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CASE REPORTS

Space invader: pleural penetration of a hypoglossal nerve stimulator sensor lead

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The mainstay of treatment for obstructive sleep apnea is positive airway pressure therapy, which may be difficult for some patients to tolerate leading to compromised adherence and requiring alternative therapies. Hypoglossal nerve stimulation has become an option for those who meet implantation criteria. Implantation of the device is an ambulatory surgical procedure and is generally well-tolerated, though rare adverse events have been reported. We report an unusual complication of hypoglossal nerve stimulation in a patient who had initial success with this therapy. After 3 years of treatment, the sensor lead penetrated into the pleural space. Components of the hypoglossal nerve stimulation were explanted, and a new sensor lead and generator were reimplanted. The new device was activated, and therapy was successfully resumed. This case demonstrates that there is a potential for a delayed complication of sensor lead penetration into the pleural space, which has only rarely been reported.

Keywords: obstructive sleep apnea, hypoglossal nerve stimulator, upper airway stimulator, adverse events, pleura

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INTRODUCTION

Obstructive sleep apnea (OSA) affects millions worldwide and causes not only excessive daytime sleepiness but also detrimental effects on cardiovascular health, neurocognitive performance,[1] mood[2] and quality of life. It is defined as repetitive collapse of the upper airway during sleep, most commonly in the retropalatal and retrolingual regions.[3] Positive airway pressure is the mainstay of OSA therapy. However for patients who have difficulty tolerating positive airway pressure therapy, hypoglossal nerve stimulation (HGNS) has become a feasible alternative. The Inspire HGNS (Inspire Medical Systems, Golden Valley, MN) consists of a generator that is implanted subcutaneously in the upper anterior chest, an electrode cuff that is placed on the distal branches of hypoglossal nerve, and a pressure sensor, which is inserted between the intercostal muscles to detect inspiratory effort, triggering the stimulator.[4] The implantation is performed as an ambulatory surgical procedure and is generally well tolerated. Although postoperative complications can occur, there is a paucity of reports in the literature that describe them. We report a case of pressure sensor penetration into the pleural space 3 years into therapy and subsequent successful surgical correction.

REPORT OF CASE

An 86-year-old man presented with frequent nocturnal awakenings as well as daytime fatigue and sleepiness. He was diagnosed with severe obstructive sleep apnea by polysomnography, which demonstrated an apnea-hypopnea index of 37.3 events/h, comprising obstructive apneas and hypopneas. Other medical history included chronic kidney disease stage 3, which did not require dialysis. In general, the patient had an active lifestyle though excessive daytime sleepiness often interfered with daily activities. The patient was treated with continuous positive airway pressure. However, he could not tolerate this therapy due to chronic nasal congestion and mask discomfort, and the patient expressed interest in pursuing HGNS therapy. His body mass index was 26 kg/m^2, and subsequent drug-induced sleep endoscopy (DISE) showed anterior-posterior collapse of his airway. Having met the inclusion criteria for HGNS by body mass index, severity of obstructive sleep apnea, age, and appropriate anatomy under DISE exam, he underwent implantation of the Inspire device without complication. As per standard protocol, 1 month after implantation, electrode impedances and sensor lead function were checked and found to be within normal parameters. Sensory and functional thresholds were determined with appropriate signals from the sensor observed and adequate anterior tongue movement. Therapy was initiated. After several adjustments, the patient had an excellent response to HGNS with improvement of apnea-hypopnea index to 9.7 events/h and resolution of OSA-related symptoms. He continued with nightly therapy and was seen at 6-month intervals with assessments of the medical device including impedances and sensor lead function. No abnormalities were noted on these routine evaluations.

After about 3 years of HGNS therapy, patient complained of pruritis on his right chest wall at the location of the pressure sensor, often scratching the area, and over a 2-week time period, he noticed the gradual appearance of a nontender and immobile,

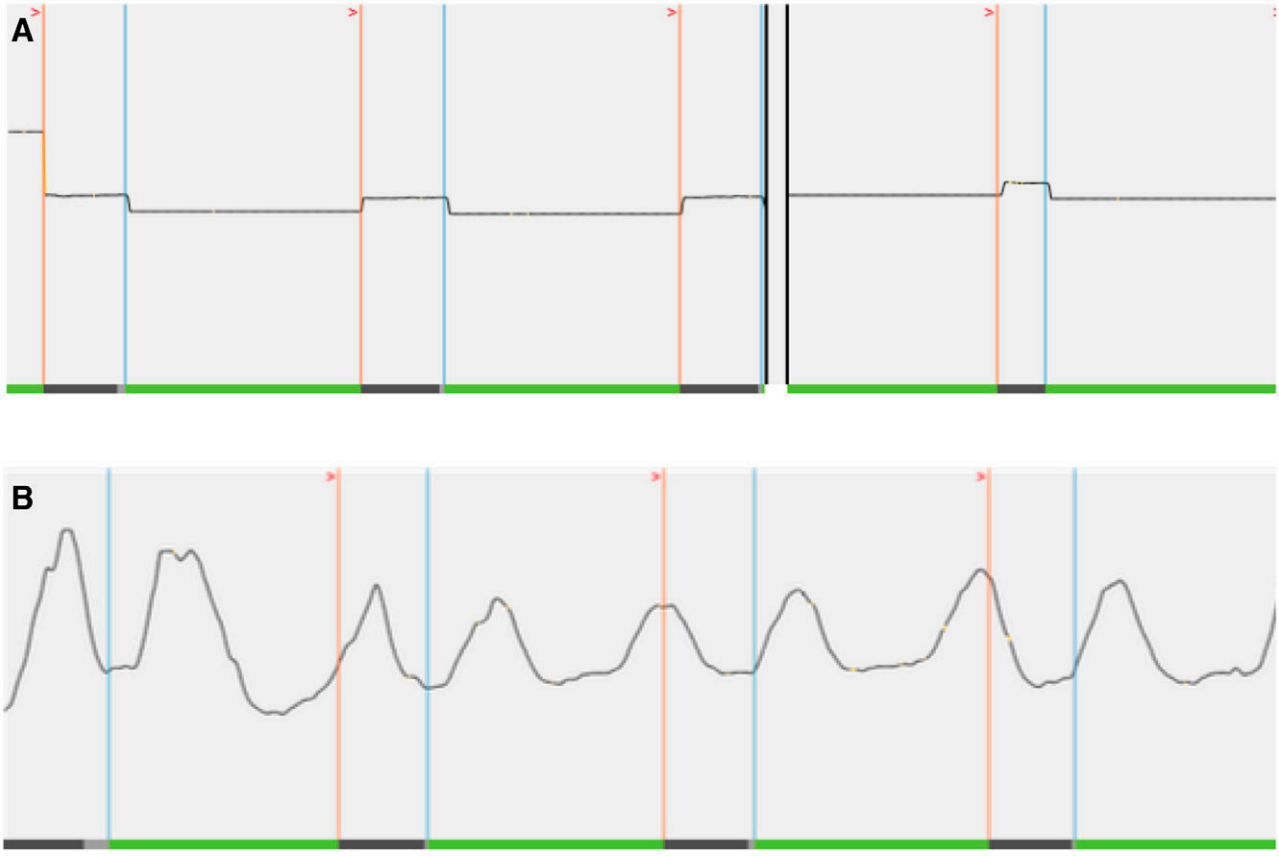
superficial mass. Though patient had an active lifestyle, he reported no impacts or athletic activities that would strain this area. He denied chest pain, shortness of breath, hemoptysis, fever, chills, and unintentional weight loss. On physical examination the mass did not appear erythematous or indurated. The patient reported no difference in his sleep quality, alertness during the day, and sensation of device stimulation. The HGNS was evaluated, and blunting of the sensor lead signal was observed (**Figure 1A**) although continued clinical resolution of OSA symptoms and appropriate anterior tongue motion with stimulation was noted. A computed tomography scan of the chest showed that the sensor lead from the HGNS had penetrated through the intercostal muscles and entered the pleural space. Coiling of the wire in the subcutaneous tissue was observed with a heterogenous, asymmetric mass (**Figure 2**) surrounding the wire. Because of potential risk of pleural infection and pneumothorax, surgical repair with removal and replacement of the device was discussed with the patient. The patient was agreeable to replacing the HGNS, and otolaryngology was asked to reevaluate the Inspire device.

Surgery was performed to replace the sensor lead and the pulse generator was replaced at the same time only to upgrade the device. A new sensor lead was placed within the fourth intercostal space, and the old sensor lead was coiled and surrounded by reactive tissue with the probe itself extending down between the ribs and into the pleural space. The old sensor lead within the fifth intercostal space was carefully dissected from the fibrous tissue. Sutures that were holding the lead in place were intact and subsequently cut and removed. Slowly the sensor lead was removed from the pleural space with no pneumothorax or evidence of air leakage. There were no complications from the removal. The generator was also removed from fibrous tissue and replaced without incident. The new sensor lead and the prior stimulation lead were connected to the new generator and tested intraoperatively with good performance. The patient returned to clinic for reactivation of HGNS 1 month after the second implantation. Sensor lead function and impedances were within acceptable parameters (**Figure 1B**). Prior HGNS settings were programmed into the new generator and tested with appropriate tongue protrusion. The patient has since been doing well on these settings with continued resolution of his OSA-related symptoms.

DISCUSSION

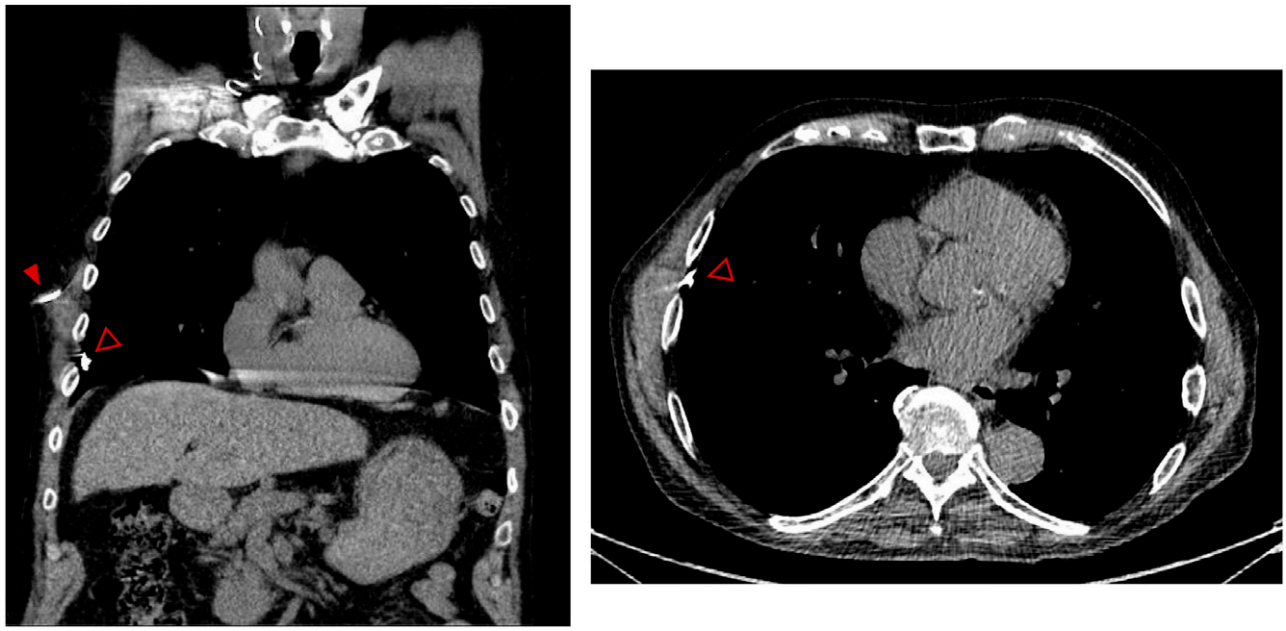
The Inspire hypoglossal nerve stimulator was approved by the US Food and Drug Administration for treatment of OSA in 2014 and

Figure 1—Sensor lead testing of the HGNS.



(A) Sensor lead within the pleural space. Sensor lead testing of the HGNS after patient had noticed a superficial lesion on his chest. The sensing by the HGNS has been blunted with each breath. **(B)** Sensor lead within the intercostal space. Sensory testing of the HGNS after replacement of sensor lead, which shows appropriate pattern and detection of respiration. HGNS = hypoglossal nerve stimulator.

Figure 2—Chest CT of the HGNS.



CT of the chest demonstrates a superficial mass from coiling of the wire (closed red arrowhead) with penetration of the sensor lead through the intercostal muscle into the pleural space (open red arrowheads) in both coronal and axial views. CT = computed tomography, HGNS = hypoglossal nerve stimulator.

is rapidly becoming an accepted therapeutic alternative for patients who are intolerant of positive airway pressure or other traditional therapies. Though more invasive than traditional therapies and requiring surgery under general anesthesia for implantation, HGNS is an effective therapy that can reduce both apnea-hypopnea index as well as improve excessive daytime sleepiness and other OSA-related symptoms. These effects are sustained up to 5 years postimplantation.[5,6] There are 3 components of the device, which includes the generator, the sensor lead, and the stimulation/electrode cuff. The sensor lead is a small pressure transducer inserted between the internal and external intercostal muscles, typically in the fifth intercostal space. It has the ability to sense inhalation. The stimulation lead with an electrode cuff is placed distally on the medial division of the hypoglossal nerve in order to innervate the genioglossus muscle. The generator coordinates inspiratory effort detected by the pressure sensor with activation of the hypoglossal nerve stimulator, thus maintaining upper airway patency.[4]

The implantation procedure is generally well-tolerated and can be done in an ambulatory setting. Initial reports of adverse events occurred in less than 2% of cases and occurred within the first 30 days post-implantation. They were usually mild, including pain at the incision site and muscle soreness.[5] Most resolved spontaneously and did not require surgical intervention. Since Food and Drug Administration approval of HGNS, there have been additional reports of more serious adverse events. Implantable medical devices, particularly electronic stimulation devices in other parts of the body such as the occipital nerve stimulator or the trigeminal nerve stimulator, can potentially have mechanical complications such as hematoma, lead fracture, and lead migration.[7] A recent retrospective review examined HGNS complications with the most common adverse event being infection, though hematoma, device migration, hypoglossal nerve palsy, pain, and fractured leads were also reported though more rarely. Many of these complications required surgical revision or complete explantation of the device.[8] However, current listed potential adverse events on the Food and Drug Administration Summary of Safety and Effectiveness template for HGNS does not include lead penetration into the pleural space.[9] This case adds pleural space penetration to this body of literature. The pleural space is sterile, and any foreign bodies within the space can lead to potential pneumothorax, hemothorax, and pleural infection. In our patient, there may have been several factors that could have contributed to pleural invasion of the sensor lead, including age and chronic kidney disease—both of which can lead to muscle weakness and sarcopenia.[10] The atrophy of the intercostal muscles may have made the site more vulnerable to penetration. Additionally, the patient was manipulating the site prior to the appearance of the right chest wall mass, suggesting that the repetitive scratching may have caused lead migration. Unconscious or conscious movements by the patient may unintentionally manipulate migration of the lead, which is known as twiddler's syndrome. Similar cases of lead dislodgement and even lead fractures have been reported in other implantable devices such as deep brain stimulators,[11] implantable cardiac devices,[12] and even HGNS in which twiddler's syndrome caused a 270-degree rotation of the implantable pulse generator resulting in malfunctioning of the device.[13]

The need for explantation and reimplantation of HGNS components is not common but at times may be necessary, such

as in the case of infection. These procedures tend to be technically challenging due to fibrous and extensive scar tissue formation, with reimplantation being associated with a higher complication rate.[14] Caution is needed particularly when explanting the stimulator lead as the electrode cuff is in close proximity to the hypoglossal nerve, and improper removal can potentially cause nerve damage.

In the present case, the patient did not tolerate positive airway pressure therapy, and the benefit of continued OSA treatment with HGNS as well as preventing potential infection and complications of a foreign body within the pleural space, outweighed the risks of explantation and reimplantation. The patient's procedure was successful and without complications.

Potential adverse events should be checked in the immediate postoperative period, initial activation and at every subsequent follow up visit. Abnormal sensor lead signaling, such as blunting as seen with our patient, can be a sign of lead fracture or penetration, and should signal to the clinician that further investigation of HGNS may be warranted. As HGNS becomes more widely used as an alternative therapy for OSA, it is important for clinicians to be aware of benefits as well as risks of this therapy, including the risk of adverse events—both immediate and delayed—that may require surgical correction.

ABBREVIATIONS

HGNS, hypoglossal nerve stimulator
OSA, obstructive sleep apnea

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DISCLOSURE STATEMENT

All authors have seen and approved the manuscript. Work for this study was performed at the Donald and Barbara Zucker School of Medicine at Hofstra-Northwell. The authors report no conflicts of interest.