Multiple Cranial Neuropathies and First Bite Syndrome after Carotid Endarterectomy
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Abstract

First Bite Syndrome (FBS) is characterized by severe pain in the parotid region after the first bite of a meal. It is a well-described complication of carotid body tumor, cervical schwannoma, and other parapharyngeal space surgery.\textsuperscript{1-3}

We present the first case report of multiple cranial neuropathies, as well as, First Bite Syndrome following carotid endarterectomy (CEA). A 70 year-old man developed left facial weakness, hoarseness and severe pain localizing to the left parotid gland, ear, and pharynx following CEA. He was diagnosed with a paresis of the left marginal mandibular branch of the facial nerve, a left vocal fold paresis with severe glottic incompetence, and FBS. The patient was treated with injection laryngoplasty and speech therapy for a left vocal fold paresis, botulinum toxin (BNT) injection of the parotid gland for the FBS, with significant improvement but not complete resolution of his symptoms 17 months after the CEA.

Introduction

First Bite Syndrome (FBS) is characterized by severe pain in the parotid region after the first bite of a meal. The etiology of FBS is unknown, however it is thought to be the result of the loss of sympathetic innervation to the parotid gland, with a subsequent denervation supersensitivity related to uninhibited parasympathetic stimulation. Symptoms typically last a few seconds and improve with each subsequent masticatory movement, but recur with each subsequent meal. The pain may radiate to the jaw or ear, and may occur when salivating or thinking about food.\textsuperscript{4} Symptoms vary from mild to severe, and can result in severe disability and weight loss.

Case Report

A 70 year-old male with carotid stenosis underwent a left CEA. The vascular surgeon noted that the carotid bulb was high, and partial transection of the posterior belly of the digastric muscle with mobilization of the hypoglossal nerve was necessary to obtain adequate exposure.

Postoperatively, the patient reported persistent severe (10/10) left sided facial pain during the first few bites of meals. The patient also reported hoarseness, left lower lip weakness, and post-prandial left-sided otalgia. Left marginal mandibular weakness was noted on physical examination. Videostroscopy revealed a left vocal fold paresis (Figure 1) with severe glottic incompetence, and hypersensitivity of the left hemilarynx. Laryngeal EMG revealed an absence of fibrillations or fasciculations and giant wave activity was noted. A MRI of the neck with contrast revealed only post-operative changes.

The patient was treated with gabapentin and opioid analgesics without improvement of his FBS symptoms for three months after the CEA. He noted a 20 lb weight loss, and expressed extreme emotional distress. He underwent ultrasound guided abobotulinumtoxinA (Dysport\textsuperscript{TM}) injection of 280 units into the superficial and deep lobes of the parotid gland with a subsequent reduction in pain (1/10). No facial weakness or dry mouth was noted. Five months later, he noted a return of the pain (4/10) and was injected with 50 units of onabotulinumtoxinA (Botox\textsuperscript{®}), with a similar reduction in pain (1/10). Seventeen months post-operatively, he reports 2/10 pain, when eating fruits.

Discussion

First Bite Syndrome is a known postoperative complication of parapharyngeal space surgery due to damage to the cervical sympathetic chain. Although the exact etiology is unknown, Netterville proposed that FBS is due to loss of sympathetic innervation to the myoepithelial cells of the parotid gland, with a subsequent denervation hypersensitivity related to uninhibited parasympathetic stimulation and myoepithelial spasm.\textsuperscript{5,6}

BNT injection of the parotid gland has previously been reported to be an effective treatment for FBS.\textsuperscript{7}

BNT has been postulated to ameliorate the symptoms of FBS through the inhibition of acetylcholine degranulation into the synaptic cleft of motor neurons, thereby selectively paralyzing the myoepithelial fibers. In fact, Kawashima et al\textsuperscript{8} reported absence of FBS in patients who had loss of both sympathetic and parasympathetic innervation. Although An important additional pathway for BNT pain relief in FBS may also involve inhibition of substance P, CGRP, or glutamate from afferent nerve terminals, which has been demonstrated in animal studies.\textsuperscript{9}

Conclusion

We report the first case of multiple cranial neuropathies and FBS following CEA. FBS is an uncommon complication of CEA which should be considered when parotid pain associated with eating is noted. BNT is a new treatment modality for FBS that shows promise, although the exact mechanism of action is poorly understood.

References

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