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The Guilleminault (CG) Triad: Ankyloglossia, Mouth Breathing, and High-Arched Palate in Sleep-Disordered Breathing

Garrett Sherman
Southern California Ear
Max Beranek
Fabian Peralta
Rebecca Maginot
Nina Yoshpe
ninayoshpe@gmail.com

Southern California Ear

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The Guilleminault (CG) Triad: Ankyloglossia, Mouth Breathing, and High-Arched Palate in Sleep-Disordered Breathing

Authors

Garrett Sherman1, Max Beranek1, Fabian Peralta2, Rebecca Maginot3, Nina Yoshpe1,*

Affiliations

1 Southern California Ear, Nose, and Throat, Long Beach, CA, USA

2 Facultad de Estomatología, Universidad Peruana Cayetano Heredia, Lima, Peru

3 [Affiliation for Rebecca Maginot — please provide full institutional name, city, country]

Corresponding author

Nina Yoshpe

Southern California Ear, Nose, and Throat

433 E Wardlow Rd, Long Beach, CA 90807, USA

Email: Ninayoshpe@gmail.com

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Abstract

Obstructive sleep apnea is a highly prevalent sleep disorder with substantial health and economic consequences. Dr. Christian Guilleminault (1938–2019), who coined the term "obstructive sleep apnea," championed an interdisciplinary, prevention-focused approach to sleep-disordered breathing and, throughout his career, repeatedly linked three anatomical findings — ankyloglossia, mouth breathing, and high-arched palate — to sleep-disordered breathing. This review honors his contribution by consolidating that recurring association into a single, easily recognized clinical construct that we term the "CG Triad." We conducted a bibliometric review of Dr. Guilleminault's publications in the Scopus and PubMed databases, using his author identifier combined with the search terms "ankyloglossia," "mouth breathing," and "high-arched palate," and extracted article counts and citation metrics for each condition. Fifty-nine relevant articles were identified — six addressing ankyloglossia, fifty-two addressing mouth breathing, and seven addressing high-arched palate — with substantial overlap across the three conditions. His work spanned the validation of tongue-mobility grading scales, the description of the short lingual frenulum as a phenotype for pediatric obstructive sleep apnea, the consequences of mouth breathing and the central role of myofunctional reeducation, and the craniofacial characterization of the high, narrow palate together with expansion therapies, including rapid maxillary expansion and distraction osteogenesis maxillary expansion. Across five decades, his research consistently connected ankyloglossia, mouth breathing, and high-arched palate to sleep-disordered breathing and emphasized early identification and prevention beginning in childhood. Formalizing these three findings as the "CG Triad" offers clinicians across disciplines — pediatricians, dentists, orthodontists, otolaryngologists, and others — a simple, memorable tool for the early recognition

and prevention of obstructive sleep apnea, reflecting the multidisciplinary philosophy that defined Dr. Guilleminault's career.

Keywords Sleep apnea, Mouth breathing, Ankyloglossia, High-arched palate, Sleep, Orofacial development

Background

One of the most prevalent sleep disorders is obstructive sleep apnea (OSA), first described in 1975 by Dr. Christian Guilleminault (affectionately known to his colleagues as "CG"). OSA causes sleep disruption through airflow cessation and oxygen desaturation; when this phenomenon is paired with excessive daytime somnolence, it is classified as obstructive sleep apnea syndrome (OSAS).

Fifty years ago it was not understood how profoundly sleep apnea undermines an individual's ability to obtain restorative sleep. We now know that among adults aged 30–60 years, 24% of men and 9% of women meet criteria for OSA (Young et al. 1993), and the prevalence of OSAS is approximately 3–7% in men and 2–5% in women (Punjabi 2008). Advancing age and obesity further increase the number of cases. Beyond the suffering it causes, OSA is responsible for substantial economic losses stemming from reduced productivity, healthcare costs, and motor vehicle collisions; crashes attributable to OSA alone are estimated to cost $15.9 billion annually in the United States (Sassani et al. 2004). In this population, cardiovascular morbidity and mortality are exceedingly high (Somers et al. 2008). Given these grave consequences, dedicated and innovative specialists are essential to the field — and no specialist has done more to advance it than Dr. Christian Guilleminault.

Dr. Guilleminault was born in Marseilles, France, in 1938. He married Priscilla Grevert and had two sons, Eric and Damian. He was trained in neurology and psychiatry after graduating from the University of Paris and Geneva. He established the first sleep clinic in France in 1970 and joined Stanford University as a visiting assistant professor in 1972, becoming permanent staff in 1980. Raised in a home full of books, he was always drawn to the study of human evolution and to "helping people." He believed strongly that prevention leads to good health, often stating that "problems in sleep will cause problems in wakefulness." He was a keen observer of his patients and encouraged his trainees to pursue their own interests, reserving nights and weekends for research. Dr. Guilleminault passed away from metastatic prostate cancer in 2019, at the age of 80 (Erickson 2019).

At Stanford he received the "Unsung Hero Award" in 2017 as a constant source of inspiration and education to faculty and patients alike. He viewed sleep medicine as a team endeavor, describing trainees, fellows, clinicians, and researchers as members of an "orchestra," and he recognized the need for multidisciplinary involvement from the program's inception in 1972. He was widely regarded as an ideal clinician-scientist; "his intellectual pursuit of science was unmatched and unwavering," according to his colleague Dr. Michelle Cao **[citation to confirm]**.

Dr. Guilleminault authored or co-authored 818 documents over his career. Among his accomplishments, he coined the term "obstructive sleep apnea," co-founded *Sleep*, the official journal of the Sleep Research Society, and helped describe the apnea–hypopnea index (Erickson 2019). He conducted groundbreaking sleep apnea research with Dr. William Dement, was a founding member of the Association of Sleep Disorders Centers in 1975, and served as the first editor of *Sleep* in 1976. Dr. Dement observed that "no one has done as much in the field of sleep medicine," adding: "We worked to make sleep disorders medicine a legitimate clinical specialty,

presented courses for practicing physicians, established reimbursements for sleep testing, and worked hard to bring narcolepsy and sleep apnea to the forefront of sleep medicine practice. I feel extremely fortunate that he chose Stanford" (Erickson 2019).

It is important to keep the memory and contributions of this pioneer alive for future generations of healthcare providers, and to build upon his work. It is the aim of this paper to honor Dr. Guilleminault's contribution to sleep medicine by proposing a new triad in his name. After stringent review of his literature, three physical findings recur prominently: ankyloglossia, mouth breathing, and high-arched palate. Patients presenting with these findings may later be diagnosed with OSA. The contents of a selection of his works are discussed below to give greater context to this breakthrough research and to the proposed triad, which can serve all specialties as a simple tool for identifying anatomy associated with possible sleep-disordered breathing and for preventing its comorbidities.

Unifier of disciplines

Dr. Guilleminault was especially interested in the non-obese patient with sleep apnea, which he argued early on has a genetic component, even though no single causative gene has been identified. He recognized that the obese patient with a large neck and abdomen is easy to identify, whereas the thin patient is more easily missed. He emphasized the role of family history in the early development of sleep apnea, describing OSA as "the end of the road" (Guilleminault et al. 1995). Later in his career he focused on prevention as a means of addressing sleep apnea. He recognized that early identification often falls to the pediatrician or primary care physician — clinicians who are frequently not trained for this task — and argued that all specialties, including the pediatric dentist, otolaryngologist, cardiologist, pulmonologist, neurologist, oral surgeon, and orthodontist, must work together to fully serve these patients. The triad of ankyloglossia, high-

arched palate, and mouth breathing is readily recognizable across specialties and can facilitate the early identification of sleep disorders at any age.

Dr. Guilleminault's career was defined by clinical research and by his encouragement of a unified, cross-disciplinary response to sleep disorders. This is seen especially in his extensive research linking ankyloglossia, mouth breathing, and high-arched palate to OSA. The "CG Triad" — the grouping of these three conditions as an indicator of possible sleep-disordered breathing — is drawn directly from the cross-disciplinary connections evident throughout his writings, and provides an easy-to-remember tool for identifying the possibility of OSA.

Mouth breathing is a condition in which a patient relies on air intake through the mouth rather than the nose (Figure 1). A number of conditions can cause mouth breathing, including enlarged tonsils, enlarged adenoids, facial structure, and a deviated septum. Long-term complications include malocclusion and OSA. Dr. Guilleminault wrote extensively about the persistence of mouth breathing following tonsillectomy and adenoidectomy, recommending post-operative assessment to confirm that patients have resumed nasal breathing (Torre and Guilleminault 2018).

Ankyloglossia (tongue-tie) is a congenital anomaly that restricts movement of the tongue owing to a short lingual frenulum (Figure 2). It may result in difficulty breastfeeding in infants, as well as mouth breathing, orthodontic problems, sleep-disordered breathing, and OSA.

High-arched palate (high, narrow palate) is a condition in which the curvature above the palatal midline is abnormally pronounced; its exact root cause is unknown (Figure 3). It is associated with numerous dental conditions and with OSA.

Methods

A search for articles was carried out using the PubMed and Scopus databases for all publications by Dr. Guilleminault (alone or with co-authors) addressing ankyloglossia, mouth breathing, or high-arched palate.

Ankyloglossia

In Scopus, the Analyze author output was searched with the free term "ankyloglossia," and Boolean operators ("OR," "AND") were applied. The query (AU-ID("Guilleminault, Christian G." 35415169100) AND ankyloglossia) returned 5 articles. A parallel PubMed search ("Guilleminault C" AND "ankyloglossia") returned 4 articles, for a working total of 5 articles on this topic.

Mouth breathing

In Scopus, the Analyze author output was searched with the free term "mouth breathing," and Boolean operators were applied. The query (AU-ID("Guilleminault, Christian G." 35415169100) AND (mouth AND breathing)) returned 46 articles. A parallel PubMed search ("Guilleminault C" AND "mouth breathing") returned 22 articles, for a working total of 52 articles on this topic.

High-arched palate

In Scopus, the Analyze author output was searched with the free term "high-arched palate," and Boolean operators were applied. The query (AU-ID("Guilleminault, Christian G." 35415169100) AND (high-arched AND palate)) returned 7 articles. A parallel PubMed search ("Guilleminault C" AND "high-arched palate") returned 4 articles, for a working total of 7 articles on this topic.

After analysis across the three topics of tongue-tie, mouth breathing, and high-arched palate, a total of 59 articles were obtained.

Results

Ankyloglossia

Dr. Guilleminault authored or co-authored 6 articles related to ankyloglossia. The most-cited was "A frequent phenotype for pediatric sleep apnoea: short lingual frenulum" (Guilleminault et al. 2016), with 48 citations; the mean number of citations across the related documents was 33. These articles addressed the validation of grading scales for ankyloglossia and tongue mobility, the relationship of ankyloglossia to maxillofacial morphological development, and effective, safe treatments for the condition.

Mouth breathing

Regarding mouth breathing, Dr. Guilleminault produced 35 articles, 1 book, 3 book chapters, 2 editorials, 1 note, and 4 reviews. The most-cited work was "Sleepwalking and sleep terrors in prepubertal children: what triggers them?" (Guilleminault et al. 2003), with 327 citations; the mean number of citations across the 52 related documents was 54. Of these, 9 documents specifically discuss mouth breathing, focusing mainly on etiology, consequences, and the importance of early treatment in at-risk populations.

High-arched palate

Dr. Guilleminault produced 6 articles and 1 review regarding high-arched palate. The most-cited was "Rapid maxillary expansion for pediatric obstructive sleep apnea: a systematic review and meta-analysis" (Camacho et al. 2017), with 117 citations; the mean number of citations across the 7 related documents was 43. Of the 7 documents, 6 primarily describe high-arched palate as a morphological alteration strongly associated with OSA, emphasizing the effect of treatment and

the benefits of early intervention. Nineteen additional articles reviewed overlapped with the mouth-breathing and ankyloglossia documents.

Discussion

Ankyloglossia

Ankyloglossia (tongue-tie) is a condition in which the lingual frenulum is abnormally short. It can be asymptomatic; when symptomatic, it is associated with difficulties in sucking, swallowing, and speech, and with sleep-disordered breathing in children.

Dr. Guilleminault contributed to 5 articles on this condition. He collaborated on a study comparing measurement scales for classifying ankyloglossia against tongue mobility; in 1,052 subjects, the tongue range of motion ratio (TRMR) was found to be the best first-line tool (grade 1: TRMR > 80%; grade 2: 50–80%; grade 3: < 50%; grade 4: < 25%) (Yoon et al. 2017a). He retrospectively studied the relationship between OSA and ankyloglossia in adults (Chien and Guilleminault 2017), showing that limited tongue movement may be closely related to the development of a high-arched palate, maxillary crossbite, small mandible, and overbite; adults with a short lingual frenulum showed an increased incidence of deviated nasal septum and high-arched palate, predisposing them to crossbite and/or overbite (Yoon et al. 2017b).

He described the lingual frenulum as a “phenotype for sleep apnea” in his 2016 article (Guilleminault et al. 2016), a pivotal contribution positioning tongue-tie as a potential marker for sleep apnea. His subsequent studies grew increasingly specific in identifying the morphological changes associated with tongue-tie, exploring a possible relationship between ankyloglossia and both a longer soft palate and alterations of the hyoid bone, and confirming the association

between ankyloglossia and conditions such as high-arched palate and maxillary hypoplasia (Yoon et al. 2017b).

Finally, Dr. Guilleminault and colleagues reviewed the efficacy and safety of treatment, concluding that the combination of lingual frenuloplasty and myofunctional therapy is the most effective and safe long-term therapy for this condition. This study was published after his death (Zaghi et al. 2019).

Mouth breathing

The nose warms, humidifies, and filters inspired air; breathing through the mouth bypasses these functions. Mouth breathing may occur with nasal obstruction and in patients with skeletal facial abnormalities, and has been linked to behavioral disorders, lower-airway and asthma morbidity, distortions of jaw growth, and dental malocclusion. Under unbalanced muscular forces the maxilla narrows in width; as it narrows, the nasal cavity narrows concomitantly, causing septal deviation and increased nasal resistance. Dr. Guilleminault emphasized that the palate is the floor of the nose — if it is narrow, the nostrils are narrow and the septum may be deviated.

He collaborated on many studies examining how mouth breathing accompanies OSA and related disturbances such as snoring, sleep-disordered breathing, clenching, and myofascial tension. Much of his research examined treatments that increase or decrease mouth breathing, and he was a particular advocate of myofunctional therapy to recondition patients to breathe through the nose (Guilleminault et al. 2013a; Lee et al. 2015).

In 1998, Dr. Guilleminault described how mouth breathing may relate to abnormal nasal resistance caused by upper-airway allergies, infection, and enlarged tonsils and adenoids, noting even in his early work that these problems are often inherited, particularly in patients with a

high-arched palate (Guilleminault and Pelayo 1998). He explored how mouth breathing relates to orthodontic change, positing a close relationship among mouth breathing, tongue position, and tonsillar enlargement, and hypothesizing a self-reinforcing cycle in which mouth breathing inflames the tonsils, enlarged tonsils alter tongue position, and altered tongue position perpetuates mouth breathing.

In children with Ehlers–Danlos syndrome, he demonstrated that mouth breathing secondary to a high-arched palate aggravates orofacial growth, advocating early treatment with careful evaluation for sleep apnea (Guilleminault et al. 2013b). He emphasized both the consequences of mouth breathing and the importance of myofascial reeducation, showing that without such reeducation there is a greater probability of recurrence of sleep-related disorders (Guilleminault et al. 2013a). He also studied premature infants at 6 months post-natal age, showing that premature babies are at greater risk of developing sleep-related breathing problems — possibly a consequence of increased nasal resistance and poor muscle tone (Huang et al. 2014). He was deeply concerned about sudden infant death syndrome and devoted many projects to this problem.

Dr. Guilleminault recognized that many mouth-breathing problems can be addressed with adenotonsillectomy; however, his research emphasized the importance of long-term follow-up, as many children continue to mouth-breathe post-operatively. Because sucking, mastication, swallowing, and nasal breathing are all related to future craniofacial growth, mouth breathing may return after surgery unless myofunctional therapy is initiated (Lee et al. 2015; Guilleminault and Akhtar 2015). He stressed the importance of re-establishing nasal breathing after tonsillectomy and adenoidectomy — a goal sometimes overlooked once snoring and other symptoms resolve (Torre and Guilleminault 2018).

High-arched palate (high, narrow hard palate, HNP)

In many of his studies of patients with OSA, Dr. Guilleminault described the morphological changes associated with the high-arched palate. He found that such patients frequently presented with retroposition of the mandible, a more acute nasion–sella–basion cranial base angle than expected, and inferior displacement of the hyoid bone (Jamieson et al. 1986).

He used these morphological findings to aid screening of patients with sleep apnea. While high BMI and neck circumference are readily identified, he emphasized that changes in the dimensions of the oral cavity in non-obese subjects — particularly the palatal arch and mandibular size — are critical considerations that clinicians frequently miss (Kushida et al. 1997). Integrating these facial and palatal findings, he described "long face syndrome," characterized by a long, narrow facial appearance, retrognathia, micrognathia, and a high, narrow palate, in *Neurologic Clinics* in 1996 (Chervin and Guilleminault 1996). (The related entity "adenoid facies" was described earlier by Meyer [1868], Tomes [1872], and Subtelny [1954] **[full references needed if these historical sources are to be cited]**.)

With M. Camacho and colleagues, Dr. Guilleminault studied treatment of the high-arched palate, showing improvement in morphological changes and OSA symptoms: rapid maxillary expansion in children improved the apnea–hypopnea index and low oxygen saturation in short-term follow-up (< 3 years), with longer-term benefits (> 3 years) largely reflecting growth and spontaneous resolution of OSA (Camacho et al. 2017). His group also evaluated distraction osteogenesis maxillary expansion (DOME), reporting the safety and efficacy profile of the first 20 adult patients treated at Stanford and finding significant reductions in the Epworth Sleepiness Scale, the Nasal Obstruction Symptom Evaluation, the apnea–hypopnea index, and the oxygen desaturation index in short-term follow-up (8 months) (Liu et al. 2017).

These patients were subsequently reviewed, showing reduced nasal airflow velocity and a downstream reduction of negative pressure in the pharyngeal airway (Iwasaki et al. 2020). The results of DOME led to its incorporation into the OSA treatment algorithm in 2019: by widening the palate — the floor of the nose — DOME increases the internal nasal valve angle and decreases nasal resistance in adults (Iwasaki et al. 2020). Dr. Guilleminault was also included in studies demonstrating the impact of DOME on patients with a narrow maxilla and nasal floor, with reductions in the severity of OSA, refractory nasal obstruction, and daytime somnolence and an increase in the percentage of REM sleep (Yoon et al. 2020; Liu et al. 2019).

Conclusions

Dr. Christian Guilleminault had a profound impact on the field of sleep medicine. As demonstrated across his publications, there is a significant association between ankyloglossia, mouth breathing, and high-arched palate and patients with sleep-disordered breathing. He showed that ankyloglossia may be a principal cause of limited tongue mobility, contributing to the high-arched formation of the palate and, ultimately, to upper-airway collapse and sleep-disordered breathing. He was deeply committed to early identification beginning with the pediatrician. The proposed triad is straightforward and easy for all clinicians — pediatricians, internists, dentists, orthodontists, neurologists, cardiologists, and speech therapists — to recognize.

Dr. Guilleminault thoroughly explored each of these conditions, proposing effective and safe methods of prevention, diagnosis, and treatment. He recognized the value of a multidisciplinary approach to the early identification of sleep disorders, encouraging all disciplines to work together as an “orchestra.” Through the knowledge he left us, new research directions can be

explored to transform how we evaluate patients with any of these conditions. To promote early, effective, and safe treatment for all patients at risk of developing the CG Triad, prevention and early identification are crucial. Sleep medicine is fortunate to have had such a passionate pioneer, whose work continues to stimulate research, education, and treatment worldwide.

The Sleep Research Society held its first meetings in Chicago in 1961, with Drs. Nathaniel Kleitman, Allan Rechtschaffen, and William Dement (Shepard et al. 2005). Although Dr. Guilleminault was not present at that meeting, his arrival at Stanford in 1972 began to stimulate worldwide interest in sleep medicine. He was honored by societies internationally, receiving the Distinguished Scientist Award of the Sleep Research Society and the Lifetime Achievement Award of the National Sleep Foundation (both 2005), the AASM Excellence in Research Award (1995), the William C. Dement Academic Achievement Award of the American Sleep Disorders Association (1995), the President's Award of the American Board of Sleep Medicine (1991), the Nathaniel Kleitman Award (1986), the Annenberg Award for his contributions to SIDS and apnea research (1999), and the Collegium Internationale Neuro-Psychopharmacologicum Award for Research (1996), among many others. He was nominated for a Nobel Prize in 2002. Two awards bear his name: the CG Young Investigator Endowment Fund and the Christian Guilleminault Award for Research in Sleep Medicine of the World Association of Sleep Medicine. Through his work and the legacy of the many physicians, dentists, orthodontists, speech therapists, and respiratory therapists he inspired, Dr. Guilleminault richly deserves this honor.

List of abbreviations

AASM: American Academy of Sleep Medicine

AHI: apnea–hypopnea index

BMI: body mass index

CG: Christian Guilleminault

DOME: distraction osteogenesis maxillary expansion

HNP: high, narrow palate

ODI: oxygen desaturation index

OSA: obstructive sleep apnea

OSAS: obstructive sleep apnea syndrome

REM: rapid eye movement

SDB: sleep-disordered breathing

SIDS: sudden infant death syndrome

SRS: Sleep Research Society

TRMR: tongue range of motion ratio

Declarations

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Not applicable

Consent for publication

Not applicable

Availability of data and materials

All data analyzed during this study are derived from previously published articles indexed in the PubMed and Scopus databases and are cited within this article.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

GS and MB conceived the review and performed the literature search. FP and RM extracted and analyzed the data. NY initiate and finished the manuscript. All authors read and approved the final manuscript.

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Figure

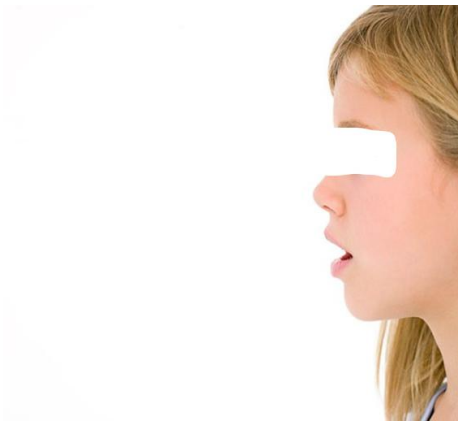


Figure 1. Mouth breathing.





Figure 2. Ankyloglossia (tongue-tie).





Figure 3. High-arched palate (high, narrow palate).