 0.05]
Oxygen nadir (%)	88 (71-91; 7)	91 (87-95; 4.3)	-4.1 [W = 12; p = 0.11]	90.0 (76-94; 7)	*
CO ₂ peak (mmHg)	51.7 (46.1-65.8; 8.6)	46.5 (39-60.2; 12.5)	9.5 [W = 16; p = 0.24]	44.3 (36-55.9; 5.6)	10.9 [W = 0; p < 0.05]

Data expressed in median, range and interquartile range

*unable to run analysis due to insufficient power, as one patient had the same median oxygen nadir before and after TPP.

AHI = apnoea-hypopnoea index; OAHl = obstructive apnoea-hypopnoea index; OAI = obstructive apnoea index; MOAI = mixed and obstructive apnoea index; CO₂ = carbon dioxide

The adverse events we noted are summarized in Table 3. These occurred in 80% of patients and the most common was pressure areas secondary to the plate that required time with the plate out. The occurrence of pressure areas is not unexpected due to the sensitivity of the neonatal oral mucosa, but it was likely more common as we developed expertise in fitting and trimming the plates. The second most common was hospital-acquired bronchiolitis, but during these respiratory infections no infant required additional respiratory support above ongoing use of the TPP. As mentioned above, an infant with an intact soft palate did not tolerate the TPP; this infant developed increased oral secretions and more obstructive episodes clinically with the plate in place. These responses were attributed to intact sensation in the soft palate. Social stressors resulted in another family discharging against medical advice (DAMA) however they attended all outpatient follow-ups and went on to complete the TPP treatment although this child had sufficient residual OSA after TPP treatment that home CPAP therapy

was recommended. Contact dermatitis (25%) secondary to steri-strips (3M™, Solventum, United States of America) were managed with topical hydrocortisone cream. Two patients developed oral candidiasis of which one had persistent symptoms requiring a prolonged course of oral nystatin, combined with regular disinfection (benzalkonium chloride) and ultrasonic cleaning of the plate. Nystatin prophylaxis is now prescribed for all patients for the duration of their TPP therapy.

Table 3
List of recorded adverse events (n = 8)

Mucosal pressure area	6/8 (75%)
Bronchiolitis~	3/8 (38%)
Oral candidiasis	2/8 (25%)
Contact dermatitis	2/8 (25%)
Persistent OSA*	1/8 (13%)
DAMA	1/8 (13%)

DAMA = discharge against medical advice. This patient discharged against medical advice for social reasons however continued to treatment as an outpatient.

~ 2 patients had rhinovirus; 1 had influenza A

*patient had significant improvement from baseline, however had sufficient residual obstructive symptoms to warrant CPAP

Discussion

This review covers the initial period of implementing TPP therapy to treat infants with PRS at CHW, in Sydney. The data from our first ten infants demonstrated substantial resolution of OSA at the completion of their therapy, confirming its effectiveness for managing airway obstruction in PRS, even as the method was first introduced to our center, with results consistent with those from the team in Tübingen where the TPP originated and is well established [12, 16].

Other treatment options available at our hospital in Australia, include CPAP and NPA, with surgical intervention such as mandibular distraction osteogenesis (MDO) and tracheostomy reserved for infants who fail these therapies. NPAs bypass obstruction by placing a modified endotracheal tube through the nasal passage with the tip ending at the distal oropharynx. They offer an interim airway and allow the infant to feed while awaiting mandibular growth [1, 6, 8]. CPAP has been the primary, non-invasive approach traditionally offered at CHW, with worldwide evidence of demonstrating improvement and avoidance of surgical intervention[10, 17, 18]. Factors reported to limit the use of CPAP in other centers include poor patient adherence, as well as limited availability of infant masks that can form a good seal with minimal leak [6, 8]. However, at CHW we have demonstrated that the treatment is clinically effective

and most infants are able to cease therapy by age 12 months [19]. Elsewhere in Australia, Queensland Children's Hospital reported 96% success rates with non-surgical management (primarily NPA and CPAP) of their patients with PRS over a 5-year period[1]. Perth Children's Hospital recently reported implementing a standardized approach to managing upper airway obstruction in PRS [14].

The advantages of the TPP that led us to implement the therapy in Australia include its ability to relieve upper airway obstruction, promote mandibular advancement via the velopharyngeal spur, and to be managed by parents in the community. The velopharyngeal spur facilitates early oral feeding but also offers an alternate option to avoid surgical intervention via MDO [16]. The TPP also allows greater freedom of movement than a CPAP device that is dependent on access to a power source[12, 20]. However, effective implementation of this treatment pathway requires a multi-disciplinary team of orthodontists, pediatric dentists, neonatologists, sleep physicians, ear nose and throat surgeons, speech pathologists, dietitians and specialized nursing staff experienced in managing PRS. It also requires access to specialized technicians and equipment such as 3D scanners and printers and an accredited dental laboratory to fabricate the TPP. The team then needs to develop the expertise to manage the workflow and any issues that arise during treatment.

We have now developed a clinical model of care and workflow for our center. Inclusion criteria are designed to maximize patient safety and minimize adverse events, which for our cohort were present in 8 patients (Table 3). Similar to Tübingen, mucosal pressure areas were most common [16]. These were managed conservatively with prescribed plate-free time and relieving the pressure areas on the plate without discontinuation of treatment. As highlighted earlier, one patient with an intact soft palate had worsening signs of obstruction within 1 week of starting TPP, thought to be due to intact sensation in the soft palate resulting in irritation and increased oral secretions so we now include an intact palate as an exclusion criterion. It is anticipated that with increasing expertise and experience our team will be able to investigate novel initiatives to overcome some of these problems, as has occurred in Tübingen [21]. However, parent preferences are an increasing component of management-decision pathways, and as previously mentioned our center maintains expertise in the use of nasal CPAP in the home. One significant change that has already occurred in our center is a reduction in the requests for MDO, with a total of 7 patients between 2019–2023, compared to only 1 over the study period. Ongoing throughput of sufficient patient numbers will be important for maintaining and developing our expertise.

Limitations to this study are primarily due to our small patient cohort, limiting statistical power. It also increased the impact of outlying events such as one patient having an intercurrent viral infection at the time of their pre-discharge PSG causing 'worse' parameters (Table 2). Nonetheless, this study indicates the capacity for a tertiary center that is remote from Europe, such as CHW, to successfully implement this highly specialized therapy. Moving forward, we expect to continue to offer the TPP as a non-invasive treatment option for infants with PRS and significant airway dysfunction, who are referred to our center for management. Progressive expansion of expertise within our specialist teams will facilitate the workflow. Long-term, we can also follow-up and review of patient outcomes to provide objective evidence of whether the significant improvements in upper airway obstruction are maintained.

Future plans and conclusions

This study demonstrates that we have been able to successfully establish a TPP service in Australia with assistance and guidance from Professor Poets and his team in Tübingen. While this report focuses on patient outcomes regarding upper airway obstruction, ongoing data collection and further studies will assess secondary outcomes such as growth, feed establishment and long-term outcomes. With the implementation of TPP therapy in other centers, data from larger patient numbers should enable future reviews and cohort studies comparing TPP against other therapies as well as consolidating data regarding the utility of TPP as a suitable management option for patients with PRS.

Abbreviations

CHW

The Children's Hospital at Westmead

CO₂

carbon dioxide

CPAP

continuous positive airway pressure

DAMA

discharge against medical advice

LOS

length of stay

MAI

mixed apnoea index

MDO

mandibular distraction osteogenesis

MOAI

mixed and obstructive apnoea index

NPA

nasopharyngeal airway

OAHl

obstructive apnoea–hypopnoea index

OAI

obstructive apnoea index

PRS

Pierre Robin Sequence

SCHN

Sydney Children's Hospital Network

TLA

tongue–lip adhesion

TPP
Tübingen palatal plate
VOUS
variant of unknown significance

Declarations

Author contributions

All authors contributed to the study conception and design. Material preparation, data collection and analysis were performed by Nathan Lieu. The first draft manuscript was written by Nathan Lieu and all authors reviewed and edited previous versions and approved the final manuscript. Supervision was provided by Karen Waters.

Disclosure statement:

All authors have seen and approved the manuscript. This study was performed at the Children's Hospital at Westmead, Australia. The authors report no conflicts of interest.

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