SEVERE OBSTRUCTIVE SLEEP APNEA IN CHILDREN: A CASE REPORT

T Marcovici[1, 2], A Albu[2], D Zabos[3], R Horhat[1, 3], OBelei[1, 2], L Olariu[1, 2], O Marginean[1, 2]

Abstract

Adenotonsillar hypertrophy is a common cause of obstructive sleep apnea in children. Affecting ventilation and sleep architecture, recurrent respiratory obstruction can lead to metabolic, cardiovascular, neurobehavioral and somatic consequences. Purpose. To present the etiology and management of nocturnal and diurnal respiratory disorders in a pre-school patient coming from rural area. Materials and Methods. A female patient aged 4 years and 9 months was assessed by an interdisciplinary team. The evaluation included: case history, physical examination, laboratory tests and specialty consults. Case presentation. The patient presented multiple sleeping disorders (apnea, noisy and difficult breathing, intense snoring, orthopnea, frequent awakening) and diurnal symptoms (hypersomnia, attention deficit, nasal obstruction, diminished appetite, dysphagia, nasal voice). At admission, the clinical examination showed failure to thrive, adenoid face, sternal depression, noisy and difficult mouth breathing, severe substernal and intercostal retractions. Important adenotonsillar hypertrophy (grade IV) was confirmed by otolaryngology examination. Sleep polygraph showed very severe obstructive sleep apnea. Extracapsular tonsillectomy and adenoidectomy were performed under high anesthetic and surgical risk. Intra and postoperative outcome was positive, with the remission of the upper respiratory airway obstruction symptoms. Conclusions. Despite suggestive symptoms the patient presentation was late. Neurobehavioral manifestations and somatic disorders suggest the long-term evolution of upper respiratory airway obstruction. The extreme severity of sleep apnea was revealed by polygraph study. The sleep functional evaluation has contributed decisively to the therapeutic management and the favorable evolution of the case.

Keywords: obstructive apnea, sleep, child

Introduction

Obstructive sleep apnea (OSA) is a potentially lethal pathology that remains underdiagnosed in children. [1, 2, 3] Its etiology is polymorphic and should be systematically sought in order to initiate appropriate therapy. [2, 4, 5, 6, 7] Adenotonsillar hypertrophy is a common cause of obstructive sleep apnea in children. [1, 3, 4, 8] Affecting ventilation and sleep architecture, recurrent respiratory obstruction can lead to metabolic, cardiovascular, neurobehavioral and somatic consequences. [4, 9, 10, 11, 12, 13, 14]

Materials and Methods

A female patient aged 4 years and 9 months, coming from a rural area, has been evaluated in our clinic by: history, physical examination, paraclinical explorations: biological (hematological, biochemical, immunological, bacteriological); imaging (cardiopulmonary radiography, cardiac ultrasound); functional (electrocardiogram, sleep polygraph); specialty consults (otorhinolaryngology, pediatric cardiology, pediatric pneumology, pediatric orthopedics, somnology).

Case presentation

The girl was hospitalized in august 2018 for sleep apnea, restless sleep, nasal obstruction, constant noisy and difficult mouth breathing, excessive daytime sleepiness, attention deficit, diminished appetite, and difficulties in swallowing solid foods, failure to thrive. She is the third child of an affirmative healthy couple, from a physiological pregnancy carried to full term, born by caesarean section, with a birth weight of 4000g, birth length = 52 cm and an Apgar score 9. She was artificially fed with goat milk since birth and diversified incorrectly. Rickets prevention and vaccination were performed according to the national immunization schedule. The patient's personal history reveals recurrent upper airway infections and adenoidectomy performed in august 2017.

At admission, the patient was afebrile, with a relatively influenced general condition and failure to thrive (weight = 15 kg, height =111 cm, BMI = 12.2 kg/m2, less than the 5th percentile) (Figure 1), adenoid faces, pale skin, poor subcutaneous cellular tissue, noisy mouth breathing, nasal voice, speech difficulties using short sentences and excessive sleepiness. She presented inter and subcostal retractions and important depression of the stern during inspiration.

[1] "Victor Babeş" University of Medicine and Pharmacy, Timisoara, Department of Pediatrics
[2] Clinical Emergency Hospital for Children"Louis Turcanu", Timisoara, Department of Pediatrics
[3] Clinical Emergency Hospital for Children"Louis Turcanu", Timisoara, Department of ENT

E-mail: t_marcovici@yahoo.com, alexandraandreea.albu@yahoo.com, cabmedz@gmail.com, raluhorhat@gmail.com, oana22_99@yahoo.com, tunealaura@yahoo.com, omarginean@ymail.com

Respiratory frequency was normal (22 breaths per minute), oxygen saturation around 96-97% and increased heart rate (128 beats per minute). Cardiopulmonary auscultation was normal. Blood pressure range was normal for age (105/60 mmHg). Oropharyngeal examination revealed important adenotonsillar hypertrophy, with large cryptic tonsils extended to the midline

The ENT exam described grade IV tonsillar hypertrophy (with the absence of the oropharyngeal isthmus); cryptic tonsils, hyperplasia of the rinopharyngeal lymphoid tissue with the complete obstruction of choanae; small bilateral auditory conduct for the biological age, retracted tympanic membranes.

The biological evaluation revealed atopic status (peripheral blood eosinophilia and elevated serum immunoglobulin E levels and mild allergic response to mites), inflammation, hyper-immunoglobulin G and hypergammaglobulinemia, post streptococcal inflammatory syndrome and vitamin D deficiency (Table 1, 2 and 3). Chest X-ray interpretation showed the sunken appearance of the chest (pectus excavatum) (Figure 2). The physical examination and paraclinical cardiac evaluation (electrocardiogram test – ECG, cardiac ultrasound) showed normal heart function. The somnological evaluation was achieved by using the sleep-related breathing disorder scale from the questionnaire on sleeping in children (22 questions) and polygraph sleep study The score calculated on the basis of the questionnaire was suggestive for a sleep-related breathing disorder. The sleep study has shown an extremely severe obstructive sleep apnea, with an apnea-hypopnea index = 112.9/h, desaturation index = 58.4/h, 74.5 obstructive apnea/h, 23.7 mixt apnea/h, 8 central apnea/h and 6.8 hypopnea/h (Figure 3). Respiratory events occurred in all positions adopted during sleep. The patient has persistently snored throughout the evaluation, adopted the seating position repeatedly and has woken frequently throughout the assessed period. She presented several abnormalities of the periphery oxygen saturation during sleep (average SaO2 = 90%, maximum SaO2 = 100%, minimum SaO2= 51%). In 16.7% of the time spent in bed, her oxygen saturation was less than 90%.

ENT consult recommended adenotonsillectomy. The Extracapsular tonsillectomy and adenoidectomy was performed the 5th day after admission, under high anesthetic and surgical risk. The postoperative care plan was complex: parenteral nutrition + oral; antibiotics, steroid anti-inflammatories, anti-hemorrhagic agents, analgesics, probiotics and roborants.

The intra and postoperative outcome was positive. The child regained appetite and became cheerful, the general condition improved, sleep became peaceful, without any respiratory events. Antistreptolysin O (ASO) titer decreased (1000 UI/mL). The hospitalization period lasted for 13 days.

Discharge recommendations included hygienic-dietary treatment (adequate nutrition, sleep hygiene, avoiding dorsal decubitus during sleep, avoiding exposure to environmental emissions), medication (antibiotics, nebulization with hypertonic saline, analgesics if needed, probiotics, oral vitamin D supplementation), physiotherapy (breathing exercises and Valsalva maneuver), heliotherapy and thalassotherapy.

Multidisciplinary team follow-up, motorization of nutritional status and sleep polygraph reevaluation after 6-8 weeks were recommended, but the patient did not return in the clinic as scheduled.

Discussions

Pathophysiological factors involved in obstructive sleep apnea (OSA) can be divided into anatomical factors that reduce airway caliber and those that promote increased upper airway collapsibility. [4, 6, 7, 15] Children with OSA experience partial or complete obstruction of the upper airway during sleep and they achieve to reopen them through short awakenings or hypercapnic ventilatory response. [15] Increased respiratory effort, alveolar hypoventilation, intermittent hypoxia and hypercapnic cause sleep fragmentation. [1, 12] The altered architecture can lead to dysfunction of the prefrontal cortex, affecting cognitive behavior, executive function and learning ability. [1, 2] The type of pediatric OSA is influenced by individual susceptibility, environmental and lifestyle conditions (diet, physical/intellectual activity). According to phenotype, OSA in children can be classified as: type I (adeno-tonsillar hypertrophy + OSA); type II (obesity + OSA); type III (craniofacial or neuromuscular disorders + OSA). [4, 16]

Genetic predisposition, inflammation and atopic status may be implicated in the etiopathogenesis of OSA in children.[2,8] Recurrent viral and bacterial infections as well as environmental exposure to irritants (cigarette smoke, allergens) would favor excessive adeno-tonsillar tissue proliferation and OSA worsening.[8]

The major consequences of pediatric OSA are neurobehavioral and metabolic disorders, cardiovascular pathology and somatic growth impairment. [1, 10, 11, 14, 16] The complications of OSA results from the interaction between intermittent hypoxia, hypercapnic, repeated variations of intrathoracic pressure and episodic awakenings. [12, 16]

Sleep fragmentation, associated with intermittent hypoxia and hypercapnic has negative effects on the prefrontal cortex and affects the executive functions in children with OSA. [1]. Intermittent hypoxia exacerbates neuronal apoptosis and affects the brain regions responsible for memory and learning. Academic performance can be affected in the long term. Excessive daytime sleepiness is the consequence of sleep fragmentation. [1, 2]

Some children with OSA (≤ 5%) fail to thrive as a consequence of anorexia, dysphagia, decreased nocturnal secretion of growth hormone, intermittent hypoxia and increased respiratory effort during sleep. [1, 2, 16, 17, 18] A sunken chest in children with OSA could be a consequence of chronic upper airway obstruction and difficult breathing, under the conditions of a compliant and immature chest. [1]

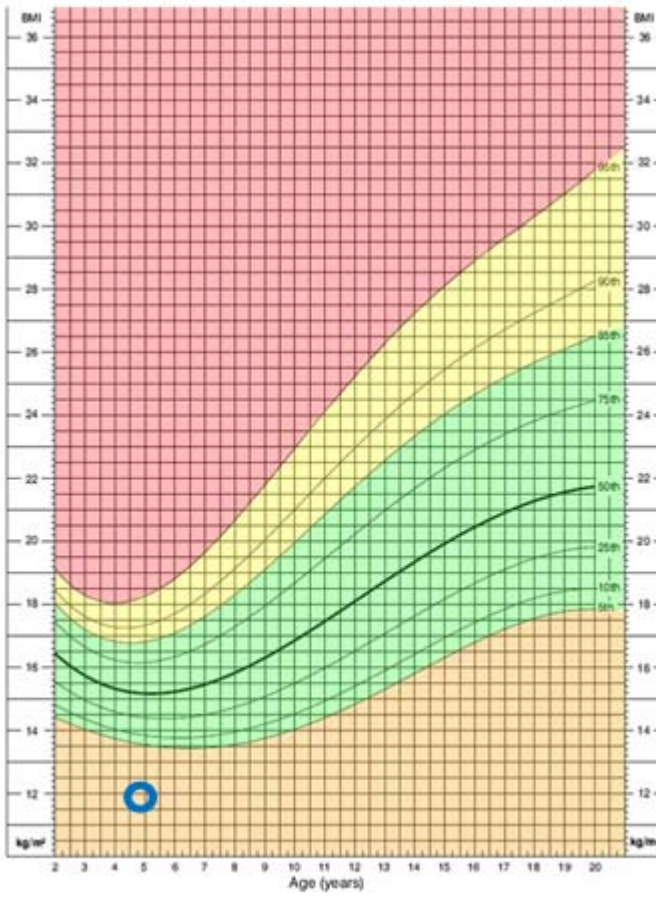

Figure 1. Evaluation of the nutritional status of the subject

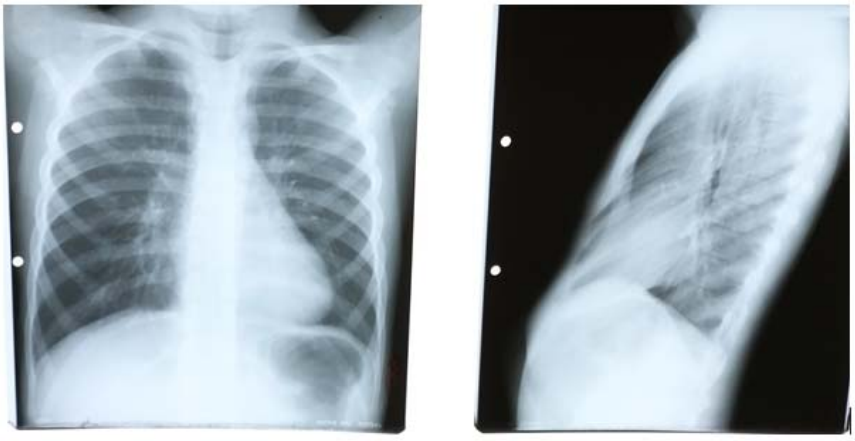

Figure 2. Chest x-ray (posteroanterior, lateral) of the presented case

WBC - Leukocytes	10,11	4 - 12	10^3/ul
RBC - Erythrocytes	4,17	3,9 - 5,3	10^6/ul
HGB - Hemoglobin	12,0	10,7 - 14,1	g/dL
HCT - Hematocrit	35,6	30 - 43	%
MCV - Mean corpuscular volume	85,4	72 - 88	fL
MCH-mean corpuscular hemoglobin	28,8	23 - 31	pg
MCHC - mean corpuscular hemoglobin concentration	33,7	32 - 36	g/dL
PLT – Thrombocytes	439	150 - 450	10^3/ul
Neutrophils	4,65	1,5 - 8	10^3/ul
Lymphocytes	3,59	1,5 - 10	10^3/ul
Monocytes	1,16	0,1 - 1	10^3/ul
Eosinophils	**0,58**	0,08 - 0,48	10^3/ul
Basophils	0,13	0 - 0,1	10^3/ul
Percent of neutrophils	46,0	18 - 44	%
Percent of lymphocytes	35,5	42 - 61	%
Percent of monocytes	11,5	2 - 8	%
Percent of eosinophils	**5,7**	2 - 4	%
Percent of basophils	1,3	0 - 1	%

Table 1. Full blood count – pacient case report results

ALT - alanine transaminase	16	<39	U/L
AST - aspartate transaminase	22	< 52	U/L
Creatinine	28	25 - 55	umol/L
C-Reactiva Protein	0,44	0 - 5	mg/L
ESR - Erythrocyte Sedimentation Rate	**29**	2-13	mm/h
ASO - Antistreptolysin O	**3204**	0 - 200	iU/mL
Total Protein	75,2	60 - 80	g/L
Albumin	57,4	59,8 - 72,4	%
Alpha1-globulin	2,6	1 - 3,2	%
Alpha 2-globulin	9,6	7,4 - 12,6	%
Beta-globulin	9,4	7,5 - 12,9	%
Gamma-globulin	**21,0**	8 - 15,8	%
IgA-Immunoglobulin A	1,99	0,27 - 1,95	g/L
IgG-Immunoglobulin G	**19,79**	5,04 - 14,65	g/L
IgM- Immunoglobulin M	1,24	0,24 - 2,1	g/L
Vitamin D (25-hydroxi)	**23,92**	30 - 100	ng/ml
Ionic Calcium	1.09	1,05 - 1,3	mmol/L
Total Calcium	2,46	2,3 - 2,75	mmol/L
Alkaline Phosphatase	132	< 269	U/L
Phosphor	1,27	1,1 - 2	mmol/L
Magnezium	0,78	0,7 - 1,05	mmol/L
Nasal secretion	negative culture result		
Pharyngeal exudate	negative culture result		

Table 2. Biological, biochemical, imunological and bacteriological evaluation of the patient

Allergen-Specific Immunoglobuline E (IgE)	
Herb pollen	0 - Undetectable
Birch pollen	0 - Undetectable
Black wormwood	0 - Undetectable
Derm. pteronyssinus	0 - Undetectable
Derm. farinae	**1 – Extremely low**
Cat hair/epithelium	0 - Undetectable
Dog hair/epithelium	0 - Undetectable
Horse hair/epithelium	0 - Undetectable
Cladospo. herbarum	0 - Undetectable
Aspergilius fumigatus	0 - Undetectable
Alternaria alternata	0 - Undetectable
Egg White	0 - Undetectable
Egg Yolk	0 - Undetectable
Milk	0 - Undetectable
Cod Fish	0 - Undetectable
Alpha lactalbumin	0 - Undetectable
Beta lactoglobulin	0 - Undetectable
Casein	0 - Undetectable
Bovine albumin	0 - Undetectable
Wheat	0 - Undetectable
Rice	0 - Undetectable
Soya	0 - Undetectable
Peanuts	0 - Undetectable
Almonds	0 - Undetectable
Carrot	0 - Undetectable
Potato	0 - Undetectable
Apple	0 - Undetectable
Total Immunoglobulin E 0 – 60 UI/ml	**715,6**

Table 3. Atopic status evaluation of the presented patient

The question are is used frequently to assess for OSA risk in children aged 2 to 18 years. [10, 14, 19] Preoperative sleep study (polysomnography/ polygraph) diagnoses sleep apnea and identifies severe OSA that amplifies risks of perioperative adenotonsillectomy complications. [2, 14, 20]

Characterized by adenotonsillar hypertrophy, case presented belongs to OSA type I. The important morbidity due OSA is somatic (failure to thrive, adenoid faces, sunken chest) and neurobehavioral (hyperactivity, attention deficit, fragmented and restless sleep, diurnal hypersomnia) [1, 16].

In children with moderate / severe OSA and adeno-tonsillar hypertrophy, ablation of adenoid vegetation and tonsillectomy is the first line therapy. [2, 4, 8, 18] In severe OSA the anesthetic and operative risk is important, co-morbidities increasing intra and perioperative risk. Recurrent hypoxemia enhances the sensibility to opioids directly proportional with the severity of OSA. Failure to thrive and severe OSA are risk factors for postoperative complications. [18] Pulmonary edema with severe respiratory obstruction can occur during the first postoperative hours and orotracheal intubation or non-invasive ventilation might be needed.

Up to 75% of operated patients continue having residual OSA. The longer OSA remains untreated, the less is probable the complete resolution of symptoms. [1, 16, 21]

Late diagnosis of OSA involves higher risks of developing cardiovascular, metabolic and somatic complications, as well as cognitive disorders with decreased scholar abilities. Often the quality of life of both child and family is affected. [4, 22].

Due to the persistence of OSA in a significant number of children, post-operative re-assessment through questionnaire and sleep study is mandatory. [14, 18]

Conclusions

Patient presentation and diagnosis were late despite suggestive symptoms. Neurobehavioral manifestations and somatic disorders revealed the long-term evolution of upper respiratory airway obstruction.

Specifying the extreme severity of sleep apnea by polygraph study has contributed decisively to the therapeutic management and the favorable evolution of the case.

Preventing the complications of obstructive sleep apnea on the growth and development of the child is imperiously needed and is based on early diagnosis and appropriate therapy of the disease.

References

1. Cataletto ME, Serebrisky D. Childhood Sleep Apnea, 2016, https://emedicine.medscape.com/article/1004104.
2. Tan HL, Gozal D, Kheirandish-Gozal L. Obstructive sleep apnea in children: a critical update. Nat Sci Sleep, 2013, 5: 109-123.
3. Davidson Ward SL, Perez IA. Treatment Options in Obstructive Sleep Apnea. In: Sheldon S, Kryger M, Ferber R, Gozal D.(ed.). Principles and Practice of Pediatric Sleep Medicine (Second Edition), 2014: 255-262.
4. Oros M. Sindrom de apnee în somn de tip obstructive la copil. In: Mihaicuta S. Curs practic de somnografie. Editura Victor Babeş, Timisoara, 2010:115-124.
5. Andreas S. Obstructive sleep apnoea: clinical aspects. In: Simonds AK, de Becker W (ed.) Respiratory Sleep Medicine 2012: 32-34.
6. Dehlink E, Tan HL. Update on pediatric obstructive sleep apnoea. Thorac Dis,2016, 8(2):224-35.
7. Verbraecken J. Obstructive sleep apnoea: epidemiology and risk factors. In: Simonds AK, de Becker W (ed.) Respiratory Sleep Medicine 2012: 25-28.
8. Kheirandish-Gozal L. Novel pharmacological approaches for treatment of obstructive sleep apnea. In: Sheldon S, Kryger M, Ferber R, Gozal D.(ed.). Principles and Practice of Pediatric Sleep Medicine (Second Edition), 2014:295-300.
9. Tauman R, Gozal D. Obstructive sleep apnea in children. Expert Rev Resp Med, 2011, 5(3):425-40.
10. Andreas S. Obstructive sleep apnoea:consequences. In: Simonds AK, de Becker W (ed.) Respiratory Sleep Medicine 2012: 35-38.
11. Hoyos CM, Melehan KL, Grunstein RR, Phillips CL. Does obstructive sleep apnea cause endothelial dysfunction? A critical review of the literature. Sleep Med Rev, 2015, 20: 15-26.
12. Atkeson A, Jelic S. Mechanisms of endothelial dysfunction in obstructive sleep apnea. Vasc Health Risk Manag, 2008, 4(6):1327-1335.
13. Gunn S, Khatwa UA. Obstructive Sleep Apnea in Adolescence. In: Sleep Disorders in Adolescents. A Clinical Casebook; Editors: Sanjeev V. Kothare, Rebecca Quattrucci Scott. Springer International Publishing, 2017, 45-59.
14. Chervin RD, Weatherly RA, Garetz SL, Ruzicka DL, Giordani BJ, Hodges EK,. Dillon JE, Guire KE. Pediatric Sleep Questionnaire Prediction of Sleep Apnea and Outcomes. Arch Otolaryngol Head Neck Surg, 2007, 133(3):216-222.
15. Verbraecken J, de Backer W. Obstructive sleep apnoea: pathophysiology. In: Simonds AK, de Becker W (ed.) Respiratory Sleep Medicine 2012: 29-31.
16. Capdevila OS, Kheirandish-Gozal L, Dayyat E, Gozal D. Pediatric obstructive sleep apnea: complications, management and long-term outcomes. Proc Am Thorac Soc, 2008, 5(2):274-282.
17. Nachalon Y, Lowenthal N, Greenberg-Dotan S, Goldbart AD. Inflammation and growth in young children with obstructive sleep apnea syndrome before and after adenotonsillectomy. Mediators Inflamm. 2014;2014:146893. doi:10.1155/2014/146893.
18. Beck SE. A 2½-year-old girl with snoring, restless sleep, and severe obstructive sleep apnea. In: Pediatric Sleep Pearls, 2017:133-136.
19. Moturi S, Avis C. Assessment and treatment of common pediatric sleep disorders. Psychiatry, 2010, 7(6):24-37.
20. Nixon GM, Davey MJ. Sleep apnoea in the child. Aust Fam Physician, 2015, 44:352–55.
21. Marcus CL, Brooks LJ, Draper KA, Gozal D, Halbower AC, Jones J, Schechter MS, Sheldon SH, Spruyt K, Ward SD, Lehmann C, Shiffman RN. Diagnosis and management of childhood obstructive sleep apnea syndrome. Pediatrics, 2012, 130 (3):576 - 584.
22. Brunetti L, Francavilla R, Scicchitano P, Tranchino V, Loscialpo M, Gesualdo M, Zito A, Fornarelli F, Sassara M, Giordano P, Miniello VL, Ciccone MM. Impact of sleep respiratory disorders on endothelial function in children. ScientificWorldJournal. 2013 Dec 26;2013:719456. doi: 10.1155/2013/719456. PubMed PMID: 24453895; PubMed Central PMCID: PMC3888735.

Correspondence to:

Marcovici Tamara
Clinical Emergency Hospital for Children "Louis Turcanu"
Timisoara, Dr. Iosif Nemoianu Street, 2, 300011,
Phone: 0040256203394/Fax: 000256-201.976
E-mail: t_marcovici@yahoo.com