Case Report

Modified Surgical Approach to Hypoglossal Nerve Stimulator Implantation in the Pediatric Population

Sarah N. Bowe, MD; Gillian R. Diercks, MD, MPH; Christopher J. Hartnick, MD, MS Epi

Upper airway stimulation with the hypoglossal nerve stimulator is a promising treatment modality for severe obstructive sleep apnea in carefully selected patients with Down syndrome. The pediatric population presents a greater variety in body habitus, including thorax size. A modified surgical approach, utilizing a medially placed, single chest incision, instead of two separate chest incisions, provides an alternative that is particularly useful for pediatric patients with small stature. As this technology is evaluated for Food and Drug Administration clearance in the pediatric population, it is important to consider modifications in surgical technique, partnering prior surgical experience with the technical support of company representatives.

Key Words: Obstructive sleep apnea, hypoglossal nerve stimulator; upper airway stimulation, surgery, approach, technique, traditional, modified, pediatric, children, adolescents.

INTRODUCTION

The hypoglossal nerve stimulator (Inspire Medical Systems, Maple Grove, MN) is an implantable device that senses respiratory patterns and delivers electrical impulses to the medial segment of the hypoglossal nerve during inspiration, resulting in tongue protrusion. Upper airway stimulation (UAS) with this device has proven effective in managing adults with moderate to severe obstructive sleep apnea (OSA). Children with Down syndrome (DS) have a high incidence of OSA, with estimated rates between 30% and 60%. There is often persistence of disease following tonsillectomy and adenoidectomy, as well as frequent noncompliance with positive pressure ventilation. As a result, an Food and Drug Administration (FDA)-approved trial was organized to evaluate the safety and efficacy of UAS in the pediatric DS population (ClinicalTrials.gov NCT02344108). Implantation in the first patient was well tolerated, with significant improvement in OSA severity and eventual tracheostomy decannulation.

The pediatric population presents a greater variety in body habitus, including the size of the thorax. Previously, stimulator placement has been performed through three incisions. One neck incision is made for placement of the hypoglossal nerve stimulation lead, and the implantable pulse generator (IPG) and pleural sensing lead are placed through two separate chest incisions. Herein, we present our technique, utilizing one chest incision for placement of both the IPG and sensing lead. This incision has been relocated to a more medial position that provides direct access to the IPG pocket, as well as a more recognizable approach to the costal cartilage for pediatric otolaryngologists familiar with rib graft harvest. As a result, our modified surgical approach provides an alternative option that may be preferable to the traditional approach, particularly for pediatric patients with small stature.

CASE REPORT

A 10-year-old female with DS and severe OSA (apnea-hypopnea index 21 events/hour) was enrolled in the ongoing pediatric FDA trial. Her body mass index was 15.5 kg/m², placing her in the 26th percentile for her age. As noted, two separate chest incisions are used in the traditional approach (Fig. 1A). The IPG is implanted in a subcutaneous pocket in the right midinfraclavicular region, and the pleural sensing lead is placed between the internal and external intercostal muscles through an incision made in the region of the 4th to 6th intercostal spaces. Due to her size, there were concerns from the surgical staff and device technical representative that the infraclavicular incision would be located immediately over the IPG, increasing the risk of extrusion. In our patient, the lower chest incision was in close proximity to the inferior edge of the IPG pocket. As a result, there was an
opportunity to use this incision for placement of both the IPG and sensing lead (Fig. 1B).

Previously, the sensing lead incision has spanned from the anterior axillary line to the inferolateral border of the pectoralis major (Fig. 1A).\(^1\) Dissection is carried down through the serratus anterior to expose the external and internal intercostal muscles. A tunnel is extended medially between these muscles, ending at the midclavicular line. The sensor is placed facing the pleura and detects respiratory cycles via pressure variations.\(^1\) Again, due to the patient’s body habitus, concerns arose that the sensing lead would potentially suffer from ventricular motion interference due to the close proximity of cardiac structures. Therefore, the incision was relocated to a medial position, starting at the nipple line and extending 2.5 cm medial and lateral to this location (Fig. 1B). The nipple line identified the approximate location of the bony-cartilaginous junction of the rib, which served as the starting point of sensing lead placement. Dissection was carried down through the inferior aspect of the pectoralis major, exposing the external and internal intercostal muscles. A tunnel was then dissected laterally, placing the lead at a greater distance from the heart and decreasing potential interference with respiratory pressure change sensation (Fig. 2). Furthermore, the medial incision provided direct access to the pectoralis major, facilitating development of the IPG pocket superiorly.

The remaining portions of the procedure were performed in the manner described previously, although no tunneling was necessary between the sensing lead and the IPG, as they shared the same incision.\(^2\) Institutional review board approval from the Massachusetts Eye and Ear Infirmary was obtained.

**DISCUSSION**

Hypoglossal nerve stimulation offers a promising treatment modality for management of persistent severe OSA in carefully selected patients from the pediatric DS
population. Further long-term study is actively underway to evaluate the safety and efficacy of UAS over the first year after device implantation.

The current study is approved for patients with DS between 10 and 21 years old. This case report presents the youngest and smallest patient to be implanted with the Inspire UAS System. Due to the patient’s small thorax, there were notable concerns with the traditional surgical approach, including possible stress on the IPG incision with wound breakdown, as well as potential interference in pleural sensing due to ventricular motion artifact. From our experience with costal cartilage harvest, we adjusted the sensing lead incision to a more medial location, which not only provided a more familiar approach to the rib surface, but also allowed direct access to the pectoralis major for development of the IPG pocket. As a result, as this technology is evaluated for FDA clearance in the pediatric population, it is important to consider modifications to the traditional surgical approach, partnering our own surgical experience with the technical support and guidance of company representatives.

Acknowledgments
The authors sincerely thank our partner, Luke Lozier, Therapy Development Director at Inspire Medical Systems, for his technical support and guidance provided during planning and execution of this project.

BIBLIOGRAPHY