CASE REPORT

First bite syndrome: a complication of carotid endarterectomy

Mary Jiayi Tao, Graham Roche-Nagle

Toronto General Hospital,
Toronto, Ontario, Canada**Correspondence to**Dr Graham Roche-Nagle,
graham.roche-nagle@uhn.ca

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SUMMARY

First bite syndrome (FBS) is an uncommon pain syndrome characterised by severe cramping or spasm in the parotid region with the first bite of each meal. The proposed pathogenesis is sympathetic denervation of the parotid gland secondary to iatrogenic injury with resultant cross-stimulatory parasympathetic hypersensitivity response. FBS is a potential sequela of surgeries involving the infratemporal fossa, parapharyngeal space and/or deep lobe of the parotid gland, however, only four cases of FBS secondary to carotid endarterectomy have been documented to date. We present a case and management of a 77-year-old man who developed FBS after an ipsilateral carotid endarterectomy, to raise awareness of this complication among surgeons who operate in the neck region.

BACKGROUND

First bite syndrome (FBS) is a relatively uncommon entity characterised by sudden onset of intense pain localised to the parotid region. The pain is triggered by the first bite of each meal and it typically lasts for a few seconds. Although it gradually disappears with subsequent mastication, it will recur at the first bite of the next meal. This clinical syndrome has been described as a complication of parapharyngeal space surgeries and parapharyngeal space tumours. Presently, only four cases of FBS secondary to carotid endarterectomy (CEA) have been documented in the literature. Given the high rate of CEA procedures performed worldwide, surgeons who operate in the neck region should be aware of this complication and should consider FBS a potential consequence of injury to the sympathetic chain or plexus.

CASE PRESENTATION

A 77-year-old man presented with a history of left-sided amaurosis fugax while on holiday. An urgent carotid duplex demonstrated a 95–99% stenosis of the left proximal internal carotid artery (ICA). He agreed to a left CEA under general anaesthetic. At operation, he was found to have extensive internal carotid disease extending high above the common carotid artery bifurcation. Dissection continued cephalad, necessitating retraction of the digastric muscle. Arteriotomy demonstrated a lengthy plaque containing a tight stenosis and plaque ulceration. This was duly endarterectomised following shunt insertion and a bovine pericardial patch was placed. The patient recovered well and was discharged home day 1 after surgery.

At his 3-week clinic follow-up, his wound was healed and he had no new neurological symptom. However, he did report sharp pain at the ipsilateral parotid region radiating to the ear occurring after the first bite of food, lasting 10–15 s, and resolving despite further eating. This started a few days after the operation. The symptoms repeated when starting to eat food and on occasion the patient documented the smell of certain foods such as strawberries triggering the symptoms. Physical examination was normal.

TREATMENT

After a literature review and discussion with ear, nose and throat colleagues, a diagnosis of FBS was made. Treatment options were discussed and the patient wished to be managed conservatively.

OUTCOME AND FOLLOW-UP

The patient's symptoms resolved at 8 months.

DISCUSSION

FBS is a relatively uncommon entity characterised by sudden onset of intense pain localised to the parotid region. The pain may radiate through the upper neck and auricular regions and will typically last 5 s before gradually disappearing with subsequent mastication, and will invariably recur at the first bite of the next meal.¹ The clinical syndrome characteristically presents 5–7 days postoperatively on the ipsilateral surgical side.² The symptoms are typically the worst with the first meal of the day and are precipitated by sialagogues such as bitter food and citrus fruits.^{3–4} The pain can be so debilitating that it may interfere with an individual's ability to eat and consequently their quality of life.²

The current description of FBS was defined by Netterville *et al*,⁵ in 1998, as a postoperative complication of parapharyngeal space surgeries and parapharyngeal space tumour. Presently, only four cases of FBS secondary to CEA have been documented in the literature.^{3–6–8}

Two main aetiological factors have been identified thus far. The main class being postoperative syndromes, as more than 95% of published cases of FBS correspond to complications following surgeries,⁴ particularly parapharyngeal space dissections, resection of the deep lobe of the parotid gland and sacrifice of the sympathetic chain.⁹ In fact, Linkov *et al*⁹ reported a 9.6% (n=45) cumulative incidence in 499 patients who had undergone such procedures, and found deep lobe parotidectomy to be the most common cause of FBS in this cohort. Malignant tumours arising from the

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Figure 1 Cervical sympathetic innervations of the parotid gland.

parapharyngeal space, ipsilateral submandibular gland and parotid gland have also been shown to be responsible for FBS in some patients.⁴

Netterville *et al*⁵ hypothesised that FBS is secondary to loss of the cervical sympathetic innervations of the ipsilateral parotid gland (figure 1). The parotid gland receives dual innervation by sympathetic and parasympathetic nerves, which acts collaboratively to cause contraction of myoepithelial cells.¹⁰ Loss of the sympathetic innervation to the parotid gland will cause denervation hypersensitivity to the salivary myoepithelial cells.⁵ Mastication will induce the release of parasympathetic neurotransmitters resulting in a supramaximal response to the myoepithelial cells. It is speculated that this supramaximal response, or contractile response, is what is responsible for the intense head and neck pain and/or cramps experienced by patients with FBS.⁴ This theory is consistent with findings by Chiu *et al*² and Kawashima *et al*,¹¹ as both found evidence to suggest that loss of sympathetic innervation to the parotid gland from the superior cervical ganglion (SCG) is a likely aetiological factor in FBS. Anatomically, SCG is 3 cm long and lies posterior to the carotid sheath at the level of the second and third cervical vertebrae, between the ICA and the longus capitis muscle (figure 1).² The postganglionic sympathetic fibres supplying the parotid gland originate from the SCG and run along the external carotid artery (ECA) as the external carotid plexus.¹ Therefore, surgical interventions and manipulations around the ECA, such as ligation, resection or mobilisation, may potentially disrupt the sympathetic fibres and ultimately result in FBS.

The diagnosis of FBS is a clinical one, based on history findings and a normal physical examination. Differentiation of FBS from temporomandibular joint dysfunction can be appreciated if palpation of the ipsilateral joint or mechanical joint movement does not elicit pain. Gastro-oesophageal reflux disease can be excluded based on the absence of digestive and pharyngeal findings. Glossopharyngeal neuralgia and stylalgia are both unrelated to eating and can be appreciated by targeted palpation in the subtonsillar region and at the styloid process, respectively.⁴

If FBS is suspected to be a postoperative complication, additional investigations will not be required. In contrast, if suspicion of FBS arises in the absence of a recent surgery, thorough

investigations of the parapharyngeal space and deep lobe of the parotid gland are needed to detect possible malignancies. Repeated imaging should be performed as tumours may only become visible several months after the onset of any symptoms.⁴

There are a variety of potential treatments available for alleviating FBS, however, there is no general consensus on the best management option to date. Dietary modifications are completely ineffective, as are pharmacological therapy involving non-steroidal anti-inflammatory medications, anaesthetic sprays, local anaesthetic blocks, oral analgesics, and codeine and other narcotics.^{2 12 13} Anticonvulsants (carbamazepine, pregabalin, gabapentin) used alone or in conjunction with amitriptyline may decrease the intensity and duration of pain symptoms,^{2 12 14} although high doses of gabapentin may cause drowsiness,¹² and there is conflicting evidence on the efficiency of carbamazepine

Learning points

- First bite syndrome (FBS) is an uncommon pain disorder characterised by severe cramping or spasm in the parotid region with the first bite of each meal that can be a postoperative complication of any head and neck surgery, including carotid endarterectomies.
- The proposed pathogenesis of FBS is sympathetic denervation of the parotid gland secondary to iatrogenic injury with resultant cross-stimulatory parasympathetic hypersensitivity response.
- The diagnosis of FBS is a clinical one, based on history findings and a normal physical examination; if FBS is suspected to be a postoperative complication, additional investigations will not be needed; if FBS arises in the absence of a recent surgery, thorough investigations are needed to detect possible malignancies.
- A large proportion of FBS cases are self-resolving after several months to 1 year, although pharmacological, surgical or laser ablation therapies can be considered.

by itself.^{2 14 15} Parotidectomy in the context of a malignant tumour will relieve pain symptoms of FBS though partial parotidectomies are known to be a cause of FBS, particularly if the residual gland is the deep lobe.^{9 16} Other surgical interventions including tympanic neurotomy and parotid gland parasympathectomy have been shown to be unsuccessful.²⁻⁴ Intraparotid injection of botulinum toxin A (BTA) can be effective at providing temporary analgesia and improvements in quality of life in some patients, as it blocks acetylcholine release at the neuromuscular junction, thus preventing contraction of salivary myoepithelial cells.^{8 13 17 18} Most recently, Amin *et al*¹⁹ described the first documented use of laser tympanic plexus neurectomy for ablation of the parasympathetic innervation to the parotid gland, which demonstrated promising initial responses. Before proceeding with pharmacological, surgical or laser ablation therapies, it is important to note that a large proportion of FBS cases are self-resolving after several months to 1 year,²⁻⁴ thus making conservative management appropriate and effective, as was the case with our patient.

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Contributors GR-N identified the case and was involved in the management of the patient; MJT wrote and edited the case report.

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