



ORIGINAL ARTICLE

Non-invasive ventilation in complex obstructive sleep apnea – A 15-year experience of a pediatric tertiary center



I.C. Girbal*, C. Gonçalves, T. Nunes, R. Ferreira, L. Pereira, A. Saianda, T. Bandeira

Respiratory, Sleep and Ventilation Unit, Department of Pediatrics, Santa Maria Hospital - CHLN, Academic Medical Center of Lisbon, Lisbon, Portugal

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KEYWORDS

Noninvasive ventilation;
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Abstract

Introduction: Obstructive sleep apnea (OSA) affects approximately 1–3% of pediatric population and is associated with significant morbidity. As adenotonsillar hypertrophy (ATH) is its primary cause in children, elective adenotonsillectomy is the first treatment of choice. Noninvasive ventilation (NIV) has been increasingly considered as an option, mainly for children with complex diseases, ineligible or waiting for surgeries, or after surgery failure.

Objectives: To describe the experience in the management of children with complex OSA, and to evidence the feasibility and advantages of NIV.

Methods: This was a retrospective cohort study of 68 children on NIV, in whom complex OSA was the main indication for ventilation, in a Pediatric Respiratory Unit at a University Hospital between January 1997 and March 2012. Demographic and clinical data were collected on the underlying diagnosis, therapeutic interventions prior to NIV, NIV related issues and outcome.

Results: Forty (59%) children were male, median age at starting NIV was 6 years and 7 months, with interquartile range (IQR) of 15–171 months. Twenty-two (32%) were infants and 25 (37%) adolescents.

The most common diagnosis was congenital malformations and genetic disorders in 34 (50%) patients. Nine patients had cerebral palsy, 8 were post treatment for central nervous system tumors and 6 had inborn errors of metabolism. Three children had ATH and three obesity. The majority of patients (76%) had exclusively obstructive OSA and started CPAP. Ten patients had minor complications. Twenty-two patients stopped NIV due to clinical improvement, 8 were non-compliant and 8 patients died. NIV median duration was 21.5 months (IQR: 7–72).

Conclusions: NIV is feasible and well tolerated by children with OSA associated with complex disorders, and has been shown to have few complications even in infants and toddlers.

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* Corresponding author.

E-mail address: inescarmogirbal@gmail.com (I.C. Girbal).

PALAVRAS-CHAVE

Ventilação não invasiva;
Apneia obstrutiva do sono;
Crianças

Ventilação não-invasiva em apneia obstrutiva do sono complexa – Uma experiência de 15 anos de um centro pediátrico terciário

Resumo

Introdução: A apneia obstrutiva do sono (OSA) afeta aproximadamente 1–3% da população pediátrica e está associada com uma morbidade significativa. Como a hipertrofia adenotonsiliana (ATH) é a sua principal causa, a adenocele eletiva é a primeira opção terapêutica. A ventilação não invasiva (VNI) tem sido cada vez mais considerada como uma opção, principalmente para as crianças com doenças complexas, inelegíveis, à espera de cirurgias ou após falência do tratamento cirúrgico.

Objetivos: Descrever a experiência no tratamento de crianças com OSA complexo e avaliação da viabilidade e das vantagens do NIV.

Métodos: Estudo de coorte retrospectivo de 68 crianças em NIV, onde o complexo OSA foi a principal indicação para ventilação, numa Unidade Respiratória Pediátrica de um Hospital Universitário entre janeiro de 1997 e março de 2012. Os dados demográficos e clínicos recolhidos englobaram o diagnóstico subjacente, as intervenções terapêuticas prévias à NIV, as intercorrências relacionadas com a NIV e a evolução clínica.

Resultados: Quarenta (59%) crianças eram do sexo masculino, idade média no início da NIV foi de 6 anos e 7 meses, com intervalo interquartil (IQR) de 15–171 meses. Vinte e duas (32%) eram crianças e 25 (37%) adolescentes.

O diagnóstico mais comum foram as malformações congénitas e as doenças genéticas em 34 (50%) pacientes. Nove pacientes tiveram paralisia cerebral, 8 sequelas de pós-tratamento para tumores do sistema nervoso central e seis sofriam de erros inatos do metabolismo. Três crianças sofriam de ATH e três de obesidade. A maioria dos pacientes (76%) sofria exclusivamente de OSA obstrutiva e iniciou CPAP. Dois doentes sofriam complicações menores. Vinte e dois doentes pararam o NIV, devido a melhoria clínica, 8 eram não-conformes e 8 morreram. A duração média do NIV foi 21,5 meses (IQR: 7–72).

Conclusões: A NIV é uma terapia viável e bem tolerada em crianças com OSA e doenças complexas, com poucas complicações, incluindo em bebés e crianças pequenas.

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Introduction

Pediatric obstructive sleep respiratory disorder has a broad clinical spectrum, from simple snoring to the most severe obstructive sleep apnea (OSA).

OSA occurs in 1–3% of children,^{1–3} and is defined by airway obstruction, either partial and prolonged or complete and intermittent, interfering with sleep patterns and ventilation.

Adenotonsillar hypertrophy (ATH) is the most frequent cause in children, but craniofacial malformations, as well as neuromuscular diseases and obesity have been increasingly considered.⁴

In childhood, OSA has important repercussions on somatic growth, the cardiovascular system and neurocognitive development.^{5–7} Early diagnosis and intervention are essential to prevent complications and improve quality of life.^{8,9}

Identification of clinical criteria (respiratory pauses, hard or noisy breathing or excessive daytime somnolence) and predisposing clinical conditions¹⁰ support the diagnosis of OSA, but polysomnography (PSG) is currently considered the gold standard.^{11,12}

As the main cause of OSA in childhood is ATH, adenotonsillectomy is the most commonly recommended treatment for uncomplicated cases.^{10,13,14} There is, however, growing evidence of surgery failure¹⁵ and there are a significant number of children with craniofacial defects who need alternative therapies. These children represent a group with complex OSA, requiring innovative surgical and medical approaches.

In children with chronic conditions or complex OSA, NIV has been increasingly used since 1984,^{16,17} when surgery is contraindicated or ineffective, or as a supportive treatment until appropriate stage for surgical intervention, allowing for clinical stabilization and growth.^{18–20}

NIV efficacy and tolerance in children have been reported in a few studies^{18,21,22} and clearly the experience of qualified teams with appropriate support facilities warrants high quality care.^{10,23,24}

The main objective of this study is to describe our experience in a tertiary Portuguese pediatric center in the management of children with complex OSA, for whom noninvasive positive-pressure ventilation was an effective therapeutic alternative.

Methods

A retrospective case series study was performed through chart review of all cases of children submitted to NIV, in which complex OSA was the main indication for ventilation. Eligible cases were identified from internal clinical database in the period of January 1997 to March 2012. Children with other indications for NIV were excluded.

Demographic data were collected, including primary and secondary clinical diagnoses, sleep studies results, modes of ventilation, adherence, complications and efficacy related to NIV. Clinical data concerning sleep disordered breathing including difficult or noisy breathing, respiratory pauses, unsettled sleep, excessive sweating, excessive daytime somnolence, morning headache and failure to thrive were assessed. All patients underwent sleep studies: continuous transcutaneous monitoring of O₂ and CO₂ (Tc CO₂), complemented with capillary blood gases from the beginning through 2004, on an inpatient basis, formal sleep studies at the sleep

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