The “vagal ansa”: a source of complication in vagus nerve stimulation

Chittur Viswanathan Gopalakrishnan, MS, MCh,¹ John R. W. Kestle, MD,¹ and Mary B. Connolly, MB, FRCP(C)²

¹Division of Neurosurgery, Department of Surgery, University of British Columbia and BC Children's Hospital; and ²Division of Neurology, Department of Paediatrics, University of British Columbia and BC Children's Hospital, Vancouver, British Columbia, Canada

A 16-year-old boy underwent vagus nerve stimulation for treatment-resistant multifocal epilepsy. During intraoperative system diagnostics, vigorous contraction of the ipsilateral sternomastoid muscle was observed. On re-exploration, a thin nerve fiber passing from the vagus to the sternomastoid was found hooked up in the upper electrode. Detailed inspection revealed an abnormal course of the superior root of the ansa cervicalis, which descended down as a single nerve trunk with the vagus and separated to join the inferior root. The authors discuss the variation in the course of the ansa cervicalis and how this could be a reason for postoperative neck muscle contractions.

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THE ansa cervicalis, located in the anterior triangle of the neck, is formed by the anterior rami of the first 3 or 4 cervical spinal nerves. This nerve loop innervates the infrahyoid muscles.⁵ It is frequently encountered during surgical procedures of the neck. Variations in origin, branching patterns, course, and innervation to the muscles have been described.¹¹ This makes it important for surgeons who operate in and around this area to be familiar with the anatomy.^{1,15}

Vagus nerve stimulation (VNS) is a palliative procedure for patients with treatment-resistant epilepsy and has shown a positive benefit in reducing seizure frequency. There have been complications reported either due to the surgical procedure as such or due to hardware-related problems.^{6,8,14,16} Side effects are mainly related to stimulation, are usually reversible, and tend to decrease over time. They seldom necessitate the removal of the device. Some of the common transient symptoms described include hoarseness, cough, and dysphagia, which are usually well tolerated.

We describe an intraoperative complication noticed during testing of the VNS system after placement of the electrodes and the pulse generator. To the best of our knowledge, it has not been previously reported. We also discuss the physiology behind its occurrence and precautions to be observed in the future.

Case Report

A 16-year-old male patient presented with treatment-resistant multifocal epilepsy following encephalopathy at 14.5 years of age. Extensive investigations for an infectious, inflammatory, and immune etiology were negative. Findings on MRI were initially normal and evolved to show increased T2 signal in the left hippocampus, bilateral periventricular regions, left thalamus, and right occipital lobe. Video-encephalographic monitoring showed multifocal onset of seizure activity arising independently in both hemispheres. A brain biopsy was recommended, but the family declined. The patient's seizures did not re-

ABBREVIATION VNS = vagus nerve stimulation.

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spond to any of the antiepileptic medications, and gradually he developed frequent episodes of nonconvulsive status epilepticus. VNS was therefore recommended. We follow the standard procedure for implantation of the VNS Therapy system (Cyberonics Inc.). Once the carotid sheath is entered, the nerve is identified and prepared for approximately 3 cm. The nerve is held up using a vessel loop while the electrode and anchor helices are applied. The lead is tunneled subcutaneously to the chest and connected to the pulse generator. The intraoperative system diagnostics (lead test) is performed prior to closure of the incisions. In this case, during the test, we observed rapid contractions of the sternomastoid muscle. This prompted us to explore the electrode implantation site again, and we found that a thin nerve fiber passing from the vagus to the mid-belly of the sternomastoid muscle was becoming entangled with the upper electrode (Fig. 1). Once the nerve fiber was released, we could appreciate that the superior root of the ansa cervicalis was closely approximated to the vagus proximal to the electrode site. The nerve fiber to the sternomastoid appeared to arise from the fused “vagal ansa” segment and was inadvertently hooked up in the upper electrode, causing contractions during stimulation. Once this nerve was released, further testing did not reveal any contractions in the muscle belly. The postoperative course was uneventful, and the patient was discharged the same day.

Six months following implantation, there has not been a significant decrease in the frequency of the patient's seizures, but their duration has decreased.

Discussion

The ansa cervicalis consists of 2 roots: the superior and the inferior. The superior root, also called descendens hypoglossi, is constituted by the ventral ramus of C-1, whereas the inferior root, also known as descendens cervicalis, is usually derived from the ventral rami of C-2 and C-3. The loop formed by the union of these roots is called the ansa cervicalis, the Latin term “ansa” meaning “handle of a cup.” This loop innervates the infrahyoid muscles, which play a major role in swallowing and phonation.¹³ In human specimens, the ansa cervicalis shows a great degree of variation in its origin and distribution.^{7,11}

The fibers from C-1 run with the hypoglossal nerve for a distance of about 3–4 cm and then separate where the hypoglossal nerve turns anteriorly around the origin of the occipital artery to descend as the superior root. This root generally courses in front of the external carotid artery but can also run in front of the internal carotid artery.^{4,11} The superior root joins the inferior root, formed by the fibers of C2–3, at the level of the omohyoid tendon on the lateral surface of the internal jugular vein to form the ansa cervicalis loop (Fig. 2). The inferior root may lie medial to the internal jugular vein in roughly 15% of the cases.⁴ Variability in the origin of inferior root of the loop has been frequently reported in literature.^{4,7,11,12}

The sternomastoid is supplied by the accessory nerve and by branches from the ventral rami of the second, third, and sometimes the fourth cervical spinal nerves. Distribution of fibers from the ansa is variable, but usually the

FIG. 1. Line drawing demonstrating the variation in the course of the ansa cervicalis. Note the branch from the vagal-ansa trunk supplying the sternomastoid caught in the upper electrode. X = cranial nerve X (vagus nerve).

FIG. 2. Line drawing showing the most frequent morphological anatomy of the ansa cervicalis nerve loop. m. = muscle; mm. = muscles; X = cranial nerve (CN) X (vagus nerve); XII = CN XII (hypoglossal nerve).

superior root innervates the superior belly of the omohyoid, and occasionally it may send a branch to the sternomastoid, thyrohyoid, and upper portions of the sternohyoid and sternothyroid. The inferior root gives off branches to the inferior belly of the omohyoid muscle and the lower parts of the sternohyoid and sternothyroid muscles.^{2,10}

The vagus nerve, after exiting the jugular foramen, runs vertically down within the carotid sheath, first lying between the internal jugular vein and internal carotid artery and then between the same vein and the common carotid artery. Thus, it is situated in close proximity to the ansa cervicalis and its branches.

Communications between the ansa cervicalis and the vagus nerve have been reported frequently in the dissection room. In a detailed analysis by Banneheka et al.,³ microscopic dissection of these communications revealed false or pseudo-communications consisting of only connective tissue and true communications involving nerve fiber exchange. Kikuchi⁹ coined the term “vagal ansa” to describe a situation in which the superior root of the ansa cervicalis descends as a common trunk with the vagus. Fibers of the inferior root of the ansa, which to the naked eye seem to be communicating with the vagus nerve, are actually in communication with the superior root fibers traversing with the vagus. Therefore, the so-called vagal innervation of the infrahyoid muscles is a misnomer and actually represents true innervation from the ventral cervical rami. In our patient, the “vagal ansa” appeared to provide a motor branch to the sternomastoid resulting in contraction with stimulation.

This article highlights the importance of knowing the anatomy of the ansa cervicalis though it is not commonly encountered during implantation of a VNS device. Variations in its anatomy are being increasingly reported due to its popularity in reinnervation techniques. It is the ideal

nerve for nerve reconstruction in the neck because sacrificing the ansa does not cause serious functional or cosmetic sequelae. Quantitative data on the recurrent motor fibers within the ansa cervicalis nerve loop structure are lacking.⁵ The fact that the superior root can occur with the vagus nerve in a single connective tissue sheath makes it prudent to identify the anatomy prior to electrode insertion. It is imperative that we keep an eye on the surgical site during intraoperative testing of the device and look for contractions in the infrahyoid musculature, which could be an annoying complication, if present, after surgery. Occurrence of such contractions makes it necessary to determine whether the ansa or any of its branches is hooked up by the electrodes and gradually release them. The fact that such a complication has not been reported earlier in literature, though VNS is commonly performed worldwide, reiterates the fact that it is a rare event, but at the same time it cannot be ignored.

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Author Contributions

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Correspondence

Chittur Viswanathan Gopalakrishnan, Division of Pediatric Neurosurgery, BC Children's Hospital, 4480 Oak St., Rm. K3-159, Vancouver, BC V6H 3V4, Canada. email: doc_gopal@yahoo.com.