



apresentam

# **IDENTIFICAÇÃO E TRATAMENTO DA SEQUÊNCIA DE PIERRE ROBIN NA APS**

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Hospital Infantil Joana de Gusmão – Florianópolis - SC



Cirurgia Plástica Pediátrica



# **Sequência de Pierre Robin - SPR**

Diagnóstico e tratamento para profissionais da saúde primária

# SPR - histórico

## GLOSSOPTOSIS DUE TO ATRESIA AND HYPOTROPHY OF THE MANDIBLE

PIERRE ROBIN, M.D.  
PARIS, FRANCE

The article by Eley and Farber<sup>1</sup> of Boston published in the AMERICAN JOURNAL OF DISEASES OF CHILDREN in 1930 seems to me a precise demonstration of the conception which I presented for the first time in 1926 on hypotrophy of the mandible and its consequence, congenital or acquired glossoptosis. Dr. Armand-Delille thought this study interesting and useful and suggested my publishing in the same journal a short notice of our combined research in order to record the results of our experience.

For many years I have studied dysmorphism of the faciocranio-vertebral skeleton, in particular that of the mandible. Hypotrophy of this bone is a determining cause of glossoptosis, which through the respiratory insufficiency that it produces leads to a certain physical backwardness which persists throughout infancy, childhood and adult life.

In 1902 I first showed to different societies in Paris an apparatus, the "monobloc," which allows the reestablishment of the normal relation between the two maxillary bones when they have lost their equilibrium. At the same time the two dental arches come together normally, normal mastication becomes possible and the child is again able to breathe through his nose.

The great majority of children with hypoplasia of the mandible breathe through the mouth. At that time, I was interested only in children having both temporary and permanent teeth. So my treatment concerned only children over 6 or 7 years of age. Most of these children had adenoids, but though the adenoids were removed once or several times they continued to breathe through the mouth and the adenoid facies persisted. Such children show, as a rule, a retracted lower jaw, and because of the retraction of the mandible the floor of the mouth and the tongue are pushed back, obstructing the passage of air when the mouth is closed. The difficulty of breathing is increased because the base of the tongue presses on the epiglottis, and the glottis therefore becomes closed.

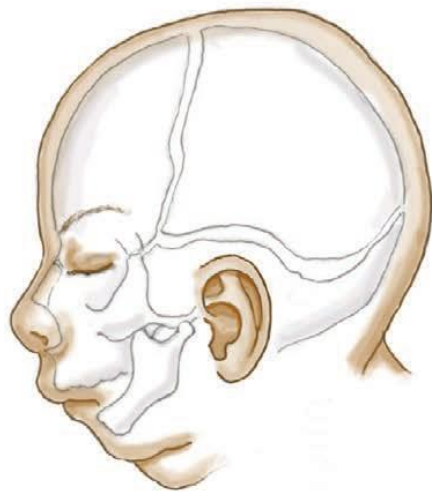
This explains why a child who has had adenoids removed is still unable to breathe through the nose, since he still suffers from glossoptosis. I demonstrated that if one forces such a child to propel the

1. Eley, R. Cannon, and Farber, Sidney: Hypoplasia of the Mandible (Micrognathia), Am. J. Dis. Child. **39**:1167 (June) 1930.

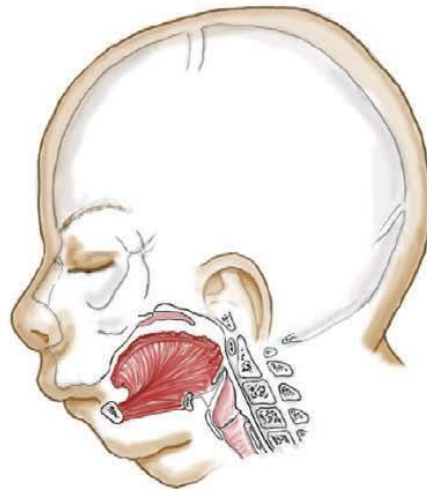
Artigo original da primeira  
descrição da Sequência de  
Pierre Robin, por Pierre  
Robin, MD

# SPR - diagnóstico

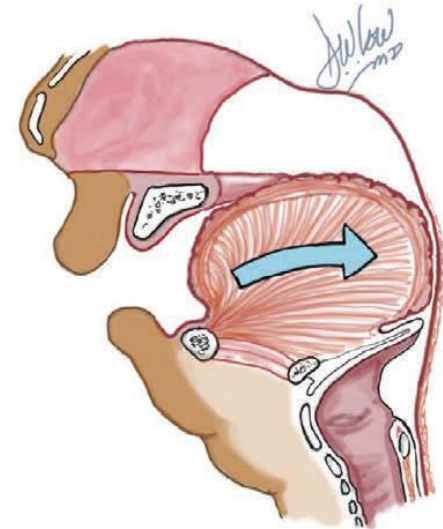
Volume 142, Number 2 • Surgery for Pierre Robin Sequence



micrognathia



glossoptosis



airway obstruction

**Fig. 1.** Pierre Robin sequence triad: micrognathia, glossoptosis, and upper airway obstruction.

TRÍADE CLÍNICA PARA O DIAGNÓSTICO

RETRO MICROGNATIA | GLOSSOPTOSE | DESSATURAÇÕES

# SPR - diagnóstico

To Distract or Not to Distract: An Algorithm for Airway Management in Isolated Pierre Robin Sequence

Richard B. Schaefer, M.D., James A. Stadler, III, and Arun K. Gosain, M.D.

*Milwaukee, Wis.*

DIAGNÓSTICO DA  
DISCREPÂNCIA MAXILO  
MANDIBULAR

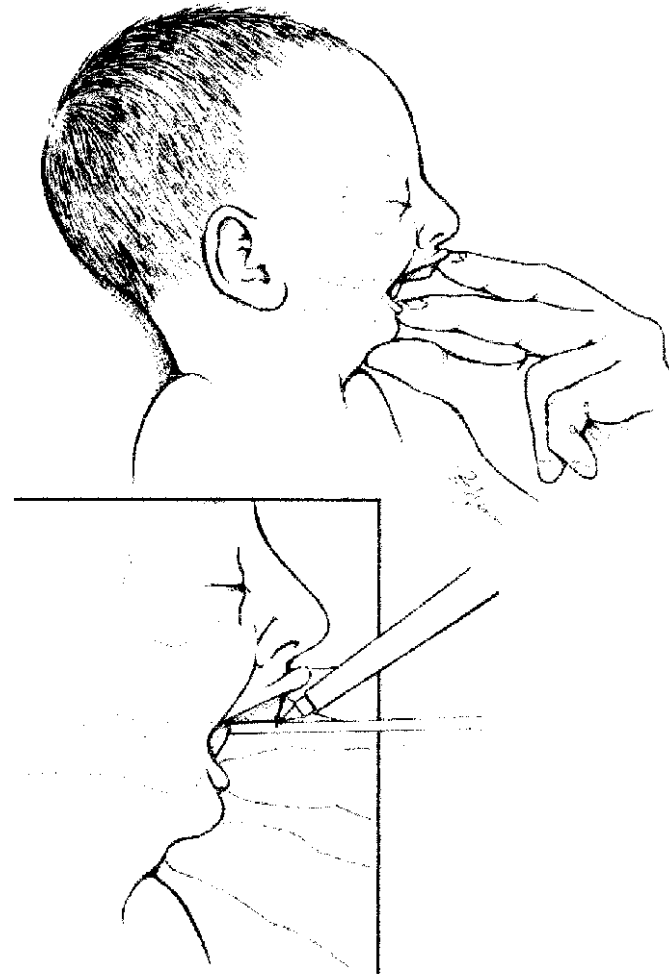


FIG. 1. Maxillary-mandibular discrepancy measurement. The mandible is brought into passive occlusion with the patient in an upright position.

# SPR - diagnóstico

Journal of Cranio-Maxillo-Facial Surgery xxx (2015) 1–8



ELSEVIER

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Journal of Cranio-Maxillo-Facial Surgery

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Reformatted 3D airway imaging in patients with airway obstruction and micrognathia

Cathy R. Henry <sup>a,\*</sup>, Arlen D. Denny <sup>b</sup>

<sup>a</sup> Penn State Hershey Medical Center Plastic Surgery, 500 University Drive, Mail Code H071, Hershey, PA 17033, United States

<sup>b</sup> Department of Plastic Surgery, Medical College of Wisconsin, Children's Hospital of Wisconsin, 9000 W. Wisconsin Avenue, Milwaukee, WI 53225, United States

that is position related. Polysomnography is obtained to quantify hypoxia and apnea, differentiating central versus obstructive or anatomical causes. Bronchoscopy is performed to evaluate the airway anatomy and the tissue dynamics (Marques et al., 2010).

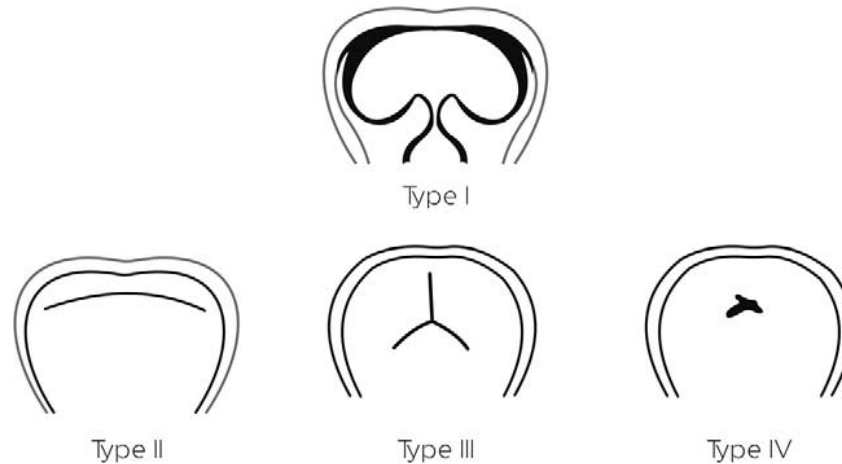


# SPR - diagnóstico

*The Journal of Craniofacial Surgery* • Volume 00, Number 00, Month 2017

## Pierre Robin Sequence: An Evidence-Based Treatment Proposal

*Oswaldo J. Gómez, MD, Oscar I. Barón, MD, and Martha L. Peñarredonda, MD*



**TABLE 1.** Sher Classification of Nasofibrolaryngoscopy Findings

Type	Description
Type I	Most frequent. Obstruction caused by posterior displacement of the tongue abutting the posterior pharyngeal wall.
Type II	Obstruction following posterior/upward displacement of the tongue abutting the soft palate and the upper portion of the oropharynx.
Type III	Pharyngeal obstruction resulting from prolapse of the pharynx middle wall.
Type IV	Obstruction owing to circular constriction of the pharynx caused by tongue and lateral pharyngeal wall movement.

Source: Elaboration based on.<sup>26</sup>

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## a importância da polissonografia como parâmetro de gravidade e avaliação de resultado do tratamento

setting (Cruz et al., 1999; Linz et al., 2011; Jarrahy, 2012; Frawley et al., 2013; Handley et al., 2013). Polysomnography is critical for characterizing the severity of the airway obstruction both before and after intervention (Freed et al., 1988; Denny, 2014; Cote et al.,

2015). Patients found to have central apnea will not improve with treatment of the tongue base obstruction. The post-intervention results are an objective measure that can help not only in assessing adequacy of treatment, but also in comparison of techniques.

# SPR - diagnóstico

ORIGINAL ARTICLE

## The Pierre Robin Mandible is Hypoplastic and Morphologically Abnormal

Elizabeth G. Zellner, MD,\* Russell R. Reid, MD, PhD,<sup>†</sup> and Derek M. Steinbacher, MD, DMD<sup>‡</sup>

From the \*Division of Plastic and Reconstructive Surgery, The Hospital for Sick Children, University of Toronto, Toronto, ON, Canada; <sup>†</sup>Section of Plastic and Reconstructive Surgery, Department of Surgery, University of Chicago, Chicago, IL; and <sup>‡</sup>Section of Plastic and Reconstructive Surgery, Department of Surgery, Yale University School of Medicine, New Haven, CT.

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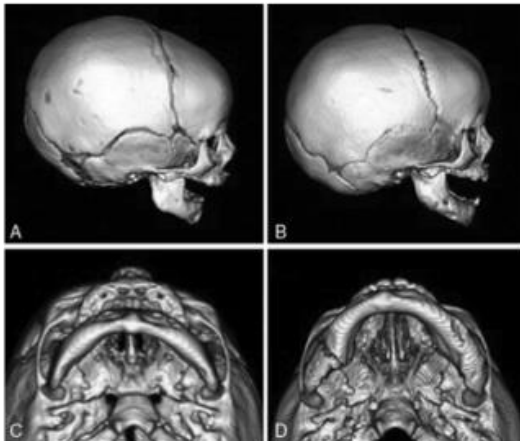


FIGURE 1. 3D computed tomographic (CT) images pre- and post-mandibular distraction osteogenesis. (A) Lateral and (B) Worms Eye view 3D CT of a Pierre Robin sequence patient preoperatively; (C) Lateral and D: Worms Eye view 3D CT of the same patient postoperatively.

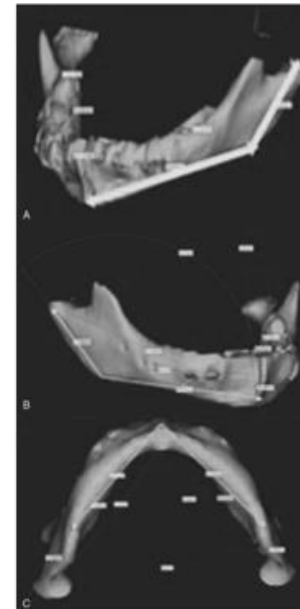


FIGURE 2. (A) Distances measured included the ramus length (condylar head to gonion) and the body length (gonion to pogonion). Three-dimensional angles measured included (B) mandibular angle (condylar head to gonion to pogonion) and (C) anterior symphyseal angle (gonion to pogonion to contralateral gonion).

# SPR – diagnóstico diferencial

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Reformatted 3D airway imaging in patients with airway obstruction and micrognathia

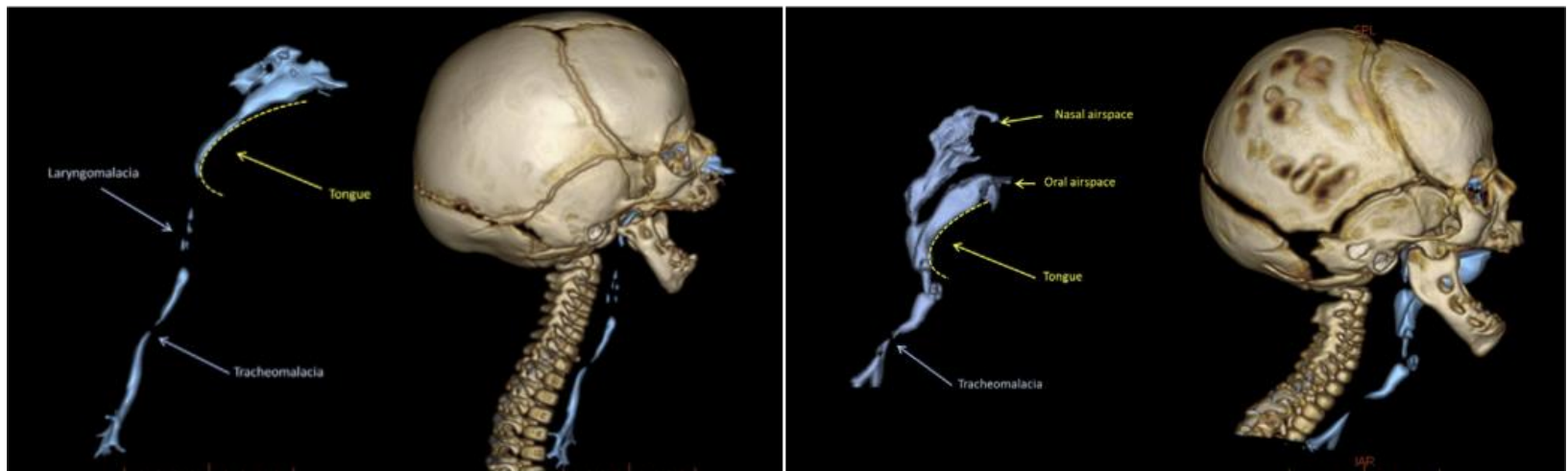
Cathy R. Henry <sup>a,\*</sup>, Arlen D. Denny <sup>b</sup>

<sup>a</sup> Penn State Hershey Medical Center Plastic Surgery, 500 University Drive, Mail Code H071, Hershey, PA, 17033, United States

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choscopy. Most valuable is the non-invasive identification of airway abnormalities that would make mandibular distraction osteogenesis ineffective in providing correction of airway obstruction in Pierre Robin patients.

- ▶ diagnóstico diferencial da obstrução da via aérea



# SPR – diagnóstico diferencial

## diagnóstico diferencial na SPR

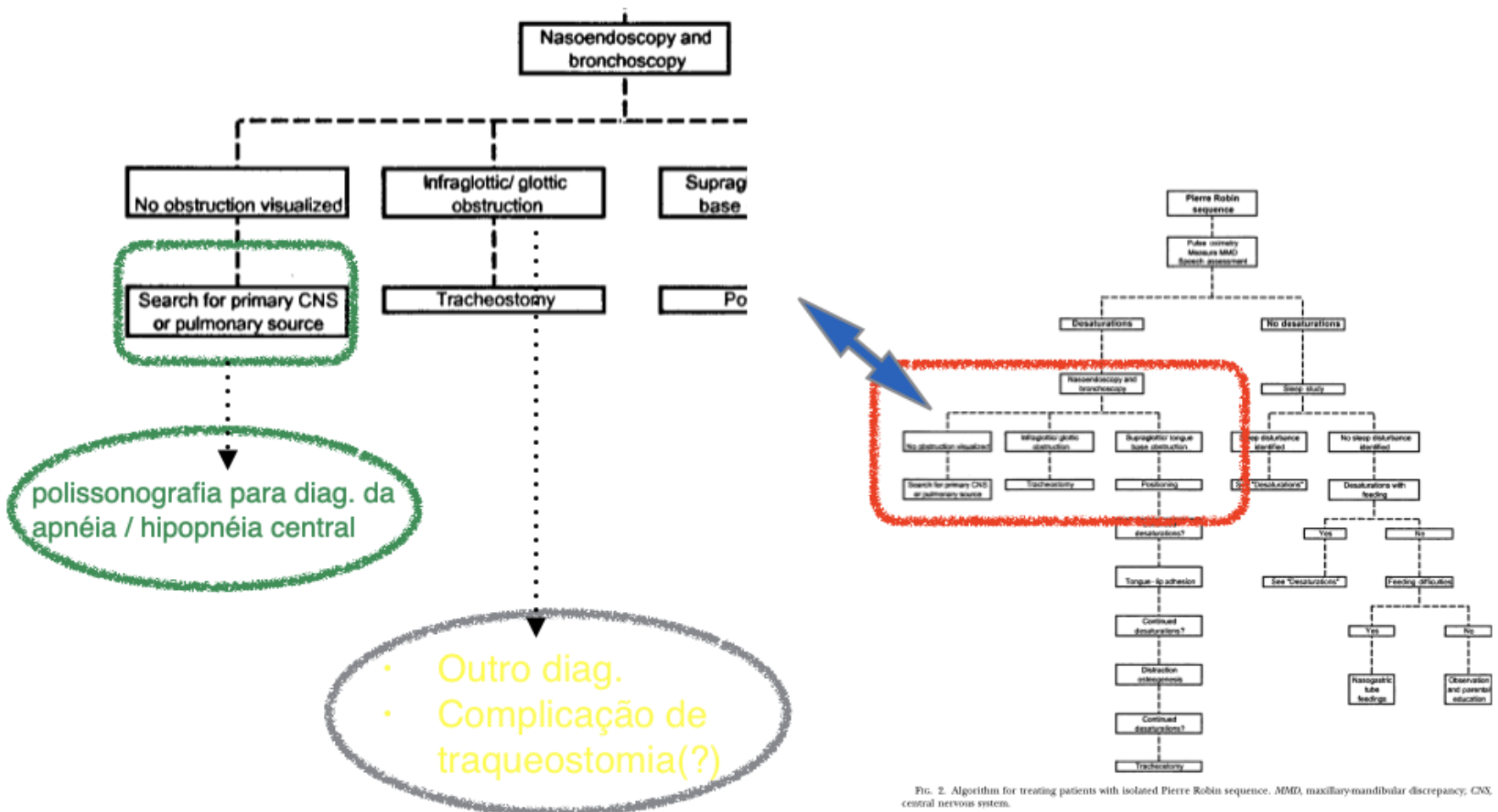


Fig. 2. Algorithm for treating patients with isolated Pierre Robin sequence. MMD, maxillary-mandibular discrepancy; CNS, central nervous system.

# SPR - diagnóstico | tratamiento | seguimiento

Journal of Cranio-Maxillo-Facial Surgery xxx (2015) 1–8



Reformatted 3D airway imaging in patients with airway obstruction and micrognathia

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► PRS = equipe multidisciplinar

Proper evaluation should occur in multi-disciplinary setting with pediatric specialists including craniofacial surgery, pulmonology, critical care, general pediatrics, anesthesia, otolaryngology, and speech pathology/feeding specialist, gastroenterology, and radiology, as well as genetics and ophthalmology if Stickler syndrome suspected (Mackay, 2011; Cladis et al., 2014). Clinical

# SPR - fisiopatologia

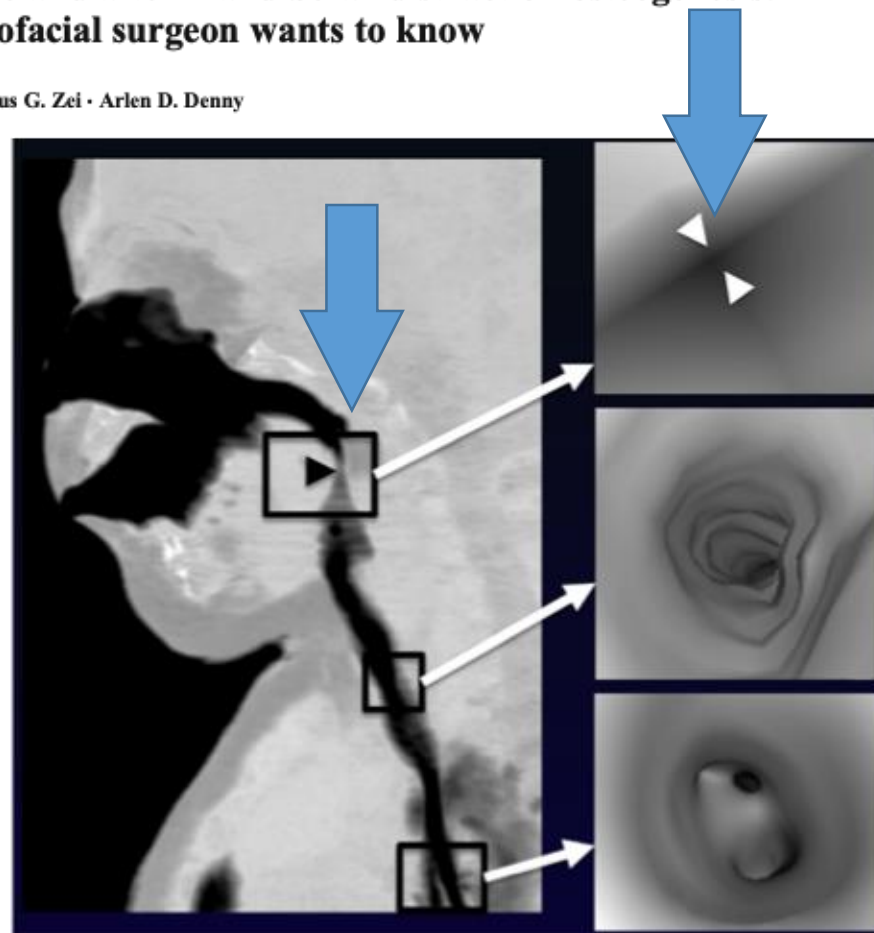
Pediatr Radiol  
DOI 10.1007/s00247-015-3323-y

PICTORIAL ESSAY

## Imaging neonates and children with Pierre Robin sequence before and after mandibular distraction osteogenesis: what the craniofacial surgeon wants to know

Arthur B. Meyers · Markus G. Zei · Arlen D. Denny

**Fig. 3** Imaging in a 2-week-old boy with Pierre Robin sequence with oropharyngeal airway obstruction but no distal airway obstruction. Virtual tracheoscopic images can be reformatted from the same CT dataset. **a** Sagittal CT image is shown with corresponding reformatted virtual tracheoscopic images at various levels. Note the areas of near-complete narrowing in the oropharynx at the level of the tongue base (*white arrowheads*). The narrowing is caused by posterior displacement of the tongue (*black arrowhead*), which is caused by the hypoplastic mandible. The remainder of the airway is patent, making this boy a good candidate for mandibular distraction osteogenesis to alleviate airway compromise





# SPR - fisiopatologia

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intervention occurs. Patients are either unable to feed due to the amount of effort that it takes to breathe, or they cannot consume enough calories to offset the work of breathing. They have significant respiratory distress with eventual metabolic collapse (Denny, 2009; Costa et al., 2014). In about 70% of these patients, the





# SPR – medidas iniciais



POSICIONAMENTO PRONADO PARA MINIMIZAR A OBSTRUÇÃO PELA BASE DA LÍNGUA

# SPR – medidas iniciais



CÂNULA NASOFARÍNGEA PARA MANUTENÇÃO DA VIA AÉREA PÉRVEA

# SPR – medidas iniciais

## Seqüência de Robin – protocolo único de tratamento

Robin sequence: a single treatment protocol

Ilza L. Marques<sup>1</sup>, Telma V. de Sousa<sup>2</sup>, Arakem F. Carneiro<sup>3</sup>, Suely P. de B. A. Peres<sup>4</sup>, Marco A. Barbieri<sup>5</sup>, Heloisa Bettiol<sup>6</sup>



Figura 8 - Lactente com intubação nasofaríngea: frente e perfil

CÂNULA NASOFARÍNGEA PARA MANUTENÇÃO DA VIA AÉREA PÉRVEA

# SPR – conduta | tratamento

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Milwaukee, Wis.

PLASTIC AND RECONSTRUCTIVE SURGERY, 

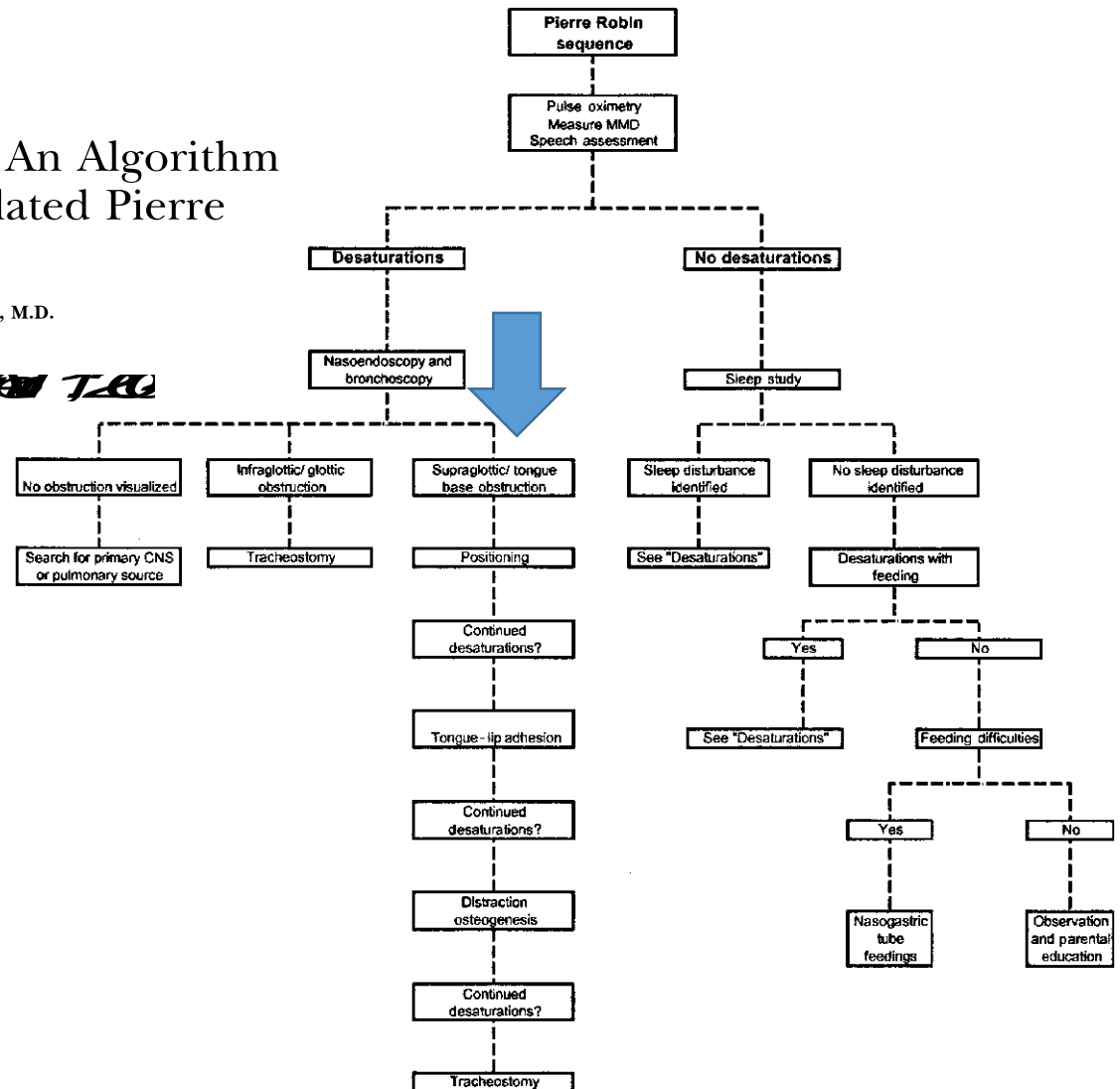


FIG. 2. Algorithm for treating patients with isolated Pierre Robin sequence.  Maxillary-mandibular discrepancy;  central nervous system.

# SPR – condotta | trattamento

## Mandibular Distraction for Micrognathia in Neonates

Carrie E. Zimmerman, BS,\* Laura S. Humphries, MD,\* Tulsi Roy, MD,\* Russell R. Reid, MD, PhD\*

\*Department of Surgery, Section of Plastic Surgery, University of Chicago Medical Center, Chicago, IL

### American Board of Pediatrics Neonatal-Perinatal Content Specification

- Know the associations and clinical features and management of macroglossia and hypoplastic mandible, including the Pierre Robin syndrome.

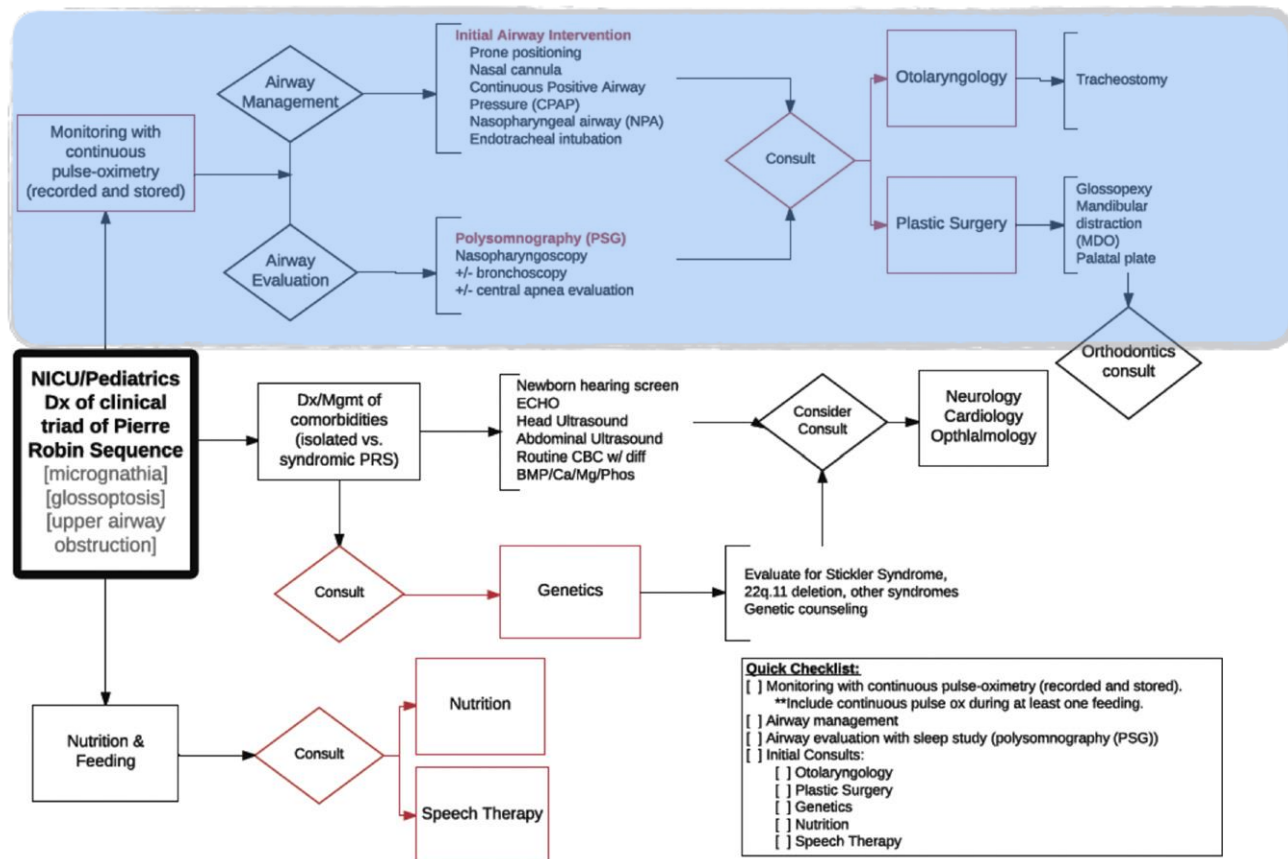


Figure 2. University of Chicago Hospital clinical evaluation and management guidelines for patients with Pierre Robin sequence (PRS). BMP=basic metabolic panel; CBC=complete blood cell count; ECHO=echocardiography.

# SPR – conduta | tratamento

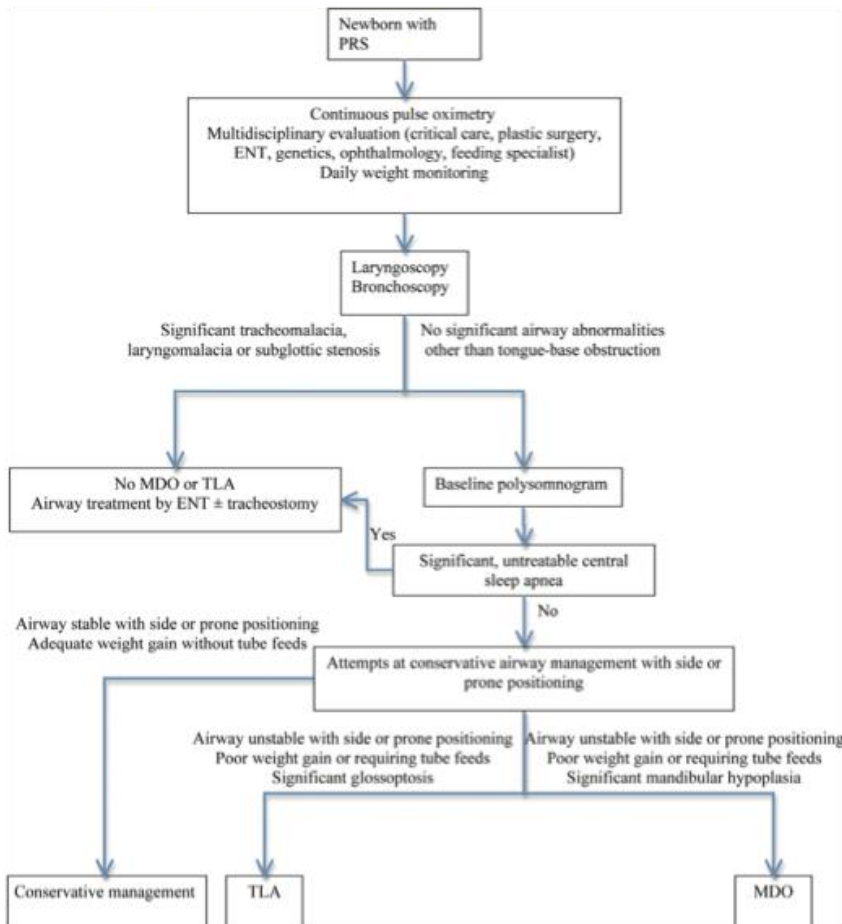
## Airway and Feeding Outcomes of Mandibular Distraction, Tongue-Lip Adhesion, and Conservative Management in Pierre Robin Sequence: A Prospective Study

Ibrahim Khansa, M.D.  
Courtney Hall, M.S., C.N.P.  
Lauren L. Madhoun, M.S.,  
C.C.C.-S.L.P.  
Mark Splaingard, M.D.  
Adriane Baylis, Ph.D.,  
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Richard E. Kirschner, M.D.  
Gregory D. Pearson, M.D.  
Columbus, Ohio

**Background:** Pierre Robin sequence is characterized by mandibular retrothia and glossoptosis resulting in airway obstruction and feeding difficulty. When conservative management fails, mandibular distraction osteogenesis, tongue-lip adhesion may be required to avoid tracheostomy. The authors' aim was to prospectively evaluate the airway and feeding outcomes of their comprehensive approach to Pierre Robin sequence, which includes conservative management, mandibular distraction osteogenesis, and tongue-lip adhesion.

**Methods:** A longitudinal study of newborns with Pierre Robin sequence treated at a pediatric academic medical center between 2010 and 2015 was performed. Baseline feeding and respiratory data were collected. Patients underwent conservative management if they demonstrated sustainable weight gain with

Plastic and Reconstructive Surgery • April 2017



**Fig. 1.** Proposed comprehensive algorithm for the treatment of Pierre Robin sequence. PRS, Pierre Robin sequence; ENT, ear, nose and throat; MDO, mandibular distraction osteogenesis; TLA, tongue-lip adhesion.

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# SPR – conduta | tratamento

## SPR - opções de tratamento

- ◆ conservador
- ◆ adesão língua-lábio
- ◆ distração mandibular osteogênica
- ◆ traqueostomia



# SPR – conduta | tratamento

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## SPR - opções de tratamento

### ◆ conservador

- ◆ posição lateral ou pronada
- ◆ alimentação em ortostatismo
- ◆ oximetria de pulso contínua
- ◆ pode ser mantido se não houver dessaturações e se conseguir ganhar peso

# SPR – conduta | tratamento

## Airway and Feeding Outcomes of Mandibular Distraction, Tongue-Lip Adhesion, and Conservative Management in Pierre Robin Sequence: A Prospective Study

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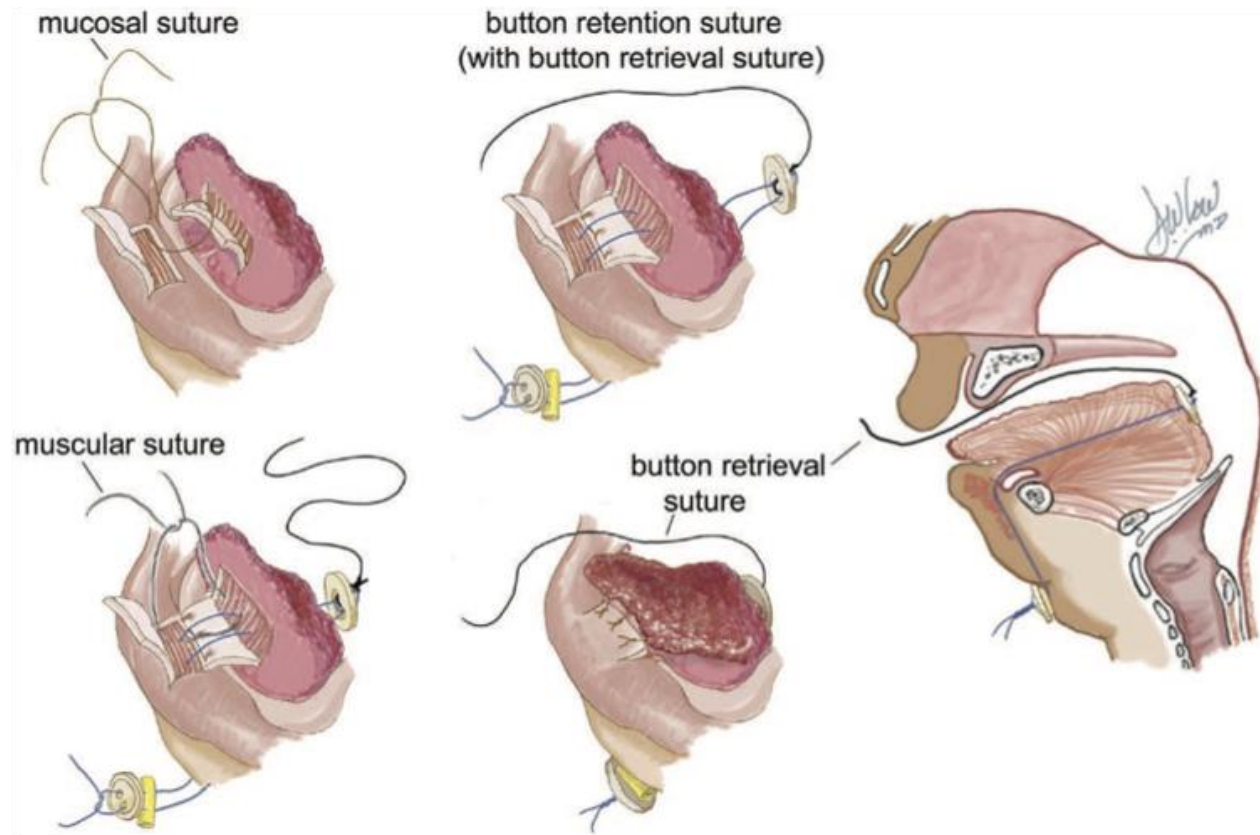
**Methods:** A longitudinal study of newborns with Pierre Robin sequence treated at a pediatric academic medical center between 2010 and 2015 was performed. Baseline feeding and respiratory data were collected. Patients underwent conservative management if they demonstrated sustainable weight gain with

## SPR - opções de tratamento

- ◆ adesão língua - lábio
  - ◆ temporário
  - ◆ atua sobre a glossoptose
  - ◆ sem efeito sobre a mandíbula
  - ◆ riscos de deiscência dos retalhos, infecção

# SPR – conduta | tratamento

## ◆ adesão língua - lábio



**Fig. 2.** Tongue-lip adhesion technique showing mucosal, button retention, muscular, and button retrieval sutures.

# SPR – conduta | tratamento

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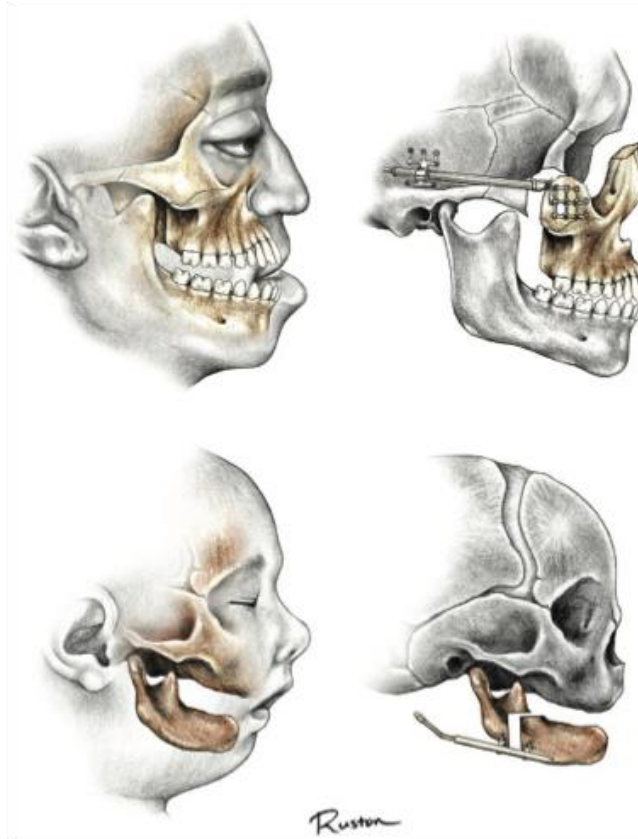
**Background:** Pierre Robin sequence is characterized by mandibular retrusion and glossoptosis resulting in airway obstruction and feeding difficulty. When conservative management fails, mandibular distraction osteogenesis and tongue-lip adhesion may be required to avoid tracheostomy. The authors' purpose was to prospectively evaluate the airway and feeding outcomes of their comprehensive approach to Pierre Robin sequence, which includes conservative management, mandibular distraction osteogenesis, and tongue-lip adhesion.  
**Methods:** A longitudinal study of newborns with Pierre Robin sequence treated at a pediatric academic medical center between 2010 and 2015 was performed. Baseline feeding and respiratory data were collected. Patients underwent conservative management if they demonstrated sustainable weight gain with

## SPR - opções de tratamento

- ◆ distração osteogênica mandibular - DMO
  - ◆ atua sobre a mandíbula
  - ◆ distratores internos ou externos
  - ◆ efetivo
  - ◆ morbidade e risco presentes, controlados

# SPR – conduta | tratamento

Plastic and Reconstructive Surgery • November 2017



distração  
osteogênica  
no  
esqueleto  
facial

**Fig. 2.** Distraction osteogenesis for craniofacial advancement and airway expansion. (Above) Midface distraction osteogenesis. A Le Fort III distraction, which advances the malar, orbital, maxillary, and nasal bones, is represented. (Below) Mandibular distraction osteogenesis. This procedure advances the dental arch with its attached tongue base, thereby expanding the retrolingual airway.

# SPR – condotta | trattamento

## Distraction Osteogenesis of the Craniofacial Skeleton

Jack C. Yu, M.D., D.M.D., Jeffrey Fearon, M.D., Robert J. Havlik, M.D., Steve R. Buchman, M.D., and John W. Polley, M.D.

*Augusta, Ga.; Dallas, Texas; Indianapolis, Ind.; Ann Arbor, Mich.; and Chicago, Ill.*

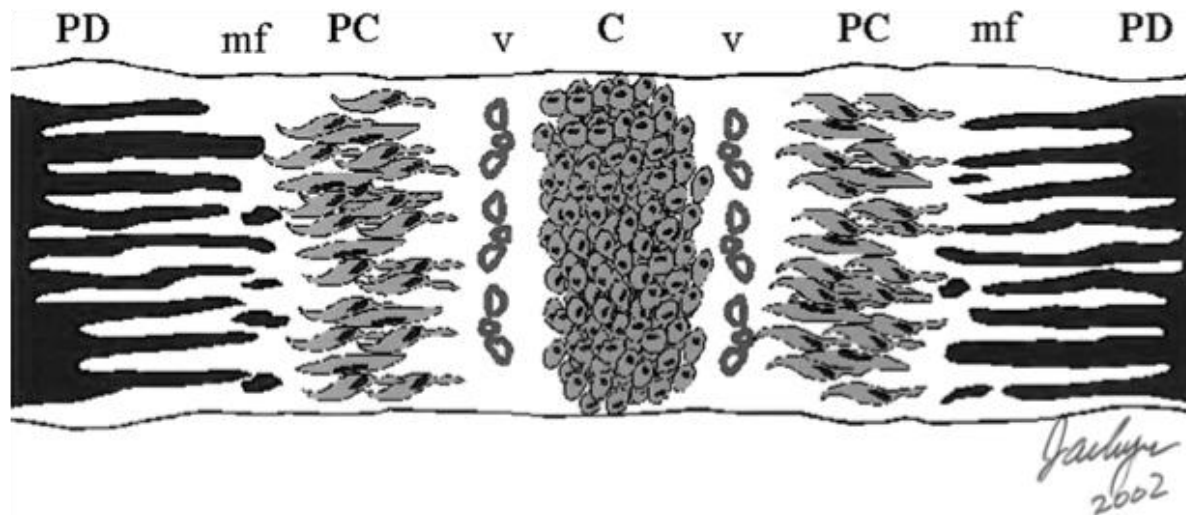


FIG. 1. A diagram of the five zones and four transition areas of the distraction gap during the middle, activation phase. The five zones are the central zone (C), the paracentral zones (PC), and the two proximal/distal zones (PD). The four transitional areas are the two areas of vasculogenesis (v) and the two areas of mineralization front (mf). The central zone is the most cellular and most blastema-like. The transitional area of mineralization front shows clear anisotropy, with the nascent trabeculae in perfect alignment with the line of tensile force.



# SPR – condotta | trattamento

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<sup>b</sup> Department of Plastic Surgery, Medical College of Wisconsin, Children's Hospital of Wisconsin, 9000 W. Wisconsin Avenue, Milwaukee, WI 53225, United States

2011). Mandibular distraction improves the airway while at the same time addressing the underlying skeletal abnormality of micrognathia (Cicchetti et al., 2012; Denny, 2009; Jarrahy, 2012).

# SPR – condotta | trattamento

## Mandibular Distraction for Micrognathia in Neonates

Carrie E. Zimmerman, BS,\* Laura S. Humphries, MD,\* Tulsi Roy, MD,\* Russell R. Reid, MD, PhD\*

*\*Department of Surgery, Section of Plastic Surgery, University of Chicago Medical Center, Chicago, IL*

### American Board of Pediatrics Neonatal-Perinatal Content Specification

- Know the associations and clinical features and management of macroglossia and hypoplastic mandible, including the Pierre Robin syndrome.

MDO remains the gold standard surgical treatment available to definitively relieve UAO in select patients with PRS. However, it carries morbidity and risk in both the early and late postoperative period. The exploration of less inva-




# SPR – condotta | trattamento

## Mandibular Distraction for Micrognathia in Neonates

Carrie E. Zimmerman, BS,\* Laura S. Humphries, MD,\* Tulsi Roy, MD,\* Russell R. Reid, MD, PhD\*

*\*Department of Surgery, Section of Plastic Surgery, University of Chicago Medical Center, Chicago, IL*

diagnosis of airway obstruction and airway management. The gold standard surgical intervention for management of symptomatic micrognathia is mandibular lengthening by distraction osteogenesis (MDO) to anteriorly reposition a retroflexed tongue and relieve obstruction. Although MDO is

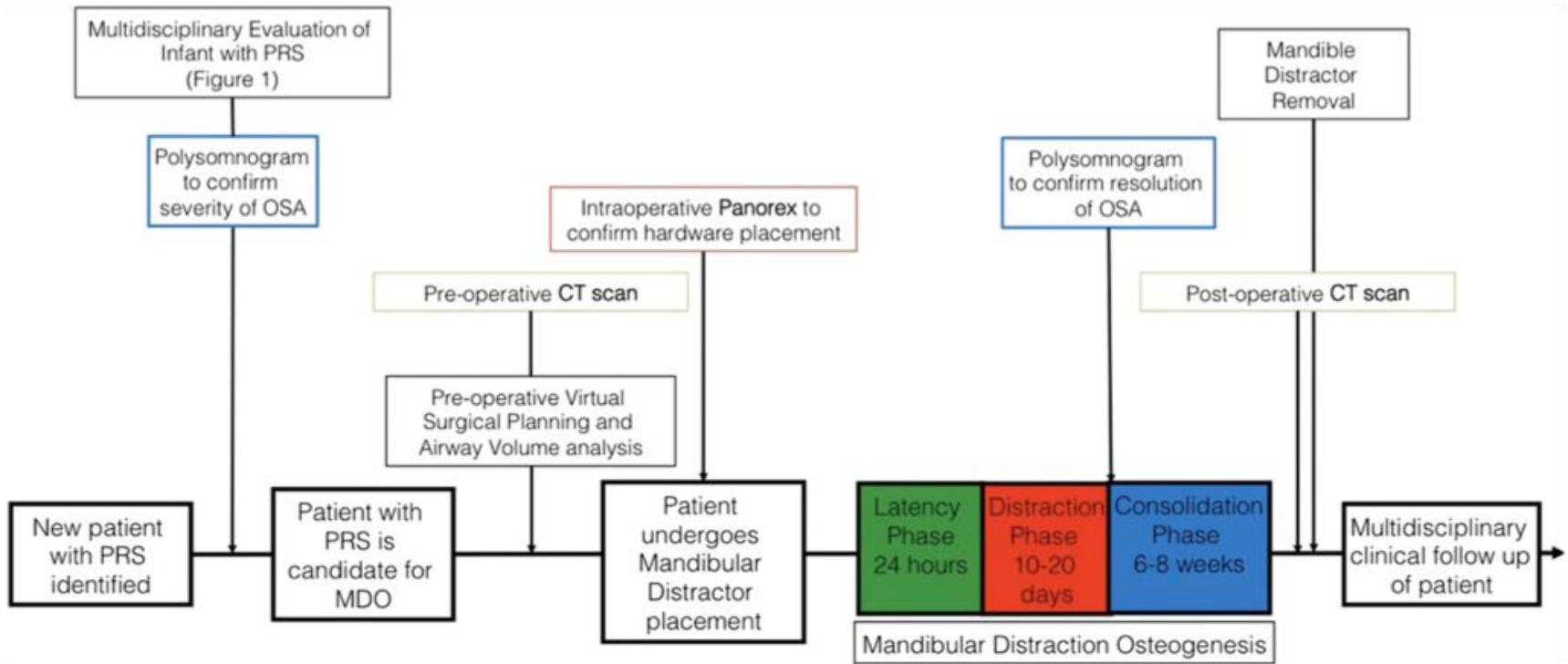


# SPR – condotta | trattamento

## Mandibular Distraction for Micrognathia in Neonates

Carrie E. Zimmerman, BS,\* Laura S. Humphries, MD,\* Tulsi Roy, MD,\* Russell R. Reid, MD, PhD\*

\*Department of Surgery, Section of Plastic Surgery, University of Chicago Medical Center, Chicago, IL



**Figure 3.** University of Chicago Hospital mandibular distraction osteogenesis timeline and protocol for patients with Pierre Robin sequence (PRS). CT=computed tomography; MDO=mandibular distraction osteogenesis; OSA=obstructive sleep apnea.

# SPR – condotta | trattamento

## Airway and Feeding Outcomes of Mandibular Distraction, Tongue-Lip Adhesion, and Conservative Management in Pierre Robin Sequence: A Prospective Study

Ibrahim Khansa, M.D.  
 Courtney Hall, M.S., C.N.P.  
 Lauren L. Madhoun, M.S.,  
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 Adriane Baylis, Ph.D.,  
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 Richard E. Kirschner, M.D.  
 Gregory D. Pearson, M.D.  
 Columbus, Ohio

**Background:** Pierre Robin sequence is characterized by mandibular retrothia and glossoptosis resulting in airway obstruction and feeding difficulty. When conservative management fails, mandibular distraction osteogenesis, tongue-lip adhesion may be required to avoid tracheostomy. The authors' purpose was to prospectively evaluate the airway and feeding outcomes of their comprehensive approach to Pierre Robin sequence, which includes conservative management, mandibular distraction osteogenesis, and tongue-lip adhesion. **Methods:** A longitudinal study of newborns with Pierre Robin sequence treated at a pediatric academic medical center between 2010 and 2015 was performed. Baseline feeding and respiratory data were collected. Patients underwent conservative management if they demonstrated sustainable weight gain with

**Table 2. Airway and Feeding Outcomes of the Patients in the Three Groups**

	Conservative	TLA	MDO
No. of patients	10	8	10
<b>Airway</b>			
Days to extubation postoperatively	—	6.9	9.6
% with reintubation postoperatively	—	25%	0%
Baseline AHI	6.1	15.2	27.7
Follow-up AHI	5.5	2.8	1.5
Mean decrease in AHI from baseline	9.8%	81.6%	94.6%
Patients with residual moderate OSA (AHI ≥ 5)	20%	12.5%	0%
Tracheostomy	0%	0%	0%
<b>Feeding</b>			

# SPR – conduta | tratamento

## Airway and Feeding Outcomes of Mandibular Distraction, Tongue-Lip Adhesion, and Conservative Management in Pierre Robin Sequence: A Prospective Study

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## SPR - opções de tratamento

### ◆ traqueostomia

- ◆ morbidade
- ◆ pode ser necessário nas comorbidades
- ◆ apnéias de origem central, outras obstruções da via aérea
- ◆ evitar sempre que possível - pode piorar prognóstico ao adicionar morbidade

# SPR - seguimiento

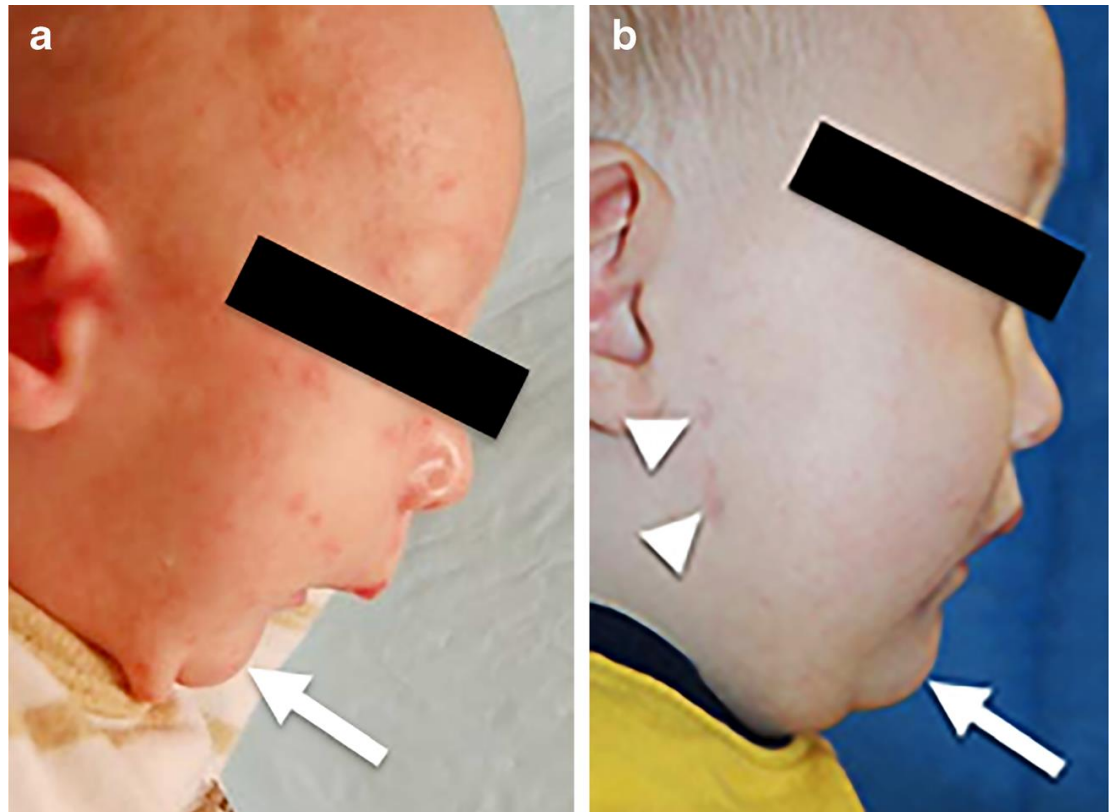
Pediatr Radiol  
DOI 10.1007/s00247-015-3323-y

PICTORIAL ESSAY

Imaging neonates and children with Pierre Robin sequence before and after mandibular distraction osteogenesis: what the craniofacial surgeon wants to know

Arthur B. Meyers & Markus G. Zei & Arlen D. Denny

**Fig. 1** Clinical photographs of an infant boy with Pierre Robin sequence and micrognathia causing episodes of complete airway obstruction and feeding intolerance. **a** Photo at 2 months old prior to mandibular distraction osteogenesis (MDO) shows marked micrognathia (*arrow*). **b** Post MDO there is improvement of the micrognathia (*arrow*). Small scars are noted from the pins of the external fixation device (*arrowheads*). This boy was able to maintain his airway after the procedure and avoided tracheostomy. Also note the boy's weight gain secondary to resolution of feeding intolerance



# SPR - seguimiento

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## Reformatted 3D airway imaging in patients with airway obstruction and micrognathia

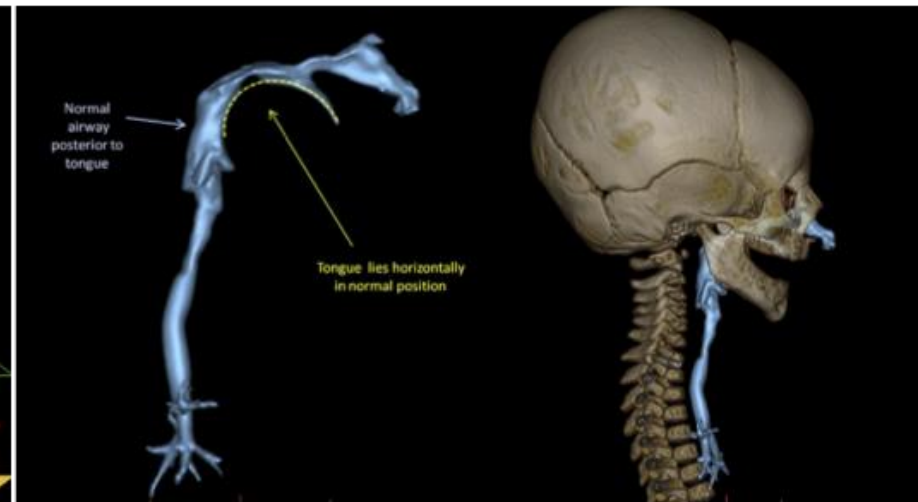
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