A Brief History of Case & Clinical Studies For OSA, Sleep & Breathing, and Tongue-Tie Disorders

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Affiliations and Disclosures

Soroush Zaghi, MD
ENT- Sleep Surgeon
Otolaryngology & Maxillofacial Surgery

- Medical Director
  - *The Breathe Institute*
- Speaker / Consultant / Board Member
  - *Academy of Applied Myofunctional Sciences*
  - *Academy of Orofacial Myofunctional Therapy*
  - *Airway Focused Dentistry Mini-Residency*
  - *ALF InterFACE Advisory Board*
  - *American Academy of Physiological Medicine and Dentistry*
  - *American Academy of Craniofacial Pain*
  - *American Laser Study Club*
  - *Buteyko Breathing Educators Association*
  - *International Association of Orofacial Myology*
  - *International Consortium of Oral Ankylofrenula Professionals*
  - *Itamar Medical: Watch PAT Home Sleep Device*
  - *Light Scalpel CO2 laser; JedMed Surgical Instruments*
  - *Myofunctional Research Company; MyoBrace; MyoMunchee*

Harvard
Medical School Alumnus

UCLA
ENT Residency

Stanford
Sleep Surgery Fellowship

Stanford-Trained Sleep Surgeon:

- Multidisciplinary perspective to advanced treatment of OSA.
- Sleep Medicine, Sleep Dentistry, Otolaryngology (ENT), Maxillofacial Surgery, and Myofunctional Sciences.
- Clinical Research and Evidence-Based Medicine.

the BREATHE INSTITUTE

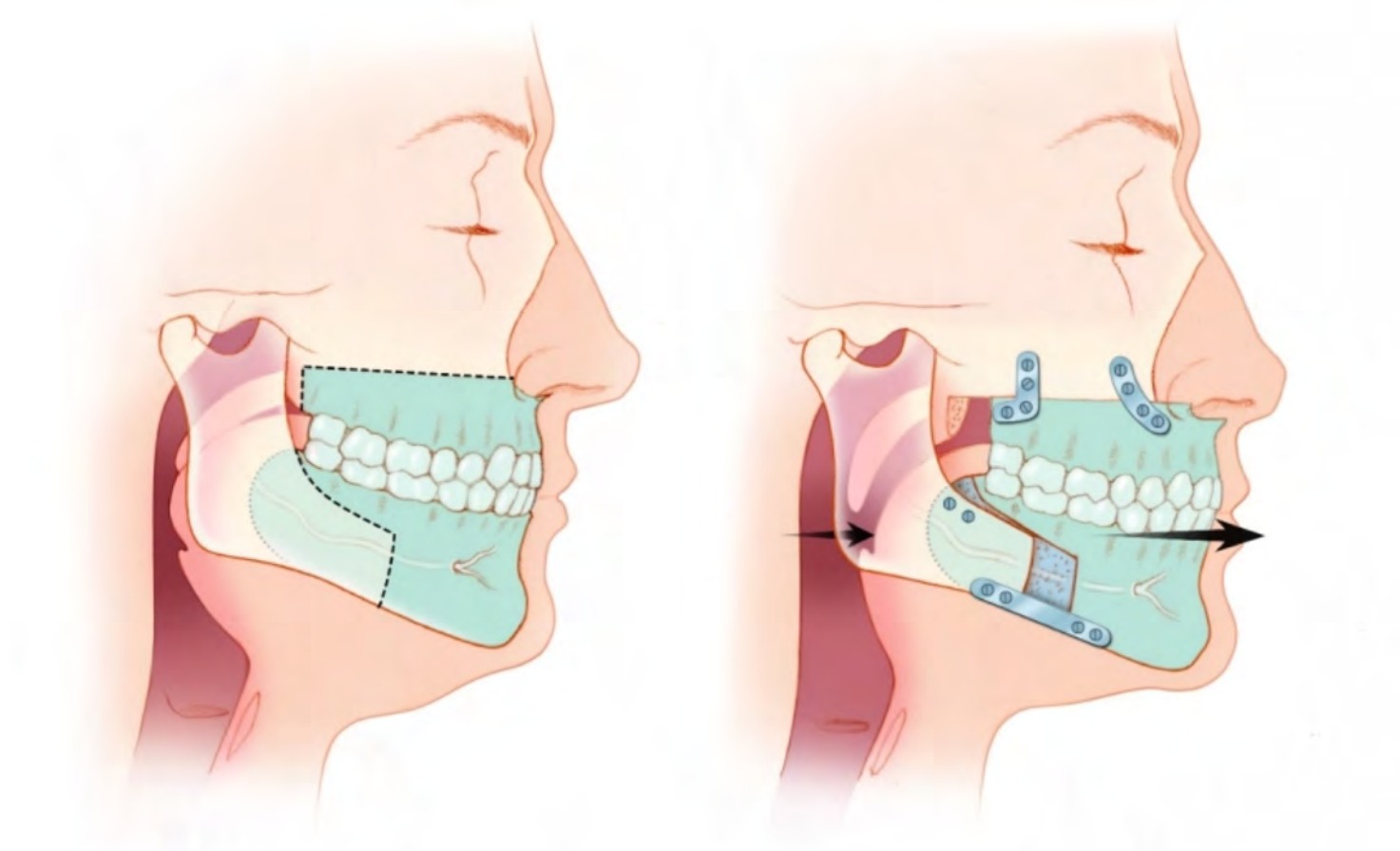
Structural & functional approach to sleep and breathing issues for children and adults.

Patient care, research, and education.

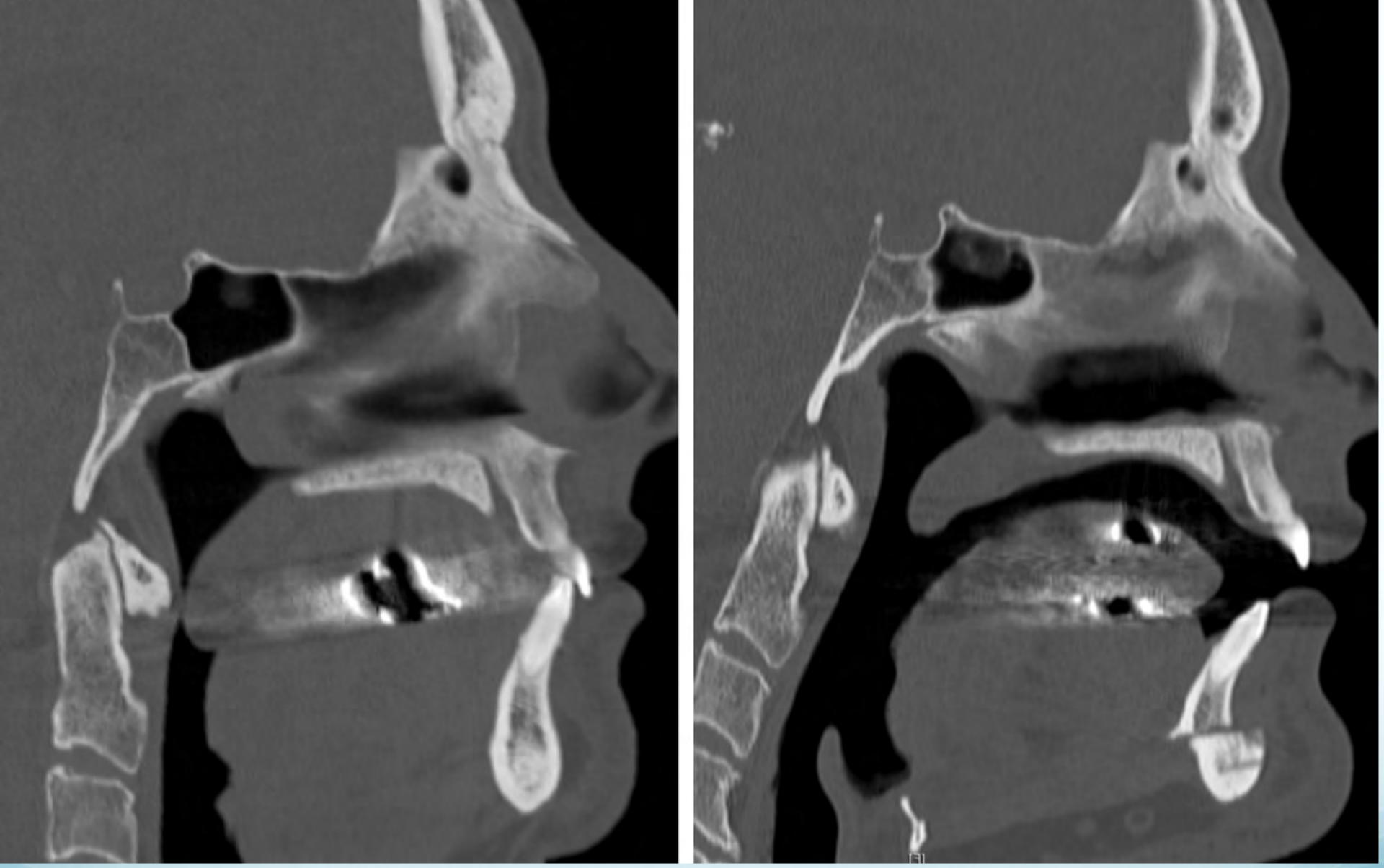
Maxillomandibular Advancement for Treatment of Obstructive Sleep Apnea: A Meta-Analysis

Soroush Zaghi, MD; Jon-Erik C. Holty, MD, MS; Victor Certal, MD; Jose Abdullatif, MD; Christian Guilleminault, DM, MD, DBiol; Nelson B. Powell, MD, DDS; Robert W. Riley, MD, MS, DDS; Macario Camacho, MD

JAMA Otolaryngology–Head & Neck Surgery. 2016 Jan 1;142(1):58-66.



Forty-fives studies with individual data from 518 patients were included



MMA orthognathic surgery successfully opens the airway and reduces the severity of obstructive sleep apnea by 65 to 80%!

Table 2. Rates of Surgical Success or Cure by Preoperative AHI Severity

Surgical Success[a]	Preoperative AHI Cohort, Events/h			
	<30 (n = 61)	30 to <60 (n = 192)	60 to <90 (n = 161)	≥90 (n = 41)
AHI cure, No. (%)	34 (55.7)[b]	88 (45.8)[b]	45 (28.0)	8 (19.5)
AHI Success-10, No. (%)	47 (77.0)[b]	140 (72.9)[b]	77 (47.8)	24 (58.5)
AHI Success-15, No. (%)	51 (83.6)[c]	169 (88.0)[c]	117 (72.7)	29 (70.7)
AHI Success-20, No. (%)	51 (83.6)[d]	176 (91.7)[d]	130 (80.7)[d]	31 (75.6)

Abbreviation: AHI, Apnea-Hypopnea Index.

[a] Surgical success is defined as a greater than 50% reduction of AHI to fewer than 20 events/h after maxillomandibular advancement (MMA) (AHI Success-20); AHI Success-15, AHI levels of fewer than 15 events/h after MMA; AHI Success-10, AHI levels of fewer than 10 events/h after MMA; and AHI cure, AHI levels of fewer than 5 events/h after MMA.

[b] $P < .001$, by Pearson χ^2 analysis.

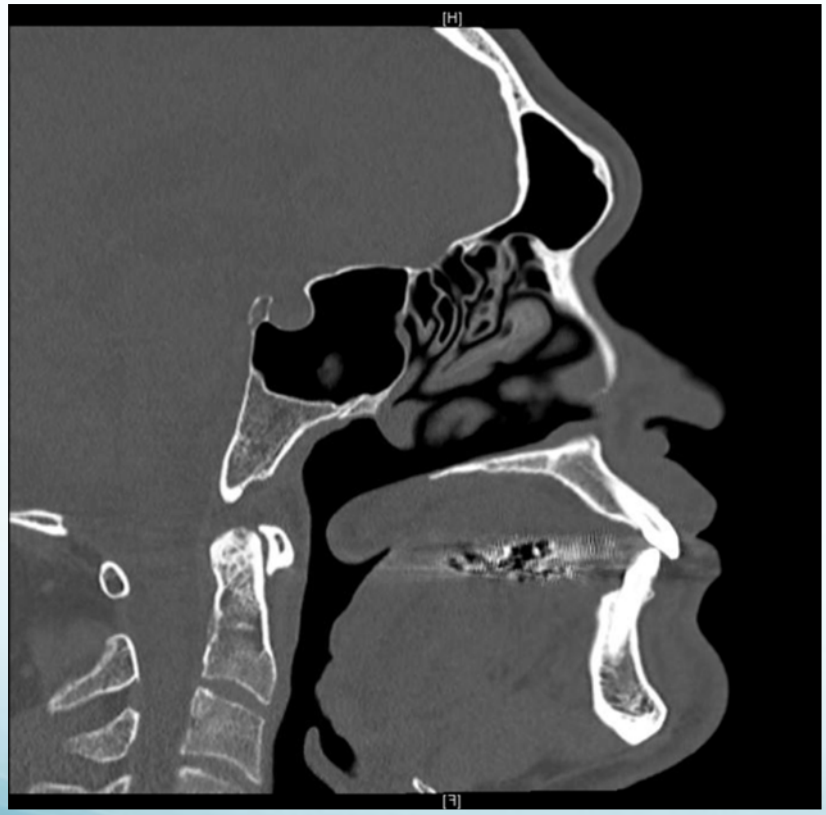
[c] $P = .009$, by Pearson χ^2 analysis.

[d] $P = .003$, by Pearson χ^2 analysis.

- Overall, surgical success rate of 85.5%.
- But, only 38.5% of patients were completely cured of sleep apnea after surgery.

Functional restriction after maxillary mandibular advancement surgery: Persistent tongue-base obstruction.

Pre-Operative CT Scan
(Before MMA Surgery)



Post-Operative CT Scan
(2 Days after MMA Surgery)

The tongue is still blocking the airway

Functional Limitation:
Weak and low tone tongue

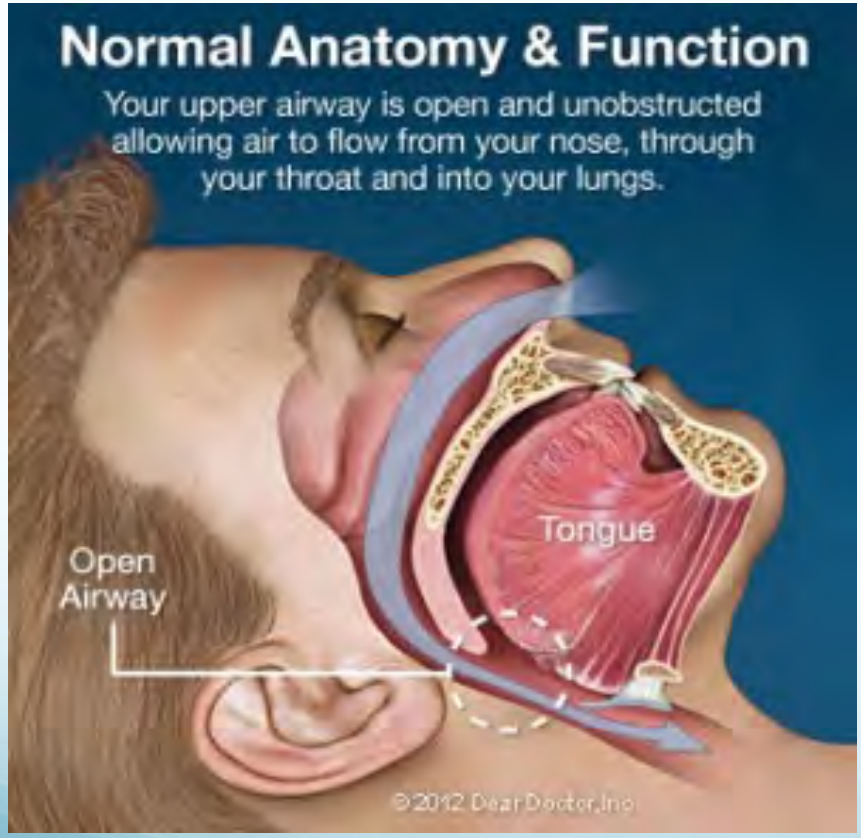

"Sleep Apnea" | Jo Koy : Lights Out

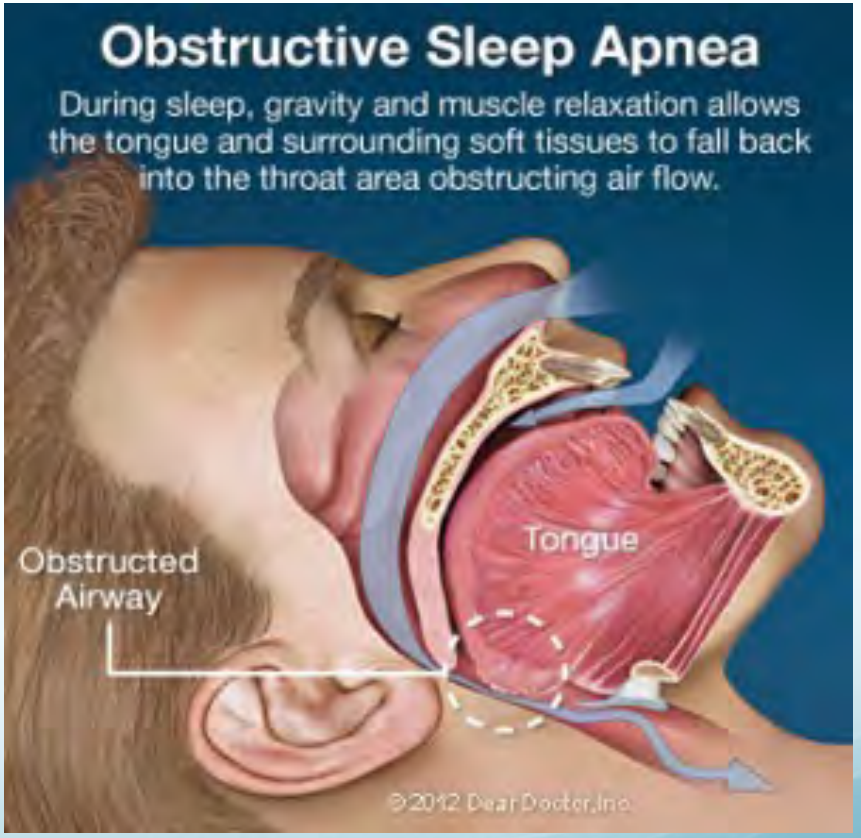

NETFLIX
STAND UP
COMEDY SPECIAL


COMEDY CENTRAL
THE JO KOY SPECIAL

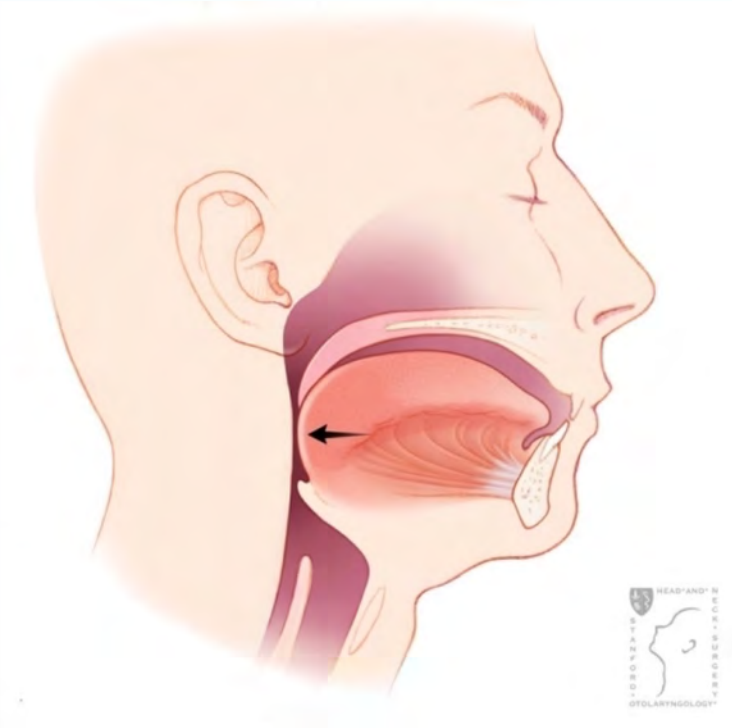

Maui

Impact of a low tone tongue in sleep-disordered breathing

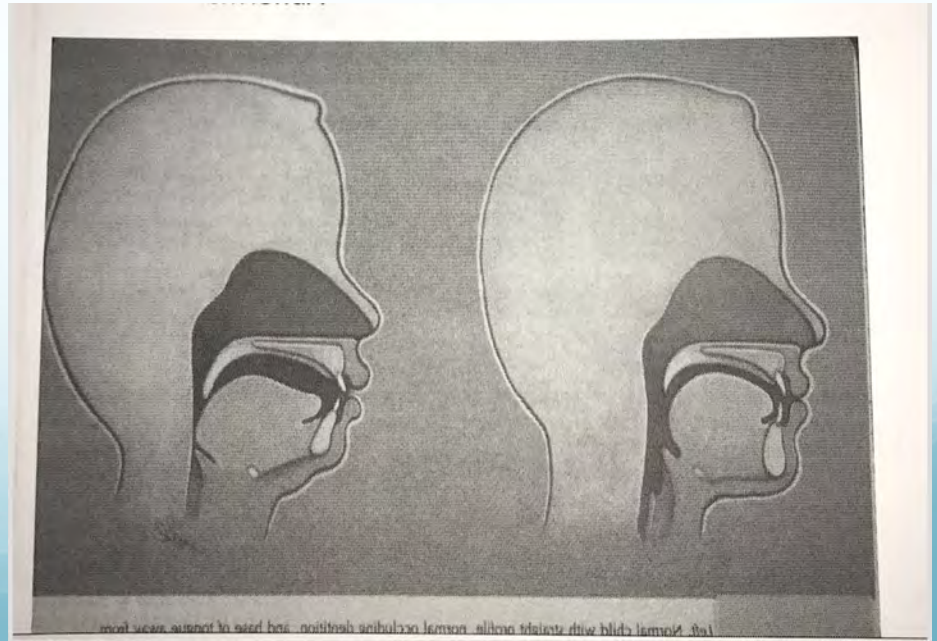




Tongue-tie may interfere with tongue mobility and range of motion.



Tongue assumes low position; may block the airway awake and during deep sleep.



Case Study: 3-year-old girl with sleep-disturbances, speech delay, open mouth breathing, trouble chewing, oral dysphagia and chronic nasal congestion found to have Grade 4 tongue-tie and Class III malocclusion.

Madelyn - 3 year-old girl with sleep-disordered breathing, swallow, and speech issues treated with myofunctional therapy and minor surgical procedure (tongue-tie and lip-tie release).

Pre-Op

Noisy mouth breathing with lips apart

Post - Op

Quiet, lips together, nasal breathing

Hindawi
Case Reports in Otolaryngology
Volume 2019, Article ID 3408053, 5 pages
https://doi.org/10.1155/2019/3408053

Case Report

Lingual and Maxillary Labial Frenuloplasty with Myofunctional Therapy as a Treatment for Mouth Breathing and Snoring

Chirag Govardhan,[1] **Janine Murdock**,[2] **Leyli Norouz-Knutsen**,[1] **Sanda Valcu-Pinkerton**,[1] **and Soroush Zaghi**[1,3]

[1]*The Breathe Institute, Los Angeles, CA, USA*
[2]*South County Pediatric Speech, Mission Viejo, CA, USA*
[3]*UCLA Health, Santa Monica, CA, USA*

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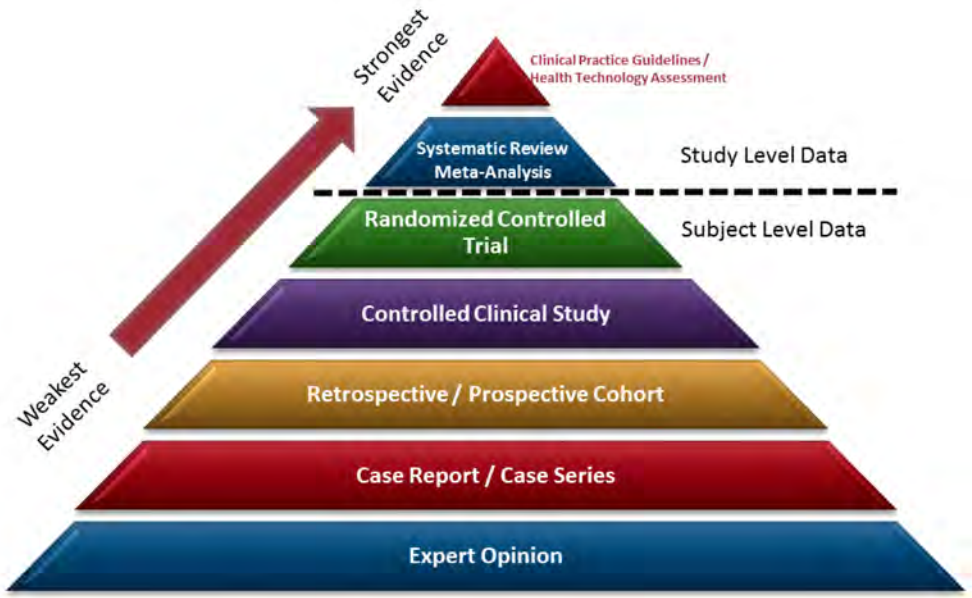
Academic Editor: Rong-San Jiang

Chronic mouth breathing may adversely affect craniofacial development in children and may result in anatomical changes that directly impact the stability and collapsibility of the upper airway during sleep. Mouth breathing is a multifactorial problem that can be attributed to structural, functional, and neurological etiologies, which are not all mutually exclusive. While therapeutic interventions (myofunctional, speech and swallowing, occupational, and craniosacral therapy) may address the functional and behavioral factors that contribute to mouth breathing, progress may sometimes be limited by restrictive lingual and labial frenum that interfere with tongue and lip mobility. This case report explores the case of a three-year-old girl with mouth breathing, snoring, noisy breathing, and oral phase dysphagia that was successfully treated with lingual and labial frenuloplasty as an adjunct to myofunctional therapy. Within four days of the procedure, the patient had stopped snoring and demonstrated complete resolution of open mouth breathing. The patient was also observed to have increased compliance with myofunctional therapy exercises. This report highlights the effectiveness of surgical interventions to improve the efficacy of myofunctional therapy in addressing open mouth posture and low tongue resting position.

Level 5 evidence: Case reports are regarded as the lowest level of evidence due to chance of bias and likelihood for alternative explanations for the outcome to be found.



Strength	Level	Design	Randomization	Control
High	Level 1	Randomized control trial (RCT)	Yes	Yes
		Meta-analysis of RCT with homogeneous results	No	
	Level 2	Prospective comparative study (therapeutic)	No	Yes
		Meta-analysis of Level 2 studies or Level 1 studies with inconsistent results	No	
	Level 3	Retrospective Cohort Study	No	Yes
		Case-control Study	No	Yes
		Meta-analysis of Level 3 studies	No	
	Level 4	Case Series	No	No
	Level 5	Case Report	No	No
		Expert Opinion	No	No
Low		Personal Observation	No	No

Summary of symptoms before release:

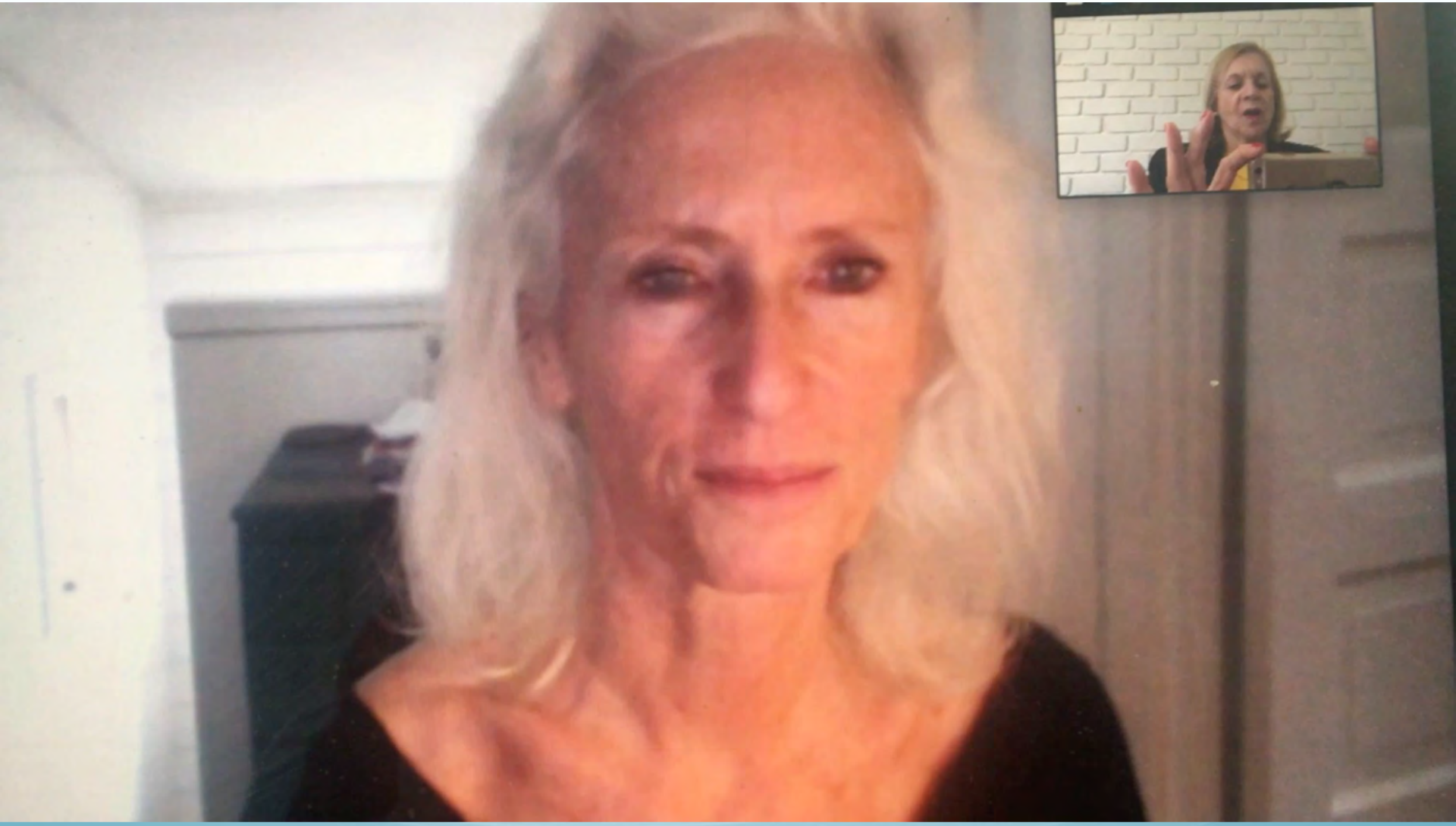
- 60 y/o female with severe headaches, jaw tension, sleep problems
- Tremendous forward head posture
- Had frenectomy when she was 7, but it did not make a huge difference
- Had palate expansion, which helped somewhat but did not resolve the issues
- Joy Moeller said her issues might resolve with a deeper (posterior) frenectomy. Dr. Zaghi evaluated and agreed.

Emotional perspective after release:

- "Nothing feels as serious, everything feels more handle-able"
- Felt "opening in the back of the neck, particularly where the neck connects to the spine".
- "The way I hold myself is completely different. This would have been impossible before the surgery"
- "In one second, everything opened up. The pressure in my jaw is gone. The pressure in the back of my head is gone."
- Dowager's hump (used to be quite large) is gone, and her spine has to adjust to that due to her prior scoliosis.
- Her spinal adjustments are close to resolving.
- Made her calmer, she is taking things easier.

"This is what normal people feel like. This is how easy it can be in the world." – Katarina W.

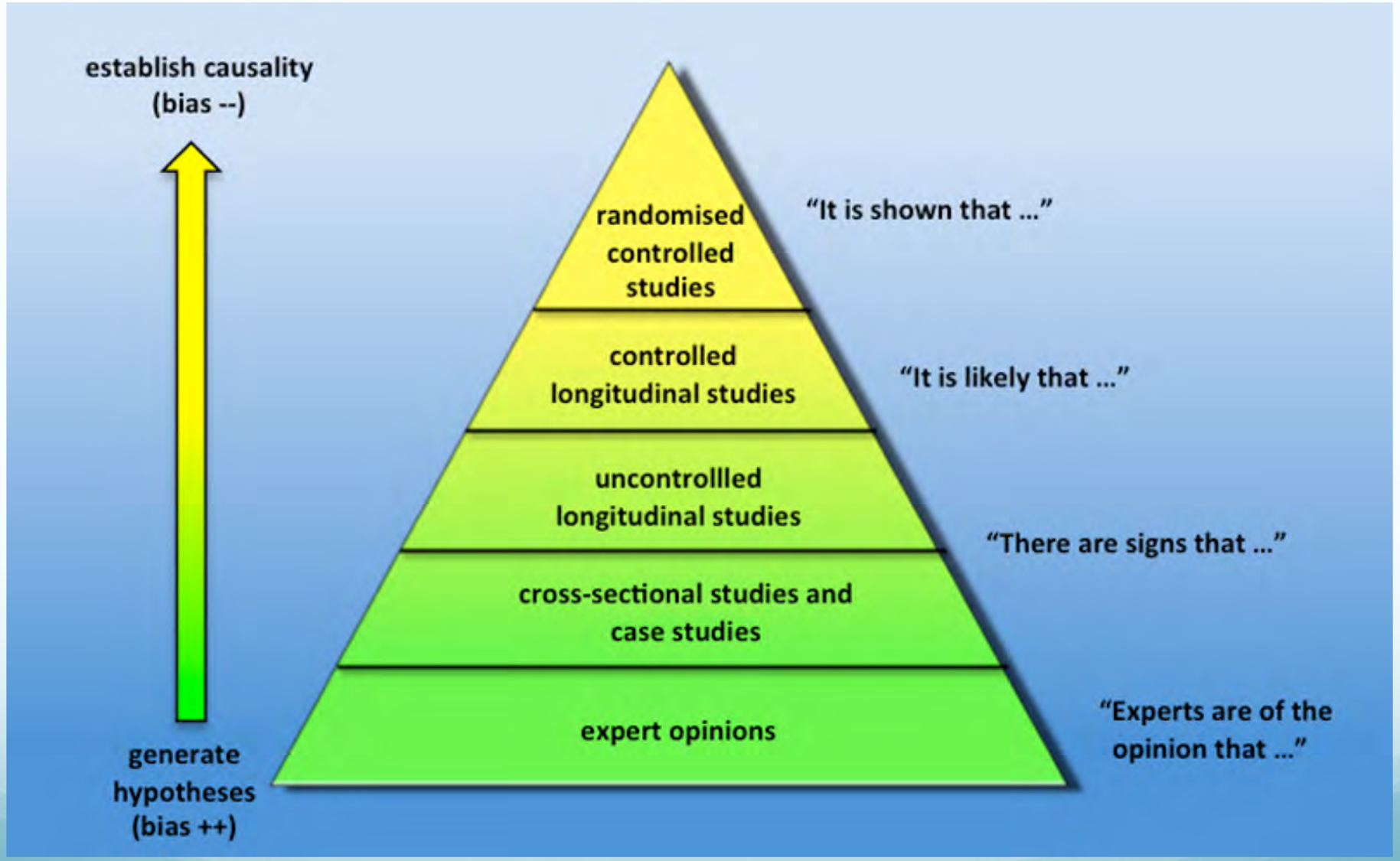
Two years later: rejuvenation of face, body, and spirit.



Feedback on my video

"Soroush,

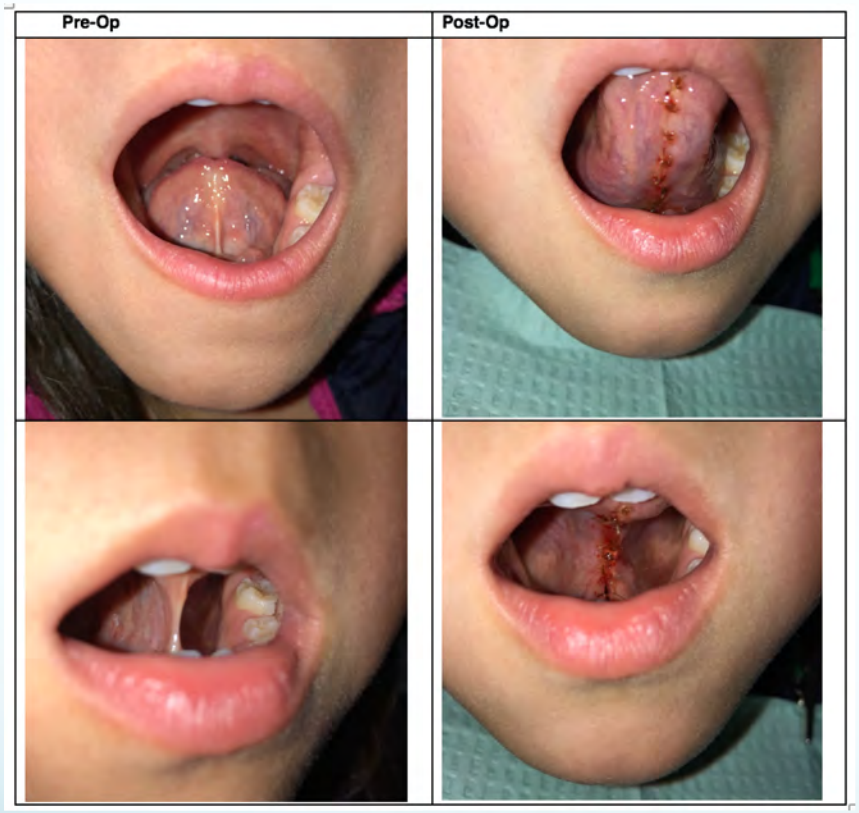
I will be honest and say that I think you are better than this 1-patient testimonial would suggest. Anecdotal experiences like this can form the basis of scientific evaluation, but it is pretty useless in itself. I would strongly recommend that you develop an evidence basis with higher-quality science than this."


establish causality
(bias --)
randomised controlled studies
"It is shown that ..."
controlled longitudinal studies
"It is likely that ..."
uncontrollled longitudinal studies
"There are signs that ..."
cross-sectional studies and case studies
expert opinions
"Experts are of the opinion that ..."
generate hypotheses
(bias ++)

<u>Case #1</u>: Madelyn 2 Months Post-Op

Thank you: Jeanine Murdock, SLP

Case #2: 11 year-old girl with sleep-disordered breathing and depressed mood issues

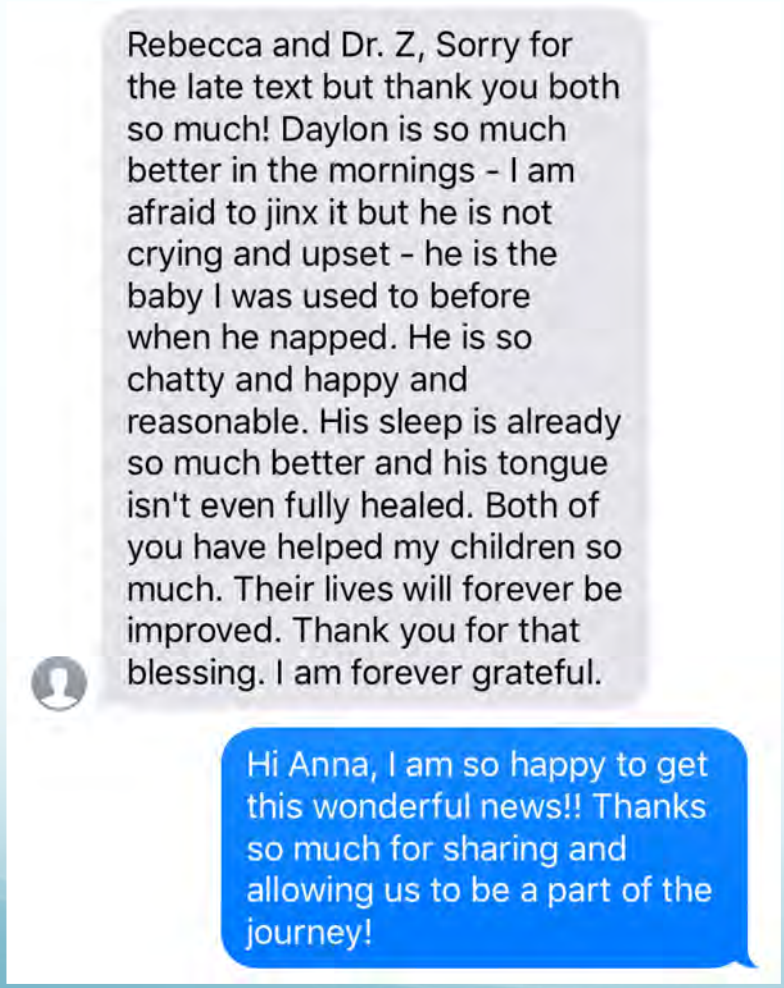


Jen,
Can you please show the day after surgery picture of Mikayla to Dr. Zaghi.
He's right she has more energy!
Thanks for your great team & amazing work.
Lori

Sent from my iPhone

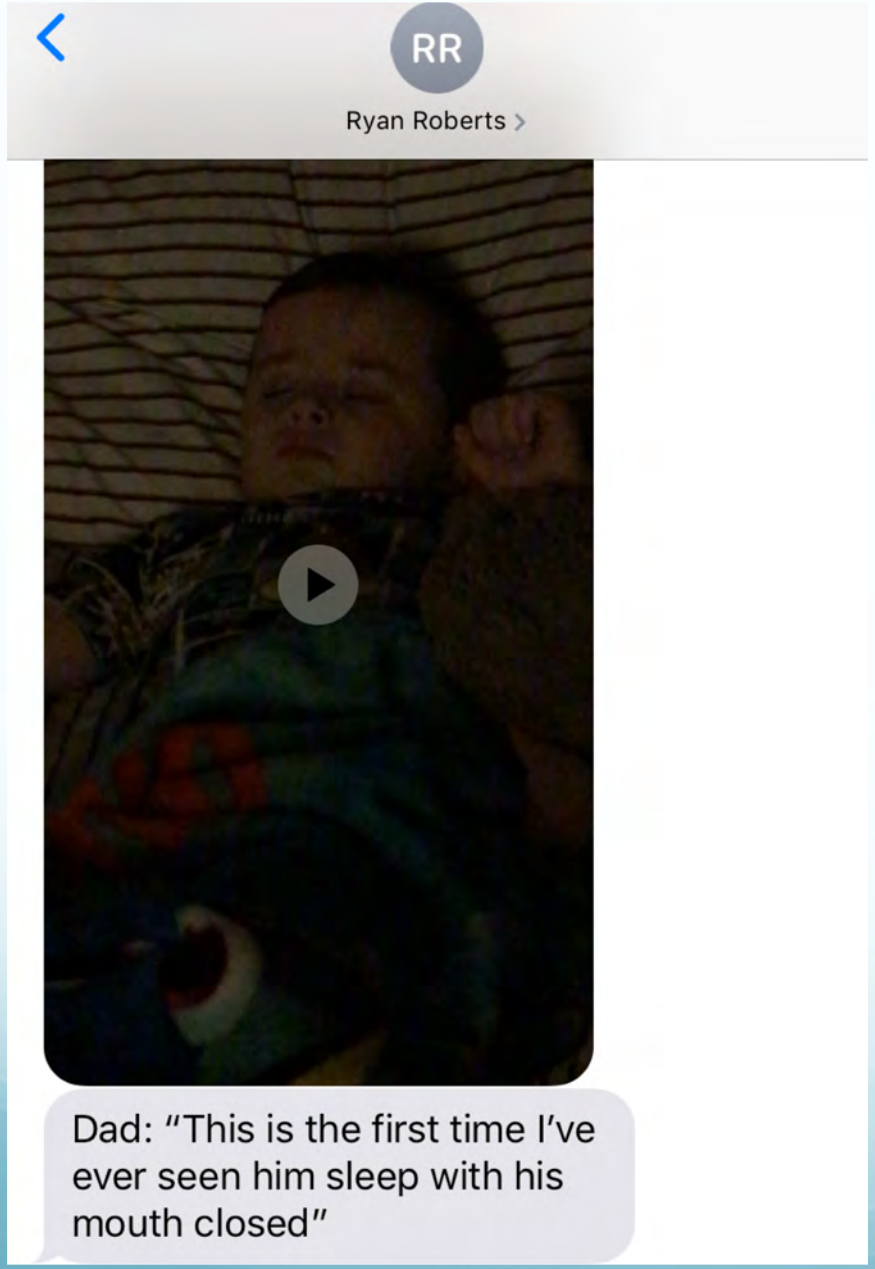
Case #3: 3-year-old boy with open mouth breathing, restless sleep, maladaptive swallow habits, and maxillofacial underdevelopment associated with oral myofascial dysfunction.

Day 5 Post-Op, 9:49 pm



Day 7 Post-Op

Case #4: Case from a colleague: 3 year-old boy with mouth breathing.



Credit: Dr. Ryan Roberts

Case #5: 7 year old female with mouth breathing, maxillofacial retrusion, and posture issues

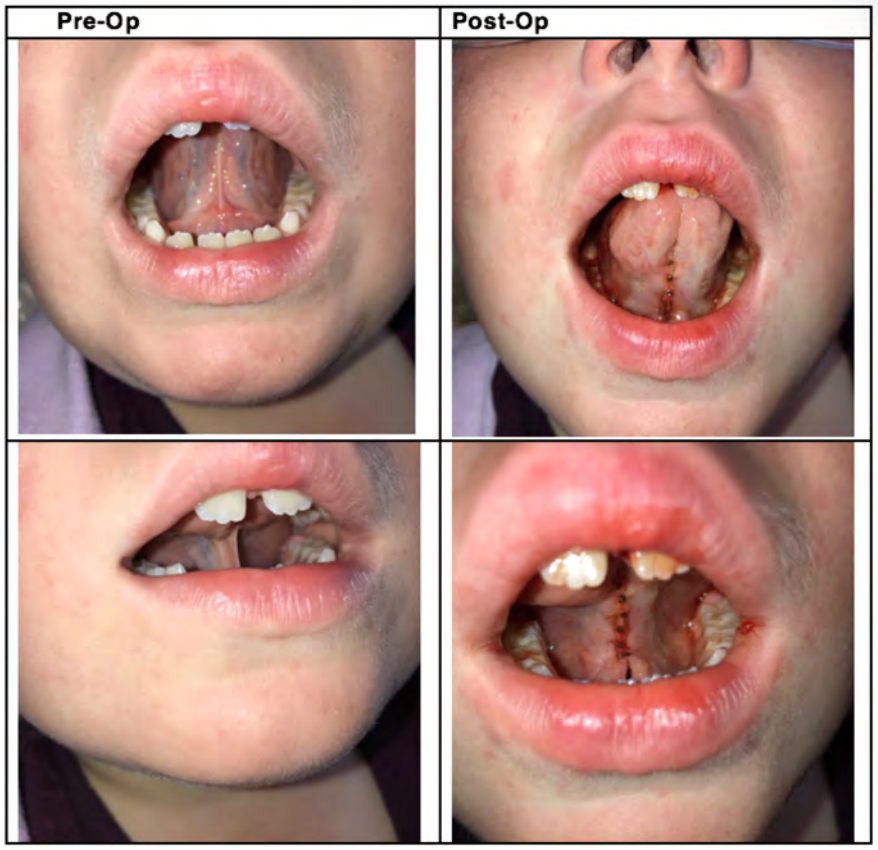


Photo Credit:
Rebecca Thorsen, SLP

December 2017

June 2018

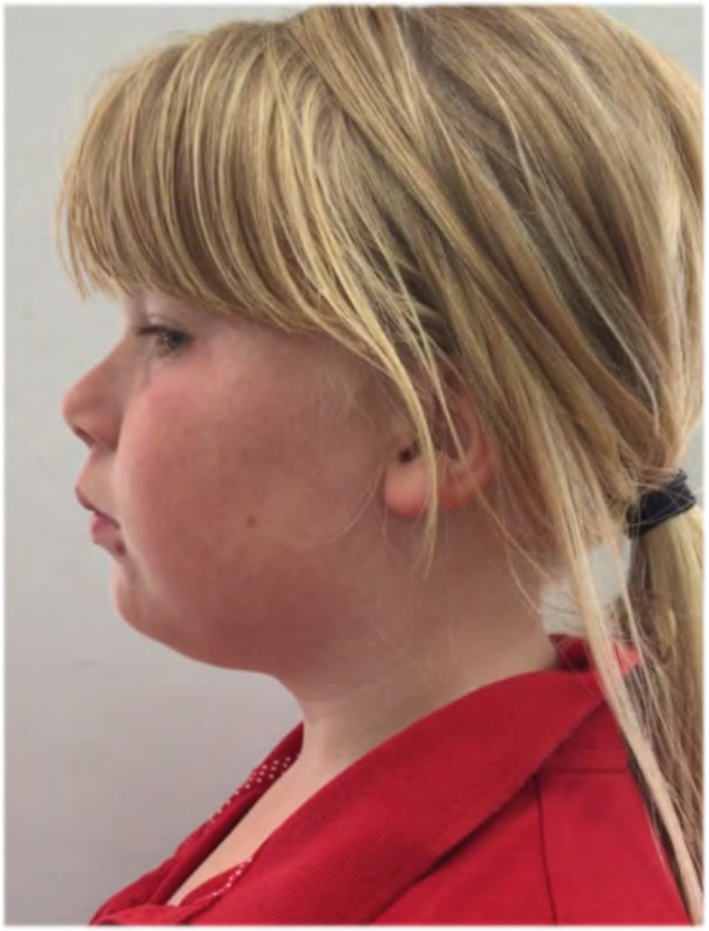


Photo Credit: Rebecca Thorsen, SLP

Level 4 Evidence:
Case series consist of **non-consecutive** collections of descriptive reports among patients who share a common treatment and outcome.

Because the cases are selected to demonstrate a common theme, case series are limited by **selection bias** and have little statistical validity.

The **internal validity** of a case series can be improved by including the results of a large number of **consecutive** cases followed over a defined period of time. A standardized data collection process can help minimize (but not eliminate) the risk of bias.

Strength	Level	Design	Randomization	Control
High	Level 1	Randomized control trial (RCT)	Yes	Yes
		Meta-analysis of RCT with homogeneous results	No	
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	Level 4	Case Series	No	No
	Level 5	Case Report	No	No
		Expert Opinion	No	No
Low		Personal Observation	No	No

2019

Level of Evidence 3
Retrospective Cohort Study

Lingual frenuloplasty with myofunctional therapy: Experience with 348 cases validating an indication of tongue-tie release for mouth breathing, sleep-disordered breathing, and craniofacial pain. [Research Manuscript, Submitted and Pending Review]

Table 3. Benefits attributed to lingual frenuloplasty with myofunctional therapy protocol.

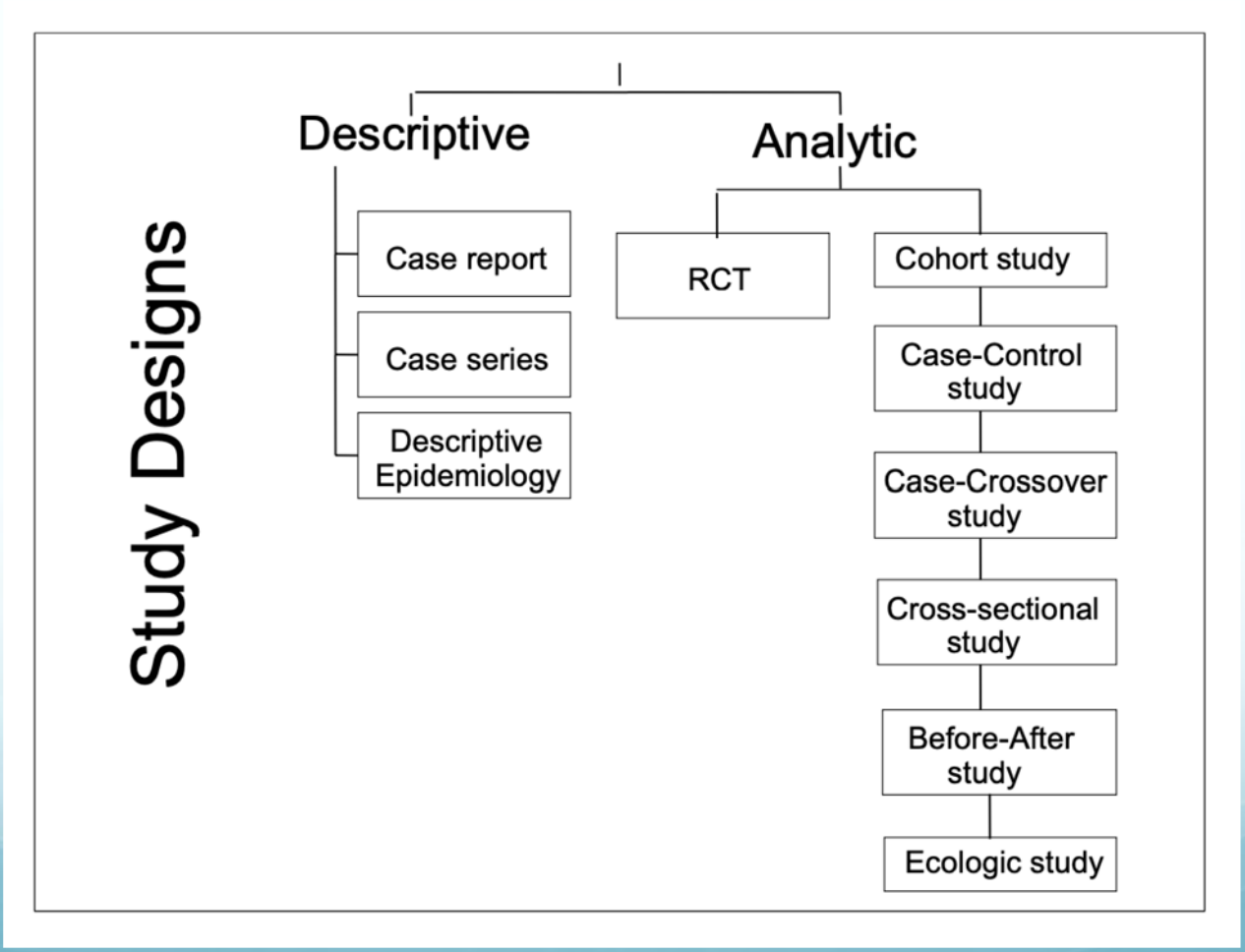
Benefits	Improved	Did Not Improve	Unsure	N/A	**Percent Improved**	**Standard Error**
Overall tongue mobility	326	12	10	-	**96.5%**	1.0%
Clenching or grinding of teeth	40	4	-	304	**91.0%**	4.3%
Ability to perform myofunctional therapy exercises	307	35	6	-	**89.8%**	1.6%
Ease of swallow	102	25	3	218	**80.3%**	3.5%
Sleep quality	195	50	11	92	**79.6%**	2.6%
Nasal breathing	174	48	4	122	**78.4%**	2.8%
Neck, shoulder, facial tension or pain	117	34	-	197	**77.5%**	3.4%
Snoring	102	38	11	197	**72.9%**	3.8%

High treatment success and rates of patient satisfaction with functional frenuloplasty

Level 3 Evidence: Analytical study that consists of **consecutive** collections of **standardized** data among patients who share a common treatment or exposure.

Most commonly, post-treatment results are compared to baseline measures.

"Quasi-experimental": Internal validity limited by the lack of a randomized control group.



Levels of Evidence for Therapeutic Studies*

Level	Type of evidence
1A	Systematic review (with homogeneity) of RCTs
1B	Individual RCT (with narrow confidence intervals)
1C	All or none study
2A	Systematic review (with homogeneity) of cohort studies
2B	Individual Cohort study (including low quality RCT, e.g. <80% follow-up)
2C	"Outcomes" research; Ecological studies
3A	Systematic review (with homogeneity) of case-control studies
3B	Individual Case-control study
4	Case series (and poor quality cohort and case-control study
5	Expert opinion without explicit critical appraisal or based on physiology bench research or "first principles"

*From the Centre for Evidence-Based Medicine, http://www.cebm.net.

Oxford Centre for Evidence-Based Medicine. OCEBM Levels of Evidence Working Group. The Oxford 2011 Levels of Evidence. 2011.**http://www.cebm.net/index.aspx?o=5653**.

ORAL MYOFUNCTIONAL THERAPY AND FRENULOPLASTY ARE NOT PROVEN TREATMENTS FOR OBSTRUCTIVE SLEEP APNEA

Posted March 17, 2018 by Dr. Kezirian & filed under Sleep Apnea.

Obstructive sleep apnea is a potentially-serious medical disorder. Patients with obstructive sleep apnea need treatments that are based on science. I am writing this post because over the last several months, I have seen a disturbing number of patients with obstructive sleep apnea who have tried and failed myofunctional therapy and/or frenuloplasty/frenectomy or who have seen something online about it and are asking for my opinion. Enough is enough. It is time for someone to speak up. **There is no proven benefit to oral myofunctional therapy or frenuloplasty for the treatment of obstructive sleep apnea in adults as it is commonly practiced in the United States. So that you do not think this is just the rant of a surgeon, I will state that I do not know of anyone respected in the sleep apnea scientific community that would disagree with this, other than one person that I will mention below.** If you are an adult and want to use exercises to treat your sleep apnea, go to Brazil for people that are using tested approaches. Do not undergo a frenuloplasty/frenectomy for obstructive sleep apnea. That is really all adults with sleep apnea need to know, but I will explain what I mean in the rest of the post.

The Laryngoscope

Publication Trends and Levels of Evidence in Obstructive Sleep Apnea Literature

Christopher J. Gouveia, MD; Soroush Zaghi, MD; Michael Awad, MD; Macario Camacho, MD; Stanley Y. C. Liu, MD, DDS; Robson Capasso, MD; Robert C. Kern, MD

Objectives/Hypothesis: Examine trends in clinical research and levels of evidence related to obstructive sleep apnea (OSA) in the medical literature. Describe the features and trends of OSA research within otolaryngology journals.

Study Design: Retrospective analysis.

Methods: Review of OSA research articles from 2006, 2011, and 2016 in four leading medical sleep and otolaryngology journals. Level of evidence was graded, and study characteristics were measured.

Results: Seven hundred eight total articles were reviewed. OSA articles significantly increased in both number and proportion of total articles in the medical sleep ($P<.001$) and otolaryngology ($P=.004$) journals. Surgically focused articles did not significantly increase in either literature. There was no significant difference between medical sleep and otolaryngology literature levels of evidence regarding OSA, and no trend toward higher levels of evidence over time. Medical sleep publications had significantly higher proportions of grant-funded ($P<.001$) and National Institutes of Health (NIH)–funded ($P<.001$) publications versus otolaryngology journals. Over time, otolaryngology journals had decreasing numbers of grant-funded and NIH-funded projects.

Conclusions: OSA research is increasingly present in medical sleep and otolaryngology literature. Levels of evidence are modest for the two specialties, and have shown no trend toward increasing over time. Concurrently, otolaryngologists are less likely to be grant funded than their medical colleagues, and sleep surgery has stagnated in the studied journals. This study encourages continued efforts to publish high-quality research on OSA. It may also help guide our specialty when setting priorities regarding research funding and support for sleep surgeons.

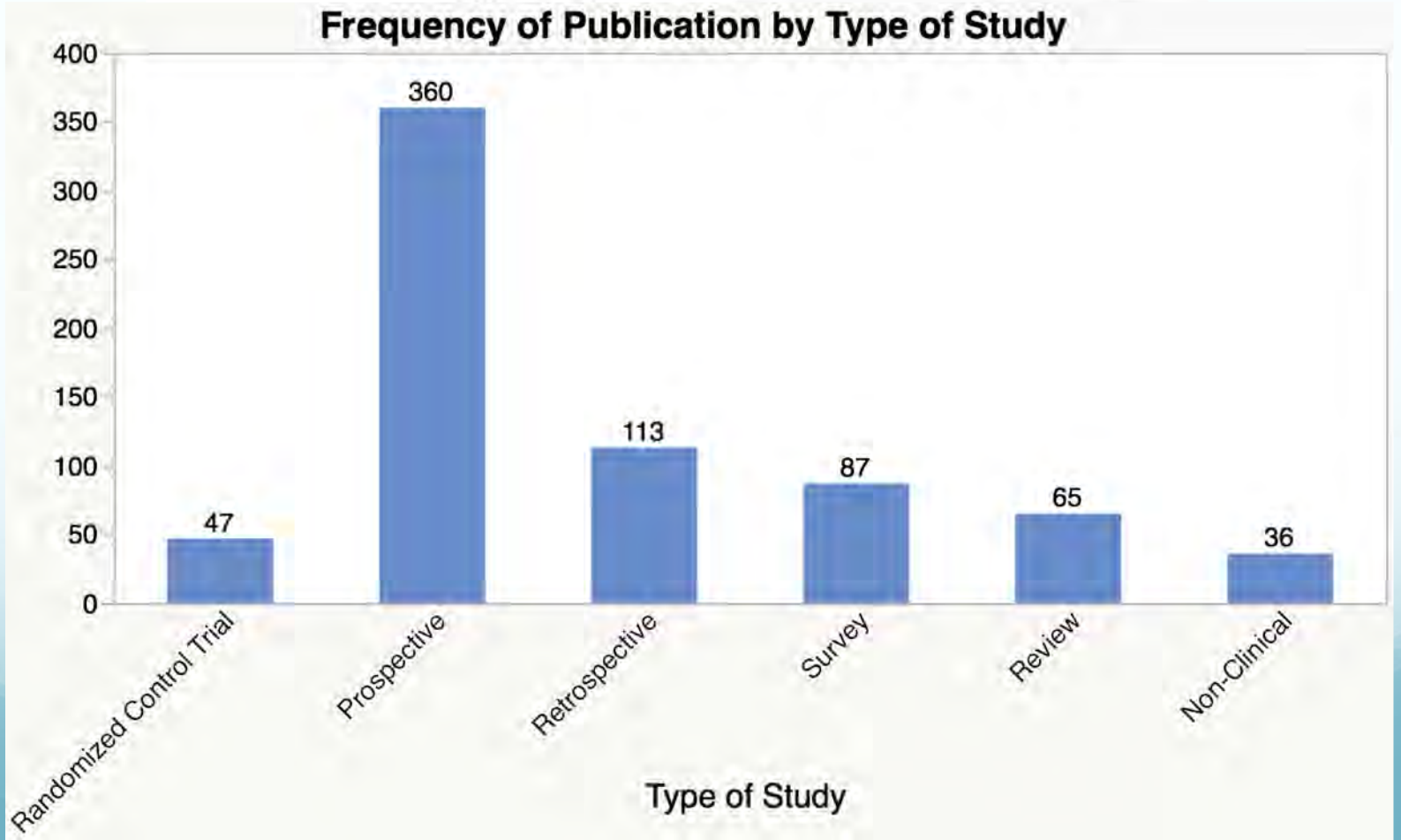
Key Words: Obstructive sleep apnea, evidence-based medicine, sleep medicine.

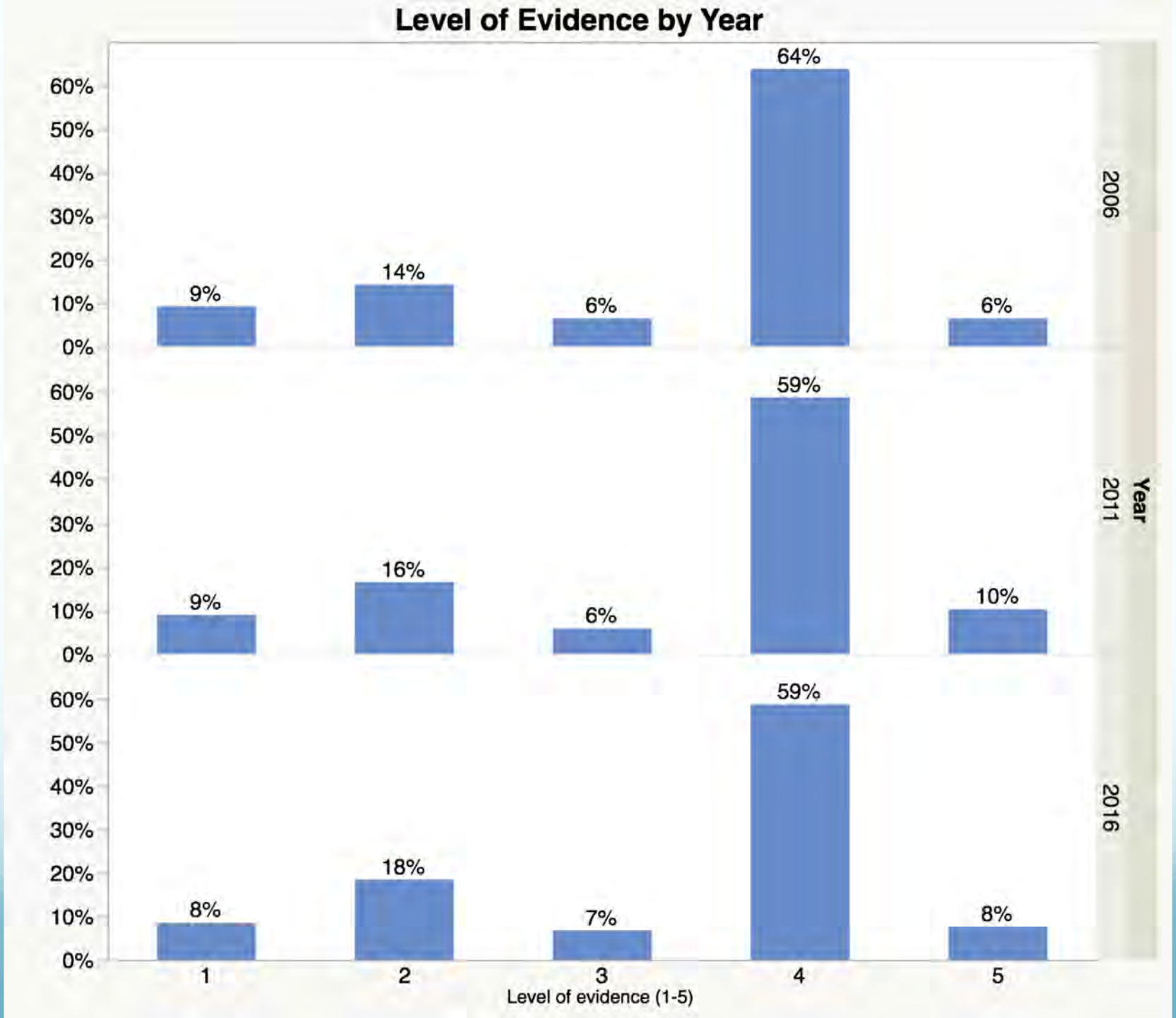
Level of Evidence: NA

***Laryngoscope*, 128:2193–2199, 2018**

Journal	Year	Sleep Apnea Publications
Sleep and Breathing	2006	20
	2011	77
	2016	106
Journal of Clinical Sleep Medicine	2006	12
	2011	26
	2016	63
Sleep Medicine	2006	19
	2011	35
	2016	50
Sleep	2006	30
	2011	32
	2016	47

Journal	Year	Sleep Apnea Publications
Otolaryngology—Head and Neck Surgery	2006	31
	2011	21
	2016	21
Laryngoscope	2006	14
	2011	13
	2016	32
Journal of the American Medical Association Otolaryngology–Head and Neck Surgery	2006	9
	2011	10
	2016	5
European Archives of Otorhinolaryngology	2006	6
	2011	11
	2016	18
All	All	708




Level of Evidence by Year
60%
50%
40%
30%
20%
10%
0%
9%
14%
6%
64%
6%
9%
16%
6%
59%
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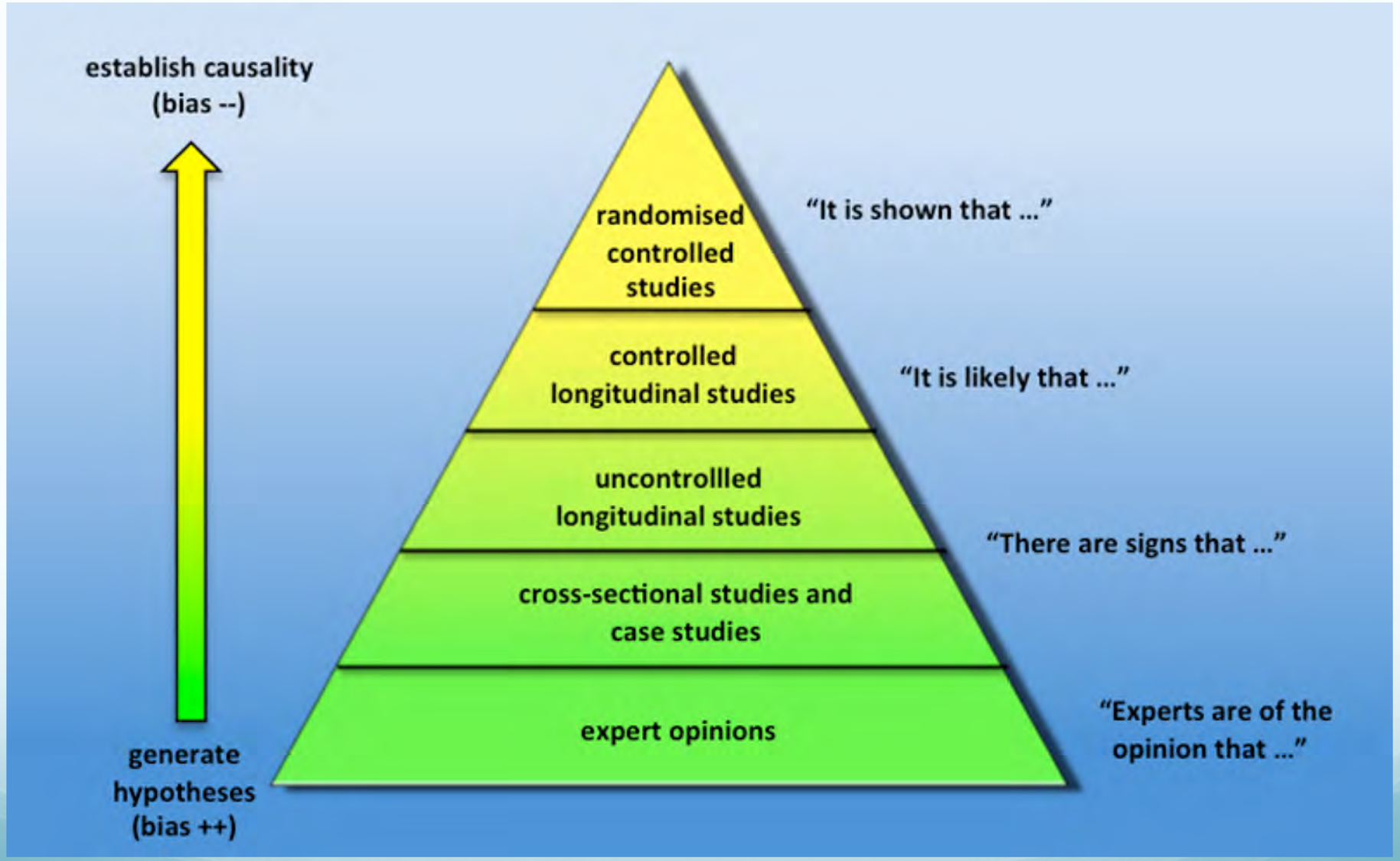
3
4
5
Level of evidence (1-5)


establish causality
(bias --)
randomised controlled studies
"It is shown that ..."
controlled longitudinal studies
"It is likely that ..."
uncontrollled longitudinal studies
"There are signs that ..."
cross-sectional studies and case studies
expert opinions
"Experts are of the opinion that ..."
generate hypotheses
(bias ++)

Literature Review: Tongue-Tie & Sleep Disordered Breathing

Evidence in children and adults

Figure 1: Example of a short lingual frenulum in a prepubertal child with obstructive sleep apnea

Stanford MEDICINE | The Stanford Center for Sleep Sciences and Medicine

Christian Guilleminault DM, MD, DBiol

Professor- Stanford University Sleep Medicine

Physician and researcher who played a central role in the early discovery of obstructive sleep apnea and is now internationally credited with the development of "Sleep Medicine" as a medical field throughout the world.

He was a founding member of the Association of Sleep Disorders Centers in 1975 and was elected as the first editor of the journal *Sleep* in June 1976. He has authored over 600+ articles in peer-reviewed medical journals and has won numerous awards for his seminal contributions.

His research over the last 10 years has been focused on identifying early risk factors that lead to occurrence of sleep-disordered-breathing and obstructive sleep apnea with the goal of prevention through early intervention.


SLEEP
SLEEP
SLEEP
SLEEP
SLEEP
SLEEP
SLEEP
SLEEP
Pierre Roble
Award
Christian Guilleminault MD
Clinical Neurophysiology of
Sleep and its Disorders

C.G. is the first to coin the terms:

"Obstructive Sleep Apnea Syndrome" (OSA)
"Upper Airway Resistance Syndrome" (UARS)

Christian Guilleminault, MD

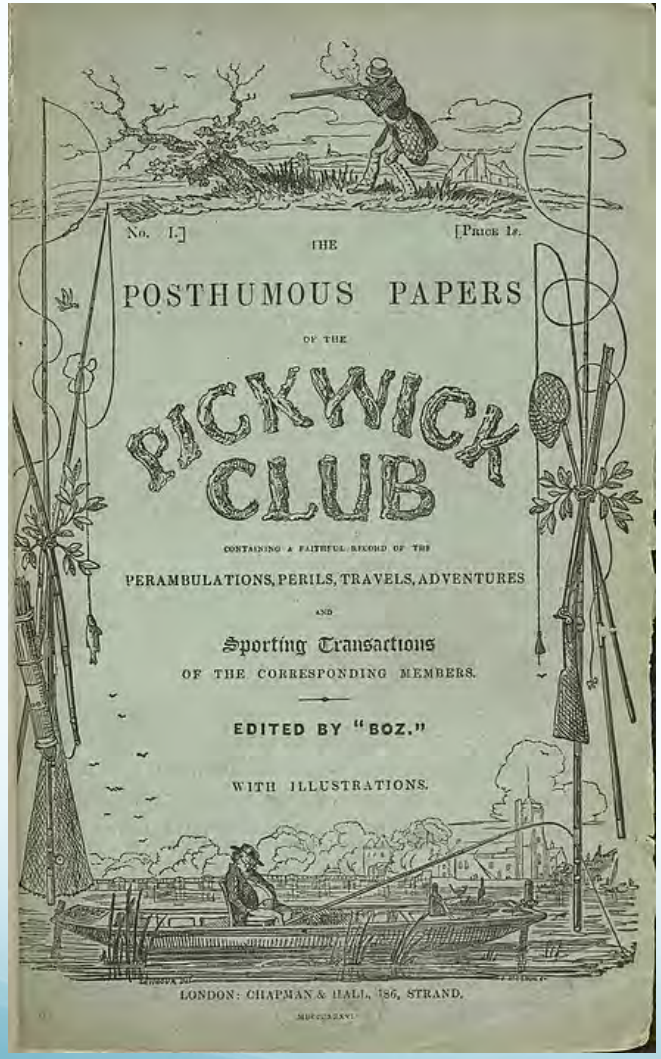
ABOUT DR. GUILLEMINAULT:
Dr. Christian Guilleminault is a physician and researcher in the field of sleep medicine who played a central role in the early discovery of obstructive sleep apnea and has made seminal discoveries in many other areas of sleep medicine. While working at the Stanford University Sleep Disorders Clinic in 1972, Guilleminault became keenly interested in reports published by Italian sleep researcher Elio Lugaresi who had reported that nocturnal hypertension was present in patients who snored. Guilleminault persuaded cardiologists John Shroeder and Ara Tilkian to spend nights in the hospital's clinical research center monitoring the systemic and pulmonary arterial blood pressure in sleeping patients. The team observed that when patients fell asleep and began snoring, prolonged pauses in their breathing (apneas) were noted that corresponded with dramatic elevations in their resting blood pressure, simulating strenuous exercise as if the patient were lifting weights.Guilleminault then went on to publish several articles illustrating dramatic improvements and reversal of sleep apnea following tracheostomies.Tracheostomy proved curative in these patients, and demonstrated reversal of cardiac arrhythmias and blood pressure abnormalities during sleep; temporarily capping these artificial airways would re-capitulate the changes of sleep apnea, further establishing the causative relationship between sleep apnea and cardiovascular abnormalities.

Guilleminault then went on to describe obstructive sleep apnea in non-obese patients, being the first to coin the term "obstructive sleep apnea syndrome" (OSAS), a term commonly used nowadays. In addition, he described the presence of OSAS in children, demonstrating its association with learning and attention problems along with cardiovascular derangements. Following this work, he went on to describe the presence of elevated upper airway resistance in children in 1982, emphasizing the symptoms of attention deficit, hyperactivity, and abnormal behavior during wakefulness and sleep, learning disabilities and sleepwalking, sleep terrors and enuresis that accompanied this form of sleep-related breathing disorder; he described the same syndrome in adults and penned the term "upper airway resistance syndrome" (UARS) in adults. Finally, working in collaboration with Dr. William C. Dement, Guilleminault established the Apnea–hypopnea index (AHI), which is still in use today to characterize the presence and severity of sleep apnea.

Guilleminault continues to be a prolific researcher in the field of sleep medicine and has authored over six hundred articles in peer-reviewed medical journals to date and has won several awards for his research in the field of sleep medicine. He was a founding member of the Association of Sleep Disorders Centers in 1975 and was elected to be the first editor of the journal Sleep in June 1976, a role in which he continued to serve until 1997. He continues to practice clinical medicine and contribute to research endeavors at the Stanford Center for Sleep Sciences and Medicine.

Charles Dickens is the first to *describe*:

sleep-disordered breathing (obesity hypoventilation syndrome)



March 30, 1836

The Posthumous Papers of the Pickwick Club (also known as ***The Pickwick Papers***) was Charles Dickens's first novel.

Chapters were issued in monthly installments following the "perambulations, perils, travels, and adventures" of various central and supporting characters.

- Joe — the "fat boy" who consumes great quantities of food and constantly falls asleep in any situation at any time of day.

- Joe's sleep problem is the origin of the medical term Pickwickian syndrome, which ultimately led to the subsequent description of obesity hypoventilation syndrome.

The Pickwick Papers and Sleep Apnea

Joe the "fat boy" from the Pickwick Papers, Illustration by Kyd (Joseph Clayton Clarke)

"The object that presented itself to the eyes of the astonished clerk, was a boy–a wonderfully fat boy–habited as a serving lad, standing upright on the mat, with his eyes closed as if in sleep."

"Sleep!" said the old gentleman, 'he's always asleep. Goes on errands fast asleep, and snores as he waits at table."

"How very odd!" said Mr. Pickwick.

"Ah! odd indeed," returned the old gentleman.

1836

Level of Evidence 5

Expert Opinion, Personal Observation, Case Report

"Finally an experience which indicated the severity of his disability led him to seek hospital care.

The patient was accustomed to playing poker once a week and on this crucial occasion he was dealt a hand of three aces and two kings. According to Hoyle this hand is called a "full house." Because he had dropped off to sleep he failed to take advantage of this opportunity. [Italics original]. A few days later he entered…hospital."

First case report of sleep-disordered breathing

<u>Level 5 Evidence:</u> Report of one individual case.

1972

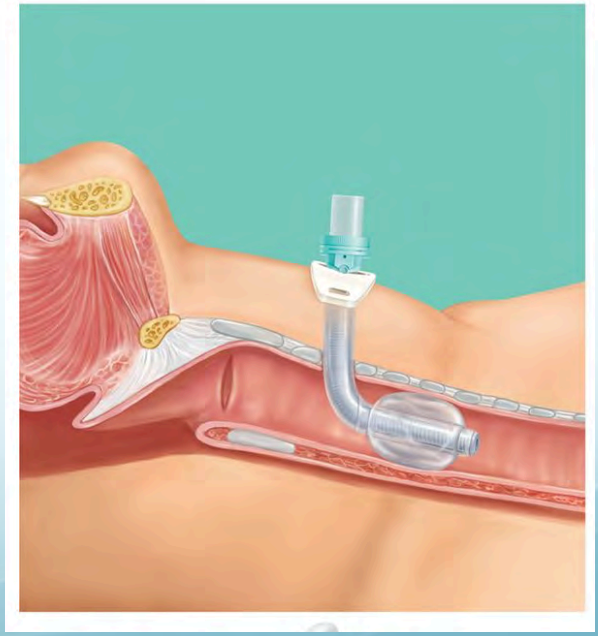
Obstructive Sleep Apnea

Level of Evidence 5
Expert Opinion, Personal Observation, Case Report

Career [edit]

While working at the Stanford University Sleep Disorders Clinic in 1972, Guilleminault became keenly interested in reports published by Italian sleep researcher Elio Lugaresi who had reported that nocturnal hypertension was present in patients who snored. Guilleminault persuaded cardiologists John Shroeder and Ara Tilkian to spend nights in the hospital's clinical research center monitoring the systemic and pulmonary arterial blood pressure in sleeping patients. The team observed that when patients fell asleep and began snoring, prolonged pauses in their breathing (apneas) were noted that corresponded with dramatic elevations in their resting blood pressure, simulating strenuous exercise as if the patient were lifting weights.[1] Guilleminault then went on to publish several articles illustrating dramatic improvements and reversal of sleep apnea following tracheostomies.[2] Tracheostomy proved curative in these patients, and demonstrated reversal of cardiac arrhythmias and blood pressure abnormalities during sleep; temporarily capping these artificial airways would re-capitulate the changes of sleep apnea, further establishing the causative relationship between sleep apnea and cardiovascular abnormalities.[3]

Pickwickian Syndrome: obesity, daytime sleepiness, loud snoring (Charles Dickens, Fat Boy in *Pickwick Papers*, 1836)



1976

"The sleep apnea syndromes."
Guilleminault, Christian, Ara Tilkian, and William C. Dement.
Annual review of medicine 27.1 (1976): 465-484.

Level of Evidence 4
Case Series

At the suggestion of a respiratory specialist (P. Sadoul) and a neurologist-sleep researcher (E. Lugaresi), the first symposium on sleep-related respiratory problems that included specialists from both areas was held in Italy in 1972 (19). As a result of this intellectual confrontation, a new concept of "sleep-induced apnea syndromes" with secondary cardiovascular consequences has rapidly evolved. In spite of crippling hypersomnolence or hemodynamic problems, the primary respiratory abnormality is typically completely undetectable when patients are fully alert, and obesity is not at all a necessary feature. This group of closely related syndromes has implications for neurologists, cardiologists, otolaryngologists, pneumologists, pediatricians, and, particularly, physicians with a special interest in sleep disorders. Because many reports involving these syndromes have been published in the European and Japanese literature, the primary goal of this review is to facilitate a greater familiarity with sleep apnea syndromes among American physicians.

It is obvious that medical centers lacking a mechanism to ensure adequate laboratory evaluation of the sleep patient who presents a serious sleep disorder will not identify or diagnose a sleep apnea syndrome, since a reliable diagnosis is heavily dependent upon standard sleep recordings (20) combined with respiratory measurements. Such a mechanism has been available at Stanford University for more than five years in the form of a sleep disorders clinic to which such patients can be referred for specialty evaluation. As a result, a uniquely large case series of sleep apnea patients has been evaluated, and a large reservoir of clinical experience with such patients has been accumulated. Because of this, and because most published reports have focused on limited areas of the problem, much of this review is based upon the Stanford University Sleep Disorders Clinic case series, particularly the clinical symptomatology, and unless otherwise specified, the reader may assume that this series is the source of specific descriptive material. Various subsamples of this case series have been described elsewhere (21–24).

At the Stanford University Sleep Disorders Clinic, between June 1972 and June 1975, we identified sleep apnea syndromes in sixty-two patients among 350 patients referred for evaluation of serious sleep problems.

1977

Level of Evidence 3

Cohort Study

Sleep-Induced Apnea Syndrome

Prevalence of Cardiac Arrhythmias and Their Reversal After Tracheostomy

ARA G. TILKIAN, M.D.*
CHRISTIAN GUILLEMINAULT, M.D.
JOHN S. SCHROEDER, M.D.
KENNETH L. LEHRMAN, M.D.
F. BLAIR SIMMONS, M.D.
WILLIAM C. DEMENT, M.D., Ph.D.
Stanford, California

Cardiac arrhythmias during wakefulness and sleep in 15 patients with sleep-induced obstructive apnea, and the effect of atropine and tracheostomy on these arrhythmias were studied by continuous overnight Holter electrocardiographic, respiratory and electroencephalographic recordings. Sleep was characterized by marked sinus arrhythmia in 14, extreme sinus bradycardia (<30 beats/minute) in six, asystole of 2.5 to 6.3 seconds in five, second degree atrioventricular (A-V) block in two, and ventricular arrhythmias—complex premature ventricular beats in 10 and ventricular tachycardia in two. Arrhythmias during wakefulness were limited to premature ventricular beats in six. Atropine administration was partially and tracheostomy highly effective in preventing the majority of these arrhythmias during sleep.

Marked sinus arrhythmia during sleep is characteristic of the syndrome of obstructive sleep apnea and is frequently accompanied by potentially life-threatening tachy- and bradyarrhythmias. Possible mechanism of production of these arrhythmias, the mode of action of tracheostomy and atropine, and the probable role of similar arrhythmias in the sudden infant death syndrome are discussed.

Cohort study of 15 patients with ”sleep-induced apnea” who would develop cardiac arrhythmias during sleep. Partial improvement with atropine; significant improvement (“near reversal”) after tracheostomy.

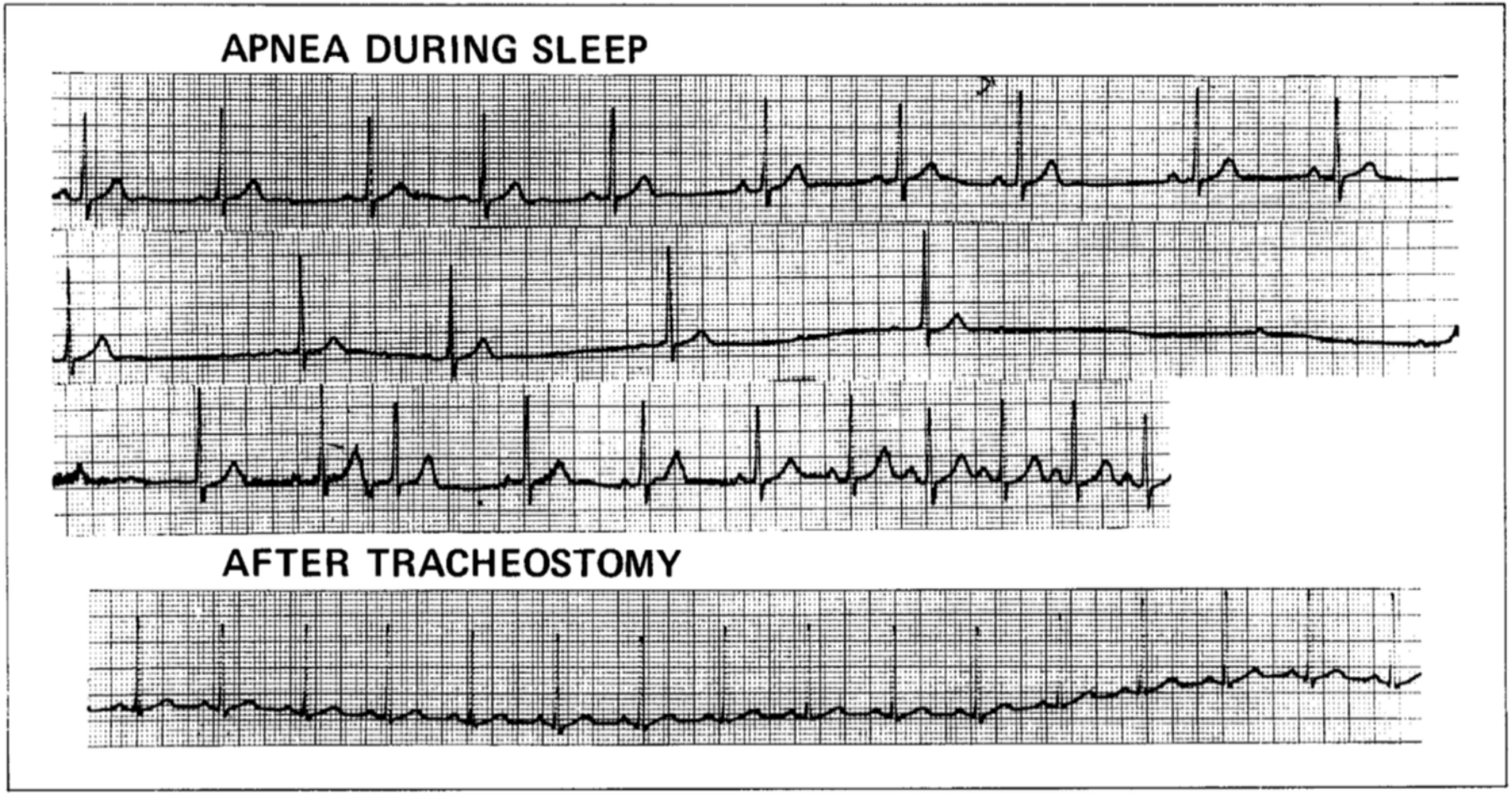


Figure 7. *Electrocardiographic recording during sleep before and after tracheostomy. Upper rhythm strip is a continuous 30 second recording and shows progressive sinus bradycardia (during apnea) and a 5.5 second sinus pause with a rapid reversal and resumption of sinus tachycardia (105 to 110/min) at onset of ventilation. Following tracheostomy, normal sinus rhythm was present during the entire recorded sleep.*

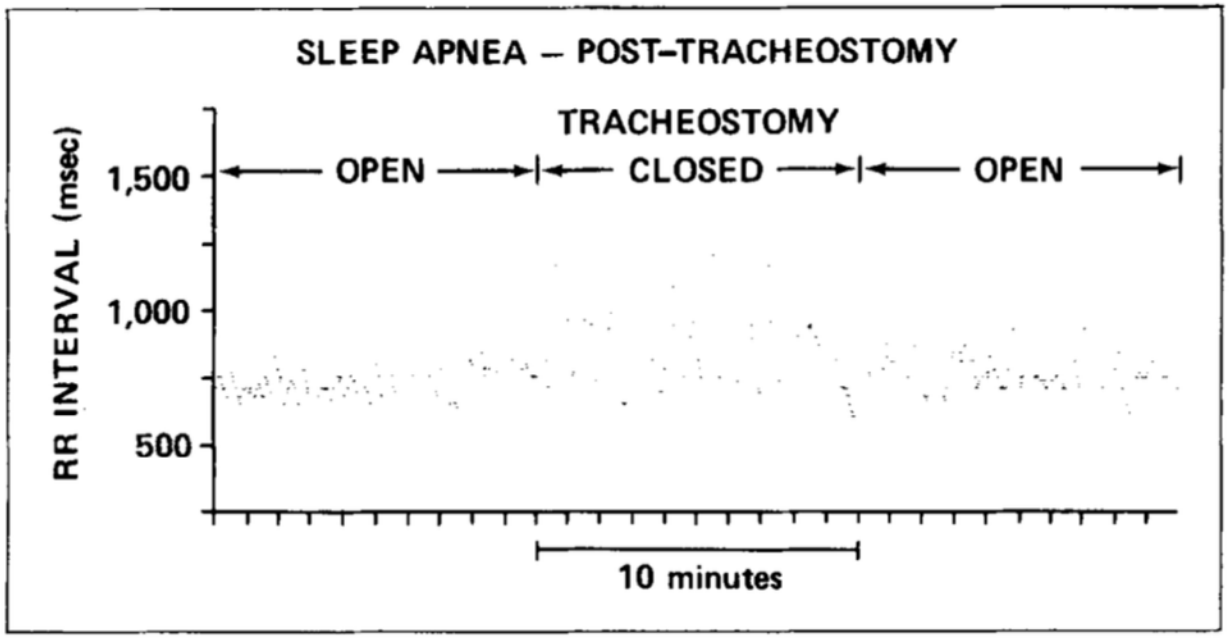


Figure 8. *Computer printout of 30 minutes of electrocardiographic recording during sleep. QRS complexes are characterized by dots indicating the RR interval. First 10 minutes are during sleep with tracheostomy open. While the recording continues, the tracheostomy is temporarily occluded for 10 minutes. The balloon cuff is deflated through the entire sleep. Marked sinus arrhythmia with sinus bradycardia immediately recurs during tracheostomy occlusion. Intermittent upper airway obstruction similar to the pretracheostomy pattern was documented during this period.*

1980

Level of Evidence 2

Controlled longitudinal studies.
No blinded randomized control studies.

Tracheostomy was the primary surgical modality utilized to treat OSA patients until the early 1980s. Definitive treatment successful in 100% of patients.

TABLE II.
Obstructive Apnea Indices Pre- and Posttracheostomy.

Study year	Study author	N	Mean Age	Mean BMI[a]	Follow-up in months	Pre-Trach Total Apnea Index	Post-Trach Obstructive Apnea Index
2013	Kumar et al[17]	1	27	40	1.0	0.0	0.0
1990	Partinen et al[12,35]	71	48.8 ± 11	34±7.7	≤1.0	69 ± 123	0.0
1989	Fletcher[9]	1	–	–	51.0	114.0	0.0
1989	Hastie[18]	1	32	46.7	6.3	25.3	1.1
1987	Fletcher[10]	8	55.4 ± 6.8	–	9.0 ± 3.2	84.6 ± 38.7	0.0
1985	Fletcher[34]	11	56.5 ± 7.4	–	(2–26)	69.6 ± 36.1	0.0
1982	Guilleminault[5]	5	45.8	27.7	3.0+	65 ± 14	2.0 ± 2.0
1981	Guilleminault[6]	4	–	–	30.0 ± 6.9	94.5 ± 19.8	0.4 ± 0.5
1980	Sugita[14]	1	40	20.9	3.0	77.0	0.0
1980	Weitzman[16]	10	47.5 ± 2.4	–	0.3 ± 0.4	96.1 ± 21.9	1.1 ± 3.3
1978	Mottai[11]	6	47.0 ± 4.0	–	7.5 ± 6.3	73.0 ± 12.2	0.0
1978	Weitzman[19]	1	67	–	0.5	96.7	4.1
Total		120	49.4 ± 10.1	34.0 ± 7.8	9.1 ± 12.4	73.0 ± 27.1	0.2 ± 1.2

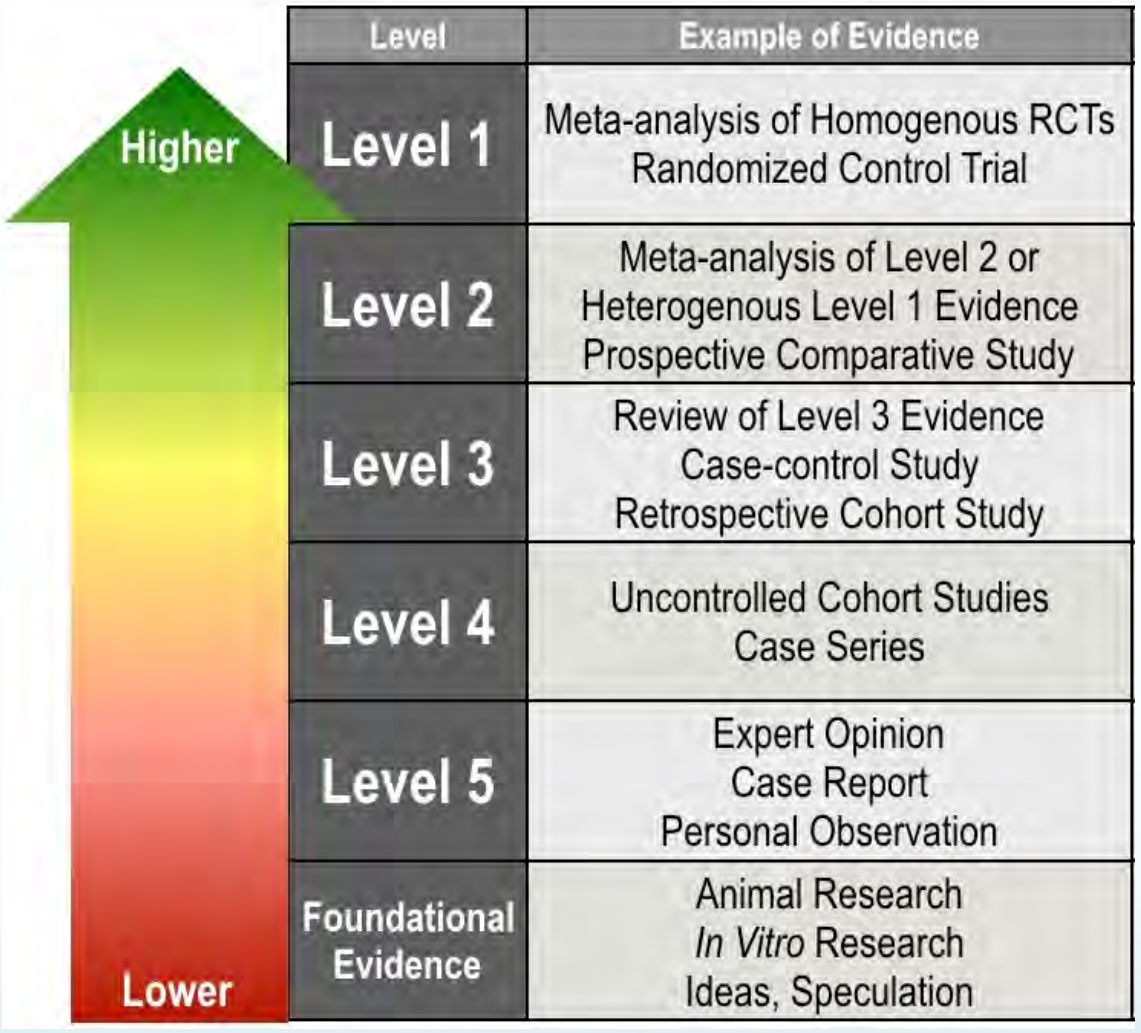
*Note the significant decrease in obstructive apnea index, $P < 0.0001$. – Denotes that the information was not available from the study.
[a]BMI = body mass index (kg/m^2); N = number of patients.

In 1980, a construction worker with a particularly severe case of obstructive sleep apnea walked into Dr. Collins Sullivan's hospital seeking help after refusing a tracheotomy procedure.

Dr. Collin had surmised that perhaps a solution to improve breathing during sleep was to use a device that could pump air from the surrounding environment and into the respiratory tract. So, Dr. Sullivan devised a breathing mask and connected it via a number of different hoses to the engine of a vacuum cleaner and was performing testing on English bulldogs, pugs and other breeds of dogs with a similarly compacted breathing mechanism.

Reference: https://blog.easybreathe.com/cpap-history-aussie-doctor-vacuum-cleaner/

The results from animal study were extremely promising – rumor has it that the dog wagged his tail, leaped outside, unearthed every bone he ever buried and, most shockingly, a nurse tech swore she saw the pooch give a double thumbs up – so Dr. Sullivan moved on to phase two: determining if this contraption would work on humans.



Level	Example of Evidence
Level 1	Meta-analysis of Homogenous RCTs Randomized Control Trial
Level 2	Meta-analysis of Level 2 or Heterogenous Level 1 Evidence Prospective Comparative Study
Level 3	Review of Level 3 Evidence Case-control Study Retrospective Cohort Study
Level 4	Uncontrolled Cohort Studies Case Series
Level 5	Expert Opinion Case Report Personal Observation
Foundational Evidence	Animal Research *In Vitro* Research Ideas, Speculation

Information begins at the bottom of the pyramid: this is where ideas and laboratory research takes place. Ideas turn into therapies and diagnostic tools, which then are tested with lab models and animals.

1981

Level of Evidence 4

Case Series

CPAP proposed as an alternative to tracheostomy based on a series of n=5 patients.

Lancet. 1981 Apr 18;1(8225):862-5.

Reversal of obstructive sleep apnoea by continuous positive airway pressure applied through the nares.

Sullivan CE, Issa FG, Berthon-Jones M, Eves L.

Abstract

Five patients with severe obstructive sleep apnoea were treated with continuous positive airway pressure (CPAP) applied via a comfortable nose mask through the nares. Low levels of pressure (range 4.5-10 cm H2O) completely prevented upper airway occlusion during sleep in each patient and allowed an entire night of uninterrupted sleep. Continuous positive airway pressure applied in this manner provides a pneumatic splint for the nasopharyngeal airway and is a safe, simple treatment for the obstructive sleep apnoea syndrome.

PMID: 6112294

Oxford Centre for Evidence-based Medicine – Levels of Evidence

1980s

"What are we to do when the irresistible force of the need to offer clinical advice meets with the immovable object of flawed evidence?

All we can do is our best: give the advice, but alert the advisees to the flaws in the evidence on which it is based."

Levels of Evidence for Therapeutic Studies*

Level	Type of evidence
1A	Systematic review (with homogeneity) of RCTs
1B	Individual RCT (with narrow confidence intervals)
1C	All or none study
2A	Systematic review (with homogeneity) of cohort studies
2B	Individual Cohort study (including low quality RCT, e.g. <80% follow-up)
2C	"Outcomes" research; Ecological studies
3A	Systematic review (with homogeneity) of case-control studies
3B	Individual Case-control study
4	Case series (and poor quality cohort and case-control study
5	Expert opinion without explicit critical appraisal or based on physiology bench research or "first principles"

Tracheostomy: Level 2 Evidence

CPAP: Level 4 Evidence

*From the Centre for Evidence-Based Medicine, http://www.cebm.net.

Oxford Centre for Evidence-Based Medicine. OCEBM Levels of Evidence Working Group. The Oxford 2011 Levels of Evidence. 2011. **http://www.cebm.net/index.aspx?o=5653**.

Oxford Centre for Evidence-based Medicine – Levels of Evidence

2018

"What are we to do when the irresistible force of the need to offer clinical advice meets with the immovable object of flawed evidence?

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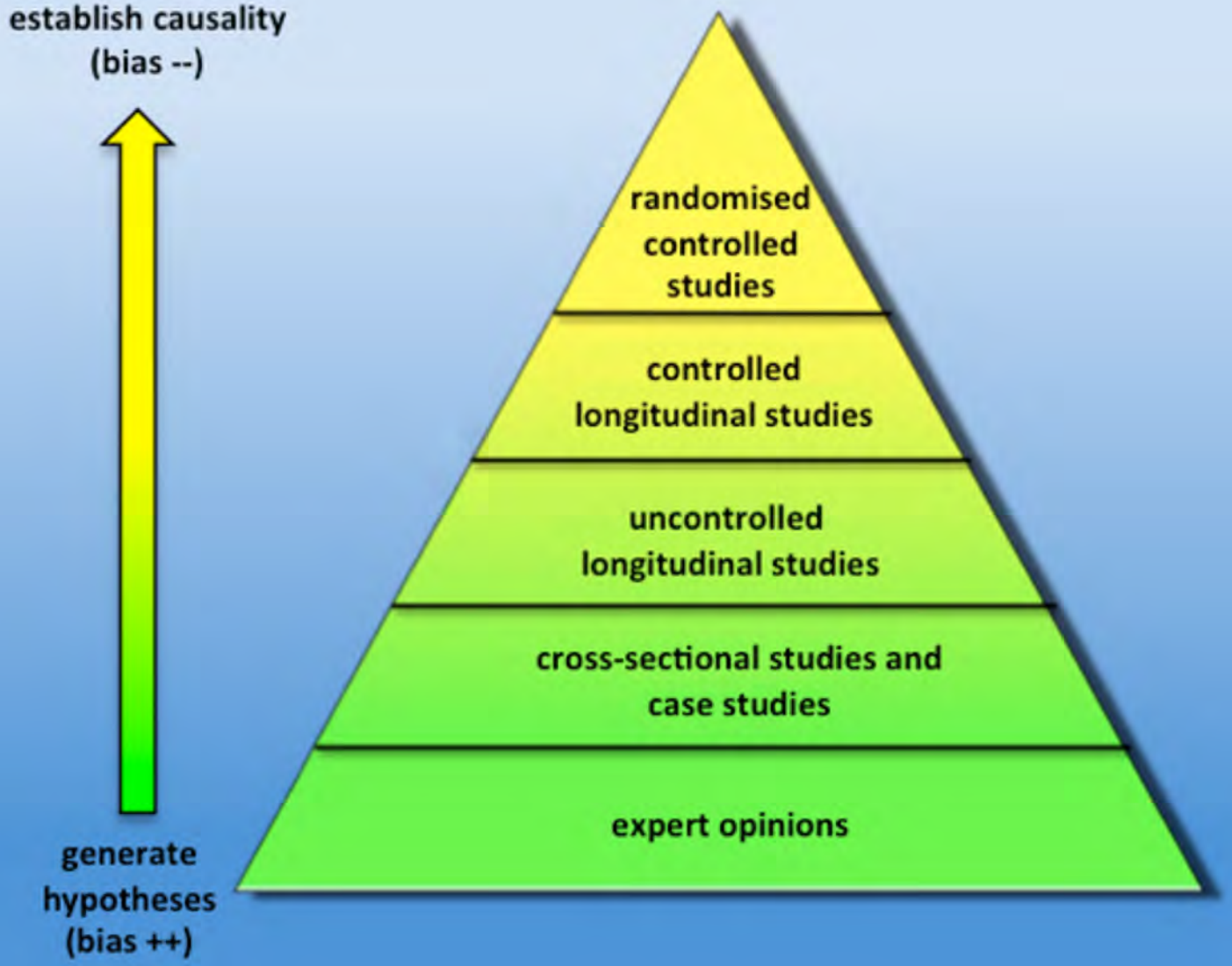
Standard of Care: Level 1/2 Evidence

Tongue-tie: Level 3-5 Evidence

*From the Centre for Evidence-Based Medicine, http://www.cebm.net.

Oxford Centre for Evidence-Based Medicine. OCEBM Levels of Evidence Working Group. The Oxford 2011 Levels of Evidence. 2011. **http://www.cebm.net/index.aspx?o=5653**.

Opinions, case reports, and case series are an opportunity for new scientific inquiry.



1976

Level of Evidence 4

Case Series

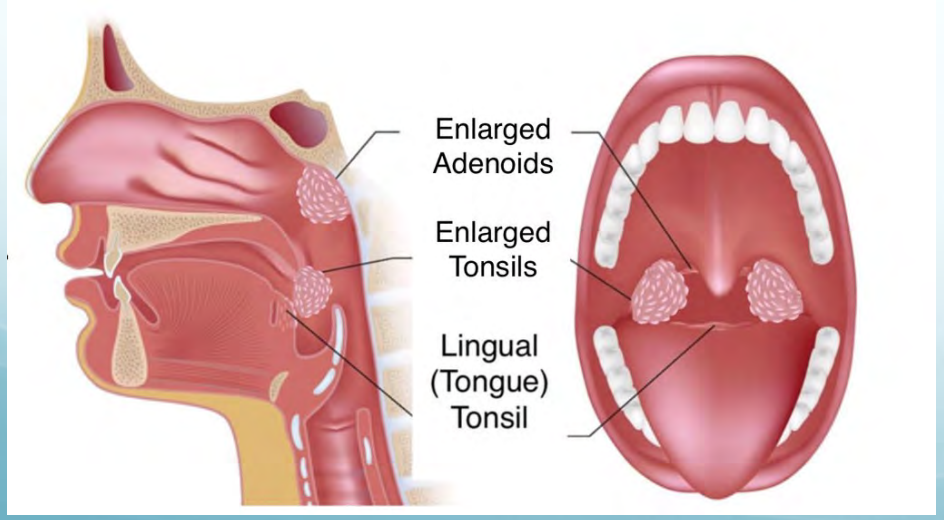
Pediatrics. 1976 Jul;58(1):23-30.

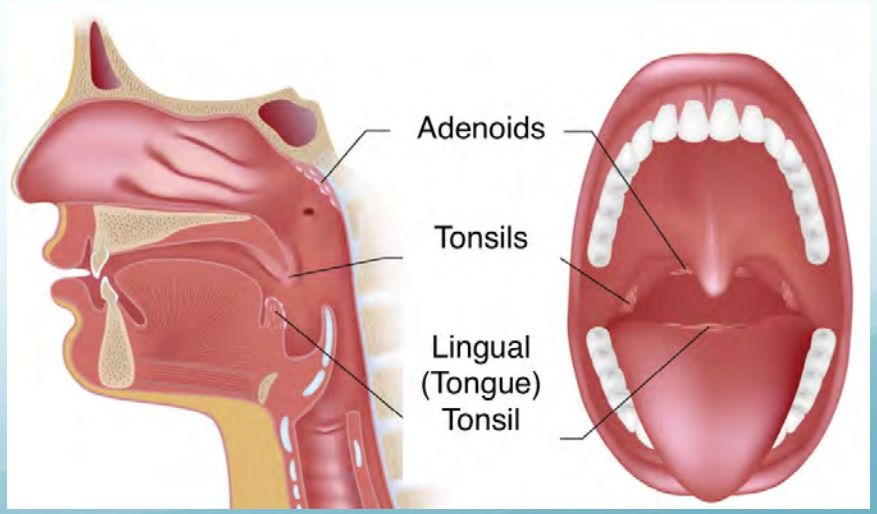
Sleep apnea in eight children.

Guilleminault C, Eldridge FL, Simmons FB, Dement WC.

Abstract

Eight children, 5 to 14 years of age, were diagnosed by means of nocturnal polygraphic monitoring with a sleep apnea syndrome similar to that seen in adults. Excessive daytime sleepiness, decrease in school performance, abnormal daytime behavior, recent enuresis, morning headache, abnormal weight, and progressive development of hypertension should suggest the possibility of a sleep apnea syndrome when any of these symptoms is associated with loud snoring interrupted by pauses during sleep. Surgery may eliminate the clinical symptomatology.





2004

Level of Evidence 3

Retrospective Study

Laryngoscope. 2004 Jan;114(1):132-7.

Sleep disordered breathing: surgical outcomes in prepubertal children.

Guilleminault C[1], Li KK, Khramtsov A, Pelayo R, Martinez S.

Author information

Abstract

OBJECTIVE: To evaluate the treatment outcomes of sleep disordered breathing (SDB) in prepubertal children 3 months following surgical intervention.

STUDY DESIGN: Retrospective investigation of 400 consecutively seen children with SDB who were referred to otolaryngologists for treatment.

METHOD: After masking the identities and conditions of the children, the following were tabulated: clinical symptoms, results of clinical evaluation and polysomnography at entry, the treatment chosen by the otolaryngologists, and clinical and polysomnographic results 3 months after surgery.

RESULTS: Treatment ranged from nasal steroids to various surgical procedures. Adenotonsillectomy was performed in only 251 of 400 cases (68%). Four cases included adenotonsillectomy in conjunction with pharyngoplasty (closure of the tonsillar wound by suturing the anterior and posterior pillar to tighten the airway). Persistent SDB was seen in 58 of 400 children (14.5%), and an additional 8 had persistent snoring. Best results were with adenotonsillectomy.

CONCLUSION: SDB involves obstruction of the upper airway, which may be partially due to craniofacial structure involvement. The goal of surgical treatment should be aimed at enlarging the airway, and not be solely focused on treating inflammation or infection of the lymphoid tissues. This goal may not be met in some patients, thus potentially contributing to residual problems seen after surgery. The possibility of further treatment, including collaboration with orthodontists to improve the craniofacial risk factors, should be considered in children with residual problems.

PMID: 14710009 DOI: 10.1097/00005537-200401000-00024

2011

Sleep Breath (2011) 15:185–193
DOI 10.1007/s11325-011-0504-2

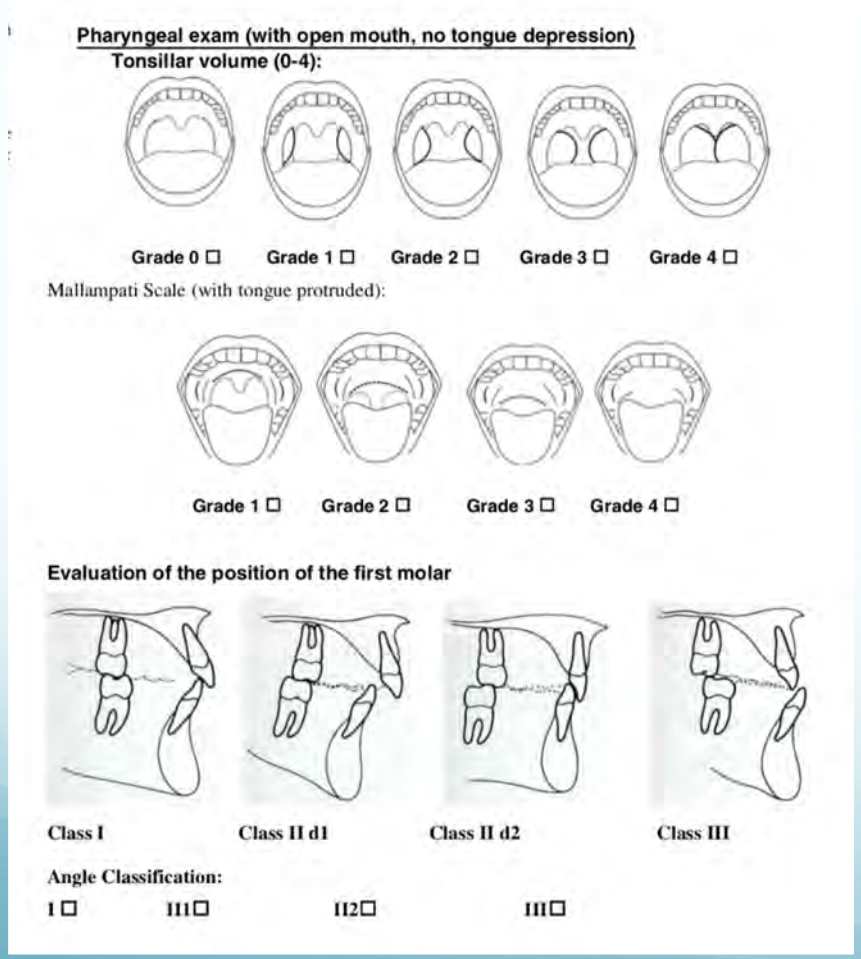
DENTAL SOCIETY ISSUE

The nasomaxillary complex, the mandible, and sleep-disordered breathing

Jee Hyun Kim · Christian Guilleminault

Level of Evidence 3

Retrospective Study



Retrospective review of 400 children (age 2-17) with sleep-disordered breathing.

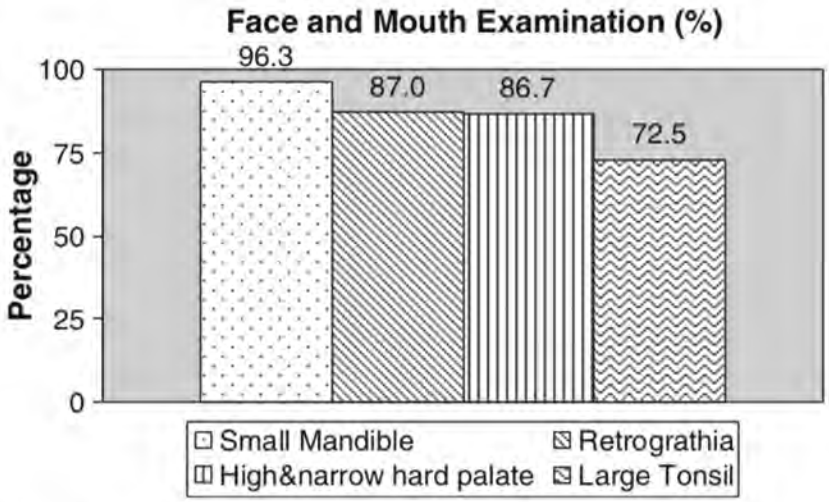


Fig. 3 Anatomical findings at clinical evaluation in total sample

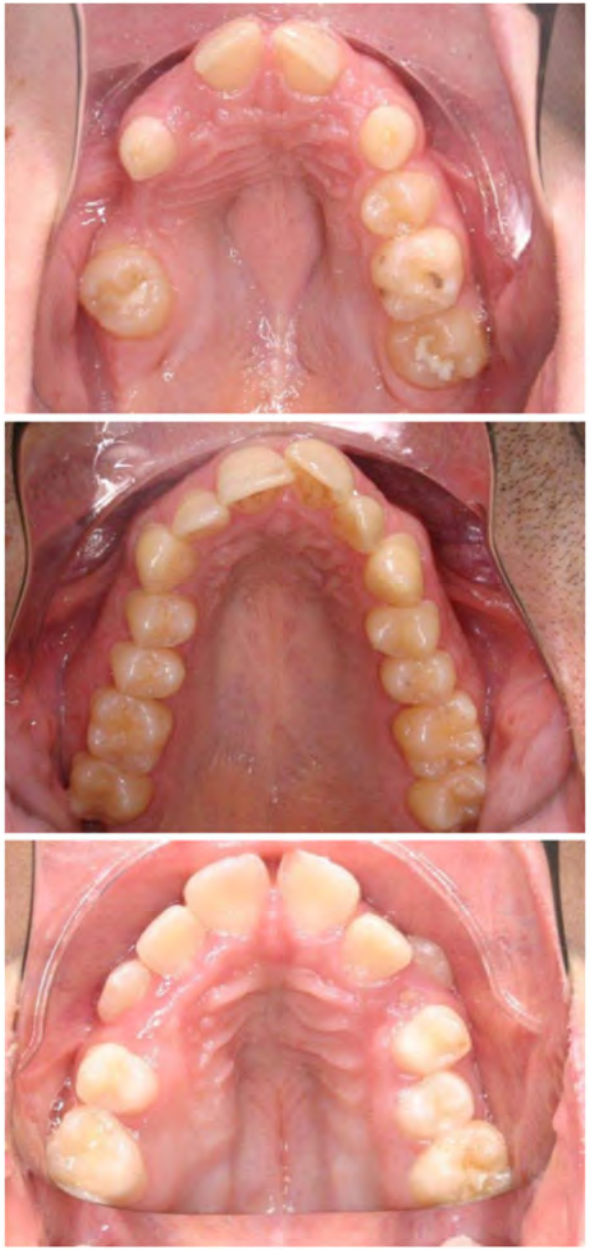


Fig. 4 Examples of high and narrow hard palate. These photos were placed in the examining room as examples of narrow and high hard palate

Fig. 5 Very narrow hard palate in a teenager who was a premature infant born at 35 weeks

2011

Sleep Breath (2011) 15:173–177
DOI 10.1007/s11325-010-0419-3

ORIGINAL ARTICLE

Adeno-tonsillectomy and rapid maxillary distraction in pre-pubertal children, a pilot study

Christian Guilleminault · Pierre-Jean Monteyrol · Nelly T. Huynh · Paola Pirelli · Stacey Quo · Kasey Li

Received: 22 March 2010 / Revised: 12 August 2010 / Accepted: 7 September 2010 / Published online: 17 September 2010

Abstract
Introduction When both narrow maxilla and moderately enlarged tonsils are present in children with obstructive sleep apnea, the decision of which treatment to do first is unclear. A preliminary randomized study was done to perform a power analysis and determine the number of subjects necessary to have an appropriate response. Thirty-one children, 14 boys, diagnosed with OSA based on clinical symptoms and polysomnography (PSG) findings had presence of both narrow maxillary complex and enlarged tonsils. They were scheduled to have both adeno-tonsillectomy and RME for which the order of treatment was randomized: group 1 received surgery followed by orthodontics, while group 2 received orthodontics followed by surgery. Each child was seen by an ENT, an orthodontist, and a sleep medicine specialist. The validated pediatric sleep questionnaire and PSG were done at entry and after each treatment phase at time of PSG. Statistical analyses were ANOVA repeated measures and *t* tests.
Results The mean age of the children at entry was 6.5± 0.2 years (mean ± SEM). Overall, even if children presented improvement of both clinical symptoms and PSG findings, none of the children presented normal results after treatment 1, at the exception of one case. There was no significant difference in the amount of improvement noted independently of the first treatment approach. Thirty children underwent treatment 2, with an overall significant improvement shown for PSG findings compared to baseline and compared to treatment 1, without any group differences.
Conclusion This preliminary study emphasizes the need to have more than subjective clinical scales for determination of sequence of treatments.

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Redwood City 94063 CA, USA
e-mail: cguil@stanford.edu

Keywords Rapid maxillary expansion · Adeno-tonsillectomy · Obstructive sleep apnea · Treatment · Power analysis

2013

Level of Evidence 3

Retrospective Study

Critical role of myofascial reeducation in pediatric sleep-disordered breathing

C. Guilleminault [a,*], Y.S. Huang [b], P.J. Monteyrol [d], R. Sato [a], S. Quo [e], C.H. Lin [c]

[a] Stanford University, Sleep Medicine Division, United States
[b] Pediatric Sleep Medicine, Taiwan
[c] Cranio-Facial Center, Chang-Gung University and Memorial, Taiwan
[d] Clinique Oto-Laryngologique, Bordeaux, France
[e] Orthodontic Department, University of California, San Francisco Dental School, United States

- 24 children with sleep apnea were cured following adenotonsillectomy and/or orthodontia.
- All were referred for myofunctional therapy.
  - Patients who completed therapy: (n=11) No relapse- Average AHI= 0.5 ± 0.4
  - Patients who did not pursue therapy: (n=13) Mild relapse. Average AHI 5.3±1.5
- Absence of myofascial treatment is associated with a recurrence of SDB.
- Important role of myofunctional therapy in preventing recurrence of sleep apnea.

2014

Original Report www.enlivenarchive.org Enliven: Pediatrics and Neonatal Biology

Towards Restoration of Continuous Nasal Breathing as the Ultimate Treatment Goal in Pediatric Obstructive Sleep Apnea

Christian Guilleminault[1*], and Shannon S Sullivan[2]

[1]*Stanford University Sleep Medicine Division*

[2]*Stanford University Sleep Medicine Division, Stanford Outpatient Medical Center*

***Corresponding author**: Christian Guilleminault DM, MD, DBiol, Stanford University Sleep Medicine Division, 450 Broadway, Redwood City CA 94063, USA, Tel: 650 723 6601; E-mail: cguil@stanford.edu

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Published Date: 6th September 2014

Citation: Guilleminault C, Sullivan SS (2014) Towards Restoration of Continuous Nasal Breathing as the Ultimate Treatment Goal in Pediatric Obstructive Sleep Apnea. Enliven: Pediatr Neonatol Biol 1(1): 001.

Abstract

The interaction between oral-facial structural growth and muscle activity starts early in development and continues through childhood. Chronic oral breathing is an important clinical marker of orofacial muscle dysfunction, which may be associated with palatal growth restriction, nasal obstruction, and/ or a primary disorder of muscular or connective tissue dysfunction. It is easily documented objectively during sleep.

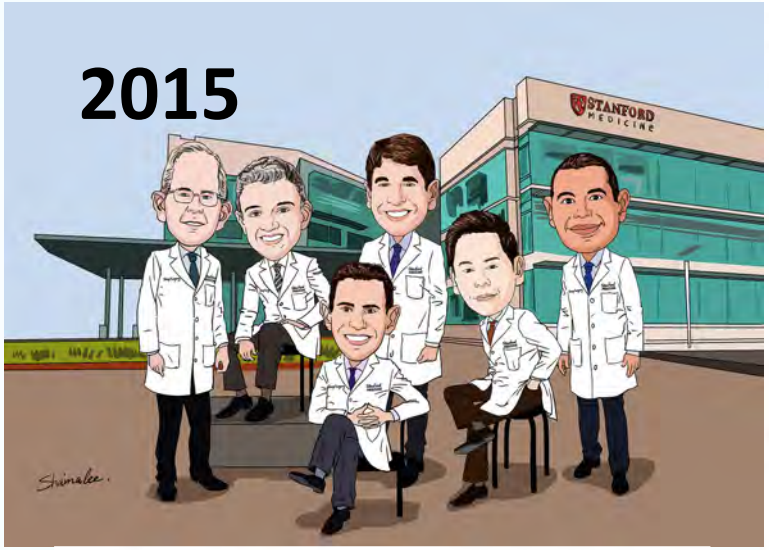
Treatment of pediatric obstructive-sleep-apnea (OSA) and sleep-disordered-breathing (SBD) means restoration of continuous nasal breathing during wakefulness and sleep; if nasal breathing is not restored, despite short-term improvements after adenotonsillectomy (T&A), continued use of the oral breathing route may be associated with abnormal impacts on airway growth and possibly blunted neuromuscular responsiveness of airway tissues.

Elimination of oral breathing, i.e., restoration of nasal breathing during wake and sleep, may be the only valid end point when treating OSA. Preventive measures in at-risk groups, such as premature infants, and usage of myofunctional therapy as part of the treatment of OSA are proposed to be important approaches to treat appropriately SDB and its multiple co-morbidities.

Keywords

Obstructive sleep apnea; Pediatrics; Oral-facial muscles; Nasal-oral functions; Myofunctional-therapy

2015



Macario Camacho June 18, 2014 at 11:53 AM

Myofunctional therapy article

To: Soroush Zaghi, Jose Abdullatif

Soroush, I would like to include you on this study. I have attached the most recent version of the article for you to review, it has comments in red that I am still addressing.

In order to include you as an author, I would like for you to perform a literature search to see if there is any additional article that we may have missed. The paper lists all the articles that Jose and I have found thus far, so you can see if you find any other ones. Please take the next few days to see if you find anything.

I should have a revised final version within a few days that I will send to all the authors for review.

Jose, would you also take one more look to see if you find any other articles (since I am listing that the systematic review was updated June 18th (today)).

I feel about 95% confident that we have all the articles, but it doesn't hurt to double check.

Thanks gentlemen,

Macario "Mac" Camacho, MD
Otolaryngology - Head and Neck Surgery
Stanford Sleep Surgery Consulting Assistant Professor 2012-2013
Stanford Sleep Medicine Fellow 2013-2014
cell: 1-(240)-535-0835

Myofunctional therap...4.docx

Myofunctional Therapy to Treat Obstructive Sleep Apnea: A Systematic Review and Meta-analysis FREE

Macario Camacho, MD, Victor Certal, MD, Jose Abdullatif, MD, Soroush Zaghi, MD, Chad M. Ruoff, MD, RPSGT, Robson Capasso, MD, Clete A. Kushida, MD, PhD

Sleep, Volume 38, Issue 5, 1 May 2015, Pages 669–675,
https://doi.org/10.5665/sleep.4652
Published: 01 May 2015 **Article history**

Abstract

Objective:

To systematically review the literature for articles evaluating myofunctional therapy (MT) as treatment for obstructive sleep apnea (OSA) in children and adults and to perform a meta-analysis on the polysomnographic, snoring, and sleepiness data.

Data Sources:

Web of Science, Scopus, MEDLINE, and The Cochrane Library.

Review Methods:

The searches were performed through June 18, 2014. The Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) statement was followed.

Results:

Nine adult studies (120 patients) reported polysomnography, snoring, and/or sleepiness outcomes. The pre- and post-MT apneahypopnea indices (AHI) decreased from a mean ± standard deviation (M ± SD) of 24.5 ± 14.3/h to 12.3 ± 11.8/h, mean difference (MD) -14.26 [95%

MYOFUNCTIONAL THERAPY TO TREAT OSA: REVIEW AND META-ANALYSIS

Myofunctional Therapy to Treat Obstructive Sleep Apnea: A Systematic Review and Meta-analysis

Macario Camacho, MD[1]; Victor Certal, MD[2]; Jose Abdullatif, MD[3]; Soroush Zaghi, MD[4]; Chad M. Ruoff, MD, RPSGT[1]; Robson Capasso, MD[5]; Clete A. Kushida, MD, PhD[1]

1. Myofunctional therapy provides a reduction in AHI of approximately 50% in adults and 62% in children.

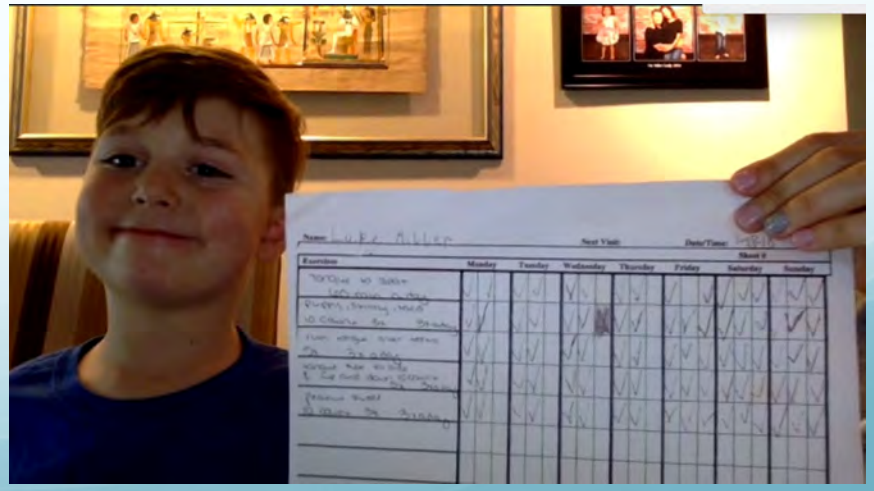
2. Improvements to daytime sleepiness and snoring.

3. Shown effective in children and adults of all ages studied thus far.

   Youngest patient: 3 years old

   Oldest patient: 79+ years old.

4. Important role in preventing relapse.

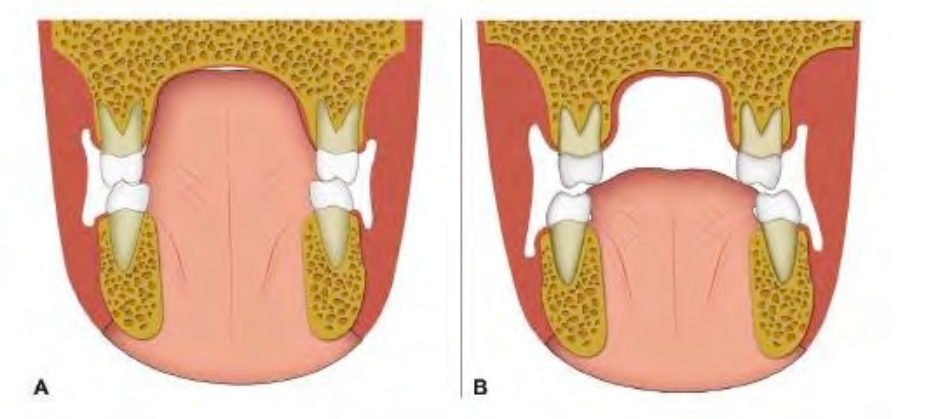


Stanford MEDICINE | The Stanford Center for Sleep Sciences and Medicine

Therapy: Goals of Treatment

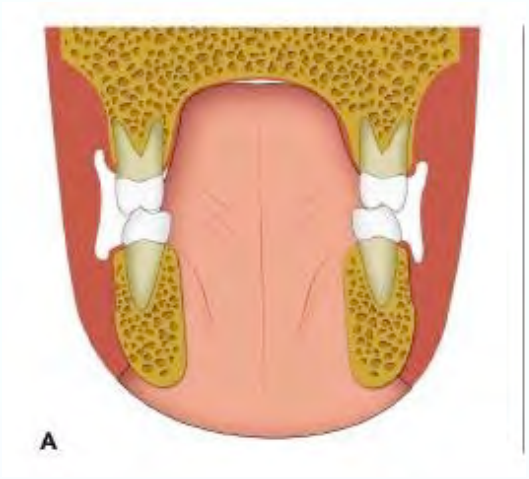
- **Lips together**

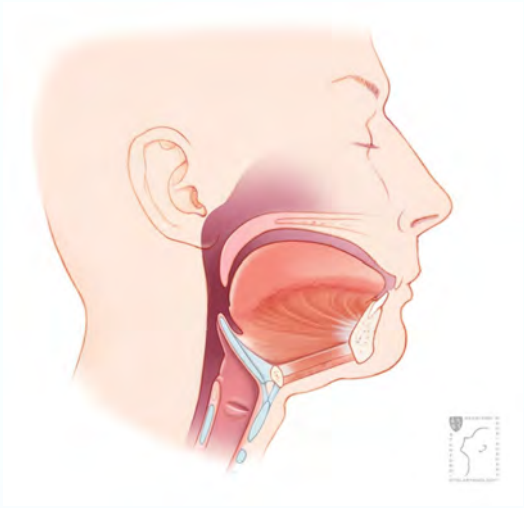
- **Tongue up on the palate**

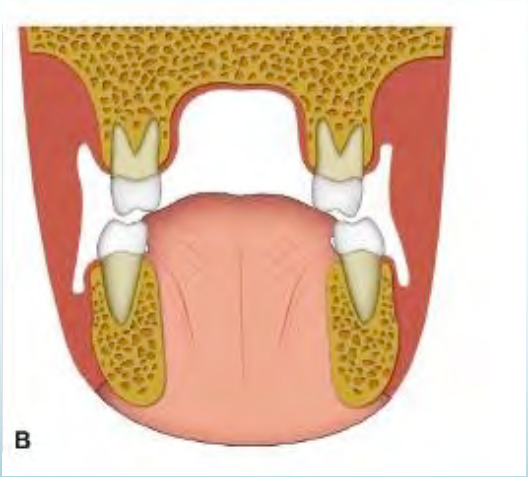


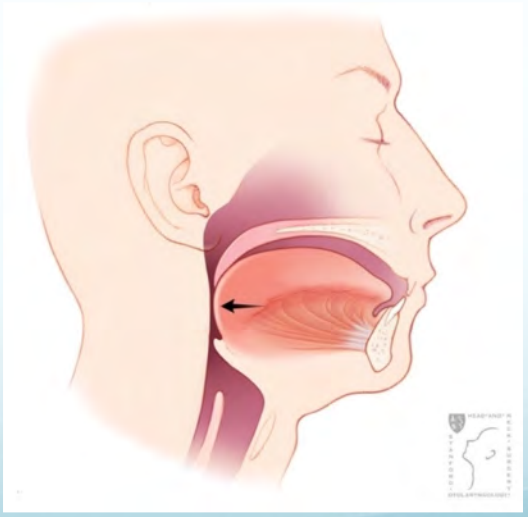
- ***Only* Nasal Breathing**

Principle of Proper Tongue Positioning: Tongue should rest completely at the roof of the mouth to maintain optimal airway function.







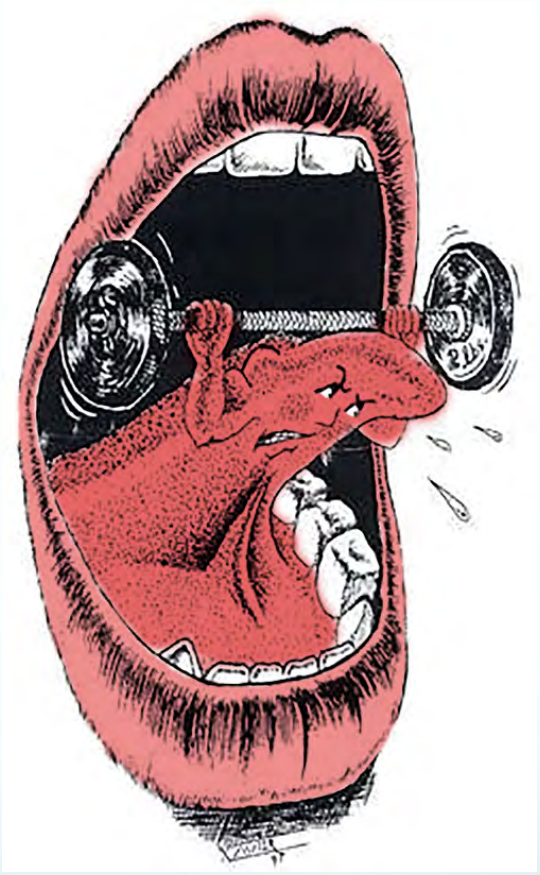


Tongue loses tone and assumes a posterior – inferior position in the airway.

Myofunctional Therapy

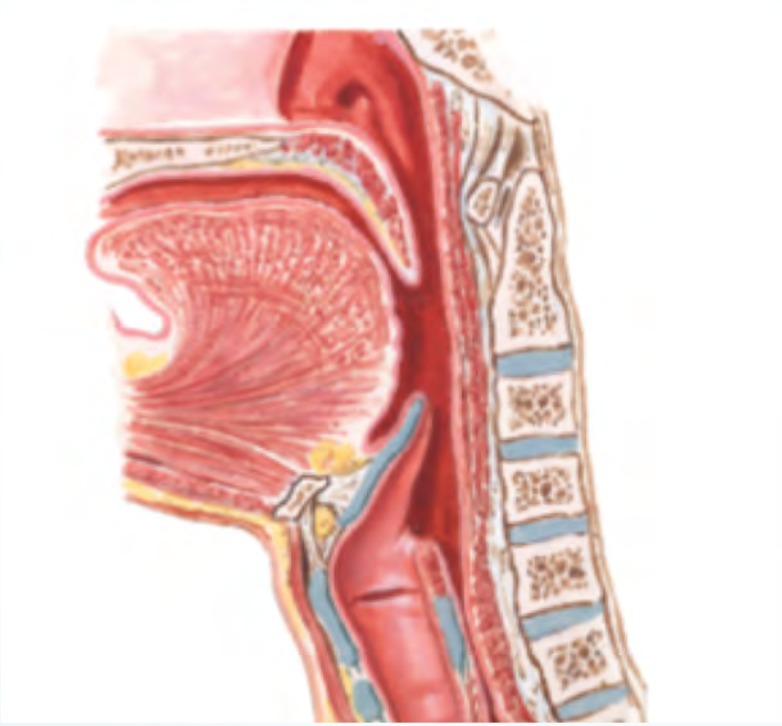
Exercise 4: Push Tongue Right: Push your tongue forward and push it to the right and hold for 10 seconds, then relax. Repeat 10 times

Exercise 5: Push Tongue Left: Push your tongue forward and push it to the left and hold for 10 seconds, then relax. Repeat 10 times.

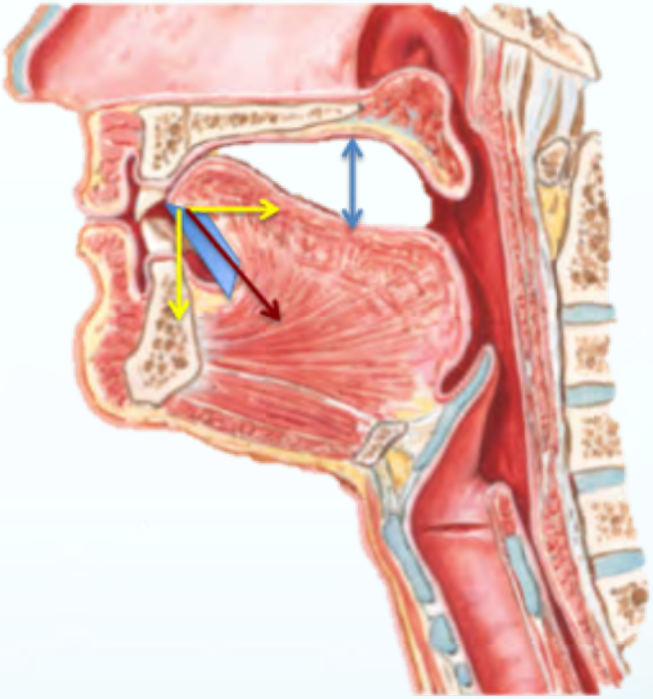


Oral myofunctional therapy is an individualized program of isometric (static) and isotonic (dynamic) strength and pattern retraining exercises of the tongue and orofacial muscles (for patients with sleep, teeth-grinding, breathing, posture, orthodontic relapse, cervical neck tension, and/or jaw pain issues) to correct maladaptive oral habits and help restore ideal resting oral posture.

Ideal Tongue Position: tongue suctioned up to the roof of mouth



Normal / Optimal
Entire tongue fills the palate



Posterior Tongue-Tie
Limited mobility of the posterior tongue

Reference Credit: http://www.paragonhealth.net.au/blog/tongue-tie-in-adults

Ideal Resting Tongue Position:

Entire tongue suctioned UP in the roof of the mouth

Complete
Lingual Palatal Suction

5 days old infant with naturally optimal tongue mobility

Structural Restrictions

→ **Tongue-tie can interfere with goals of myofunctional therapy.**

2015

ERJ open research

ORIGINAL ARTICLE
SLEEP

A frequent phenotype for paediatric sleep apnoea: short lingual frenulum

Christian Guilleminault, Shehlanoor Huseni and Lauren Lo

Affiliation: Stanford University Sleep Medicine Division, Redwood City, CA, USA.

Correspondence: Christian Guilleminault, Stanford University Sleep Medicine Division, 450 Broadway, (Pavillion C 2nd floor), Redwood City, CA 94063, USA. E-mail: cguil@stanford.edu

ABSTRACT A short lingual frenulum has been associated with difficulties in sucking, swallowing and speech. The oral dysfunction induced by a short lingual frenulum can lead to oral-facial dysmorphosis, which decreases the size of upper airway support. Such progressive change increases the risk of upper airway collapsibility during sleep.

Clinical investigation of the oral cavity was conducted as a part of a clinical evaluation of children suspected of having sleep disordered breathing (SDB) based on complaints, symptoms and signs. Systematic polysomnographic evaluation followed the clinical examination. A retrospective analysis of 150 successively seen children suspected of having SDB was performed, in addition to a comparison of the findings between children with and without short lingual frenula.

Among the children, two groups of obstructive sleep apnoea syndrome (OSAS) were found: 1) absence of adenotonsils enlargement and short frenula (n=63); and 2) normal frenula and enlarged adenotonsils (n=87). Children in the first group had significantly more abnormal oral anatomy findings, and a positive family of short frenulum and SDB was documented in at least one direct family member in 60 cases.

A short lingual frenulum left untreated at birth is associated with OSAS at later age, and a systematic screening for the syndrome should be conducted when this anatomical abnormality is recognised.

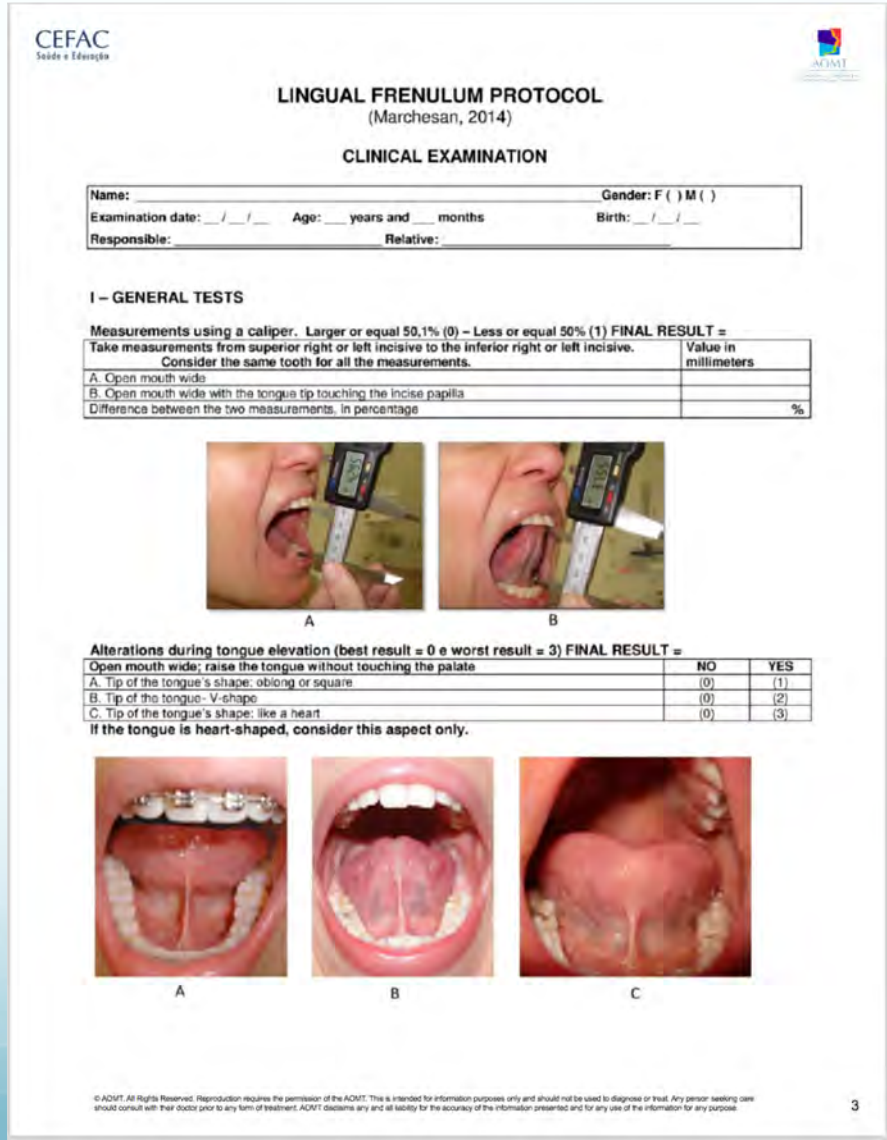
@ERSpublications
A short lingual frenulum left untreated at birth is associated with obstructive sleep apnoea syndrome at a later age http://ow.ly/6kMQ30163nG

Acknowledgements

We thank Soroush Zaghi (Stanford University Sleep Medicine Division, Redwood City, CA, USA) for his help with the statistical analyses.

Study Design

- 150 pediatric patients with OSA
  - Short frenulum (n=70)
  - Normal frenulum (n=80)



CEFAC
Saúde e Educação

LINGUAL FRENULUM PROTOCOL
(Marchesan, 2014)

CLINICAL EXAMINATION

Name: _____ Gender: F ( ) M ( )
Examination date: __/__/__ Age: ___ years and ___ months Birth: __/__/__
Responsible: _____ Relative: _____

I – GENERAL TESTS

Measurements using a caliper. Larger or equal 50,1% (0) – Less or equal 50% (1) FINAL RESULT =

Take measurements from superior right or left incisive to the inferior right or left incisive. Consider the same tooth for all the measurements.	Value in millimeters
A. Open mouth wide	
B. Open mouth wide with the tongue tip touching the incise papilla	
Difference between the two measurements, in percentage	%

A B

Alterations during tongue elevation (best result = 0 e worst result = 3) FINAL RESULT =

Open mouth wide; raise the tongue without touching the palate	NO	YES
A. Tip of the tongue's shape: oblong or square	(0)	(1)
B. Tip of the tongue- V-shape	(0)	(2)
C. Tip of the tongue's shape: like a heart	(0)	(3)

If the tongue is heart-shaped, consider this aspect only.

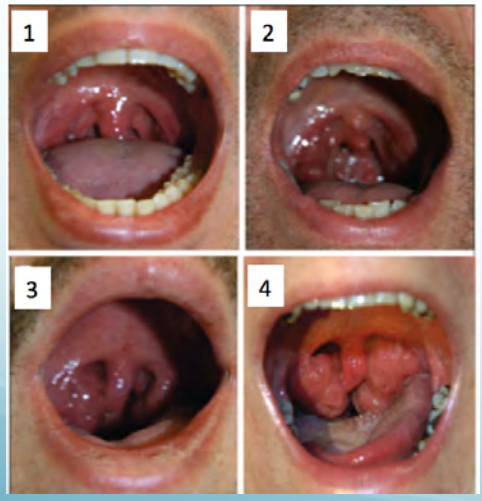
A B C

Level 3 Evidence: Case - Control Study

Study Design

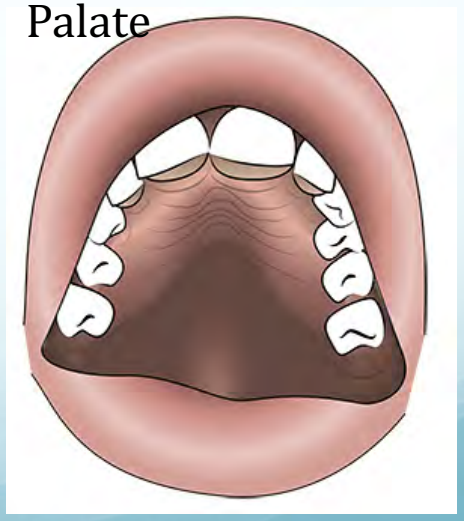
- Other Physical Exam Findings
  - Tonsil Size: Grade 1-4
  - Mallampati Tongue Position: Grade 1-4
  - High Arched Palate: Yes/No

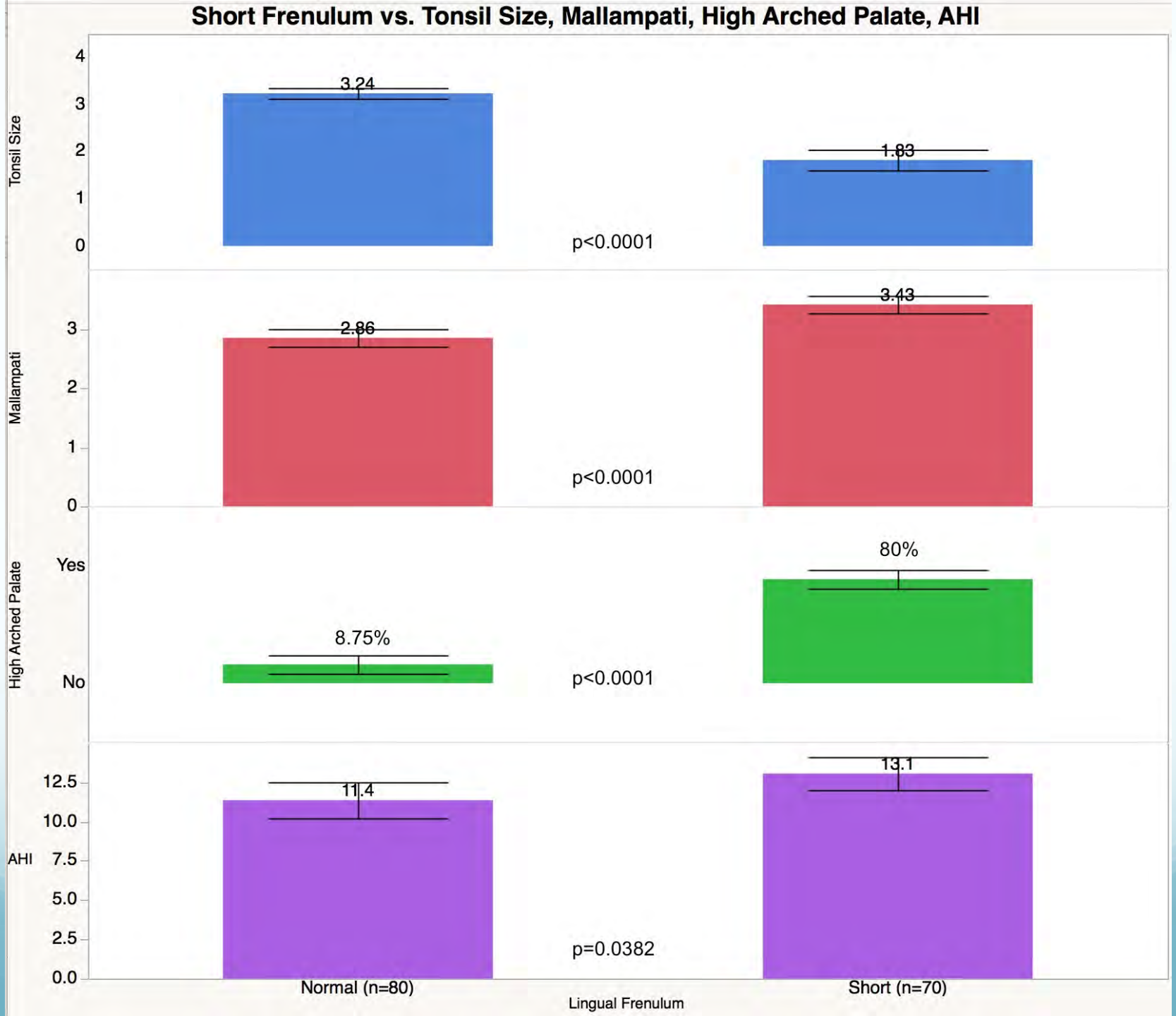
Tonsil Size



Mallampati Tongue Position

High Arched Palate




Short Frenulum vs. Tonsil Size, Mallampati, High Arched Palate, AHI
Tonsil Size
4
3
2
1
0
3.24
1.83
p<0.0001
Mallampati
3
2
1
0
2.86
3.43
p<0.0001
High Arched Palate
Yes
No
8.75%
80%
p<0.0001
AHI
12.5
10.0
7.5
5.0
2.5
0.0
11.4
13.1
p=0.0382
Normal (n=80)
Short (n=70)
Lingual Frenulum

2017

ORIGINAL ARTICLE

WILEY Orthodontics & Craniofacial

Ankyloglossia as a risk factor for maxillary hypoplasia and soft palate elongation: A functional – morphological study

A. J. Yoon[1] | S. Zaghi[2,3] | S. Ha[4] | C. S. Law[1] | C. Guilleminault[5] | S. Y. Liu[2]

[1]Sections of Pediatric Dentistry and Orthodontics, Division of Growth and Development, UCLA School of Dentistry, Los Angeles, CA, USA

[2]Division of Sleep Surgery, Department of Otolaryngology, School of Medicine, Stanford University, Stanford, CA, USA

[3]UCLA Health, Santa Monica, CA, USA

[4]UCLA School of Dentistry, Los Angeles, CA, USA

[5]Sleep Medicine Division, Stanford Outpatient Medical Center, Redwood City, CA, USA

Correspondence
A. J-S. Yoon, Section of Pediatric Dentistry and Orthodontics, Division of Growth and Development, UCLA School of Dentistry, Los Angeles, CA, USA.
Email: jungdds@gmail.com

Structured Abstract

Objectives: To characterize associations between restricted tongue mobility and maxillofacial development.

Setting and Sample Population: Cross-sectional cohort study of 302 consecutive subjects from an orthodontic practice.

Material and Methods: Tongue mobility (measured with tongue range of motion ratio [TRMR] and Kotlow free tongue measurement) was correlated with measurements of the maxillofacial skeleton obtained from dental casts and cephalometric radiographs.

Results: Tongue range of motion ratio and Kotlow measures of restricted tongue mobility were associated with (i) ratio of maxillary intercanine width to canine arch length, (ii) ratio of maxillary intermolar width to canine arch length and (iii) soft palate length. Restricted tongue mobility was not associated with hyoid bone position or Angle's skeletal classification.

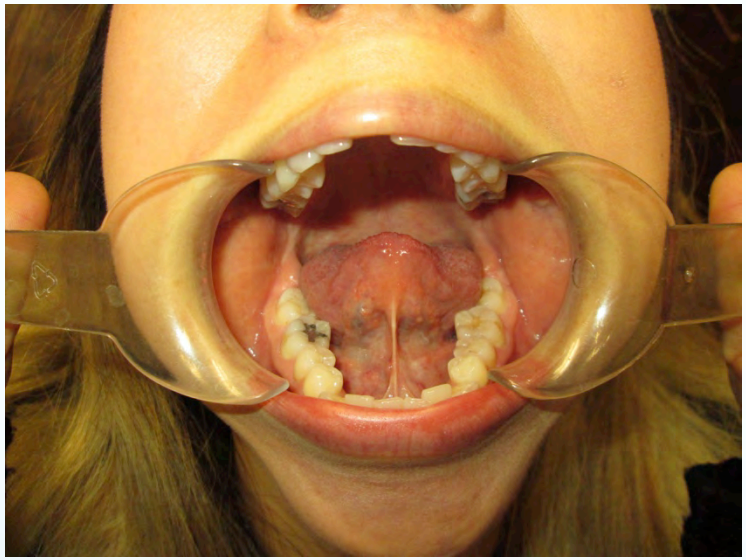
Conclusions: Restricted tongue mobility was associated with narrowing of the maxillary arch and elongation of the soft palate in this study. These findings suggest that variations in tongue mobility may affect maxillofacial development.

KEYWORDS
ankyloglossia, frenulum, maxillofacial development, myofunctional dysfunction

Level 3 evidence: Cross-sectional cohort study

Question:

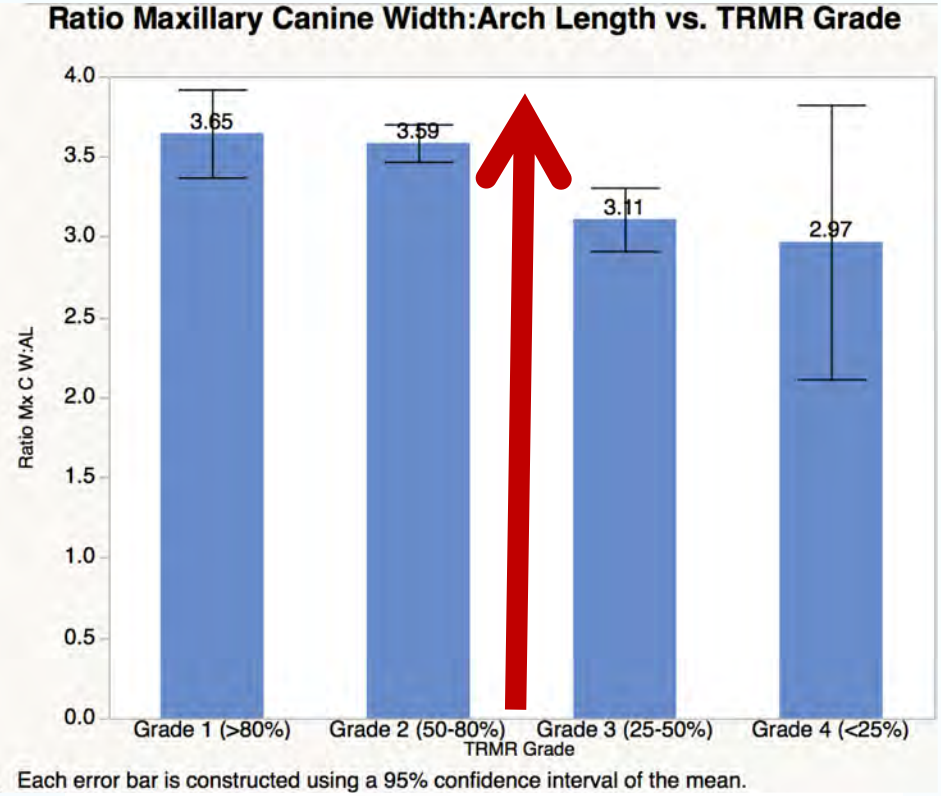
Could altered tongue mobility affect development of the maxillary arch?

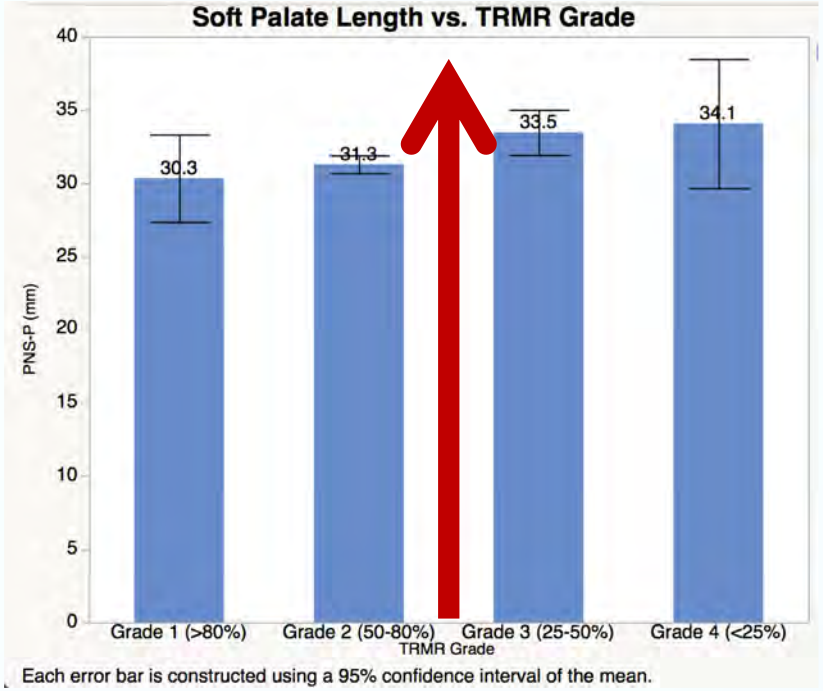


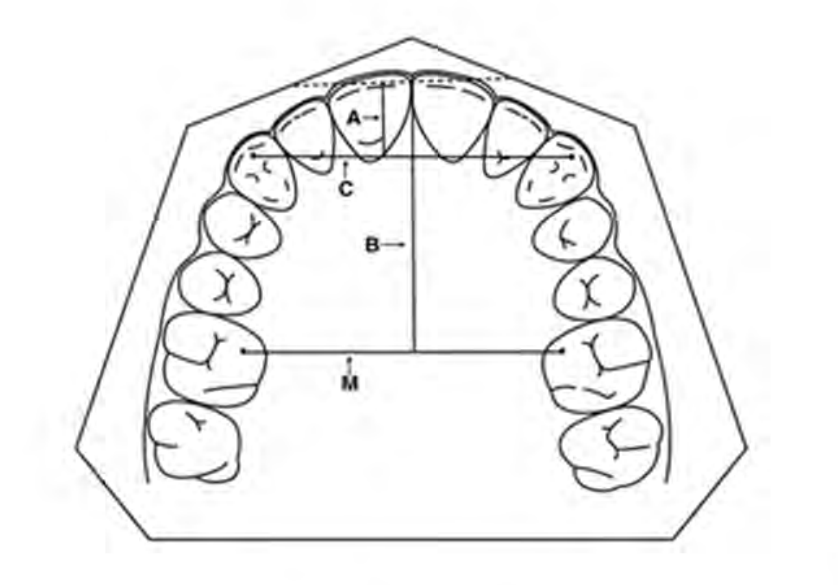
Restricted Tongue Mobility

V-Shaped Maxillary Arch

Level 3 evidence: Cross-sectional cohort study


Ratio Maxillary Canine Width:Arch Length vs. TRMR Grade
Ratio Mx C W:AL
4.0
3.5
3.0
2.5
2.0
1.5
1.0
0.5
0.0
3.65
3.59
3.11
2.97
Grade 1 (>80%)
Grade 2 (50-80%)
Grade 3 (25-50%)
Grade 4 (<25%)
TRMR Grade
Each error bar is constructed using a 95% confidence interval of the mean.


Soft Palate Length vs. TRMR Grade
PNS-P (mm)
40
35
30
25
20
15
10
5
0
30.3
31.3
33.5
34.1
Grade 1 (>80%)
Grade 2 (50-80%)
Grade 3 (25-50%)
Grade 4 (<25%)
TRMR Grade
Each error bar is constructed using a 95% confidence interval of the mean.


A
C
B
M


S
N
ANS
SN-Mn
PNS
A
Go
P
B
H-Mn line
H
Me

Functional Classification of Ankyloglossia Based on Tongue Range of Motion Ratio (TRMR)

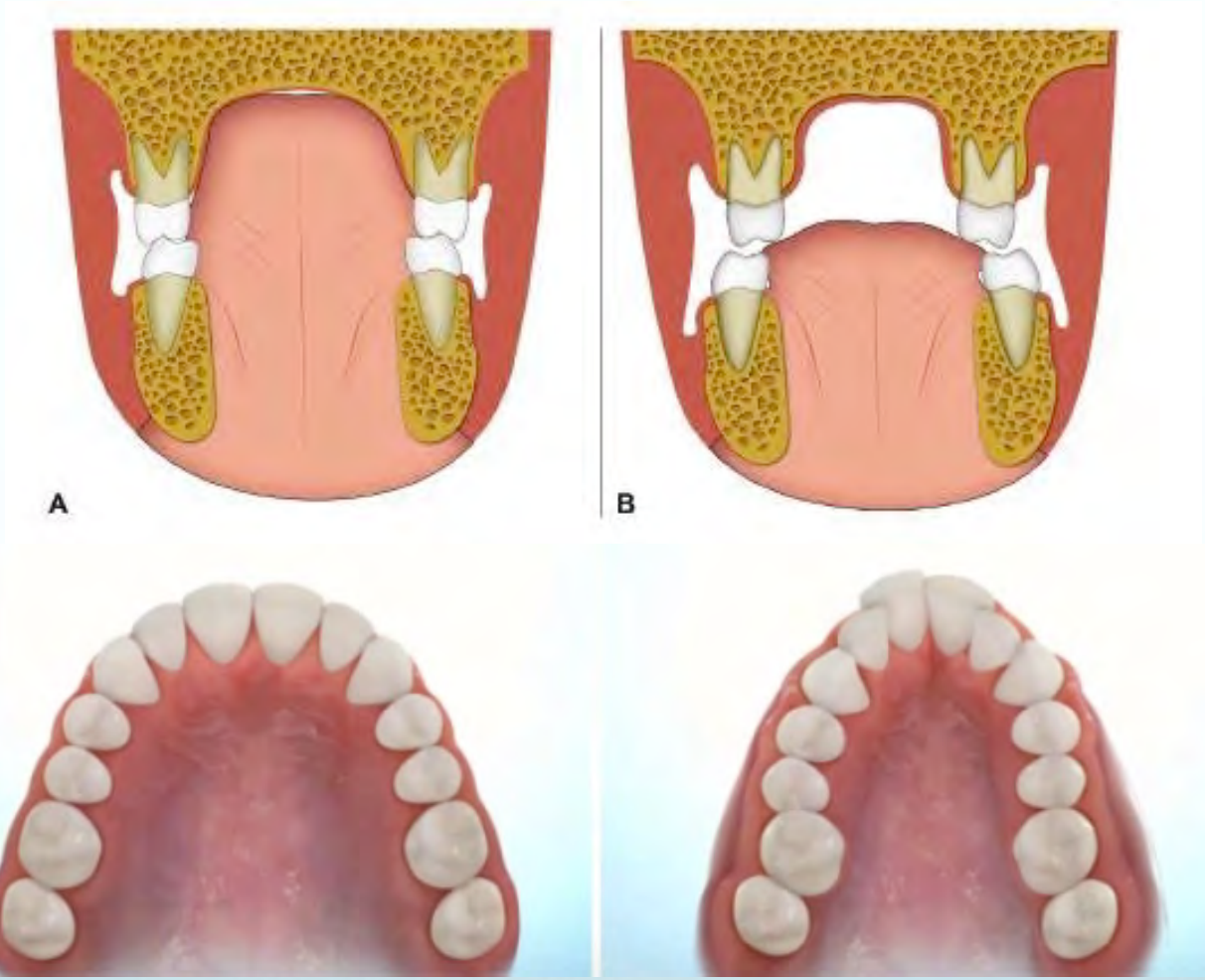
Grade 1 Functioning: TRMR > 80%

Grade 2 Functioning: TRMR 50-80%

Grade 3 Functioning: TRMR < 50%

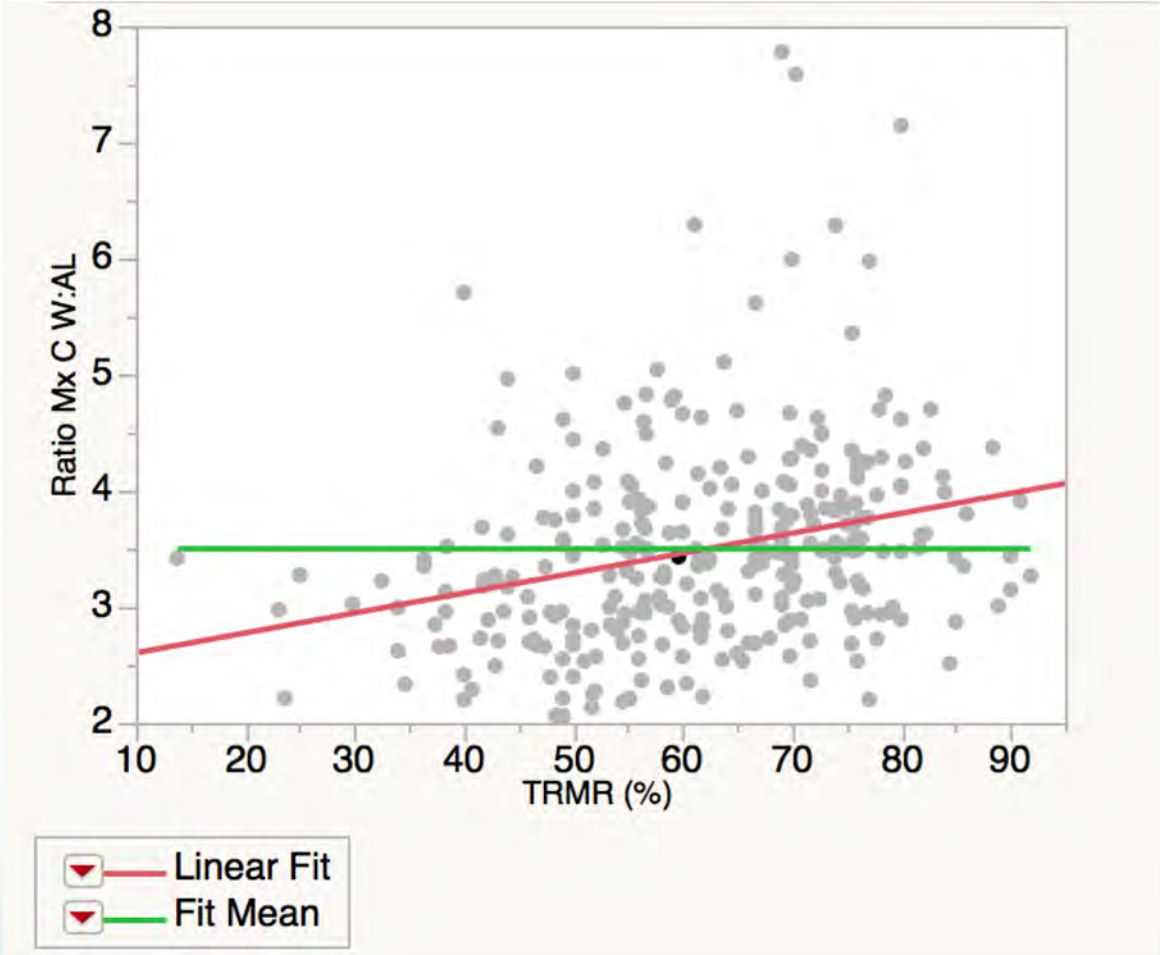
Grade 4 Functioning: TRMR < 25%

Conclusion: Restricted tongue mobility is associated with narrow V-shaped maxillary arch



Level 3 evidence showing that Grade 3+ functional ankyloglossia is associated with alterations of orofacial morphology

Maxillofacial Development is Multifactorial



Restricted tongue mobility accounts for 7.6% of the variance in narrowness of the maxillary arch. Other factors: mouth breathing, oral habits, tongue tone, oral resting posture, nutrition, chewing, swallowing as well as other genetic and environmental factors environmental play a critical role in >90% of cases.

2018

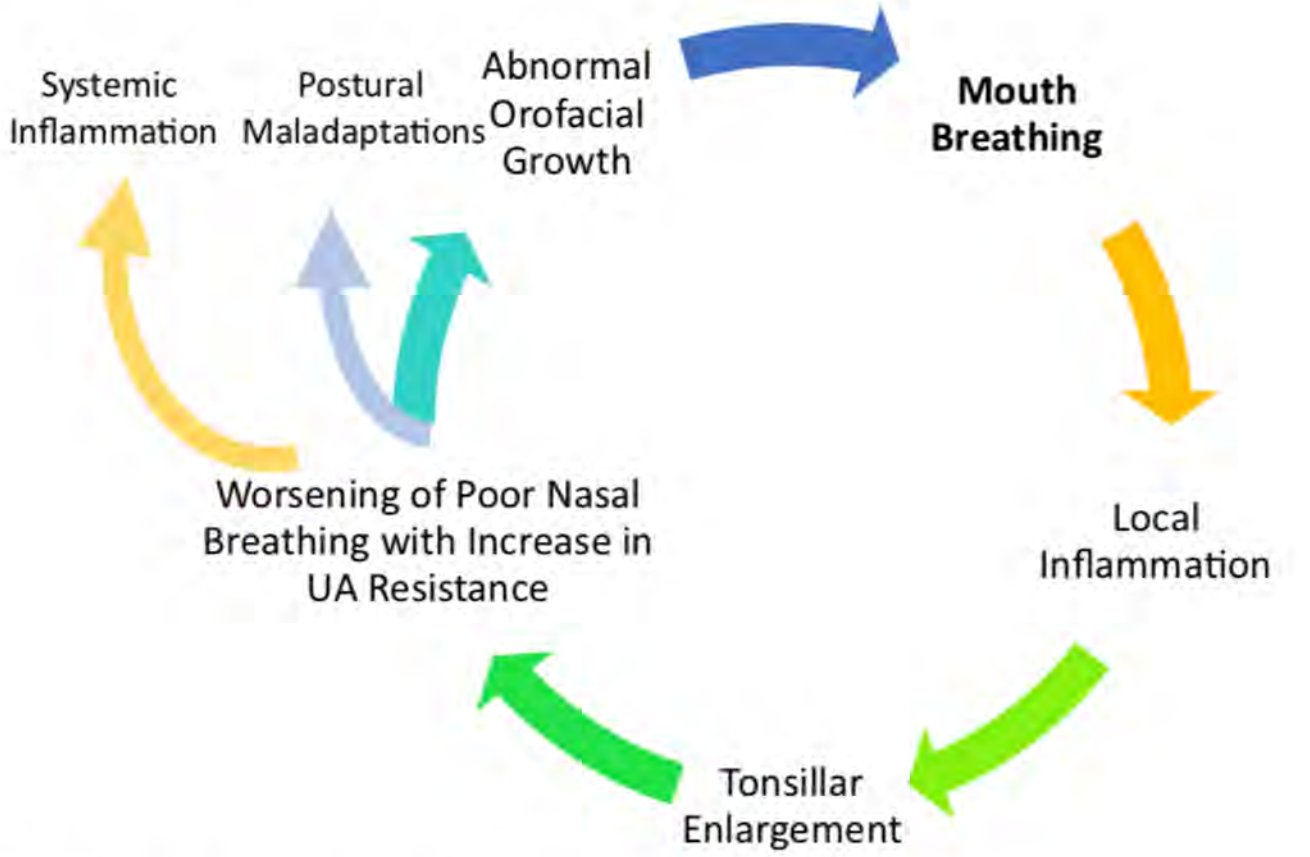


Fig. 13. Mouth-breathing and its negative impacts.
The many negative impacts of mouth breathing are summarized in the Schema with indication of the induced vicious cycles.

Guilleminault C et al., The nocturnal-polysomnogram and “non-hypoxic sleep-disordered-breathing” in children, Sleep Medicine, https://doi.org/10.1016/j.sleep.2018.11.001

Mouth Breathing and Tongue Position

- High Tongue Position

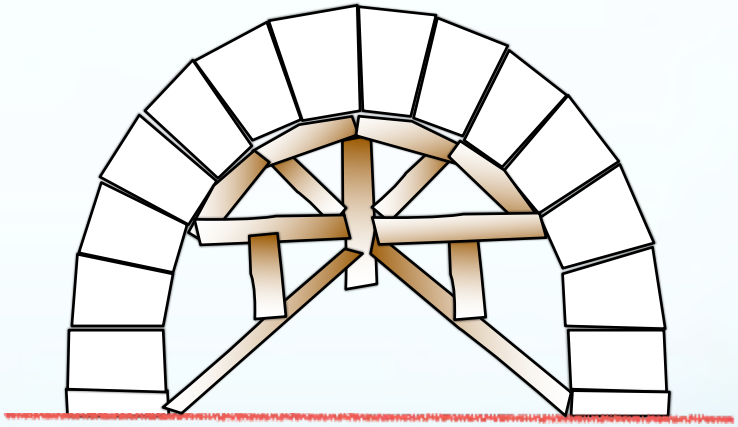
Nasal Breathing

- Low Tongue Position

Mouth Breathing

Tongue as scaffold for maxillary arch

U-Shaped Arch

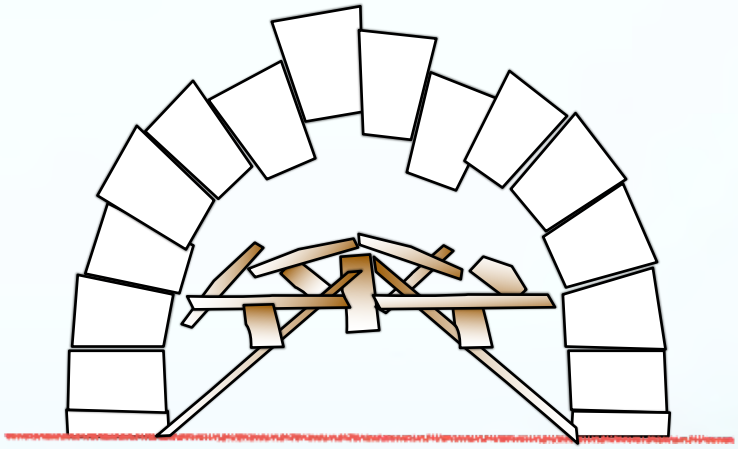


Animation Credit:
Nathan Devery and Nora Ghodousi- Zaghi, DDS

Animation Credit:
Barry Raphael, DMD

Low tongue position → dysfunctional scaffold

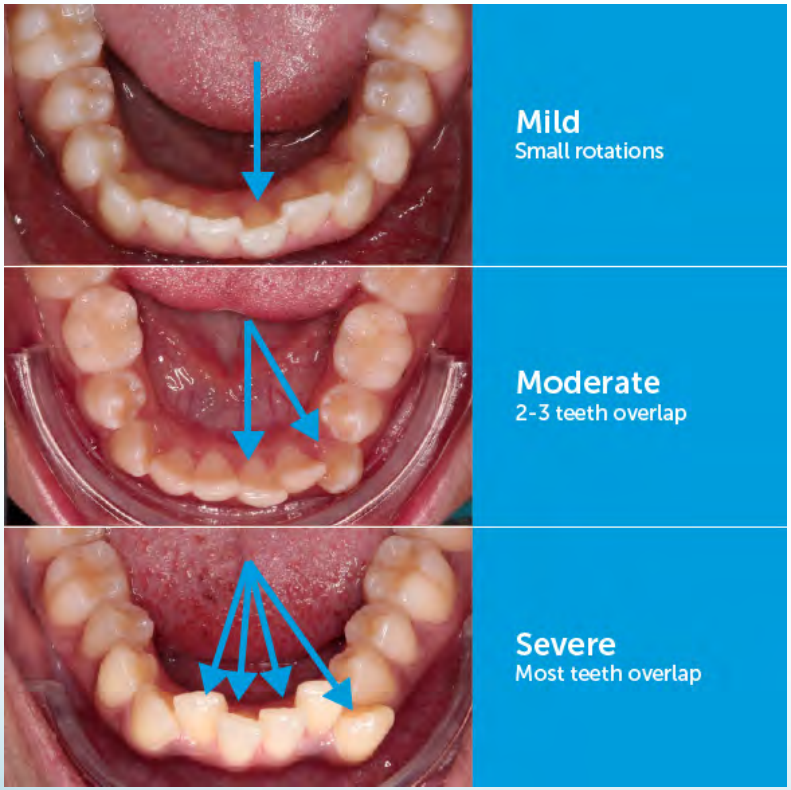
V-Shaped Arch



Animation Credit:
Nathan Devery and Nora Ghodousi- Zaghi, DDS

Animation Credit:
Barry Raphael, DMD

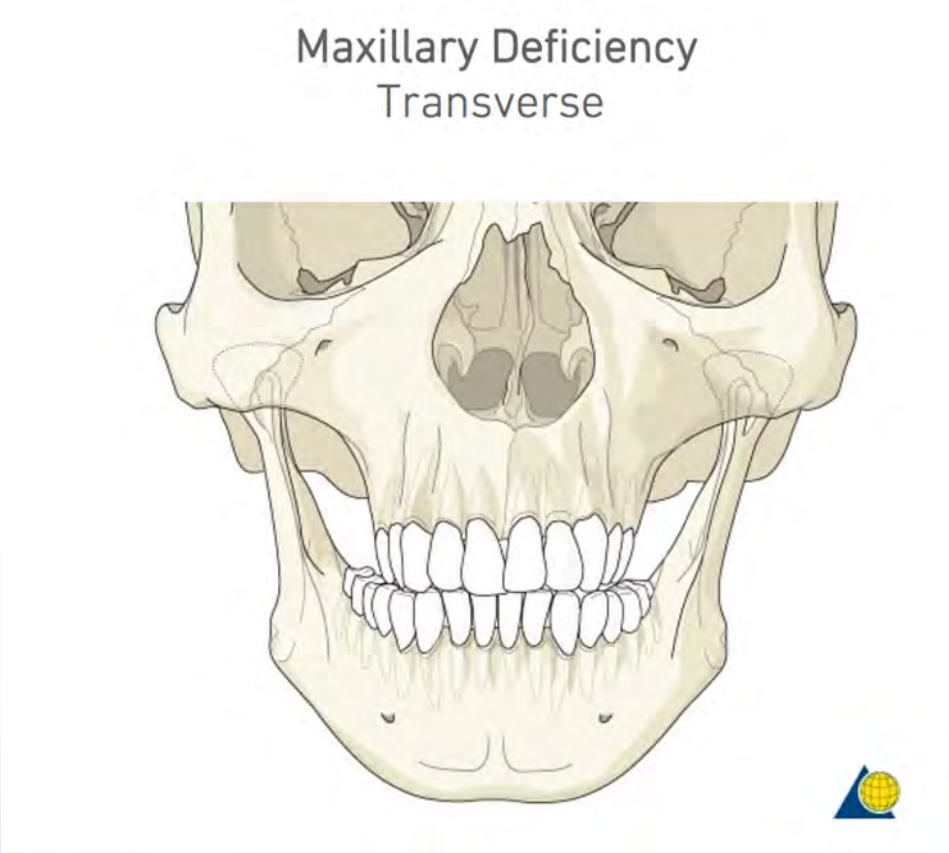
Dental Crowding

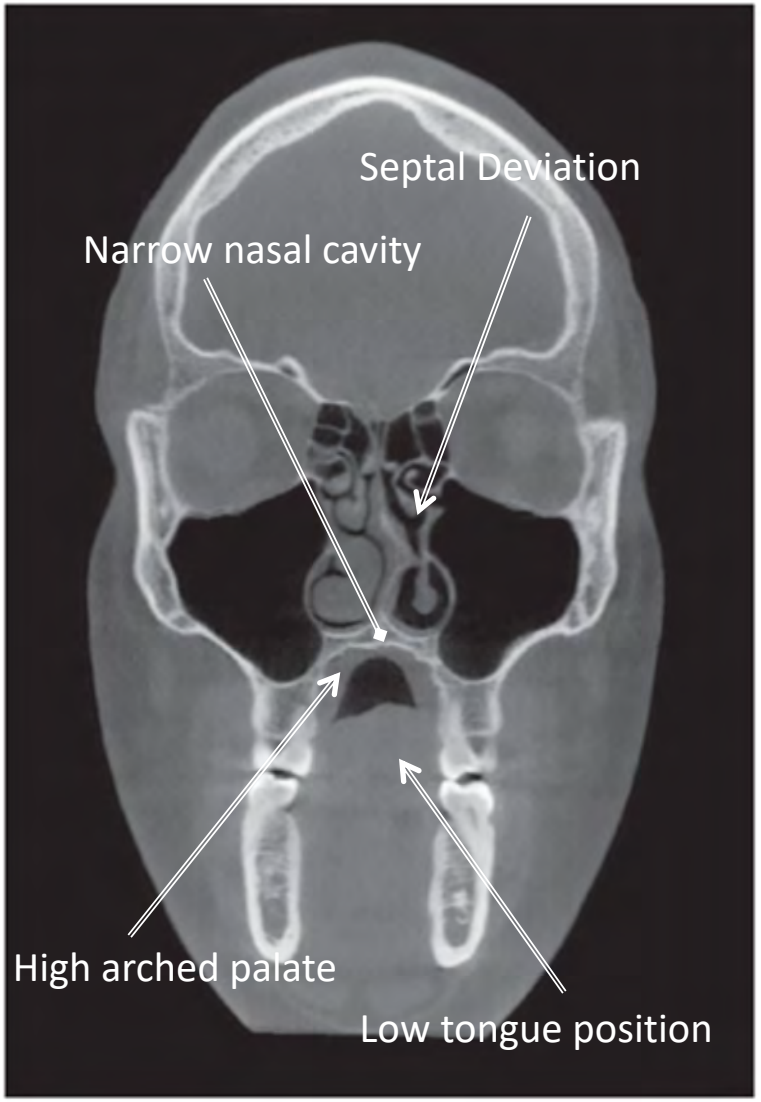


Skeletal Deficiencies: V-Shaped High Arched Narrow Palate

Tongue position is integral to development of maxillary morphology

High Arched Palate → Narrow Nasal Cavity

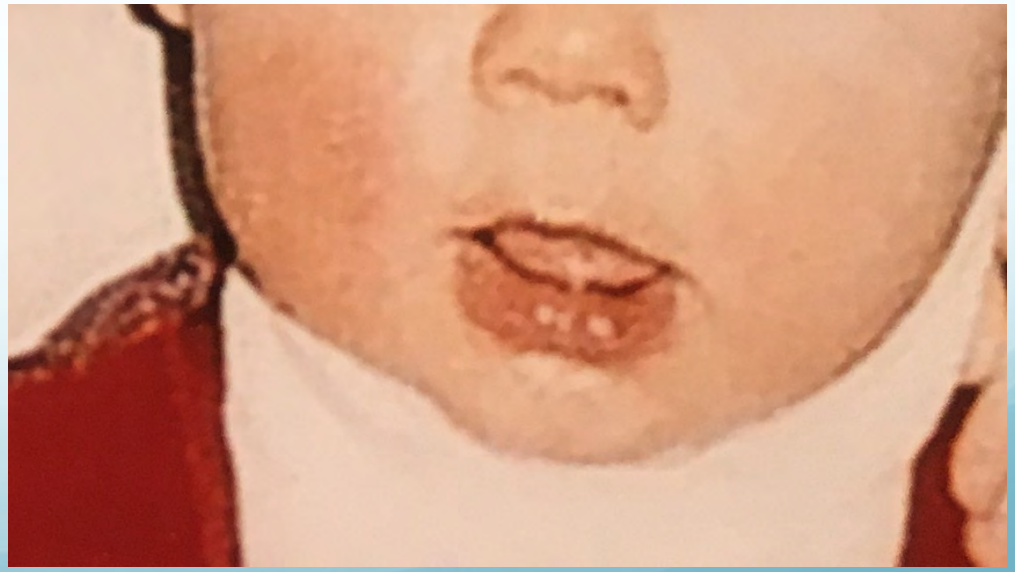




Yoon, A. J., Zaghi, S., Ha, S., Law, C. S., Guilleminault, C., & Liu, S. Y. (2017). Ankyloglossia as a risk factor for maxillary hypoplasia and soft palate elongation: A functional–morphological study. *Orthodontics & craniofacial research*, *20*(4), 237-244.

Trevor - 23 year-old with:

- Recurrent sinus infections.
- Tightness in his neck and shoulders
- TMJ pain
- Sleep issues
- Anxiety/depression due to chronic fatigue.



2015

Level of Evidence 4

Case Series

Huang et al. Int J Pediatr Res 2015, 1:1

International Journal of Pediatric Research

Research Article: Open Access

Short Lingual Frenulum and Obstructive Sleep Apnea in Children

*Yu-Shu Huang[1], Stacey Quo[2], J Andrew Berkowski[3] and Christian Guilleminault[3]**

[1]Chang Gung Memorial Hospital and University Pediatric sleep laboratory, Taiwan

[2]UCSF Dental School, USA

[3]Stanford University Sleep Medicine Division, USA

***Corresponding author:** Christian Guilleminault, Stanford University Sleep Medicine Division, 450 Broadway Pavilion C, 2nd floor, Redwood City, CA 94063, USA, E-mail: cguil@stanford.edu

We identified 27 patients with association of short lingual frenulum and SDB.

The mean age was 11.4 years (range: 2 to 16 years).

Children presented with symptoms of SDB such as snoring, poor sleep, and fatigue but also a history of symptoms associated with short lingual frenulum such as problems with speech, swallowing or suction, particularly early in life.

Table 1: Disease characteristics at entry and after first treatment

	At entry		After 1st treatment	
	n	(%)	n	(%)
Demographics (n=27)				
Boys	18	(63%)		
Mean Age (years) (SD)	11.4 ± 5.2			12.3 ± 4.6
Disease characteristics				
Overall symptoms	27	(100)	9	(90)
Fatigue	27	(100)	10	(37)
EDS	9	(35)	1	(4)
Poor sleep	18	(67)	9	(33)
Snoring	20	(74)	2	(7.5)
Speech problems+	13	(48)	2	(7.5)
Swallowing problems+	7	(26)	0	(0.0)
Chewing problems+	6	(22)	1	(3.7)
Tonsil scale				
0/1	8(30)			18(66.6)
2	9(33)			9(33)
3	5(18.5)			0 (0.0)
4	5(18.5)			0 (0.0)
Mouth breathing	27(100)			25 (92.5)
PSG findings				
AHI, mean ±SD	12 ± 4.6			3 ± 2
SaO2nadir, mean ±SD	89 ± 2.5			94 ± 1.6
Flow limitation, mean ±SD)	73 ± 11			31 ± 9
Mouth breathing (%TST)	78 ± 14			61 ± 16

+ indicates that symptom was reported during pre-pubertal period but not present necessarily at time of evaluation

n= 10 children with large tonsils referred to ENT for T&A and frenectomy

n=8 children with normal sized tonsils referred to ENT for frenectomy.

n=9 children referred to orthodontists for rapid maxillary expansion (RME) and need for frenectomy was also mentioned.

==========

n= 8 / 10 children had tonsils and frenectomy performed

n= 5/ 8 children had isolated frenectomy

n= 0/9 children referred for RME had frenectomy performed

Results: "Short Lingual Frenulum and OSA in Children"

n= 8 / 10 children had tonsils and frenectomy performed

n= 5/ 8 children had isolated frenectomy

n= 0/9 children referred for RME had frenectomy performed

There was an overall improvement by clinical evaluation and PSG in all children whether frenectomy was performed or not.

However, only ***two*** children (both treated with tonsillectomy and frenectomy) had complete resolution of abnormal mouth breathing.

All others (92.5%, 25/27) persisted with abnormal mouth breathing, including ***all five*** children that were treated with isolated frenectomy.

These 25 patients with residual mouth breathing were referred for myofunctional therapy (+ frenectomy if not yet performed).

==========

There were 11 patients who followed up after completing 4-6 months of therapy :

n=4 children with isolated frenectomy

n= 2 children with prior tonsillectomy who then proceeded with frenectomy

n= 5 children with orthodontics who then proceeded with frenectomy

All 11/ 11 patients achieved 100% improvement to mouth breathing and sleep symptoms.

Conclusion: "Short Lingual Frenulum and OSA in Children"

Frenectomy for short lingual frenulum in isolation or following T&A ***helps but is commonly insufficient*** to resolve all abnormal breathing patterns during sleep among children with OSA.

Frenectomy alone may not be sufficient to restore normal nasal breathing function during sleep, particularly if the frenulum-related problem has lingered over years.

Myofunctional therapy may be needed after frenulum surgery to to restore normal nasal breathing function.

2019

Laryngoscope Investigative Otolaryngology Open Access

Lingual frenuloplasty with myofunctional therapy: Exploring safety and efficacy in 348 cases.

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Date Submitted by the Author:	n/a
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Specialty Area:	Speech language pathology < Laryngology, Sleep < Pediatric airway < Pediatrics, Quality of Life, Snoring < Sleep Medicine, Outcomes/cost effectiveness < Clinical < Allergy/Rhinology
Abstract:	Abstract Background: Ankyloglossia (tongue-tie) is a condition of altered tongue mobility due to the presence of restrictive tissue between the undersurface of the tongue and the floor of mouth. Potential implications of restricted tongue mobility (such as mouth breathing, snoring, dental clenching, myofascial tension) remain underappreciated due to limited peer-reviewed evidence. Here we explore the safety and efficacy of lingual frenuloplasty and myofunctional therapy for the treatment of these conditions in a large and diverse cohort of patients with restricted tongue mobility. Methods: 420 consecutive patients (ages 29 months to 79 years) treated with myofunctional therapy and lingual frenuloplasty for indications of mouth breathing, snoring, dental clenching, and/or myofascial tension

Lingual frenuloplasty with myofunctional therapy: Experience with 348 cases exploring saftey and efficacy of tongue-tie release for mouth breathing, snoring, dental clenching, and myofascial tension. [Research Manuscript, Submitted]

Table 1. Patient-reported satisfaction with lingual frenuloplasty and myofunctional therapy treatment protocol.

Satisfaction:	Number	Percent Total	
A (very satisfied)	250	71.8%	**Overall Satisfied: 91.1%**
B (somewhat satisfied)	67	19.3%	
C (neutral)	21	6.0%	
D (somewhat dissatisfied)	10	2.9%	**Overall Dissatisfied: 2.9%**
F (very dissatisfied)	0	0.0%	

Table 2. Health-related quality of life following lingual frenuloplasty and myofunctional therapy treatment protocol.

Health-Related Quality of Life:			
A (much better)	137	39.3%	**Overall QOL Improved: 87.4%**
B (somewhat better)	167	48.0%	
C (neutral)	42	12.1%	
D (somewhat worse)	2	0.6%	**Overall QOL Worse: 0.6%**
F (much worse)	0	0.0%	

Table 3. Benefits attributed to lingual frenuloplasty with myofunctional therapy protocol.

Benefits	Improved	Did Not Improve	Unsure	N/A	Percent Improved	Standard Error
Overall tongue mobility	326	12	10	-	**96.5%**	1.0%
Clenching or grinding of teeth	40	4	-	304	**91.0%**	4.3%
Ability to perform myofunctional therapy exercises	307	35	6	-	**89.8%**	1.6%
Ease of swallow	102	25	3	218	**80.3%**	3.5%
Sleep quality	195	50	11	92	**79.6%**	2.6%
Nasal breathing	174	48	4	122	**78.4%**	2.8%
Neck, shoulder, facial tension or pain	117	34	-	197	**77.5%**	3.4%
Snoring	102	38	11	197	**72.9%**	3.8%

Table 4. Patient reported risks and complications.

Risks/ Complications	Reported	Not Reported	Percent Reported	Standard Error
Pain	157	191	**45.1%**	2.7%
--- Pain for longer than 7 days	5	343	**1.4%**	0.6%
Bleeding	44	304	**12.6%**	1.8%
--- Prolonged bleeding >24 hours	7	341	**2.0%**	0.8%
Numbness of the tongue-tip	17	331	**4.9%**	1.2%
--- Numbness >2 weeks	9	339	**2.6%**	0.9%
Salivary gland issues	12	336	**3.4%**	1.0%
--- Complaints> 2 weeks	3	345	**0.9%**	0.5%
Second stage release procedure to further improve tongue mobility after initial improvement	12	336	**3.4%**	1.0%
Revision surgery to excise scarring that resulted in worse mobility than prior to initial release	11	337	**3.2%**	0.9%

High rates of patient satisfaction and treatment success.
Low risk of minor complications.

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Case: 47-year-old female loud snoring and breathing interruptions during sleep associated with fragmented sleep and excessive daytime sleepiness. She was diagnosed with obstructive sleep apnea in April 2015; she reports that CPAP is intrusive and cumbersome to use. There is a history of forward head posture as well as neck and shoulder tension.

July 2017: Baseline

Sleep Study Report

Sleep Summary

Start Study Time:	11:52:42PM
End Study Time:	4:03:41AM
Total Recording Time:	4 hrs, 10 min
Total Sleep Time	**3 hrs, 10 min**
% REM of Sleep Time:	21.8

Respiratory Indices

	REM	NREM	All Night
pRDI:	63.5	31.5	**38.3**
pAHI:	63.5	16.5	**26.6**
ODI:	47.2	7.7	**16.2**

Indices are calculated using technically valid sleep time of 3 hrs, 9 min.
pRDI/pAHI are calculated using oxi desaturations ≥ 3%

Oxygen Saturation Statistics

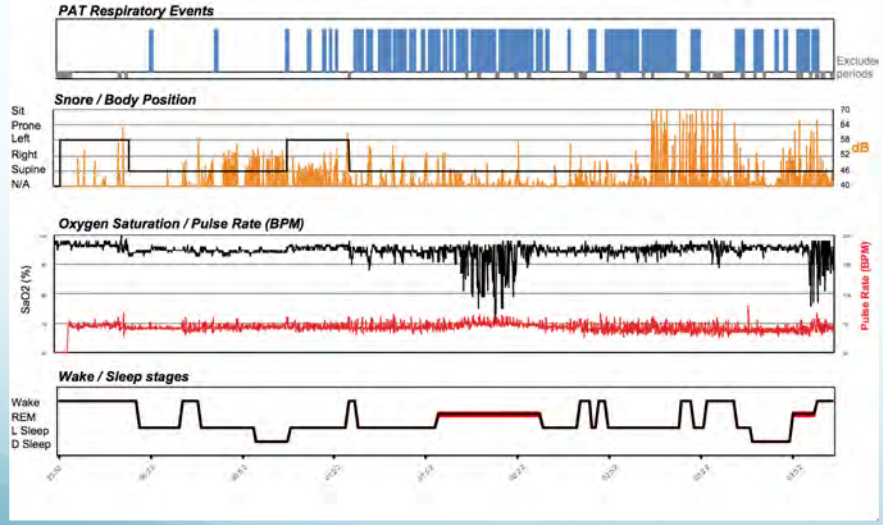
Mean: 94 Minimum: 70 Maximum: 98
Mean of Desaturations Nadirs (%): 88

Oxygen Desatur. %:	4-9	10-20	>20	Total
Events Number	33	16	2	51
Total	64.7	31.4	3.9	100.0

Oxygen Saturation	<90	<88	<85	<80	<70
Duration (minutes):	6.2	5.0	3.4	1.4	0.0
Sleep %	3.2	2.6	1.8	0.7	0.0

Pulse Rate Statistics during Sleep (BPM)

Mean: 70 Minimum: 57 Maximum: 101



Dec 2017: 5 months after MFT + Frenuloplasty

Sleep Study Report

Sleep Summary

Start Study Time:	12:07:26AM
End Study Time:	6:27:03AM
Total Recording Time:	6 hrs, 19 min
Total Sleep Time	**5 hrs, 27 min**
% REM of Sleep Time:	24.5

Respiratory Indices

	REM	NREM	All Night
pRDI:	19.7	11.0	**13.1**
pAHI:	18.2	6.3	**9.2**
ODI:	12.1	2.7	**5.0**

Indices are calculated using technically valid sleep time of 5 hrs, 25 min.
pRDI/pAHI are calculated using oxi desaturations ≥ 3%

Oxygen Saturation Statistics

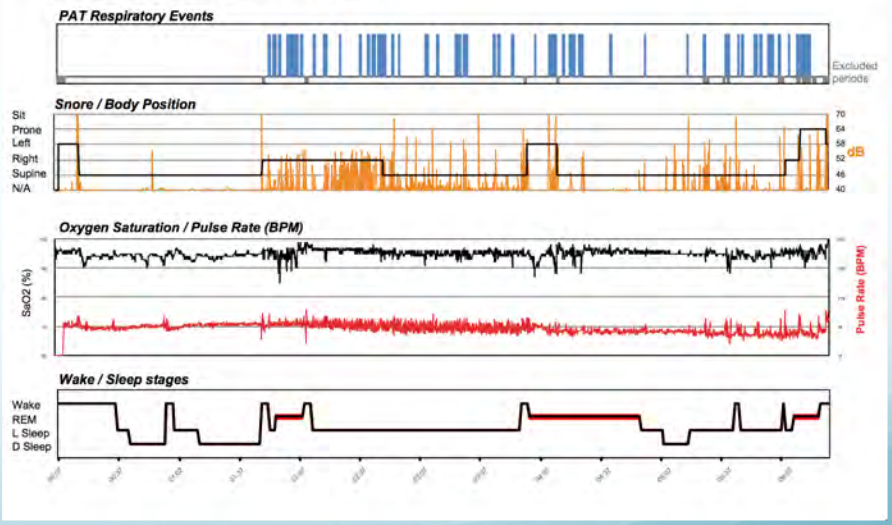
Mean: 94 Minimum: 85 Maximum: 99
Mean of Desaturations Nadirs (%): 91

Oxygen Desatur. %:	4-9	10-20	>20	Total
Events Number	26	1	0	27
Total	96.3	3.7	0.0	100.0

Oxygen Saturation	<90	<88	<85	<80	<70
Duration (minutes):	2.1	0.3	0.0	0.0	0.0
Sleep %	0.7	0.1	0.0	0.0	0.0

Pulse Rate Statistics during Sleep (BPM)

Mean: 73 Minimum: 52 Maximum: 93

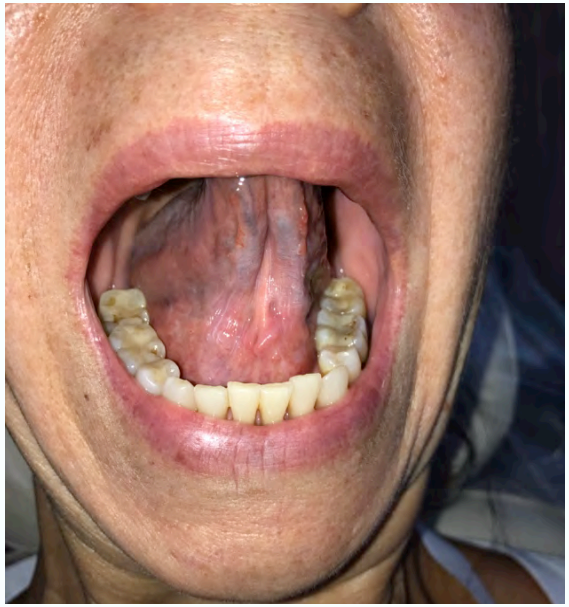


Improvements of 65 – 80% in RDI, AHI, ODI, & time spent below 90% SpO2.

60 year-old female with WORSE sleep apnea after frenuloplasty

Baseline

AHI = 17

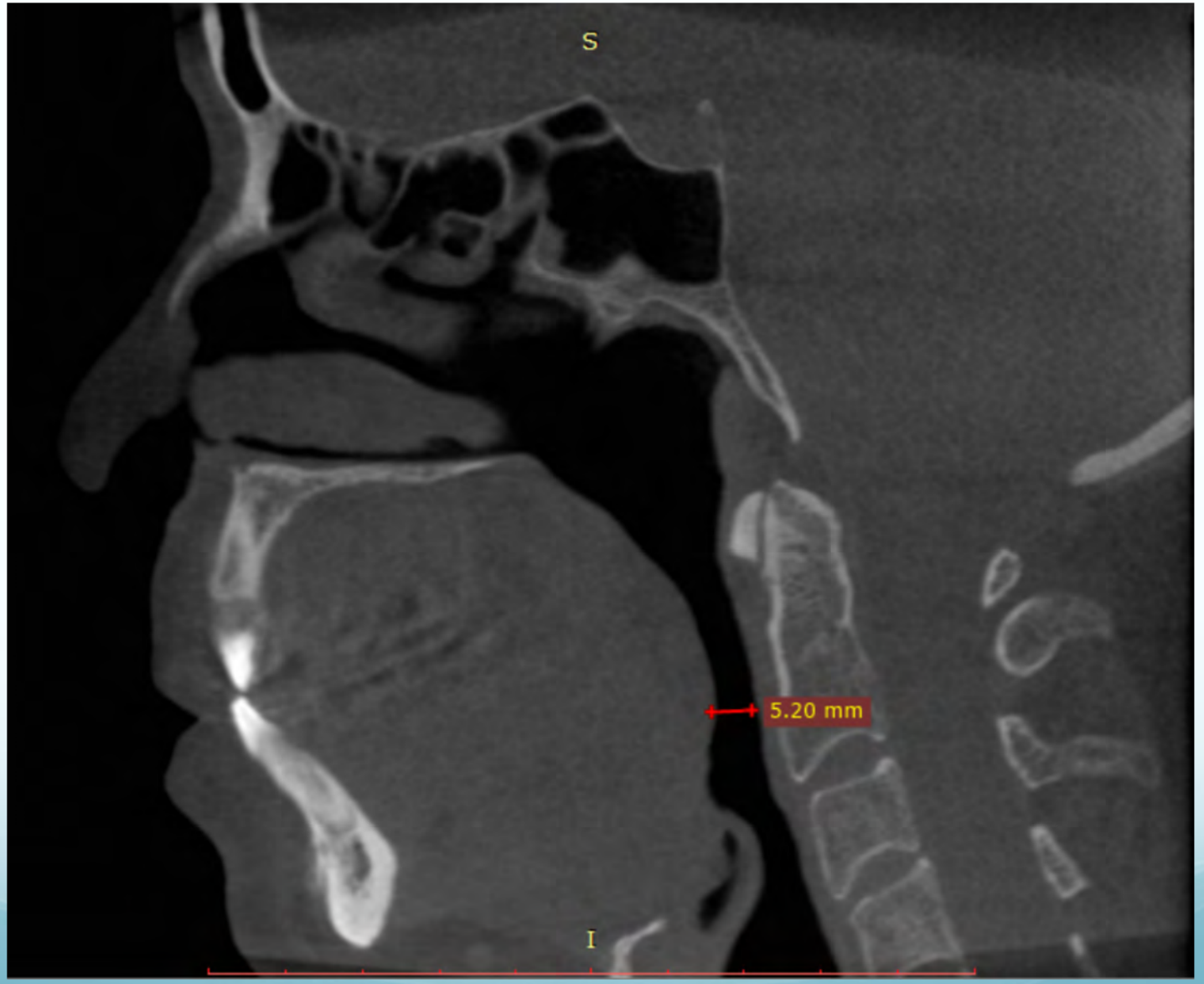


6 Weeks

AHI = 56

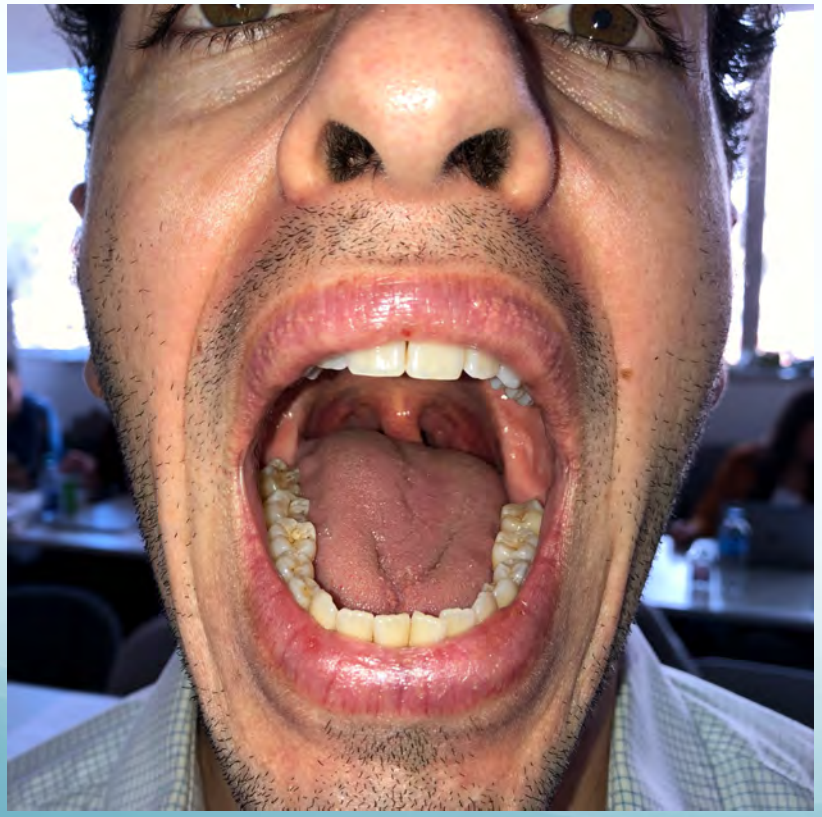
But Worsened Sleep Apnea…..

Very Narrow Posterior Airway Space

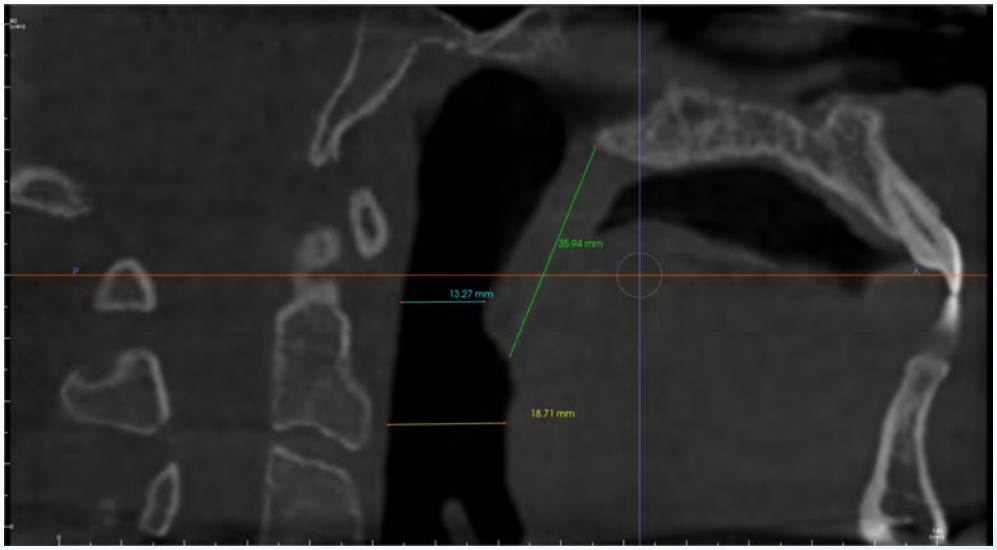


2020+: Upcoming Research: Patient selection and modifying treatment protocol to further improve success rate and reduce risks /complications.

Case: 36-year-old male with with face/neck tension, headaches, and open mouth breathing. He notices tension in his neck and face, as well as headaches, while trying to keep his tongue up to the roof of the mouth.



PRE-OP: Cone beam CT showing ***low*** tongue posture but adequate tongue space and posterior airway space (13-18mm)



POST-OP: Cone beam CT showing ***high*** resting tongue posture

“Check out my new tongue rest position, took this CT today, 3 days post op, could be a good before and after for you. Lost a few sutures toward the tip of the tongue and definitely a bit sore but otherwise doing great. Thanks again for a great course and frenuloplasty,” - Jeremy Montrose DMD

Pre-Op	Post-Op
	
	

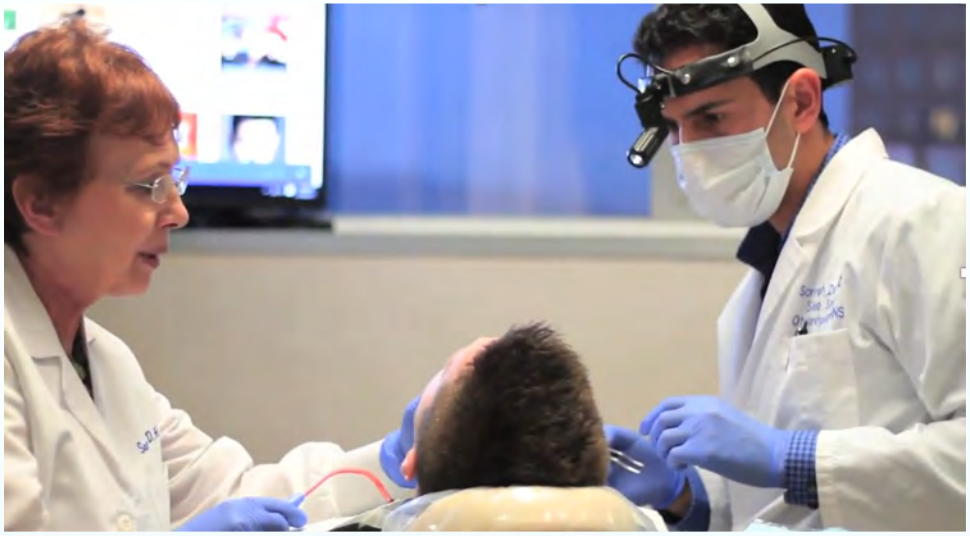
Summary of Literature Review

- 1972: Discovery of OSA in Adults
- 1976: First descriptions of pediatric OSA
- 1977-80: Use of Tracheostomy for adult OSA
- 1981: Introduction of CPAP for adult OSA
- 1995: CPAP for Pediatric OSA
- 2001: Pediatric OSA different than adult OSA.
- 2004: T&A alone is not sufficient for treatment.
- 2011: Tonsils and orthodontics also not sufficient.
- 2015: Tonsils, orthodontics, frenectomy still not sufficient.
- 2013-15: Myofunctional therapy and nasal breathing re-education as the ultimate missing link.

Summary of Literature Review

- 2013-15: Myofunctional therapy and nasal breathing re-education as the ultimate missing link.
- 2016-17: Short lingual frenulum is very common in pediatric and adult OSA.
- 2016-7: Short lingual frenulum is a risk factor for high arched narrow palate.
- 2018-9: Frenuloplasty + myofunctional therapy to optimize outcomes.
- 2019-20: Patient selection and modifying treatment protocol to further improve success rate and reduce risks /complications.

Why we do it → ***How*** we do it



Part 3: Functional Approach To Frenuloplasty

Frenuloplasty: **Do it right the first time…**

59 year-old female with a long history of headaches, chronic sinus infections, forward head posture, jaw tension, and cervical neck discomfort.

- Prior “incomplete” frenectomy at age 7.
- Maxillary expansion that was completed 3-4 years ago→ resolved her sinus infections.
- Now presenting with worsening sleep quality over the last few years and especially the past 4-5 months.


'TONGUE –TIE' RISK
TO MOM & BABY?!
THE DRS

Thank you!

Soroush Zaghi, MD
Otolaryngology (ENT) - Sleep Surgeon
Nasal Breathing, Snoring, and Sleep Apnea
Tongue-Tie and Maxillofacial Development

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