3rd INTERNATIONAL PEDIATRIC

NONINVASIVE VENTILATION CONFERENCE

Necker university hospital

Paris - France

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Which therapeutic options when CPAP fails?

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Treatment of pediatric OSAS

WHO

Every child with an AHI > 5/h irrespective of the presence of morbidity,
Or AHI between 1-5/h + comorbidity (cardiovascular, neurological)

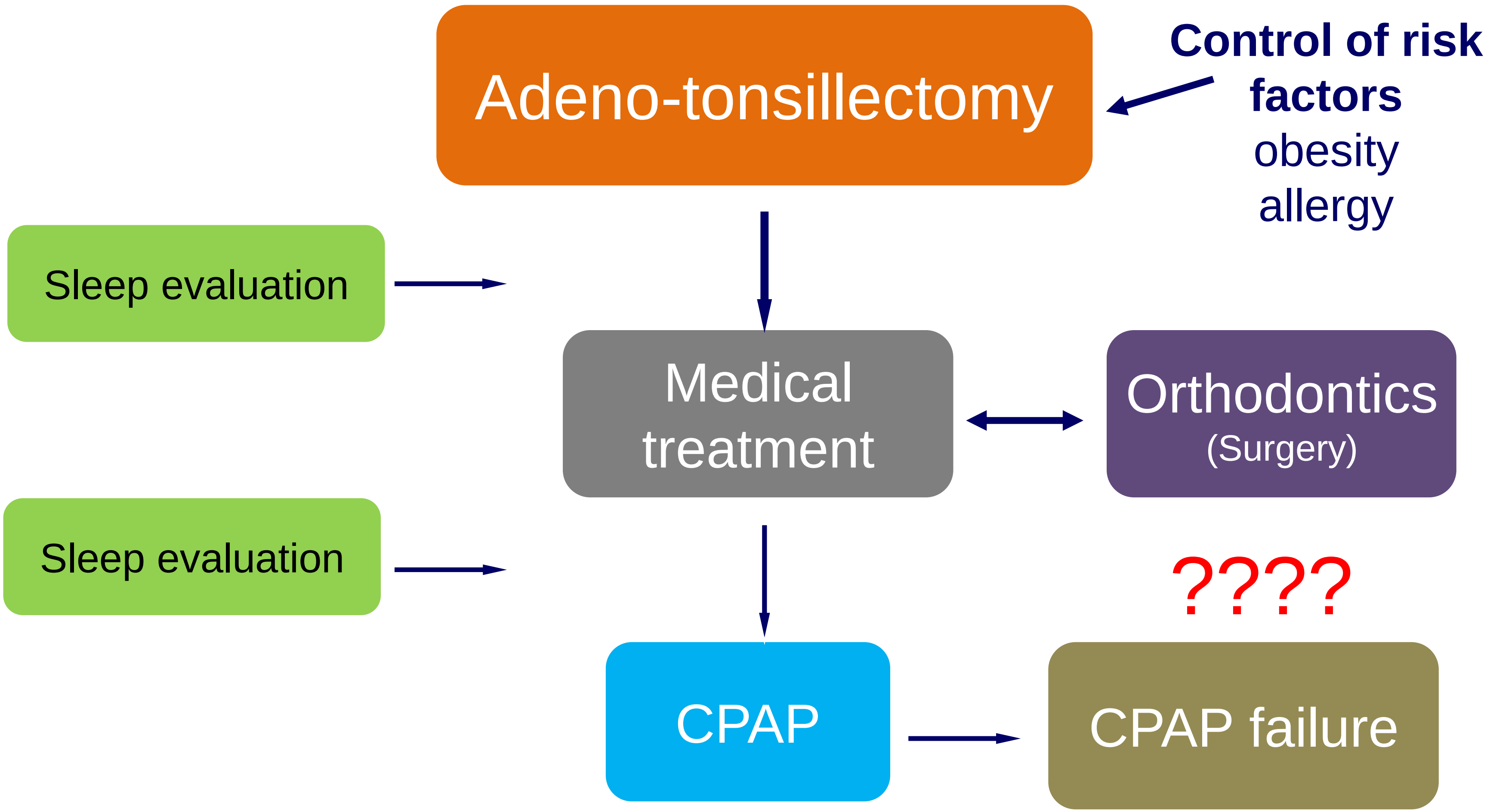
↓

HOW

A stepwise treatment approach, tailored on the severity of OSAS and on the presence of underlying diseases/comorbidity

n.b. OSAS treatment is a priority in the presence of: major craniofacial abnormalities; neuromuscular disorders; achondroplasia; Down syndrome; mucopolysaccharidoses; Prader Willi syndrome...

Stepwise treatment approach



Limits of CPAP

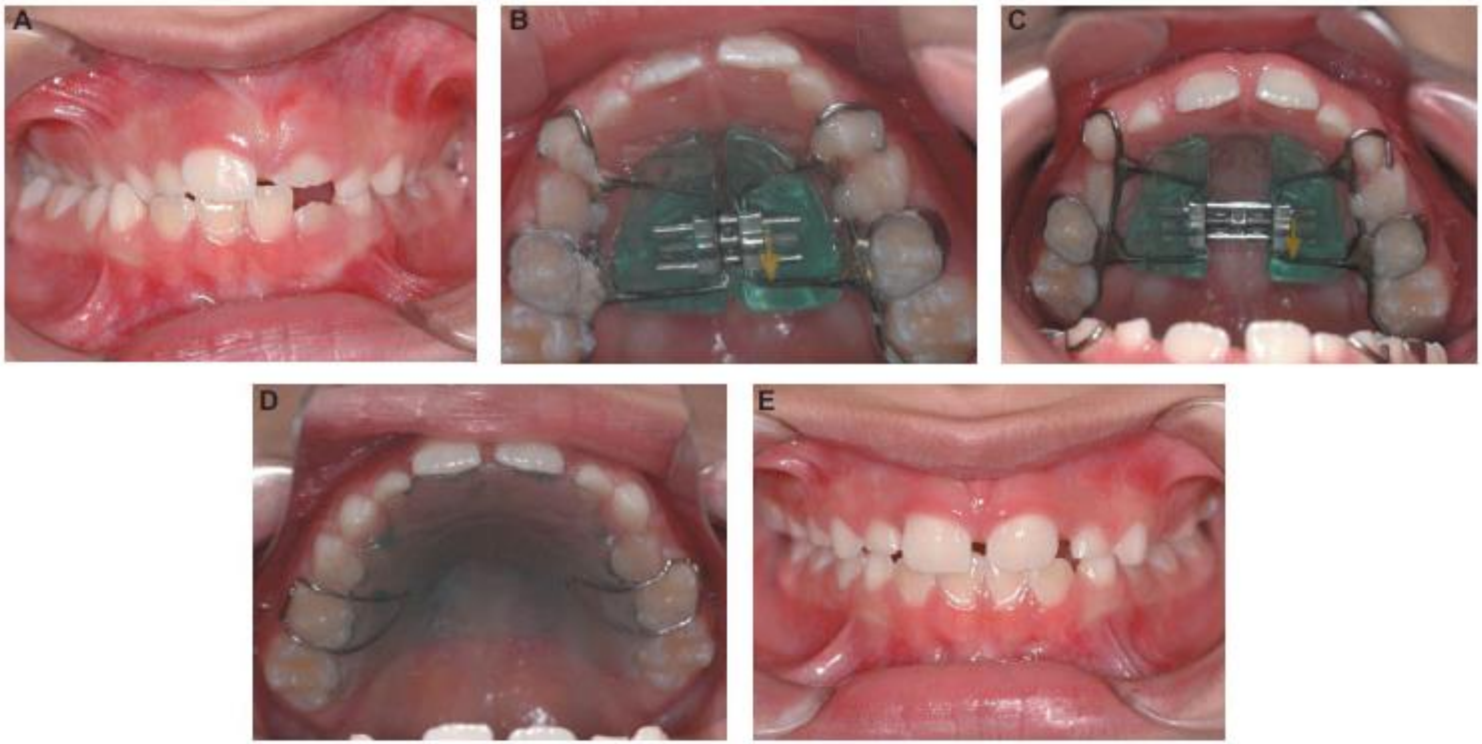
CPAP is an effective treatment for OSAS but:

- patients may not tolerate it (pressure sores, interface inadequacy...)
- patients may not be compliant: intellectual disability, default of family structure...
- patients may have too severe OSAS: CPAP dependance > ~ 18/24h

Other options?

- Revise previous therapeutic options
  - weight loss
  - mandibular advancement devices or rapid maxillary expansion
- Discuss surgery (selected patients)
  - mandibular distraction osteogenesis
  - craniofacial surgery
- High flow nasal cannula
- Tracheotomy

Rapid maxillary expansion



Craniofacial and upper airway morphology in pediatric sleep-disordered breathing and changes in quality of life with rapid maxillary expansion

Vandana Katyal,[a] Yvonne Pamula,[b] Cathal N. Daynes,[c] James Martin,[d] Craig W. Dreyer,[e] Declan Kennedy,[f] and Wayne J. Sampson[g]
Adelaide and Sydney, Australia

(Am J Orthod Dentofacial Orthop 2013;144:860-71)

Improvement of PSQ and OSA 18 questionnaire

Efficacy of rapid maxillary expansion in children with obstructive sleep apnea syndrome: 36 months of follow-up

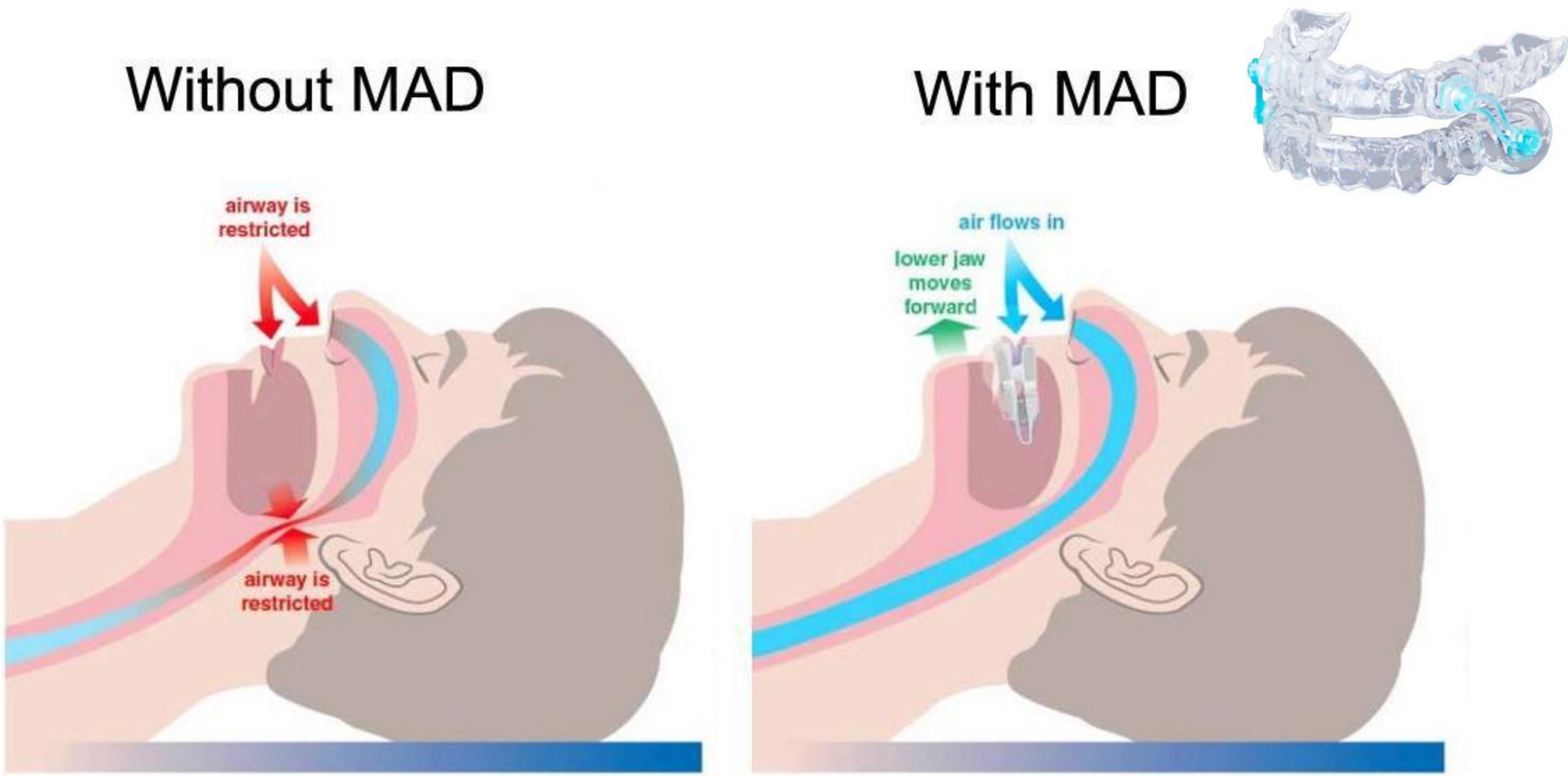
Sleep Breath (2011) 15:179–184

10 children, mean age 6.2 ± 2.1 years with high, narrow palate associated with deep bite, retrusive bite or crossbite.

	T0	T1	T2	ANOVA	Wilcoxon test		
				p value	T0 vs T1	T1 vs T2	T0 vs T2
TST (h)	6.2±11.0	7.7±0.9	6.3±1.2	0.006	0.01	0.01	NS
SE (%)	85.8±10.1	90.9±6	83.6±10.2	0.07	NS	0.06	NS
S1 (%)	5.0±4.1	2.1±1.7	3.3±2.2	0.2	0.07	0.2	0.1
S2 (%)	42.4±10.4	40.0±8.8	51.6±6.8	0.02	0.4	0.01	0.007
SWS (%)	36.4±10.7	34.4±10.4	27.9±8.1	0.02	0.3	0.03	0.01
REM (%)	15.9±6.3	23.4±5.9	17.1±5.7	0.06	0.04	0.06	NS
AHI (n/h)	6.3±4.7	2.4±2.0	2.3±1.7	0.003	0.05	NS	0.05
SaO2 (%)	95.8±1.8	97.0±2.8	97.7±1.0	0.05	0.05	NS	0.008

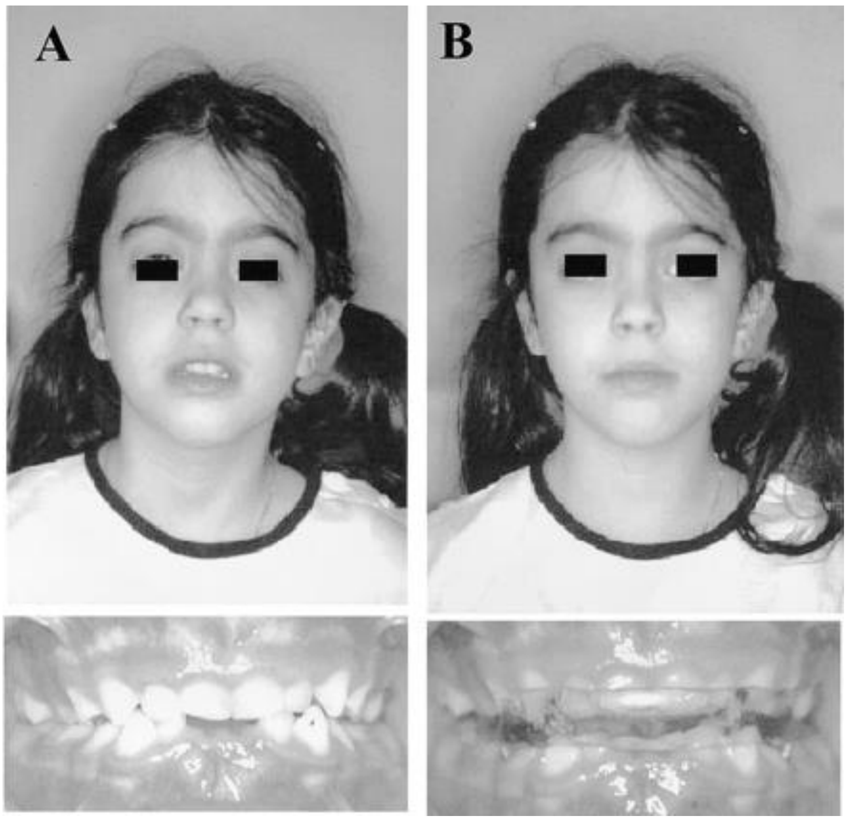
2/10 had treatment failure (persistant OSAS)

Mandibular advancement device

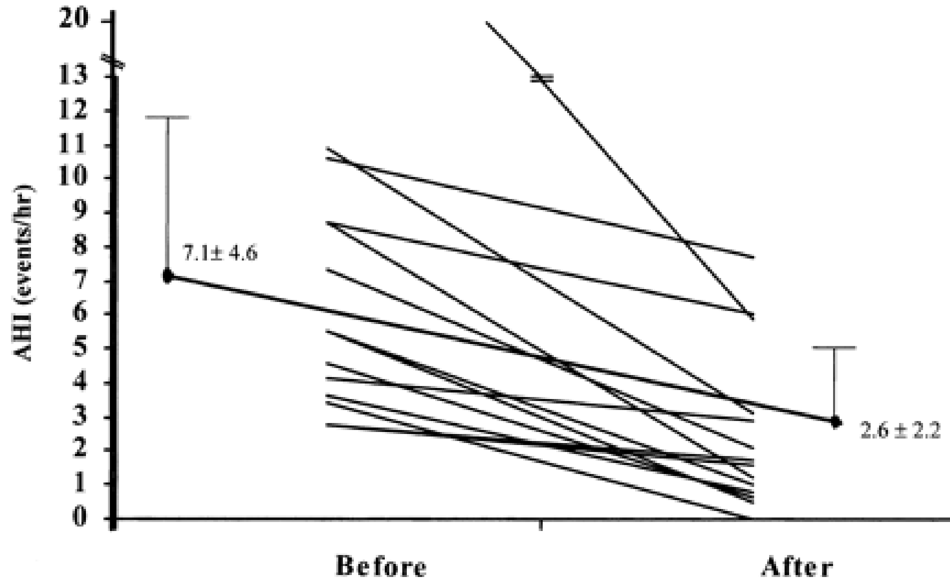


Randomized Controlled Study of an Oral Jaw-Positioning Appliance for the Treatment of Obstructive Sleep Apnea in Children with Malocclusion

MARIA P. VILLA, EDOARDO BERNKOPF, JACOPO PAGANI, VANNA BROIA, MARILISA MONTESANO, and ROBERTO RONCHETTI
Am J Respir Crit Care Med Vol 165. pp 123–127, 2002



19 children (age 7±2 yr) treated with oral appliance vs 13 non treated pts (age 7±3 years)
Evaluation after 3 months of treatment



Other options?

- Revise previous therapeutic options
  - weight loss
  - mandibular advancement devices or rapid maxillary expansion
- Discuss surgery (in selected patients)
  - mandibular distraction osteogenesis
  - craniofacial surgery
- High flow nasal cannula
- Tracheotomy

Neonatal Distraction Surgery for Micrognathia Reduces Obstructive Apnea and the Need for Tracheotomy

William Wittenborn, MD, Jayesh Panchal, MD, MBA, Jeffrey L. Marsh, MD, FACS,
Krishnamurthy C. Sekar, MD, Judith Gurley, MD

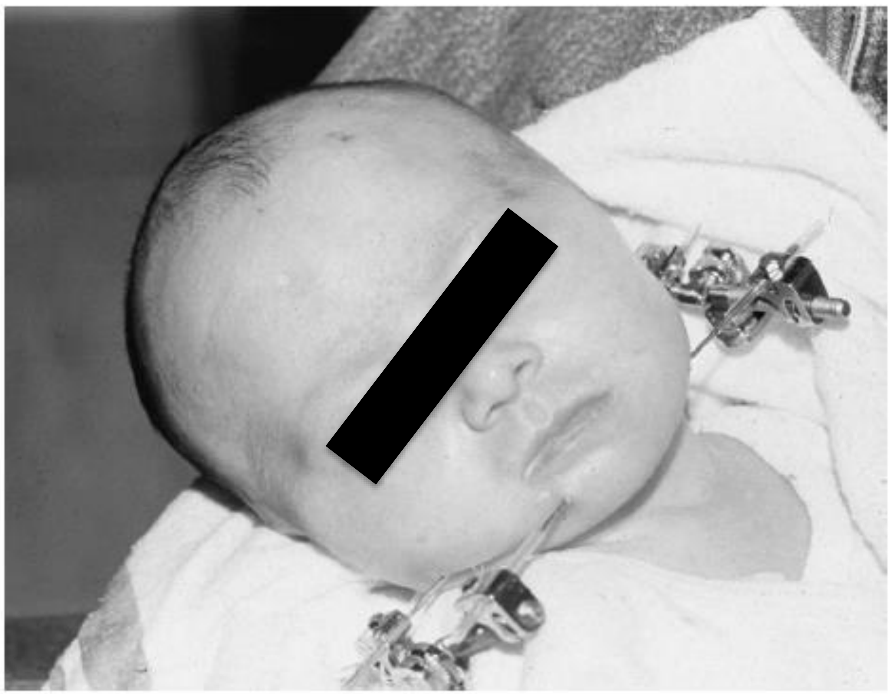


Fig 2 Photograph of the external distraction device in a neonate.

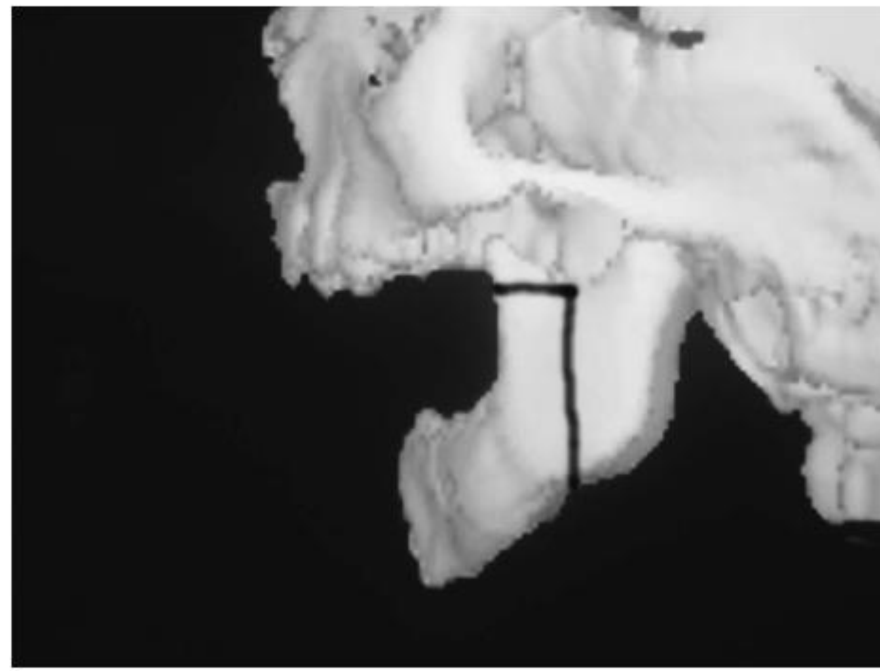


Fig 4 Site of osteotomy over the vertical ramus of the mandible.

Neonatal Distraction Surgery for Micrognathia Reduces Obstructive Apnea and the Need for Tracheotomy

William Wittenborn, MD, Jayesh Panchal, MD, MBA, Jeffrey L. Marsh, MD, FACS, Krishnamurthy C. Sekar, MD, Judith Gurley, MD

17 pts: 1 Treacher Collins, 2 Stickler,14 Pierre Robin)

NO CPAP Trial

All had osteogenesis distraction in neonatal period (mean age 29 days):

- 3 tracheotomy post surgery
- 10 pts had improvement in sleep study after surgery (AHI non specified)

Airway Management in Babies With Micrognathia: The Case Against Early Distraction

Radhika Chigurupati, DMD, and*
Robert Myall, BDS, FDSRCS, FRCD(C), MD†

Table 1. REPORTS OF MANDIBULAR DISTRACTION FOR AIRWAY OBSTRUCTION IN INFANTS AND CHILDREN

Study	Year	n	Complications	Follow-up (mos)*	Success Rate
Cohen et al[34]	1998	16	Aspiration, infection, loose pins, neuropraxia	-	87.5%
Morovic[35]	2000	7	Pin loosening	-	100%
Denny et al[36]	2000	10	Premature consolidation	-	90%
Sidman et al[37]	2001	11	Failure of device	-	90%
Monasterio et al[38]	2002	15	Infection, dislodgement of pins, reoperation	-	100%
Denny and Kalantarian[39]	2002	6	Device failure	16	84%
Izadi et al[40]	2003	15	Infection	14	90%
Chigurupati et al[41]	2004	5	Infection	14	80%
Witternborn et al[42]	2004	17	Premature consolidation	16	82%

NOTE. Some studies did not indicate the mean follow-up after distraction.

Chigurupati and Myall. Airway Management in Babies With Micrognathia. J Oral Maxillofac Surg 2005.

J Oral Maxillofac Surg
63:1209-1215, 2005

Other options?

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- Discuss surgery (in selected patients)
  - mandibular distraction osteogenesis
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- High flow nasal cannula
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Weaning from long term continuous positive airway pressure or noninvasive ventilation in children

Meriem Mastouri MD[1,2] | Alessandro Amaddeo MD[1,3,4] | Lucie Griffon MD[1] | Annick Frapin MSN[1] | Samira Touil BSc[1] | Adriana Ramirez MSc[1,5] | Sonia Khirani PhD[1,5,6] | Brigitte Fauroux MD, PhD[1,3,4]

TABLE 2 Characteristics and outcome of the weaned patients

Male/Female	38/20	
Age at CPAP/NIV initiation (years), median (range)	1.4 (0.01-16.2)	
Ventilatory mode, *n* (%)		
CPAP	50 (86%)	
NIV	8 (14%)	
Duration of CPAP/NIV (year), median (range)	1.12 (0.16-8.85)	
CPAP	0.98 (0.16-7.49)	
NIV	3.96 (0.27-8.85)	
Primary diagnosis, *n* (%)	Laryngeal obstruction[a]	9 (16%)
	Pierre Robin syndrome	6 (10%)
	Prader Willi syndrome	6 (10%)
	Treacher Collins syndrome	6 (10%)
	Bronchopulmonary dysplasia	4 (7%)
	Achondroplasia	3 (5%)
	Idiopathic OSA	2 (3%)
	Craniofaciostenosis (Crouzon, Apert)	2 (3%)
	Pycnodysostosis	2 (3%)
	Mucopolysaccharidosis	2 (3%)
	Polymalformative syndrome	2 (3%)
	Mandibular hypoplasia	2 (3%)
	Lung sequelae of viral infection/ARDS	2 (3%)
	Other[b]	10 (21%)

Pediatric Pulmonology. 2017;1–6.

Weaning from long term continuous positive airway pressure or noninvasive ventilation in children

Meriem Mastouri MD[1,2] | Alessandro Amaddeo MD[1,3,4] | Lucie Griffon MD[1] | Annick Frapin MSN[1] | Samira Touil BSc[1] | Adriana Ramirez MSc[1,5] | Sonia Khirani PhD[1,5,6] | Brigitte Fauroux MD, PhD[1,3,4]

Reason of CPAP/NIV weaning, *n* (%)	Spontaneous improvement:	33 (57%)
	Switch to oxygen therapy:	2 (3%)
	After surgery:	
	ENT surgery	14 (24%)
	Maxillofacial surgery	6 (11%)
	Neurosurgery	1 (2%)
	ENT and neurosurgery	2 (3%)

Pediatric Pulmonology. 2017;1–6.

Other options?

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  - weight loss
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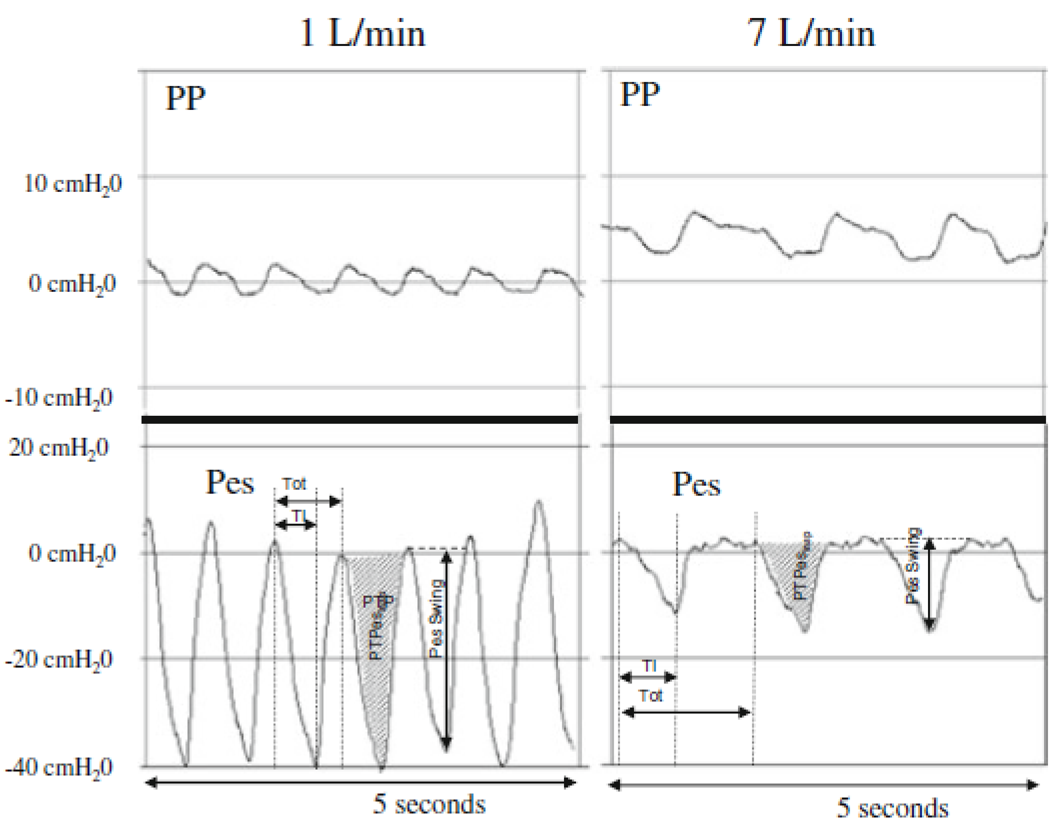
- Tracheotomy

HFNC: how does it work?

Reduction of inspiratory resistance (work of breathing)

BE de Jongh, J Perinatol. 2014; Pham TM, Pediatr Pulmonol. 2014

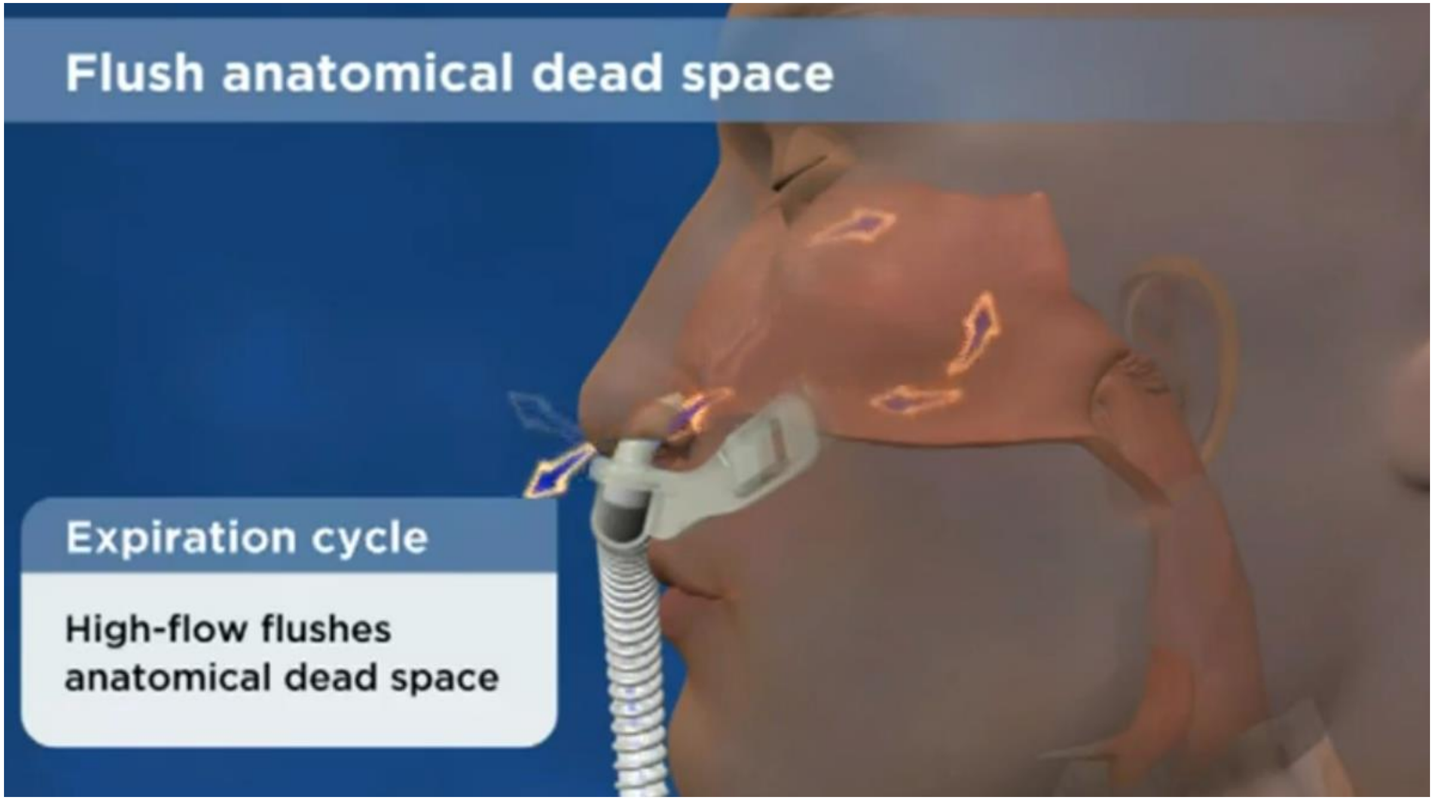


Is treatment with a high flow nasal cannula effective in acute viral bronchiolitis? A physiologic study

Intensive Care Med

HFNC: how does it work?

Washout of nasopharyngeal and intrapulmonary dead space through continual gas removal during expiration (enhance CO_2 removal) *Nahum Resp Care Clinic 2002*



HFNC: how does it work?

Provide support pressure *Arora B, Pediatr Emerg Care*

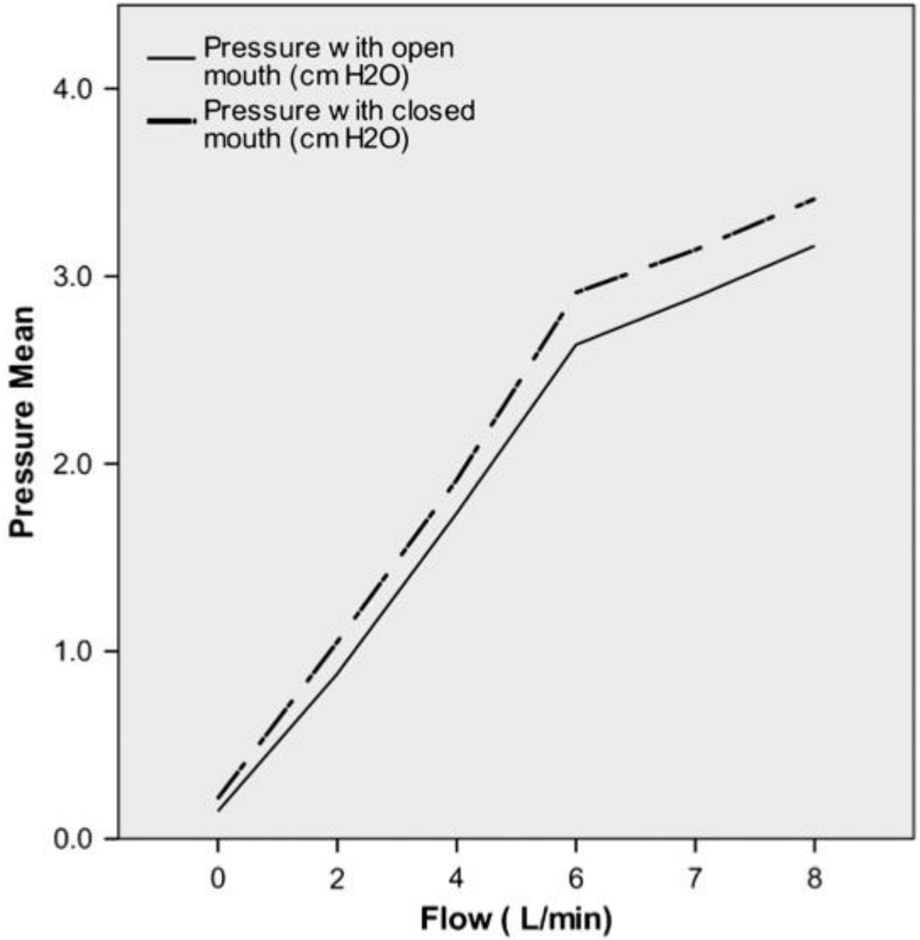


FIGURE 3. Mean pressure at different flow rates for open- and closed-mouth states.

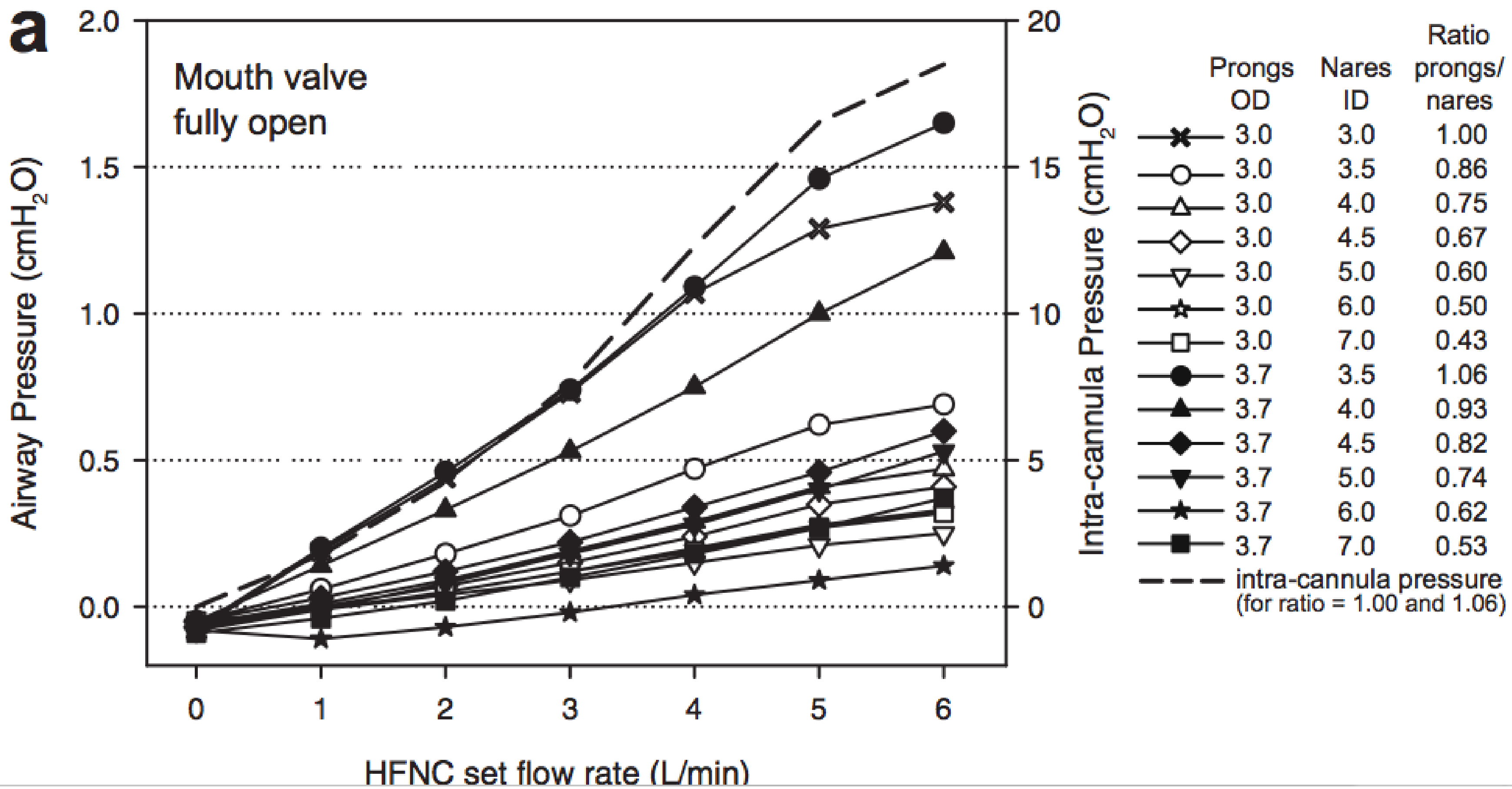
Nasopharyngeal Airway Pressures in Bronchiolitis Patients Treated With High-Flow Nasal Cannula Oxygen Therapy

Bhawana Arora, MD, Prashant Mahajan, MD, MPH, MBA,*† Marwan A. Zidan, PhD,‡ and Usha Sethuraman, MD†*

Pediatric Emergency Care • Volume 28, Number 11, November 2012

Effect of HFNC Flow Rate, Cannula Size, and Nares Diameter on Generated Airway Pressures:
An In Vitro Study

Emidio M. Sivieri, MS.E,[1,2] Jeffrey S. Gerdes, MD,[1,2,3] and Soraya Abbasi, MD[1,2,3]*



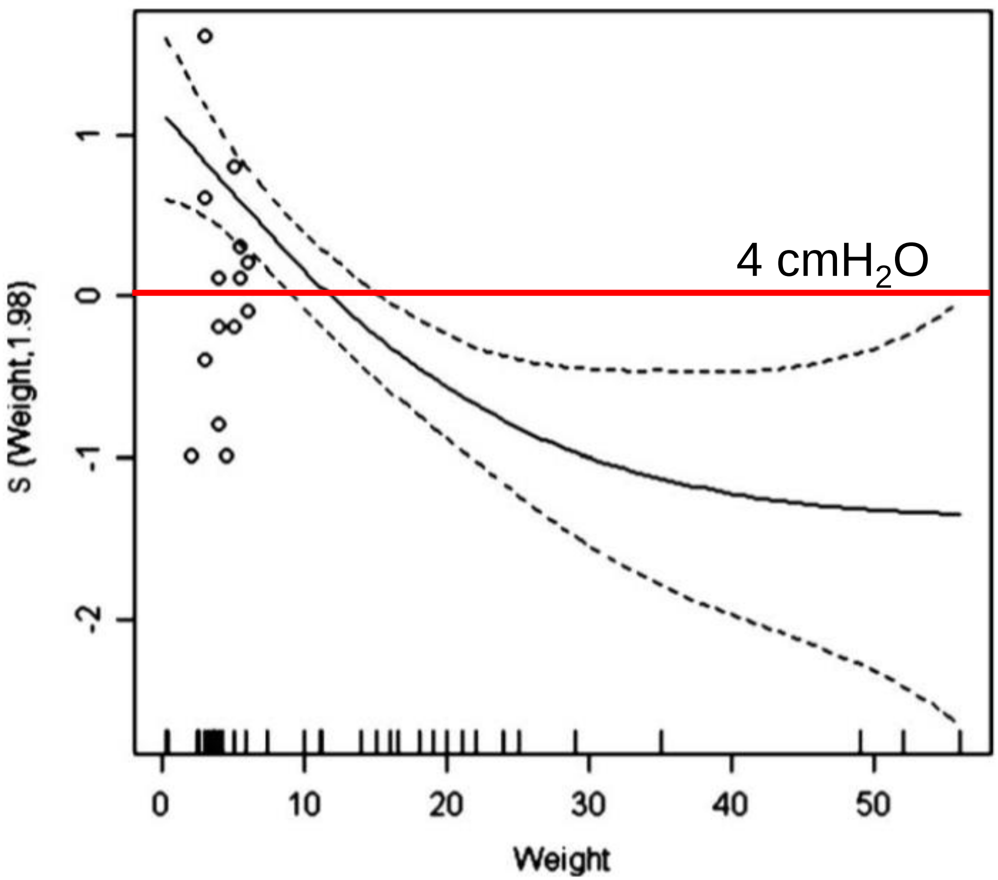
Pediatric Pulmonology 48:506–514 (2013)

Sept

Children With Respiratory Distress Treated With High-Flow Nasal Cannula

Thomas Spentzas, MD, MSc, Milan Minarik, MD, Andrea B. Patters,
Brett Vinson, CRT, LRCP, and Greg Stidham, MD

Journal of Intensive Care Medicine / Vol. 24, No. 5, September/October 2009



End expiratory airway pressure changes during HFNC from the mean airway pressure (4 ± 1.9 cmH_2O)

All patients had a positive end-expiratory pressure, with a direct relation between weight and pressure drop

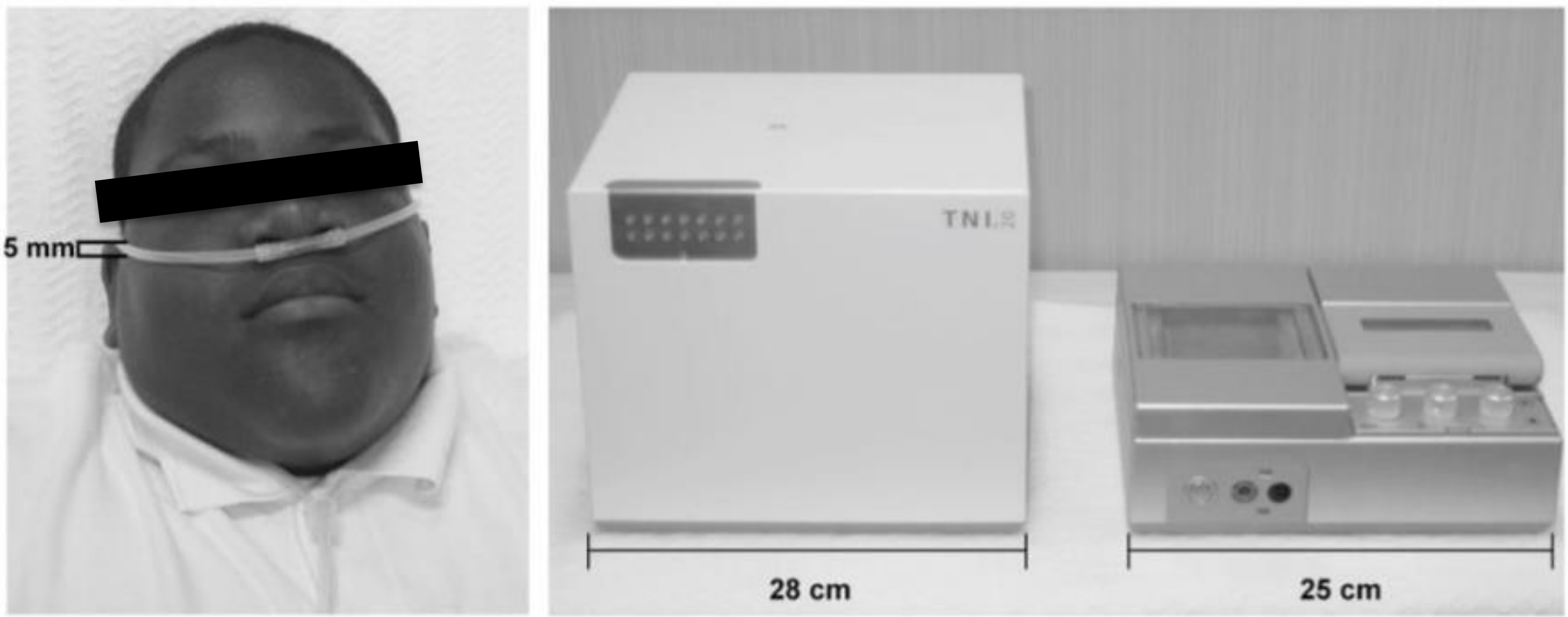
Effect of a High-Flow Open Nasal Cannula System on Obstructive Sleep Apnea in Children

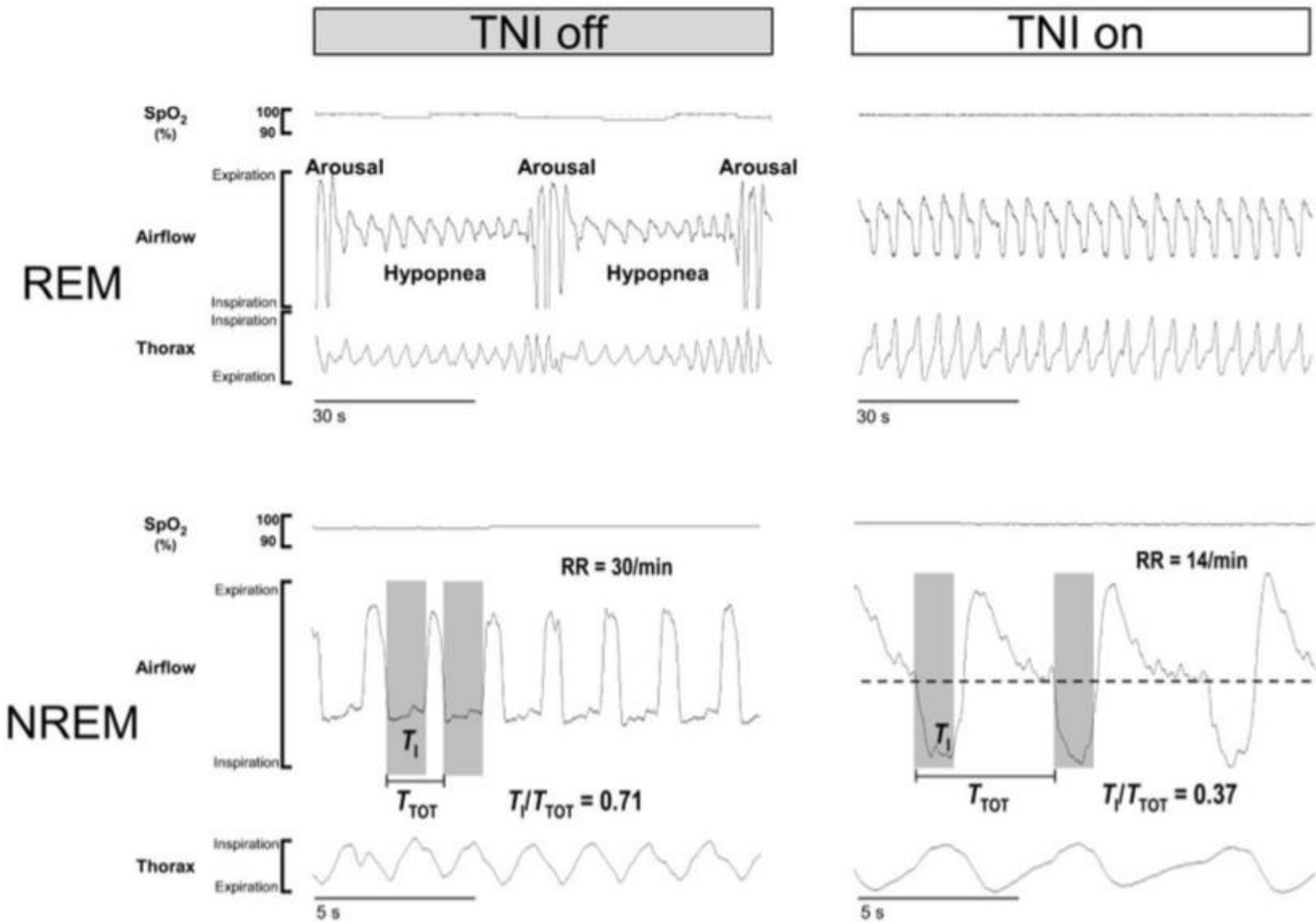
Brian McGinley, MD[a], Ann Halbower, MD[b], Alan R. Schwartz, MD[c], Philip L. Smith, MD[c], Susheel P. Patil, MD, PhD[c], and Hartmut Schneider, MD, PhD[c]

Pediatrics. 2009 July ; 124(1): 179–188. doi:10.1542/peds.2008-2824.

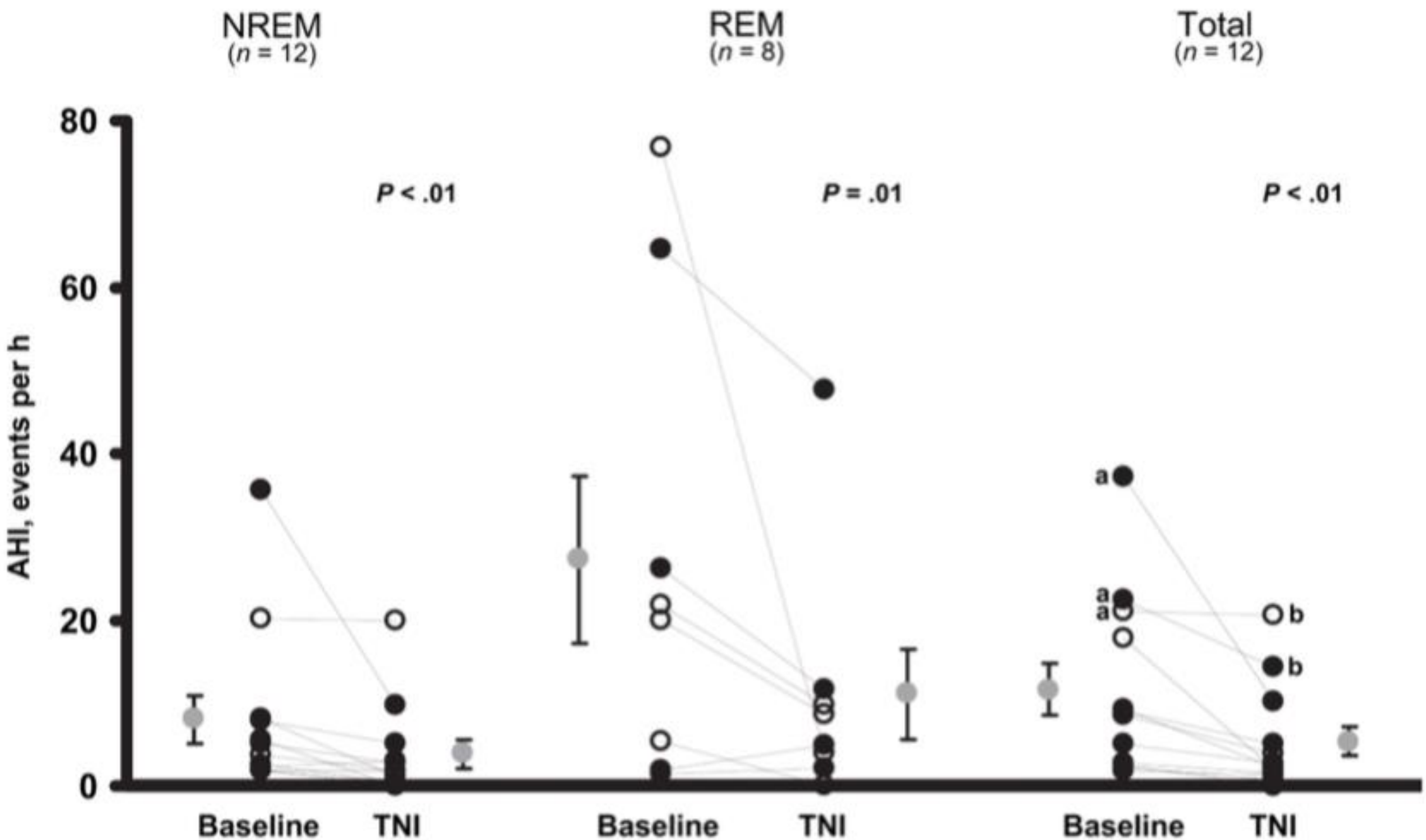
12 children, age 10±2 years, with OSAS + mean BMI 35 ± 14 kg/m^2

One night titration study with a high flow nasal cannula system





TNI off
TNI on
REM
SpO2 (%)
100
90
Arousal
Arousal
Arousal
Expiration
Airflow
Hypopnea
Hypopnea
Inspiration
Inspiration
Thorax
Expiration
30 s
30 s
NREM
SpO2 (%)
100
90
RR = 30/min
RR = 14/min
Expiration
Airflow
Inspiration
TI
TI
TTOT
TI/TTOT = 0.71
TTOT
TI/TTOT = 0.37
Inspiration
Thorax
Expiration
5 s
5 s



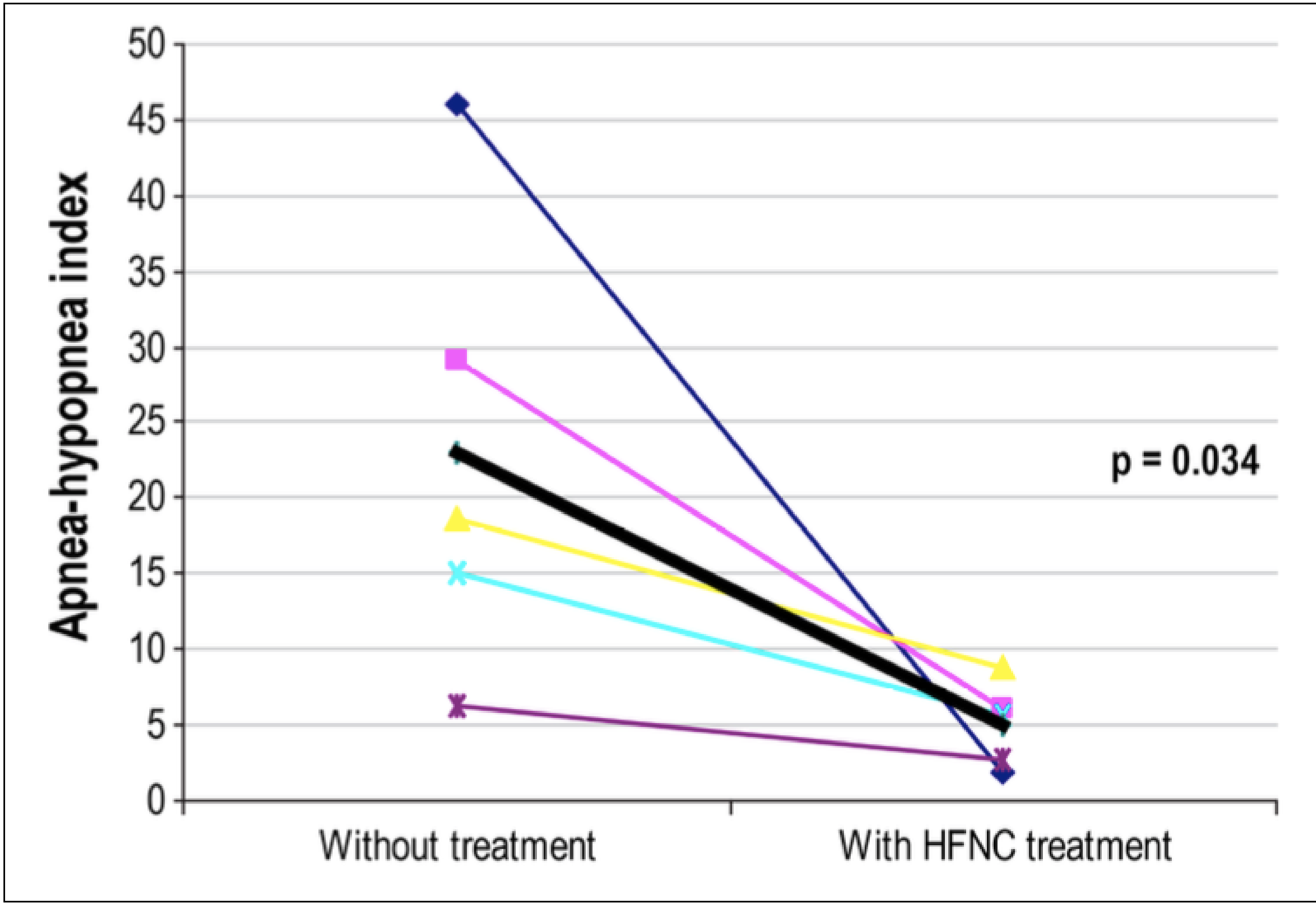
NREM
(n = 12)
REM
(n = 8)
Total
(n = 12)
80
60
40
20
0
AHI, events per h
P < .01
P = .01
P < .01
a
a
a
b
b
Baseline
TNI
Baseline
TNI
Baseline
TNI

High-Flow Nasal Cannula Therapy for Obstructive Sleep Apnea in Children

Leon Joseph, MB ChB; Shmuel Goldberg, MD; Michal S *Journal of Clinical Sleep Medicine, Vol. 11, No. 9, 2015*

5 patients with OSAS who did not tolerate CPAP:

1. Prematurity, bronchopulmonary dysplasia, *age 22 months*
2. Severe psychomotor delay, *age 15 yrs*
3. Polymalformative syndrome, *age 3 yrs*
4. Hypotonia, retrognatia, *age 2 yrs*
5. Treacher Collins, decanulation after mandibular distraction, *age 3 yrs*



➔ No data about objective adherence

High-Flow, Heated, Humidified Air Via Nasal Cannula Treats CPAP-Intolerant Children With Obstructive Sleep Apnea

Stephen Hawkins, MD[1,2]; Stephanie Huston, BS[1]; Kristen Campbell, BS[3]; Ann Halbower, MD[1,2]

[1]The Breathing Institute, Children's Hospital Colorado, Aurora, Colorado; [2]Department of Pediatric Pulmonology, University of Colorado School of Medicine, Aurora, Colorado; [3]Department of Biostatistics and Informatics, University of Colorado, Aurora, Colorado

Table 1—Demographics of study population.

Variable	Population (n = 10)
Sex, n (%)	
Female	6 (60)
Male	4 (40)
Ethnicity, n (%)	
Black	1 (10)
Hispanic	1 (10)
White	8 (80)
Age at HFNC PSG, years	10 (8, 13)
BMI percentile for age, %	88 (68, 98)
Days from diagnostic to HFNC PSG	286 (85, 386)

Proportions or median and interquartile ranges are shown. BMI = body mass index, HFNC = high-flow nasal cannula, PSG = polysomnography.

10 children intolerant to CPAP

3 Down syndrome, 5 craniofacial syndrome

AIRVO2 during a titration study.

Flow range:10 to 50 l/min.

4/10 required oxygen supplementation ??

Journal of Clinical Sleep Medicine, Vol. 13, No. 8, 2017

Table 2—Diagnostic versus optimal high-flow nasal cannula setting.

Variable	Diagnostic (n = 10)	HFNC-Optimal (n = 10)	*P* Value
TRT, min	305.5 (130.5, 372.5)	151.5 (115.0, 276.7)	.375
TST, min	280.5 (101.6, 338.1)	128.8 (114.1, 203.9)	.322
Sleep efficiency, %	88.1 (80.6, 95.0)	92.6 (83.9, 97.1)	.770
Arousal index, events/h	9.1 (5.0, 13.6)	5.5 (3.6, 7.6)	.375
Heart rate mean, bpm	87.7 (85.8, 90.9)	73.6 (66.7, 81.2)	.004
SpO_2 mean, %	91.3 (89.6, 93.5)	94.9 (92.4, 96.1)	.002
SpO_2 nadir, %	76.0 (67.3, 82.3)	79.5 (77.2, 9)	.032
ODI, events/h*	19.2 (12.7, 25.8)	6.4 (4.7, 10.7)	.013
$TcCO_2$ mean, mmHg	44.6 (39.4, 50.5)	39.3 (36.0, 46.0)	.314
$TcCO_2$ max, mmHg	52.9 (45.5, 55.5)	46.9 (45.5, 50.8)	.672
CAHI, events/h*	0.2 (0.0, 0.2)	0.1 (0.0, 0.4)	.779
OAHI, events/h*	11.1 (8.7, 18.8)	2.1 (1.7, 2.2)	.002
OAI, events/h	2.2 (0.4, 6.7)	1.0 (0.4, 2.0)	.625
OHI, events/h*	9.9 (4.7, 15.1)	0.5 (0.0, 1.6)	.002

Improvement of HR, SpO_2 and obstructive events

But…

- Predominance of hypopneas
- Lack of adherence data (children did not continue HFNC at home)
- Impossibility to mesure nasal pressure
- 4 patients required oxygen (no data about the cause of hypoxemia)

High-flow nasal cannula for children not compliant with continuous positive airway pressure

Alessandro Amaddeo [a, b, *], Sonia Khirani [a, b, c], Annick Frapin [a], Theo Teng [a],
Lucie Griffon [a, b], Brigitte Fauroux [a, b]

- Population: children aged 0-18 yrs with OSAS:
  - AHI>10/hour *and/or*
  - oxygen desaturation index > 15/hour *and/or*
  - minimal SpO_2 <90% *and/or*
  - maximal $PtcCO_2$ >50 mmHg
- Non compliant with an optimal CPAP therapy defined by a use < 2 hours/night, after at least 2 weeks of CPAP trial

Sleep Medicine 63 (2019) 24e28

High-flow nasal cannula for children not compliant with continuous positive airway pressure

Alessandro Amaddeo [a, b, *], Sonia Khirani [a, b, c], Annick Frapin [a], Theo Teng [a], Lucie Griffon [a, b], Brigitte Fauroux [a, b]

- Primary endpoint
  - objective compliance (number of hours use / night) evaluated on the device after one month as the mean of the device usage time during the 4th week of use (sole option)
- Secondary endpoints
  - objective compliance after one week as the mean of the device usage time
  - correction of OSAS on a PG with HFNC

High-flow nasal cannula for children not compliant with continuous positive airway pressure

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Lucie Griffon [a, b], Brigitte Fauroux [a, b]

- Procedure – 1
  - myAIRVO device (Fisher Paykel) with appropriate nasal cannula
  - highest tolerable flow and the largest cannula tolerated by the patient (in order to reach the highest airway pressure)

High-flow nasal cannula for children not compliant with continuous positive airway pressure

Alessandro Amaddeo [a, b, *], Sonia Khirani [a, b, c], Annick Frapin [a], Theo Teng [a], Lucie Griffon [a, b], Brigitte Fauroux [a, b]

- Procedure – 2
  - initiation during a 2 hours outpatient visit or a hospitalization
  - control visit after 1 week
  - respiratory polygraphy with HFNC after 1-3 months, when the patient tolerates the HFNC > 6h/night

High-flow nasal cannula for children not compliant with continuous positive airway pressure

Alessandro Amaddeo [a, b, *], Sonia Khirani [a, b, c], Annick Frapin [a], Theo Teng [a], Lucie Griffon [a, b], Brigitte Fauroux [a, b]

Table 1
Anthropometric data, type of nasal cannula, and final high-flow nasal cannula (HFNC) flow rate.

	Gender	Age (years)	Weight (kg)	Height (m)	Underlying disorder	Type of cannula	HFNC flow rate (L/min)
Patient 1	Female	0.1	4.9	0.60	Pierre Robin sequence	Infant	5
Patient 2	Female	1.8	9.4	0.70	Down syndrome	Pediatric	15
Patient 3	Female	6.4	19.5	1.10	Pfeiffer syndrome	Pediatric	15
Patient 4	Male	7.6	29	1.20	Down syndrome	Adult Small	20
Patient 5	Male	9.2	27	1.20	Down syndrome	Adult Medium	20
Patient 6	Female	12	76	1.50	Down syndrome	Adult Medium	20
Patient 7	Female	16.2	44	1.30	Down syndrome	Adult Medium	20
Patient 8	Female	17.3	85	1.50	Down syndrome	Adult Medium	20

High-flow nasal cannula for children not compliant with continuous positive airway pressure

Alessandro Amaddeo [a, b, *], Sonia Khirani [a, b, c], Annick Frapin [a], Theo Teng [a],
Lucie Griffon [a, b], Brigitte Fauroux [a, b]

Table 2
Respiratory polygraphy data during spontaneous breathing and with high-flow nasal cannula.

Patient	Spontaneous breathing AHI (events/h)	OAI (events/h)	OHI (events/h)	Mean SpO_2 (%)	Minimal SpO_2 (%)	ODI (events/h)	Mean $PtcCO_2$ (mmHg)	Maximal $PtcCO_2$ (mmHg)
1	27	5	18	95	77	27	43	51
2	11	1	8	97	87	15	43	47
3	13	9	4	98	85	4	39	50
4	64	19	45	98	83	31	59	66
5	64	12	52	96	82	25	48	50
Mean ± SD[a]	36 ± 26	9 ± 7	25 ± 2	97 ± 1	83 ± 4	20 ± 11	46 ± 8	53 ± 7
6	45	13	31	93	80	48	45	51
7	28	12	16	95	78	16	45	52
8	10	1	9	97	91	8	40	48
Mean ± SD[b]	33 ± 22	9 ± 6	23 ± 18	96 ± 2	83 ± 5	21 ± 14	45 ± 6	52 ± 6

Compliance at 1 month (h/night)	With high-flow nasal cannula AHI (events/h)	OAI (events/h)	OHI (events/h)	Mean SpO_2 (%)	Minimal SpO_2 (%)	ODI (events/h)	Mean $PtcCO_2$ (mmHg)	Maximal $PtcCO_2$ (mmHg)
6 h 40 min	6	2	3	99	93	5	44	48
7 h 30 min	1	0	1	96	91	6	44	46
6 h 50 min	0.5	0	0.5	97	93	5	46	48
6 h 45 min	2	0	2	96	88	9	42	49
8 h 5 min	0.5	0	0	96	89	3	48	50
7 h 10 min ± 0 h 36 min	2 ± 2	0.5 ± 1	1 ± 1	97 ± 1	91 ± 2	6 ± 2	45 ± 2	48 ± 2
1 h 30 min	Not compliant with high-flow nasal cannula							
0 h 50 min	Not compliant with high-flow nasal cannula							
1 h 15 min	Not compliant with high-flow nasal cannula							
–	–	–	–	–	–	–	–	–

- 2/8 patients did not tolerate HF
- Patient 3 required a tracheotomy after developing a tracheal stenosis following a neurosurgical intervention

HFNC: Conclusion

- HF may be efficient in mild to moderate OSAS in children (> hypopneas)
- HF may be better tolerated than CPAP, and could represent an alternative to CPAP in selected, non compliant patients
- Limitations of HFNC
  - no pressure monitoring: risk of high pressure when use with large cannula
  - no battery, alarms (security risk), no objective compliance (no in-built software)
- Future studies
  - patient selection ?
  - optimal flow rate ?

Other options?

- Revise previous therapeutic options
  - weight loss
  - mandibular advancement devices or rapid maxillary expansion
- Discuss surgery (in selected patients)
  - mandibular distraction osteogenesis
  - craniofacial surgery
- High flow nasal cannula
- Tracheotomy

IDEAS AND INNOVATIONS

Continuous Positive Airway Pressure for Upper Airway Obstruction in Infants with Pierre Robin Sequence

Amaddeo *et al*. Plastic and Reconstructive Surgery, 2016;137:609

Neonates with PRS evaluated over one year
n=44

Neonates seen as outpatients
n=7

No UAO group

Neonates hospitalized
n=37

No clinical UAO
n=17

No UAO group

Clinical UAO
n=20

Severe clinical UAO
n=9
Immediate CPAP in the NICU

Severe UAO group

Moderate clinical UAO
n=11
Sleep study with gas exchange

Tracheotomy
n=4

CPAP
n=5

Abnormal sleep study
CPAP, n=4

Moderate UAO group

Normal sleep study
n=7

Mild UAO group

Amaddeo *et al*.
Plastic and Reconstructive Surgery, 2016;137:609

Multidisciplinary team is mandatory!