Predictive factors of persistent/recurrent pediatric obstructive sleep apnea-hypopnea syndrome after adenotonsillectomy

Facteurs prédictifs de la persistance/récurrence du syndrome d'apnées-hypopnées obstructives du sommeil de l'enfant après adénoïdo-amygdalectomie

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Pediatric obstructive sleep apnea syndrome (OSAS) is a sleep-breathing disorder condition where a child experiences recurrent episodes of partial or complete upper airway collapse during sleep (1). This leads to intermittent obstructive apnea (ie; cessation of breathing) or obstructive hypopnea (ie; a significant decrease in airflow) despite persisting breathing efforts (1). According to the American Academy of Sleep Medicine, the severity of OSAS is graded based on the obstructive apnea-hypopnea index (OAHI) (mild: OAHI: 1.5 – 5 events/h; moderate: OAHI: 5 – 10 events/h; and severe: OAHI ≥ 10 events/h) (1,2). The prevalence of OSAS in the general population of children is estimated to be between 2% and 5.7% and peaks at 2-6 years of age (3). The most common phenotype of pediatric OSAS, known as phenotype 1, typically affects preschool-age children and is primarily caused by adenotonsillar hypertrophy (4). Inflammatory and infectious processes can trigger the growth of tonsillar or adenoid tissue, resulting in upper airway obstruction, mouth breathing, and subsequent OSAS in young children (5). Phenotype 2 is observed in adolescents and is often linked to obesity (4). Phenotype 3 manifests from early ages of life and is associated with morpho-structural abnormalities, craniofacial changes, or genetic syndromes (4). Children with pediatric OSAS, with or without nasal obstruction, often breathe through their mouths (6). Chronic mouth breathing during the growing period may impair the oral-facial function (including tongue hypotonia), and craniofacial morphology (adenoid facies with posteriorly rotated mandible) (6). These mechanical and functional changes can make the upper airway narrower and more likely to collapse during sleep (6).

In children, untreated or inadequately treated OSAS can lead to various negative outcomes (7). These may include sleep disturbances, daytime sleepiness, behavioral issues, cognitive dysfunction, impaired academic performance, and mood disturbances (8). In more severe cases, it can also be associated with metabolic and cardiovascular abnormalities as well as growth impairments, making it a major public health concern (1). Early recognition, assessment, and treatment can help reduce complications and enhance long-term outcomes (7). When dealing with pediatric OSAS, it is essential to comprehend the diverse and intricate nature of its pathophysiology and adopt a multidisciplinary approach for effective treatment (3). Various treatment options are available, encompassing both surgical and non-surgical interventions (4,9). Surgical options include adenotonsillectomy, which involves the simultaneous removal of the adenoids and tonsils, or the correction of craniofacial anomalies in younger children (4). Tonsil and adenoid hypertrophy are the most common causes of pediatric OSAS, even in cases involving obesity or complex disorders. Therefore, adenotonsillectomy stands out as the first-line surgical treatment for children with moderate to severe OSAS and adenotonsillar hypertrophy (8,10,11). The surgical method used for tonsillectomy can be either total tonsillectomy where the entire tonsil and its surrounding capsule are removed, or partial tonsillectomy where a part of the tonsil or its capsule is left in place (9,12).
Prior research has suggested that adenotonsillectomy can result in significant improvement in functional outcomes and polysomnographic findings for most cases (8,11)However, a technical report including 350 studies has shown that OSAS may persist or relapse in 13% to

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73% of children following adenotonsillectomy(10). A 2023 consensus statement from the American Academy of Otolaryngology-Head and Neck Surgery defined persistent/recurrent OSAS as an OAHI > 1 /hour with symptoms or > 5 /hour without symptoms after adenotonsillectomy (13). Although persistent/recurrent OSAS following adenotonsillectomy is usually associated with obesity (OSAS phenotype 2) and underlying complex disorders (OSAS phenotype 3), it can also affect otherwise children with OSAS phenotype 1. Several factors such as age, medical co-morbidities, causes and sites of upper airway obstruction, OSAS severity and surgical technique were found to be associated with an increased risk of persistent/recurrent OSAS after adenotonsillectomy. This editorial aimed to identify the predictive factors of a high risk of persistent/recurrent OSAS after adenotonsillectomy.

Children who undergo surgery at an older age (> 7 years old) are more likely to experience persistent or recurrent OSAS, possibly due to mouth breathing-related obstructive upper airway abnormalities (11). They also have a higher risk of persistent neurocognitive dysfunction even after complete normalization of OAHI (11).

Several other studies identified preoperative obesity as a significant risk factor for persistent/recurrent OSAS as well, even when multivariable modeling was used to control for confounding factors such as age and baseline OAHI (11,14). The odds ratios of persistent/ recurrent OSAS in obese children ranged in these models from 3.2 to 4.7 (10). The strong association with a higher risk of persistent/recurrent OSAS in obese children was attributed to adenoid regrowth or lingual tonsil enlargement (15)

Asthma and allergic rhinitis have also been identified as predictive factors for persistent/recurrent OSAS (16). This could be due to the decreased level of Tregs (Foxp3+ cells) and the subsequent imbalance of Th2/Th1 in allergic diseases which may increase the likelihood of adenotonsillar regrowth after partial tonsillectomy (16).

All studies that evaluated baseline OAHI identified pre-operative severe OSAS as a risk factor for persistent/ recurrent OSAS even when adjusted for other comorbidities such as obesity (10,17).

In addition, sleep endoscopy findings in 34 children with persistent OSAS after adenotonsillectomy found multiple sites of partial or complete obstruction (inferior turbinates, adenoid, tongue base, epiglottis, aryepiglottic, lingual tonsil) in 97% of cases (15). These findings suggest that multiple-site obstruction should be considered in children with persistent OSAS (15). Some morphological abnormalities such as clinically small tonsils (a Brodsky tonsil score ≤ 2), a high Mallampati score (class 3 or 4), and micrognathia were identified as potential predictors for multiple site obstruction (15).

Regarding surgical technique, the American Academy of Pediatrics recommendation suggests that "adenoidectomy or tonsillectomy alone may not be sufficient, because residual tonsillar tissue may contribute to persistent obstruction" (10). Tonsillectomy without adenoidectomy can be effective; however, a persistent obstruction due to residual lymphoid tissue was reported in the literature (8,10). When compared with total tonsillectomy, partial tonsillectomy carries an increased risk of regrowth of the tonsils (1.6% vs. 0.002) (18), particularly in younger children under three years of age. The period for tonsillar regrowth ranged from one to 18 months and may result in more recurrent OSA (19).

The natural history of childhood OSAS to early adulthood has been studied in a community-based cohort including 243 children, with a mean follow-up duration of 10 years (20). Among patients who had OSAS at baseline, more than one-third had persistent disease (OAHI ≥ 5 events/h) at follow-up (20). Only participants with baseline age > 10 years showed a significant but weak correlation between baseline and follow-up log-transformed OAHI (20). Based on these findings, authors suggested that adolescent OSAS (ie; diagnosed in late childhood/early adolescence) and more severe diseases have an increased likelihood of persisting into adulthood (20). In contrast, children OSAS, diagnosed in younger children, are less likely to correlate with adult disease (20). Among participants with OAHI < 5 events/h at baseline, nearly one-fifth develop incident OSAS with an OAHI ≥ 5 events/h at follow-up (20). Male sex, obesity, and higher body mass index z score were identified as risk factors for adult-onset OSAS in these participants (20).

In conclusion, adenotonsillectomy is associated with significant clinical and polysomnographic improvements in children with OSAS. However, special attention should be paid to the risk of persistent/recurrent OSAS after surgery. Several factors have been identified as predictive of a high risk of persistent/recurrent OSAS, including mainly older age, male sex, obesity, asthma/rhinitis, severe disease, small tonsils, a high Mallampati score, micrognathia and partial tonsillectomy. Children with predictive factors of persistent/recurrent OSAS require personalised treatment, rigorous post-surgical clinical and polysomnographic monitoring and extended follow-up into adulthood (Table 1).

Table 1. To take home message: Predictive factors of persistent/ recurrent pediatric obstructive sleep apnea-hypopnea syndrome after adenotonsillectomy.

- Old age
- Male sex
- Obesity
- Asthma/Rhinitis
- Severe disease
- Small tonsils (a Brodsky tonsil score ≤ 2)
- High Mallampati score (class 3 or 4)
- Micrognathia
- Partial tonsillectomy

References

1. Savini S, Ciorba A, Bianchini C, Stomeo F, Corazzi V, Vicini C, Pelucchi S. Assessment of obstructive sleep apnoea (OSA) in children: An update. Acta Otorhinolaryngol Ita. 2019; 39(5):289-297.
2. Sateia MJ. International classification of sleep disorders-third edition highlights and modifications. Chest. 2014;146(5):1387-1394.
3. Esposito S, Ricci G, Gobbi R, Vicini C, Caramelli F, Pizzi S, Fadda A, Ferro S, Plazzi G. Diagnostic and therapeutic approach to children

and adolescents with obstructive sleep apnea syndrome (OSA): Recommendations in Emilia-Romagna Region, Italy. Life. 2022 16;12(5):739.

4. Narayanasamy S, Kidambi SS, Mahmoud M, Subramanyam R. Pediatric sleep-disordered breathing: A narrative review. Pediatr Med 2019; 2:52.
5. Nosetti L, Zaffanello M, De Bernardi di Valserra F, Simoncini D, Beretta G, Guacci P, Piacentini G, Agosti M. Exploring the intricate links between adenotonsillar hypertrophy, mouth breathing, and craniofacial development in children with sleep-disordered breathing: Unraveling the vicious cycle. Children. 2023 21;10(8):1426.
6. Bokov P, Dahan J, Boujemla I, Dudoignon B, André CV, Bennaceur S, Teissier N, Delclaux C. Prevalence of mouth breathing, with or without nasal obstruction, in children with moderate to severe obstructive sleep apnea. Sleep Med. 2022;98:98-105.
7. Garde AJB, Gibson NA, Samuels MP, Evans HJ. Recent advances in paediatric sleep-disordered breathing. Breathe. 2022;18(3):220151.
8. Marcus CL, Moore RH, Rosen CL, Giordani B, Garetz SL, Taylor HG, Mitchell RB, Amin R, Katz ES, Arens R, Paruthi S, Muzumdar H, Gozal D, Thomas NH, Ware J, Beebe D, Snyder K, Elden L, Sprecher RC, Willging P, Jones D, Bent JP, Hoban T, Chervin RD, Ellenberg SS, Redline S; Childhood Adenotonsillectomy Trial (CHAT). A randomized trial of adenotonsillectomy for childhood sleep apnea. N Engl J Med. 2013 20;368(25):2366-76.
9. Uwiera TC. Considerations in surgical management of pediatric obstructive sleep apnea: Tonsillectomy and beyond. Children. 2021;8(11):944.
10. Marcus CL, Brooks LJ, Draper KA, Gozal D, Halbower AC, Jones J, Schechter MS, Ward SD, Sheldon SH, Shiffman RN, Lehmann C, Spruyt K; American Academy of Pediatrics. Diagnosis and management of childhood obstructive sleep apnea syndrome. Pediatrics. 2012;130(3):e714-55.
11. Bhattacharjee R, Kheirandish-Gozal L, Spruyt K, Mitchell RB, Promchiarak J, Simakajornboon N, Kaditis AG, Splaingard D, Splaingard M, Brooks LJ, Marcus CL, Sin S, Arens R, Verhulst SL, Gozal D. 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.
12. Mitchell RB, Archer SM, Ishman SL, Rosenfeld RM, Coles S, Finestone SA, Friedman NR, Giordano T, Hildrew DM, Kim TW, Lloyd RM, Parikh SR, Shulman ST, Walner DL, Walsh SA, Nnacheta LC. Clinical Practice Guideline: Tonsillectomy in Children (Update). Otolaryngol Head Neck Surg. 2019;160(1_suppl):S1-S42.
13. Ishman SL, Maturo S, Schwartz S, McKenna M, Baldassari CM, Bergeron M, Chernobilsky B, Ehsan Z, Gagnon L, Liu YC, Smith DF, Stanley J, Zalzal H, Dhepyasuwan N. Expert consensus statement: Management of pediatric persistent obstructive sleep apnea after adenotonsillectomy. Otolaryngol Head Neck Surg. 2023;168(2):115-130.
14. Guilleminault C, Li K, Quo S, Inouye RN. A prospective study on the surgical outcomes of children with sleep-disordered breathing. Sleep. 2004 1;27(1):95-100.
15. Coutras SW, Limjuco A, Davis KE, Carr MM. Sleep endoscopy findings in children with persistent obstructive sleep apnea after adenotonsillectomy. Int J Pediatr Otorhinolaryngol. 2018;107:190-193.
16. Huo Z, Shi J, Shu Y, Xiang M, Lu J, Wu H. The relationship between allergic status and adenotonsillar regrowth: A retrospective research on children after adenotonsillectomy. 2017. Sci Rep 7(1):46615.
17. Mitchell RB. Adenotonsillectomy for obstructive sleep apnea in children: Outcome evaluated by pre- and postoperative polysomnography. Laryngoscope. 2007;117(10):1844-54.
18. Odhagen E, Sunnergren O, Hemlin C, Hessén Söderman AC, Ericsson E, Stalfors J. Risk of reoperation after tonsillotomy versus tonsillectomy: a population-based cohort study. Eur Arch Otorhinolaryngol. 2016;273(10):3263-8.
19. Celenk F, Bayazit YA, Yilmaz M, Kemaloglu YK, Uygur K, Ceylan A, Korkuyu E. Tonsillar regrowth following partial tonsillectomy with radiofrequency. Int J Pediatr Otorhinolaryngol. 2008;72(1):19-22.
20. Chan KC, Au CT, Hui LL, Ng S kwan, Wing YK, Li AM. How OSA evolves from childhood to young adulthood: Natural history from a 10-year follow-up study. Chest. 2019;156(1):120-130.