Obstructive Sleep Apnea Due To Extrathoracic Tracheomalacia

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We report obstructive sleep apnea in a 3-year-old boy with tracheomalacia secondary to tracheotomy that resolved after placement of a metallic stent in the region of tracheomalacia. The tracheal location of obstruction during sleep in this case contrasts with the usual location in the pharynx or, less often, the larynx. This case also demonstrates the utility of polysom-

nography in managing decannulation of tracheostomies.

Keywords: Obstructive sleep apnea, polysomnography, stent, tracheomalacia, tracheostomy

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The site of obstruction in obstructive sleep apnea (OSA) is the pharynx or, less often, the larynx. We describe a case of OSA secondary to localized extrathoracic tracheomalacia with resolution of OSA after airway stent placement.

REPORT OF CASE

A 30-month-old boy with mild developmental delay who had undergone tracheotomy for the management of chronic lung disease of prematurity was referred for decannulation of tracheotomy. Previous attempts had resulted in acute airway obstruction. His airway evaluation revealed nasal synechiae, adenotonsillar hypertrophy, mild subglottic narrowing, and significant suprastomal tracheal collapse from the cricoid to the stoma. Adenotonsillectomy was performed at age 36 months.

Two months later, stabilization of tracheomalacia by placing suspension sutures between the suprastomal region and the sternum was not adequate, and dyspnea with inspiratory stridor persisted. Thyroid cartilage was therefore grafted in the area of tracheomalacia.

Following these procedures, during wakefulness, he had comfortable breathing at rest with oxygen saturations of 98% and had mild intermittent stridor and respiratory distress with excitement. However, during sleep he had hypoxemia, stridor, and notable respiratory distress. Polysomnography on postoperative day 10 demonstrated severe respiratory distress, snoring, and paradoxical breathing with an apnea hypopnea index (AHI) of 6.1/h (mostly obstructive events), fragmented sleep (29 arousals/h), oxygen desaturations to 75%, and a peak end-tidal CO₂ of 63 torr. Most obstructive events occurred in REM sleep. Bronchoscopy demonstrated dynamic suprastomal collapse with 50% narrowing in the area of reconstruction (**Figure 1**).

A 2-cm long, 12-mm diameter, nickel-titanium alloy airway stent was placed in the region of tracheomalacia to manage the residual tracheal obstruction because of the potential to worsen in the setting of exercise and respiratory infections (**Figure 1**). Respiratory distress and stridor resolved after stent placement. Polysomnography 4 days later showed improvement with an

AHI of 2.5/h, predominantly central apneas, 9.8 arousals/h, and a few desaturations, mostly related to central apneas.

Three months later the AHI was 0.1/h, with 4 arousals/h and no desaturations. Bronchoscopy demonstrated a well-epithelialized stent in good position (**Figure 1**). More than 24 months later, the child has no reported obstructive symptoms.

DISCUSSION

Tracheomalacia refers to weakness of the trachea such that the airway is susceptible to collapse.¹ Sleep state-independent airway obstruction during inspiration is well known with extrathoracic tracheomalacia secondary to intraluminal pressure reduction during inspiration.¹ This child, however, had predominantly sleep state-dependent airway obstruction with worsening during REM sleep. The resolution of the obstruction after stenting of tracheomalacia with no other intervention indicates that the trachea was the only region of obstruction. To our knowledge, obstructive sleep apnea resulting from obstruction only in the extrathoracic trachea without accompanying pharyngeal or laryngeal obstruction has not been reported.

The pharynx is modeled as a Starling resistor representing a collapsible segment bounded by rigid segments upstream (nasal cavity) and downstream (trachea).² In an anatomically appropriate setting, the pharyngeal airway collapses during sleep when pharyngeal muscles that distract the airway relax, resulting in airway pressure that is less than external pressure.² Upstream resistance, such as nasal obstruction, can reduce downstream pressure, which may be compensated by increased pharyngeal motor tone in subjects who do not obstruct.³ In this subject, we posit that reduced pharyngeal diameter during sleep because of reduced pharyngeal tone increased pharyngeal resistance and induced a sufficient drop in pressure upstream of the tracheomalacia to cause airway obstruction. Resistance at the nasal synechiae and the subglottic region may have exaggerated this phenomenon. Obstruction was likely highest during REM sleep because of greatest reduction in pharyngeal tone and magnified Venturi effects from greater swings in airflow during REM.^{4,5}

Figure 1

From left to right: endoscopic view of suprastomal tracheomalacia, metal stent placement, and epithelization of stent.

Tracheomalacia secondary to tracheotomy is estimated to occur in 10% of children and could be due to pressure necrosis, disruption of blood supply, or perichondritis.¹ Anterior cricoid suspension and tracheoplasty are recommended as initial interventions in the management of suprastomal tracheomalacia because of lesser complications compared to stent placement. The potential complications with stents include obstruction by granulation tissue or stenosis, stent migration, difficulty in removal, and mucus plugging.^{1,6} The evolution of stent technology may change the management approach in the future.

Before decannulation, polysomnography with a capped tracheotomy tube has been advocated in children to detect coincidental dynamic upper airway obstruction and objectively assess ventilation in the narrow pediatric airway.⁷ This evaluation may not be able to detect airway compromise due to tracheomalacia because of stenting of the malacic area by the tracheotomy tube. After decannulation, while overnight observation with pulse oximetry is usually recommended, the use of polysomnography is not systematically addressed in the literature; the American Academy of Sleep Medicine recommends clinical assessment for recurrence of sleep related breathing disorders.⁸⁻¹⁰ However, polysomnography permits robust documentation of oximetry, respiratory distress, airway obstruction, and sleep-specific events after decannulation. In this case, polysomnography allowed demonstration of obstructive sleep apnea due to extrathoracic tracheomalacia and resolution with treatment. We therefore recommend consideration of polysomnography as an assessment tool in the immediate period after decannulation.

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