Upper Airway Stimulation for Adolescents with Down Syndrome and Obstructive Sleep Apnea

Phoebe K. Yu, MD, MPH[1]; Margaret B. Pulsifer, PhD[1,2]; Julie A. Grieco, PsyD[2]; Thomas B. Kinane, MD[2]; Donald G. Keamy Jr, MD, MPH[1]; Brian G. Skotko, MD, MPP[2]; Christopher J. Hartnick, MD, MS[1]

[1] Massachusetts Eye and Ear, Boston, MA [2] Massachusetts General Hospital, Boston, MA

Introduction

- Children with Down syndrome have a high incidence of obstructive sleep apnea (OSA) and few therapy options.
- Adenotonsillectomy is the first line treatment but rarely resolves OSA in children with Down syndrome, and positive airway pressure therapy is often poorly tolerated.
- Upper airway hypoglossal nerve stimulation protrudes and stiffens the tongue to open the airway and has shown efficacy in adults with OSA, but has not been evaluated in pediatric populations.
- **Objective:** To describe the efficacy of upper airway stimulation for adolescent patients with Down syndrome and severe OSA.

Methods

- **Design:** Phase 1, single-arm, multi-center clinical trial of hypoglossal nerve stimulation in 42 adolescent patients with Down syndrome and persistent severe OSA
- **Inclusion Criteria:** 10–21 years old, apnea-hypopnea index (AHI) >10 after adenotonsillectomy, positive airway pressure intolerance, no circumferential palatal collapse
- **Outcomes:** Primary outcome of change in the apnea-hypopnea index (AHI) 12 months after surgery; secondary outcomes of other polysomnogram outcomes and patient-reported outcomes (OSA-18 quality of life survey, Epworth Sleepiness Scale)

Results

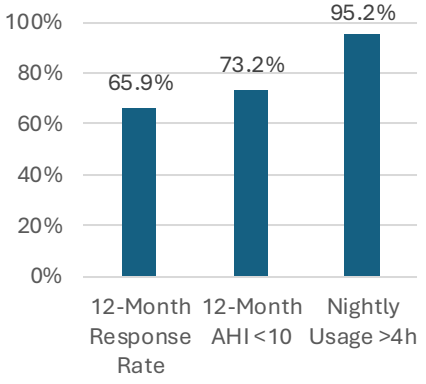
- Patient characteristics: 67% male, age 15.1 years
- The mean decrease in AHI was 12.9 events/h at 12 months (95% CI, –17.0 to –8.7 events/h).
- Quality of life on the OSA-18 improved by –34.8, and sleepiness on the Epworth improved by –5.1.
- The response rate (50% decrease in AHI) was 65.9%.
- The most common complication was temporary oral discomfort, which occurred in 5 patients (11.9%). 4 patients (9.5%) had readmissions, and 2 patients (4.8%) had reoperations.
- The mean duration of nightly therapy was 9.0 hours, with 40 patients (95.2%) using the device at least 4 hours a night.

Table 1: Polysomnographic and patient-reported outcomes.

Characteristic	Change at 12 Months	95% CI
Polysomnogram		
AHI	–12.9	–8.7, –17.0
Obstructive AHI	–12.2	–16.2, –8.2
% Time SpO2 <90%	–0.8%	–1.7%, 0.2%
SpO2 Nadir	+3.2%	1.8%, 4.7%
Patient-Reported		
OSA-18 Survey	–34.8	–42.1, –27.5
Epworth Sleepiness	–5.1	–7.4, –2.8

Summary

Fig. 1: Response and adherence rates.



- Upper airway stimulation was a safe and effective therapy for appropriately selected adolescent patients with Down syndrome and OSA.
- Objective and subjective improvements were seen at 12 months with excellent adherence.

Discussion

- Limitations: Single arm study without untreated controls, exclusion of younger children
- Our results describe a novel therapy option for treatment resistant OSA in adolescents with Down syndrome, which led to FDA approval for this indication in March 2023.

Select References: Strollo PJ et al. N Engl J Med. 2014;370(2): 139-149; Nation J, Brigger M. Otolaryngol Head Neck Surg. 2017;157(3):401-408.

We would like to acknowledge the team of investigators at the 5 sites, including: Stacey Ishman, Sally Shott, Nikhila Raol, Ryan Soose, Allison Tobey, Cristina Baldassari, Raj Dedhia