CASE REPORT

Hypoglossal nerve stimulation for obstructive sleep apnoea in a patient with previous tongue laceration

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SUMMARY

Upper airway stimulation of the tongue using an implantable neurostimulator has recently been approved for select patients with moderate to severe obstructive sleep apnoea (OSA) and intolerance to continuous positive airway pressure therapy. Effective implantation depends on the integrity of the hypoglossal nerve as well as the tongue musculature, notably the genioglossus. Prior trauma to either of these structures may be viewed as a relative contraindication to implantation. We describe a case of successful right hypoglossal nerve implantation in a patient with a history of left cardiac pacemaker placement and severe left penetrating tongue trauma with decreased mobility from contracture and deviation mimicking a hypoglossal nerve palsy. Preoperative and postoperative apnoea–hypopnoea index values were 52/hour and 5/hour, respectively. Prior soft tissue trauma to the tongue may not necessarily preclude surgical candidacy for upper airway stimulation in patients with OSA.

BACKGROUND

Hypoglossal nerve stimulation using an implantable neurostimulator has recently been approved for select patients with continuous positive airway pressure (CPAP) intolerance and moderate to severe obstructive sleep apnoea (OSA). Effective implantation depends on the integrity of the hypoglossal nerve and the tongue musculature. Prior trauma to either of these structures may be viewed as a relative contraindication to implantation. We present a case of successful hypoglossal nerve stimulator implantation in a 66-year-old man with severe OSA and history of severe penetrating tongue trauma resulting in left tongue deviation and decreased range of motion. This case report was granted exemption of review by the University of Kansas Institutional Review Board.

CASE PRESENTATION

A 66-year-old man presented to the otolaryngology clinic for surgical evaluation of his OSA. He reported that he was no longer using his CPAP machine and complained of poor tolerance for over 1 year due to persistent claustrophobia despite trying various masks. His surgical history was notable for left-sided cardiac pacemaker placement and hypertension. Physical examination revealed an overweight patient (body mass index of 29 kg/m²) and decreased tongue mobility due to scarring and contracture as well as deviation to the left of midline with possible

injury to terminal hypoglossal nerve branches or the intrinsic tongue musculature (figure 1). The patient reported that he had sustained a penetrating tongue laceration after a motor vehicle accident 50 years ago. A prior inpatient polysomnography study had been completed and indicated severe OSA, showing a baseline apnoea–hypopnoea index (AHI) of 52/hour with only four total central apnoeas and an oxygen saturation nadir of 62%.

INVESTIGATIONS

Drug-induced sleep endoscopy (DISE) was undertaken in the operating room. During DISE, patients are administered sedatives (eg, propofol) under monitored anaesthesia care to simulate sleep. Flexible nasopharyngolaryngoscopy is then performed to evaluate for patterns of collapsible upper airway obstruction. DISE for this patient revealed complete anterior–posterior collapse at the level of the velopharynx and base of the tongue with partial anterior–posterior collapse at the level of the epiglottis. Due to his sleep study parameters, inability to tolerate CPAP and findings of retroglossal collapse with lack of complete concentric collapse at the velopharynx on DISE, the patient was deemed eligible for hypoglossal nerve stimulation.

TREATMENT

The patient underwent hypoglossal nerve stimulator implantation using the Inspire upper airway stimulation system (Inspire Medical Systems, Minnesota, USA) on the right side without complication. The device prevents obstructive apnoeic events using an implantable, programmable pulse generator, a respiratory sensing lead and a neurostimulating electrode. The device is activated by the patient using a handheld remote prior to sleep. An intercostally inserted respiratory lead senses inspiration during sleep and signals the implantable pulse generator to activate the hypoglossal nerve using a stimulating electrode attached to the distal division of the hypoglossal nerve. The stimulating electrode is placed onto the hypoglossal nerve first through a cervical incision, followed by separate anterior and lateral chest wall incisions for placement of the generator and respiratory sensing lead, respectively. The generator is secured over the pectoral fascia and the respiratory lead is placed between the internal and external intercostal muscles. Both leads are then tunnelled subcutaneously to the anterior chest wall and connected to the generator.

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Figure 1 Examination revealed scarring and contracture at the left lateral and mid tongue with mild deviation to the left, mimicking a hypoglossal nerve palsy.

The hypoglossal nerve innervates both the intrinsic and extrinsic muscles of the tongue, which are responsible for its complex movements during mastication as well as gross protrusion and retraction. It is important that only tongue protrusion results from stimulation as retraction can result in obstruction during sleep. Thus, the optimal site of stimulation is found at a functional breakpoint along the nerve that separates retraction and protrusion. The distal portion of the nerve is largely responsible for tongue protrusion and passive opening of the soft palate through innervation of the genioglossus muscle and the intrinsic tongue musculature. Bipolar stimulation and monitoring of the nerve are also required during the procedure to prevent lead placement onto branches of the nerve that innervate the styloglossus and hyoglossus muscles, which cause depression and retraction.¹²

OUTCOME AND FOLLOW-UP

Postoperatively, hypoglossal nerve stimulation was titrated to 0.8 volts, where it generated right-sided tongue base protrusion (approximately 1 mm past the front teeth) and was therapeutically successful (figure 2) despite his previous tongue trauma. The patient was provided with a handheld remote to self-titrate the amplitude upwards based on fatigue and snoring symptoms for the next 30 days. The patient then came in for an overnight sleep study to confirm the therapeutic amplitude. At the titration visit, he reported subjective improvements in daytime fatigue and his Epworth Sleepiness Scale was reduced from nine preoperatively to six. Therapy compliance was high with 60 hours of usage per week. During this overnight titration study, the therapy amplitude was adjusted upwards from the minimum amplitude to find the therapeutic value, similar to a CPAP titration. The therapeutic value was determined to be 0.9 volts. At this amplitude, his AHI was reduced to five events per hour. The patient was instructed to continue efforts to exercise and maintain a healthy diet as future weight gain could worsen his sleep apnoea and impair device effectiveness.

DISCUSSION

OSA results from intermittent upper airway collapse due to decreased muscle tone and tissue redundancy. The chronic episodic hypoxaemia and arousals from sleep lead to daytime fatigue or somnolence despite satisfactory sleep hygiene and

Figure 2 Flexible laryngoscopy shows increased patency at the level of the tongue base during awake hypoglossal nerve stimulation.

schedule. Secondary sequelae of the condition include hypertension, cardiovascular disease and an increased risk of motor vehicle accidents. Obesity represents a main risk factor for OSA and its rising rates in the USA have been attributed to an increased prevalence of OSA.³

Following the Stimulation Therapy for Apnea Reduction (STAR) trial results, upper airway stimulation of the hypoglossal nerve using an implantable neurostimulator was approved in the USA for select patients with CPAP intolerance and moderate to severe OSA.⁴ Patients should have a body mass index less than 32 kg/m² and also demonstrate a lack of complete concentric or lateral collapse at the level of the velopharynx on DISE, as this finding has been associated with suboptimal AHI reduction following device implantation.⁵

The integrity of the hypoglossal nerve and functional activity of the intrinsic and extrinsic tongue musculature are critical to successful implantation. The neurostimulator is typically placed on the patient's right and relies on unilateral tongue protrusion through ipsilateral activation of the hypoglossal nerve. Our patient's tongue trauma was viewed as a possible contraindication to implantation as there were concerns that the decreased range of tongue motion and deviation to the left would prevent sufficient tongue protrusion on the stimulated side, and thus limit effectiveness of the device. A left-sided procedure was also considered, but was not performed due to the possibility of a pre-existing hypoglossal weakness and because of the patient's prior placement of a cardiac pacemaker, whose generator would be at risk for injury during surgery and could cause or receive device interference. The risk of a bilateral hypoglossal palsy had been discussed with the patient prior to implantation and no hypoglossal weakness on the right was noted postoperatively. We were able to titrate stimulation parameters to produce right-sided tongue protrusion, and achieve a desirable reduction in the patient's AHI and high therapy adherence. Thus, pre-existing contracture and deviation of the tongue may not necessarily preclude surgical candidacy for upper airway stimulation in patients with OSA.

Learning points

- ▶ Hypoglossal nerve stimulation is indicated in patients with obstructive sleep apnoea with continuous positive airway pressure intolerance and moderate to severe (apnoea–hypopnoea index 15–65/hour) sleep apnoea.
- ▶ Complete concentric collapse of the velopharynx on drug-induced sleep endoscopy is considered an absolute contraindication to device implantation.
- ▶ The integrity of the hypoglossal nerve and functional activity of the intrinsic and extrinsic tongue musculature are critical for successful upper airway stimulation.
- ▶ Prior tongue trauma with contracture and impaired mobility may be viewed as a relative contraindication to neurostimulator implantation.
- ▶ Neurostimulator parameters can be titrated to achieve a successful outcome when contralateral tongue contracture and deviation are present.

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