



Available online at
ScienceDirect
www.sciencedirect.com

Elsevier Masson France
EM|consulte
www.em-consulte.com/en



Original article

Evaluation of the efficacy of tongue-lip adhesion in Pierre Robin sequence

H. Broucqsa^a, M.-D. Lamblin^b, G. Hosana^a, P. Fayoux^{a,*}

^a Service d'ORL pédiatrique, hôpital Jeanne-de-Flandres, CHRU de Lille, avenue Eugène-Avinée, 59037 Lille, France

^b Service de neurophysiologie clinique, hôpital Jeanne-de-Flandres, CHRU de Lille, avenue Eugène-Avinée, 59037 Lille, France

ARTICLE INFO

Keywords:

Obstructive sleep apnoea
Polysomnography
Pierre Robin sequence
Tongue-lip adhesion

ABSTRACT

Objective: Tongue-lip adhesion may be used to relieve obstructive sleep apnoea in infants with Pierre Robin sequence (PRS), but only a few studies have objectively evaluated its efficacy. The purpose of this study was to evaluate the results of tongue-lip adhesion by polysomnography.

Materiel and methods: A single-centre retrospective study was conducted in infants with PRS treated by tongue-lip adhesion from 2004 to 2015, in whom at least laryngotracheal endoscopy and polysomnography were performed. The variables collected were the syndromic diagnosis, demographic data, respiratory management before tongue-lip adhesion, symptoms, and additional airway interventions. Obstructive sleep apnoea was classified into 3 groups according to severity. Polysomnography was performed one month after tongue-lip adhesion. Statistical analysis was performed with Wilcoxon signed-rank test with a limit of statistical significance of $P < 0.005$.

Results: Thirty-seven subjects in whom tongue-lip adhesion was performed at a mean age of 45 days (8 to 210 days) were included. Thirty-one patients had isolated PRS and 6 patients had associated anomalies. All patients had confirmed severe obstructive sleep apnoea. All patients required respiratory support prior to surgery: 8 intubated patients, 15 patients with noninvasive ventilation and 14 patients with nasopharyngeal airways. Eight patients had bradycardia before tongue-lip adhesion. All parameters were improved on postoperative polysomnography: oxygen saturation, hypercapnia, apnoea-hypopnoea index, bradycardia ($P < 0.005$). Only 8 patients had persistent severe obstructive sleep apnoea and required tracheostomy ($n = 5$) or noninvasive ventilation ($n = 3$). No significant correlation was observed between treatment success and any predictive variables.

Conclusion: Tongue-lip adhesion improved airway obstruction in all infants with PRS and resolved obstructive sleep apnoea in 29 patients. However, 8 patients required additional airway interventions.

© 2017 Elsevier Masson SAS. All rights reserved.

1. Introduction

Children with Pierre Robin sequence often present varying degrees of obstructive sleep apnoea syndrome (OSAS). Various studies have shown that persistent sleep apnoea syndrome is associated with a risk of cardiopulmonary and neurological complications [1,2]. Several mechanisms are responsible for pharyngeal obstruction, but glossoptosis remains the key factor [3]. Postural manoeuvres, nasopharyngeal airway and CPAP are non-surgical treatments that can be effective in a number of cases [4–7]. However, following failure of these techniques, surgical management is required to relieve obstruction. In the presence of major

obstruction, tracheostomy or mandibular distraction are sometimes essential to ensure ventilation [8,9]. Tongue-lip adhesion is a surgical procedure that relieves respiratory obstruction related to glossoptosis, based on the principle of anterior protraction of the tongue by suturing the tip of the tongue to the lower lip. It is responsible for less severe morbidity than tracheostomy or mandibular distraction [6,10]. This simple and rapid technique has few disadvantages and can allow rapid return home of the child with no need for parental education or airway devices. The efficacy of tongue-lip adhesion reported in the literature ranges between 70 and 100% depending on the author [10,11]. However, in these studies, the efficacy of this technique was assessed on the basis of oxygen saturation and clinical features with no objective evaluation of OSAS [12,13].

Preoperative polysomnographic recording is the best way of demonstrating and quantifying airway obstruction and its

* Corresponding author.

E-mail address: pierre.fayoux@chru-lille.fr (P. Fayoux).

repercussions. The initial assessment also allows evaluation of the benefit obtained at the postoperative assessment. The objective of this study was to evaluate the efficacy of tongue-lip adhesion on OSAS in children with Pierre Robin sequence based on polysomnography.

2. Materials and methods

This single-centre retrospective study included all children treated by tongue-lip adhesion for Pierre Robin sequence in the Lille University Hospital Paediatric ENT department between 2004 and 2015.

The morphological assessment comprised laryngotracheal endoscopy to evaluate the entire airway, document glossoptosis, look for any associated laryngotracheal malformation, and eliminate any extrinsic compression or underlying subglottic obstruction.

An aetiological assessment was performed in these children to confirm the clinical diagnosis of isolated Pierre Robin sequence or as part of a syndrome, including a paediatric consultation, a genetic consultation, abdominal ultrasound, echocardiography, transfontanelle ultrasound and ophthalmological examination.

Surgery was indicated in all patients with grade 3 Pierre Robin sequence according to Cole's classification [14] and after failure of at least 3 attempts of withdrawal of ventilatory assistance (intubation, CPAP, nasopharyngeal airway) or in the case of chronic respiratory distress comprising hypoxia and hypercapnia.

Preoperative assessment was based on nap polysomnography in the sleep laboratory when the child was stable. In children with respiratory distress requiring CPAP or intubation, oxygen saturation and arterial blood gases were used as reference measurements.

The following parameters were recorded for each patient: isolated Pierre Robin sequence or as part of a syndrome, age at the time of the procedure, complications of the procedure and treatment failures requiring tracheostomy, birthweight, need for jet ventilation, intubation or nasopharyngeal airway preoperatively. Preoperative and postoperative polysomnographic criteria were also recorded: apnoea-hypopnoea index, mean and maximum partial pressure of carbon dioxide (pCO_2), mean and minimum oxygen saturation and maximum duration of apnoea. Follow-up polysomnography was performed an average of 1 month after tongue-lip adhesion.

The surgical technique has been described in detail in a previous publication [15]. Briefly, the operation starts with the creation of raw zones on the ventral surface of the tongue and the posterior surface of the lower lip measuring one centimeter long by 1 to 1.5 centimeters wide. Muscles of the floor of the mouth are not released. Tongue-lip adhesion is performed by two transverse sutures largely comprising lingual and labial muscles in order to obtain sufficient resistance to traction. A retention suture is placed between the chin and the dorsal surface of the tongue for 10 days to reduce traction on the adhesion during the healing phase. The tongue-lip adhesion was released at the age of 10 months, at the same time as surgical closure of cleft palate.

For all patients, postoperative follow-up consisted of clinical assessment to evaluate the presence of dyspnoea, respiratory distress, feeding and weight gain. Respiratory follow-up consisted of continuous cardiorespiratory monitoring and arterial blood gases. Polysomnography was performed 4 weeks after the operation when the signs of respiratory distress had resolved.

The anomalies detected on polysomnography were classified into 3 stages of increasing severity, taking into account the apnoea-hypopnoea index (events per hour), heart rate (beats per minute), oxygen saturation and pCO_2 (mmHg):

- stage 1: Few or no respiratory disorders (AHI < 10/h);
- stage 2: AHI > 10/h, but no oxygen desaturation, bradycardia or hypercapnia;
- stage 3: AHI > 10/h, but desaturation and/or bradycardia and/or hypercapnia.

Analysis of morphological, endoscopic and polysomnographic parameters was performed in patients presenting failure of tongue-lip adhesion in order to identify risk factors predictive of failure. In patients with persistent signs of respiratory distress or abnormalities on cardiorespiratory monitoring or arterial blood gases, subsequent management by tracheostomy or long-term NIV was proposed.

Statistical analysis was performed by the Lille University Hospital Biostatistics department using SPSS 14 software (SPSS Inc., Chicago, IL, USA). Quantitative parameters were analyzed by a Wilcoxon signed rank test.

3. Results

Forty-eight patients with grade 3 Pierre Robin sequence were operated by tongue-lip adhesion between 2000 and 2015. Thirteen (27.1%) of these 48 patients presented Pierre Robin sequence as part of another syndrome. Eleven of these patients were excluded from the study because of missing data (7 patients) or because of associated malformations of the airways requiring additional surgical procedures (4 patients). Thirty-one of the remaining 37 patients presented isolated Pierre Robin sequence and 6 patients presented Pierre Robin sequence as part of another syndrome (16.21%), including 3 cases of multiple malformation syndromes, 1 case of Franceschetti syndrome, 1 case of Stickler syndrome and 1 case of osteochondrodysplasia. The sex-ratio was 2 girls for 1 boy. The median age at the time of tongue-lip adhesion was 45 days (range: 8–210 days). All patients had previously required respiratory management by nasopharyngeal airway ($n = 14$), CPAP ($n = 15$) or orotracheal intubation ($n = 8$). Eight patients (21.62%) presented bradycardia preoperatively. The median value for mean preoperative oxygen saturation was 95% (range: 89–97), the median value for mean preoperative pCO_2 was 53 mmHg (range: 50–60 mmHg) and the median value for mean preoperative AHI was 94 events/h (range: 64–120).

No intraoperative complications were observed and the immediate postoperative course was uneventful in 37 patients. The postoperative clinical assessment revealed resolution of signs of respiratory distress in 30 cases (81%), improvement of feeding in 22 cases (60%) and weight gain in 20 cases (54%). Postoperative cardiorespiratory monitoring and arterial blood gases demonstrated resolution of the episodes of bradycardia present before the operation.

Mean oxygen saturation was improved postoperatively in all patients with a value greater than or equal to 95% in 27 patients (72.9%). The median value for mean postoperative oxygen saturation was 96% (95–97.5; $P = 0.0007$). pCO_2 decreased postoperatively in the overall population. The median value for mean postoperative pCO_2 was 43 mmHg (range: 40–52) ($P < 0.0001$) (Fig. 1).

The apnoea-hypopnoea index (AHI) decreased in all patients. The median postoperative AHI was 27 events per hour (range: 5–65) ($P < 0.0001$) (Fig. 2).

On the postoperative assessment, 12 patients were classified as stage 1, 17 patients were classified as stage 2 and 8 patients were classified as stage 3. Multivariate analysis did not identify any risk factors predictive of failure in the stage 3 patients, except for delayed operation, which was significantly associated with a higher failure rate ($P < 0.01$). Eight patients (21.6%) presented

Download English Version:

<https://daneshyari.com/en/article/8805917>

Download Persian Version:

<https://daneshyari.com/article/8805917>

[Daneshyari.com](https://daneshyari.com)