

Managing obstructive sleep apnoea in children: the role of craniofacial morphology

Maria Fernanda Rabelo Bozzini,^{1,*} Renata Cantisani Di Francesco^{II}

^IFaculdade de Medicina da Universidade de São Paulo, São Paulo/SP, Brazil. ^{II}Faculdade de Medicina da Universidade de São Paulo, Departamento de Otorrinolaringologia, São Paulo/SP, Brazil.

Obstructive sleep apnoea syndrome is a type of sleep-disordered breathing that affects 1 to 5% of all children. Pharyngeal and palatine tonsil hypertrophy is the main predisposing factor. Various abnormalities are predisposing factors for obstructive sleep apnoea, such as decreased mandibular and maxillary lengths, skeletal retrusion, increased lower facial height and, consequently, increased total anterior facial height, a larger cranio-cervical angle, small posterior airway space and an inferiorly positioned hyoid bone. The diagnosis is based on the clinical history, a physical examination and tests confirming the presence and severity of upper airway obstruction. The gold standard test for diagnosis is overnight polysomnography. Attention must be paid to identify the craniofacial characteristics. When necessary, children should be referred to orthodontists and/or sleep medicine specialists for adequate treatment in addition to undergoing an adenotonsillectomy.

KEYWORDS: Craniofacial Morphology; Sleep-disordered Breathing; Obstructive Sleep Apnoea Syndrome; Polysomnography; Children.

Bozzini MF, Di Francesco RC. Managing obstructive sleep apnoea in children: the role of craniofacial morphology. *Clinics*. 2016;71(11):664-666

Received for publication on July 12, 2016; Accepted for publication on August 17, 2016

*Corresponding author. E-mail: mfbazzini@gmail.com

INTRODUCTION

Obstructive sleep apnoea syndrome (OSAS) is defined as a type of sleep-disordered breathing (SDB) characterized by partial and/or complete upper airway obstruction that affects normal ventilation (1). Decreased quality of life and behavioural, psychiatric, neurocognitive, cardiovascular, metabolic, endocrine, and growth abnormalities are several of the complications associated with this syndrome (2).

Obstructive sleep apnoea (OSA) is estimated to affect 1 to 5% of all children (3), and its incidence peaks are between 3 and 8 years of age (4). Multiple components are involved in SDB in children. Anatomical, craniofacial and neuromuscular factors, excess lymphoid tissues and airway inflammation are cited as the most critical components (2).

Adenoid and tonsil hypertrophy are more prevalent between 3 and 6 years of age (5), and these enlargements are strongly related to OSAS in children (1). Snoring is one of the most often reported symptoms of SDB in paediatric populations, and its prevalence ranges from 1.5 to 27.6% for different studies and populations (2).

SDB and Craniofacial Morphology

At birth, the face is approximately 40% of the adult size, and it increases to 65% by 3 years of age. Facial growth is only completed after puberty. Facial growth is influenced mainly by genetic factors, but environmental aspects such as the breathing pattern may also contribute to growth (6).

Nasal breathing in children may induce a correction of craniofacial growth and the adequate development of other functions, such as chewing and swallowing (7). To promote breathing in the presence of nasal obstructions, the head and mandible position and the tonicity of the tongue and orofacial muscles are altered. When they persist, these changes modify the equilibrium of muscle pressure on the facial bones and teeth and induce morphological dentoskeletal modifications (8).

Various craniofacial deformities are predisposing factors, including decreased mandibular and maxillary lengths, skeletal retrusion, increased lower facial height and, consequently, increased total anterior facial height, a larger cranio-cervical angle, decreased posterior airway space and an inferiorly positioned hyoid bone (1,5,8-13).

A positive correlation exists between the severity of the breathing pattern as measured by the apnoea and hypopnoea index and changes in dentofacial development. Dental arch abnormalities can be explained by long-term changes in the head, mandible and tongue positions (10). The most common dental and skeletal alterations are transverse maxillary deficiency, open bite, large overjet, retro-inclined lower incisors and protruding upper incisors (9). The prevalence of posterior crossbite is significantly more frequent in

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0/>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

No potential conflict of interest was reported.

DOI: 10.6061/clinics/2016(11)08



mouth breathers (49%) than in nose breathers (26%) (14), and children with OSA commonly have shorter inter-molar and inter-canine distances (15).

In a recent meta-analysis, Katyal (16) stated that "there is no evidence for a direct causal relationship between craniofacial structure and paediatric SDB and there is lack of intensive evidence on the critical degree of obstruction, or how long it may occur to affect growth".

Diagnosis

Children with OSAS often have cognitive problems, disturbed sleep, delayed learning, social performance disorders, bruxism and enuresis. The diagnosis is based on clinical history, a physical examination and tests confirming the presence and severity of the obstruction (9).

The gold standard for the diagnosis is overnight polysomnography, which is currently the only reliable diagnostic modality that can discern SDB from primary snoring (13). Craniofacial anatomy plays a role in OSAS, and screening for anatomical abnormalities is an important tool in its diagnosis.

Advanced technologies, such as computed tomography and magnetic resonance imaging, are currently used to evaluate the anatomical characteristics of the upper airway and craniofacial structures. However, the traditional cephalometric method is the most practical technique and is frequently used in clinical practice due to its easy access, low cost and low radiation exposure (11).

One misleading problem in assessing craniofacial morphology using a lateral cephalogram is that patients are conscious, in an upright position and have occluded teeth. Paediatric SDB typically worsens in the supine position when muscle hypotony occurs while sleeping. Further investigations are required to address the relationship between craniofacial morphology and paediatric SDB in all three dimensions (16).

Treatment

The treatment is based upon the severity of obstructive apnoea. The practice guidelines established by the American Academy of Pediatricians proposes an adenotonsillectomy as the first line of therapy for childhood OSAS (4,17,18). Several meta-analyses have reported that an adenotonsillectomy leads to significant improvements in most cases of paediatric OSAS (4,19); however, the success rates are highly variable and range from 24 to 100% (20-22).

Changing the breathing pattern from oral to nasal early in adolescence may benefit the craniofacial dimensions during growth (23). Previous studies have provided evidence for quality of life gains promptly after surgery, mainly for patients with more severe obstructive sleep disorders, and this effect is not significantly affected by gender, age or adiposity (24). An adenotonsillectomy can also efficiently improve several dental malocclusions, which benefits patients during their growth phase. In addition, this technique is mandatory for proper occlusal development (25).

However, the structural problems in many children with SDB are not fully solved after an adenotonsillectomy, and a risk exists for a slow but progressive reappearance of OSA symptoms (21). An adenotonsillectomy is associated with significant improvements in most children, but approximately 20 to 30% may exhibit residual symptoms; this percen-

tage can increase to 70% in patients with a high apnoea-hypopnoea index on preoperative polysomnography (4,17).

The recurrence is typically not immediate and may occur during the pubertal period and after the mean age at which dentoskeletal development reaches approximately 90% of its adult final growth (26). Mouth-breathing children may exhibit a dolichofacial pattern due to the mandibular plane inclination, the gonial angle and the increased posterior facial height (27). The presence of residual OSA in a large number of patients following an adenotonsillectomy is a significant public health concern and calls for further studies focusing on individual patient factors that may predict the success rates and additional therapies that may improve surgery results in these patients (19).

These findings raise important issues regarding the efficacy of an adenotonsillectomy and further suggest that overnight polysomnography should be routinely implemented (19-20). The primary justification for surgery must be to open the airways such that physiological nose breathing is possible. If the clinical follow-up after surgery shows that the child maintains mouth breathing, it is important to examine the child for nasal congestion, including septal deviations or allergic rhinitis. Adjuvant therapy, such as orthodontic maxillary expansion and/or functional training, must also be considered (28). OSA appears to be more severe in boys than in girls due to the craniofacial morphology, which may be relevant for preventing sleep apnoea in adult males. The long-term follow-up of OSA in boys is mandatory for preventing the disease in adulthood (29).

Given the impact on a child's health, determination of these predictor factors is important to prevent and manage SDB. If a health professional notices signs and symptoms SDB, the young patient should be referred to a sleep medicine specialist and to an orthodontist if dentoskeletal abnormalities are present (30).

In adults, continuous positive airway pressure therapy is the first line of treatment for patients with OSAS. This procedure prevents upper airway collapse and relieves symptoms but has poor compliance. An alternative is maxillo-mandibular advancement, which enlarges the pharyngeal space and is often performed with genioglossus advancement to decrease the amount of tongue blockage during sleep (31). The success rates and long-term stability of outcomes confirm the efficacy of maxillomandibular advancement; however, continuous follow-up is necessary to control patient lifestyle and detect possible relapses (32). The choice of this surgery should follow the failure of other treatment procedures because a longer recovery period is necessary than for other options.

Further standardization of research methods is recommended, including the establishment and acceptance of valid definitions for normal breathing, SDB and OSA (16). Paediatricians and dentists should always ask parents about their young child's nightly breathing/snoring and advise them to seek appropriate help for all children who snore and/or breathe orally.

Craniofacial morphology plays an important role in SDB in children. It is also associated with the recurrence of snoring and OSA after an adenotonsillectomy at later ages, including in teenagers and adults. Attention must be paid to identify these characteristics. When necessary, children should be referred to orthodontists and/or sleep medicine specialists for adequate treatment in addition to undergoing an adenotonsillectomy.



AUTHOR CONTRIBUTIONS

Authors make substantial contributions to conception and design of this manuscript. Bozzini MF participated in drafting and writing this review article and together with Di Francesco RC revised it critically for important intellectual content. Both authors approved the final version of the manuscript to be submitted.

REFERENCES

1. Marino A, Malagnino I, Ranieri R, Villa MP, Malagola C. Craniofacial morphology in preschool children with obstructive sleep apnoea syndrome. *Eur J Paediatr Dent*. 2009;10(4):181-4.
2. Nespoli L, Caprioglio A, Brunetti L, Nosetti L. Obstructive sleep apnea syndrome in childhood. *Early Hum Dev*. 2013;89(Suppl 3):S33-7.
3. Goldbart AD, Tal A. Inflammation and sleep disordered breathing in children: a state-of-the-art review. *Pediatr Pulmonol*. 2008;43(12):1151-60, <http://dx.doi.org/10.1002/ppul.20943>.
4. Brietzke SE, Gallagher D. The effectiveness of tonsillectomy and adenoidectomy in the treatment of pediatric obstructive sleep apnea/hypopnea syndrome: a meta-analysis. *Otolaryngol Head Neck Surg*. 2006;134(6):979-84, <http://dx.doi.org/10.1016/j.otohns.2006.02.033>.
5. Zettergren-Wijk L, Forsberg CM, Linder-Aronson S. Changes in dentofacial morphology after adeno-/tonsillectomy in young children with obstructive sleep apnoea—a 5-year follow-up study. *Eur J Orthod*. 2006;28(4):319-26, <http://dx.doi.org/10.1093/ejo/cji119>.
6. Katz ES, D'Ambrosio CM. Pathophysiology of pediatric obstructive sleep apnea. *Proc Am Thorac Soc*. 2008;5(2):253-62, <http://dx.doi.org/10.1513/pats.200707-111MG>.
7. Sousa JB, Anselmo-Lima WT, Valera FC, Gallego AJ, Matsumoto MA. Cephalometric assessment of the mandibular growth pattern in mouth-breathing children. *Int J Pediatr Otorhinolaryngol*. 2005;69(3):311-7, <http://dx.doi.org/10.1016/j.ijporl.2004.10.010>.
8. Pirilä-Parkkinen K, Löppönen H, Nieminen P, Tolonen U, Pirttiniemi P. Cephalometric evaluation of children with nocturnal sleep-disordered breathing. *Eur J Orthod*. 2010;32(6):662-71, <http://dx.doi.org/10.1093/ejo/cjp162>.
9. Juliano ML, Machado MA, de Carvalho LB, Zancanella E, Santos GM, do Prado LB, et al. Polysomnographic findings are associated with cephalometric measurements in mouth-breathing children. *J Clin Sleep Med*. 2009;5(6):554-61.
10. Pirilä-Parkkinen K, Pirttiniemi P, Nieminen P, Tolonen U, Pelttari U, Löppönen H. Dental arch morphology in children with sleep-disordered breathing. *Eur J Orthod*. 2009;31(2):160-7, <http://dx.doi.org/10.1093/ejo/cjn061>.
11. Bharadwaj R, Ravikumar A, Krishnaswamy NR. Evaluation of craniofacial morphology in patients with obstructive sleep apnea using lateral cephalometry and dynamic MRI. *Indian J Dent Res*. 2011;22(6):739-48, <http://dx.doi.org/10.4103/0970-9290.94566>.
12. Tsuda H, Fastlicht S, Almeida FR, Lowe AA. The correlation between craniofacial morphology and sleep-disordered breathing in children in an undergraduate orthodontic clinic. *Sleep Breath*. 2011;15(2):163-71, <http://dx.doi.org/10.1007/s11325-010-0345-4>.
13. Vieira BB, Itikawa CE, de Almeida LA, Sander HS, Fernandes RM, Anselmo-Lima WT, et al. Cephalometric evaluation of facial pattern and hyoid bone position in children with obstructive sleep apnea syndrome. *Int J Pediatr Otorhinolaryngol*. 2011;75(3):383-6, <http://dx.doi.org/10.1016/j.ijporl.2010.12.010>.
14. Harari D, Redlich M, Miri S, Hamud T, Gross M. The effect of mouth breathing versus nasal breathing on dentofacial and craniofacial development in orthodontic patients. *Laryngoscope*. 2010;120(10):2089-93, <http://dx.doi.org/10.1002/lary.20991>.
15. Esteller Moré E, Pons Calabuig N, Romero Vilariño E, Puigdollers Pérez A, Segarra Isern F, Matión Soler E, et al. [Dentofacial development abnormalities in paediatric sleep-related breathing disorders]. *Acta Otorrinolaringol Esp*. 2011;62(2):132-9, <http://dx.doi.org/10.1016/j.otorri.2010.10.007>.
16. Katyal V, Pamula Y, Martin AJ, Daynes CN, Kennedy JD, Sampson WJ. Craniofacial and upper airway morphology in pediatric sleep-disordered breathing: Systematic review and meta-analysis. *Am J Orthod Dentofacial Orthop*. 2013;143(1):20-30, <http://dx.doi.org/10.1016/j.ajodo.2012.08.021>.
17. Schechter MS, Section on Pediatric Pulmonology, Subcommittee on Obstructive Sleep Apnea Syndrome. Technical report: diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics*. 2002;109(4):e69, <http://dx.doi.org/10.1542/peds.109.4.e69>.
18. Fitzgerald NM, Fitzgerald DA. Managing snoring and obstructive sleep apnoea in childhood. *J Paediatr Child Health*. 2013;49(10):800-6, <http://dx.doi.org/10.1111/jpc.12421>.
19. Friedlman M, Wilson M, Lin HC, Chang HW. Updated systematic review of tonsillectomy and adenoidectomy for treatment of pediatric obstructive sleep apnea/hypopnea syndrome. *Otolaryngol Head Neck Surg*. 2009;140(6):800-8, <http://dx.doi.org/10.1016/j.otohns.2009.01.043>.
20. Tauman R, Gulliver TE, Krishna J, Montgomery-Downs HE, O'Brien LM, Ivanenko A, et al. Persistence of obstructive sleep apnea syndrome in children after adenotonsillectomy. *J Pediatr*. 2006;149(6):803-8, <http://dx.doi.org/10.1016/j.jpeds.2006.08.067>.
21. Guilleminault C, Huang YS, Glamann C, Li K, Chan A. Adenotonsillectomy and obstructive sleep apnea in children: a prospective survey. *Otolaryngol Head Neck Surg*. 2007;136(2):169-75, <http://dx.doi.org/10.1016/j.otohns.2006.09.021>.
22. Bhattacharjee R, Kheirandish-Gozal L, Spruyt K, Mitchell RB, Promchiarak J, Simakajornboon N, et al. Adenotonsillectomy outcomes in treatment of obstructive sleep apnea in children: a multicenter retrospective study. *Am J Respir Crit Care Med*. 2010;182(5):676-83, <http://dx.doi.org/10.1164/rccm.200912-1930OC>.
23. Cuccia AM, Lotti M, Caradonna D. Oral breathing and head posture. *Angle Orthod*. 2008;78(1):77-82, <http://dx.doi.org/10.2319/011507-18.1>.
24. Lee CH, Kang KT, Weng WC, Lee PL, Hsu WC. Quality of life after adenotonsillectomy for children with sleep-disordered breathing: a linear mixed model analysis. *Int J Pediatr Otorhinolaryngol*. 2014;78(8):1374-80, <http://dx.doi.org/10.1016/j.ijporl.2014.05.038>.
25. Pereira SR, Bakor SF, Weckx LL. Adenotonsillectomy in facial growing patients: spontaneous dental effects. *Braz J Otorhinolaryngol*. 2011;77(5):600-4, <http://dx.doi.org/10.1590/S1808-86942011000500011>.
26. Guilleminault C, Huang YS, Quo S, Monteyrol PJ, Lin CH. Teenage sleep-disordered breathing: recurrence of syndrome. *Sleep Med*. 2013;14(1):37-44, <http://dx.doi.org/10.1016/j.sleep.2012.08.010>.
27. Mattar SE, Valera FC, Faria G, Matsumoto MA, Anselmo-Lima WT. Changes in facial morphology after adenotonsillectomy in mouth-breathing children. *Int J Paediatr Dent*. 2011;21(5):389-96, <http://dx.doi.org/10.1111/j.1365-263X.2011.01117.x>.
28. Löfstrand-Tideström B, Hultcrantz E. Development of craniofacial and dental arch morphology in relation to sleep disordered breathing from 4 to 12 years. Effects of adenotonsillar surgery. *Int J Pediatr Otorhinolaryngol*. 2010;74(2):137-43, <http://dx.doi.org/10.1016/j.ijporl.2009.10.025>.
29. Di Francesco R, Monteiro R, Paulo ML, Buranello F, Imamura R. Craniofacial morphology and sleep apnea in children with obstructed upper airways: differences between genders. *Sleep Med*. 2012;13(6):616-20, <http://dx.doi.org/10.1016/j.sleep.2011.12.011>.
30. Huynh NT, Morton PD, Rompré PH, Papadakis A, Remise C. Associations between sleep-disordered breathing symptoms and facial and dental morphometry, assessed with screening examinations. *Am J Orthod Dentofacial Orthop*. 2011;140(6):762-70, <http://dx.doi.org/10.1016/j.ajodo.2011.03.023>.
31. Fairburn SC, Waite PD, Vilos G, Harding SM, Bernreuter W, Cure J, et al. Three-dimensional changes in upper airways of patients with obstructive sleep apnea following maxillomandibular advancement. *J Oral Maxillofac Surg*. 2007;65(1):6-12, <http://dx.doi.org/10.1016/j.joms.2005.11.119>.
32. Giarda M, Bruccoli M, Arcuri F, Benec R, Braghiroli A, Benec A. Efficacy and safety of maxillomandibular advancement in treatment of obstructive sleep apnoea syndrome. *Acta Otorhinolaryngol Ital*. 2013;33(1):43-6.