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FIRST BITE SYNDROME: OUR EXPERIENCE AT A TERTIARY CARE CENTRE

Otorhinolaryngology

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ABSTRACT

Introduction: First bite syndrome is a potential complication of surgery involving the infratemporal fossa, deep lobe of the parotid gland, parapharyngeal space, carotid body tumors, and various head and neck malignancies. It is described as acute and intense pain in the parotid region caused by the first bite of each meal which gradually improves with the subsequent bites. Various treatment strategies have been employed with no significant therapeutic benefit. This study brings out our experience of first bite syndrome at a tertiary care center. **Materials and Methods:** A retrospective study was conducted to identify the patients who developed first bite syndrome with or without Horner's syndrome at a tertiary care hospital from Jan 2020 to Dec 2022. Patient records were accessed to gather data on patient demographics, presenting symptoms, and surgical history. A total of 80 patients were included in the study. Patients were operated by Head and Neck Oncosurgeon, assisted by Vascular Surgeon. 51 of them were males while 29 were females. Once the diagnosis of FBS was made, the time of onset since surgery, duration of symptoms, and associated symptoms of Horner's syndrome were noted. **Results:** A total of 80 patients were included in the study. 51 of them were males while 29 were females. The mean age of patients was 43 years. 16.25 % of the total patients developed FBS. Out of the 80 patients, seven patients underwent excision of vagal schwannoma, two of them developed only first bite syndrome while the rest five developed first bite syndrome with Horner's syndrome. Total parotidectomy was indicated for four patients for mucoepidermoid carcinoma. Two patients underwent excision of paraganglioma and branchial fistula respectively. The remaining patients who did not develop FBS underwent excision of the carotid body tumor (8), superficial parotidectomy (23), and total parotidectomy (36). **Discussion:** The cause of FBS is largely unknown, however, Netterville et al have proposed that FBS is caused by the loss of sympathetic innervation to the parotid gland— . Netterville et al proposed that FBS is due to the damage or removal of cervical sympathetic with loss of sympathetic innervations to the parotid gland. It is interesting to note that in our study out of the 40 patients who underwent total parotidectomy only four patients developed first bite syndrome. Given the extreme rarity of this syndrome compared to the relatively high rate of external carotid artery and/or parotid gland surgery with ligation or resection of the external carotid artery, sympathetic-parasympathetic conflict on myoepithelial cells of the parotid gland is probably not the only mechanism involved in the pathogenesis of this syndrome, whose pathophysiology has not yet been fully understood– . Various treatment strategies have been proposed to relieve the pain of first bite syndrome. Non-steroidal anti-inflammatory drugs, acupuncture, anesthetic sprays or local anesthetic block and oral analgesics such as paracetamol, codeine, or narcotics, and tympanic neurectomy or auriculotemporal neurectomy have remained ineffective. **Conclusion:** The first bite syndrome is a relatively rare condition occurring as a result of a sympathetic-parasympathetic imbalance in the innervations to the parotid gland. Although most of the studies support this hypothesis there may be other factors in play owing to the rarity of this syndrome as compared to the number of neck surgeries performed. Various treatment options have been tried but none has shown a significant therapeutic benefit.

KEYWORDS

First Bite Syndrome, Horner's Syndrome

INTRODUCTION

First bite syndrome (FBS) is an uncommon entity characterized by severe cramping pain in the parotid region with the first bite of each meal[1–4]. It is known to occur as a sequela in surgeries involving the infratemporal fossa, parapharyngeal space, and deep lobe of the parotid gland. The pain gradually diminishes over the next few bites. The symptoms of FBS are often mild but can be severe enough to poorly affect the patient's quality of life. The exact etiology, risk factors, and treatment options remain poorly defined due to the rarity of its occurrence.

FBS may also be associated with Horner's syndrome in a certain group of patients which involves dissection along the carotid artery resulting from the manipulation of the sympathetic chain. Horner's syndrome, also known as oculosympathetic palsy is characterized by miosis, ptosis, enophthalmos, and anhidrosis.

In this study, we present our experience of encountering FBS in patients who underwent various neck surgeries at a tertiary care hospital. The available literature on FBS is limited to a few case series and reports. This is perhaps the largest study in the Indian setup describing first bite syndrome.

MATERIAL AND METHODS

A retrospective study was conducted to identify the patients who developed first bite syndrome with or without Horner's syndrome at a tertiary care hospital from Jan 2020 to Dec 2022. Patient records were accessed to gather data on patient demographics, presenting symptoms, and surgical history. Any patient with preoperative Horner's syndrome, previous neck surgery, and temporomandibular joint dysfunction was excluded from the study. Once the diagnosis of FBS was made, the time of onset since surgery, duration of symptoms, and associated symptoms of Horner's syndrome were noted.

A total of 80 patients were included in the study. **Patients were operated by Head and Neck Oncosurgeon, assisted by Vascular Surgeon**. 51 of them were males while 29 were females. The mean age of patients was 43 years. 16.25 % of the total patients developed FBS. Out of the 80 patients, seven patients underwent excision of vagal schwannoma, two of them developed only first bite syndrome while the rest five developed first bite syndrome with Horner's syndrome. Total parotidectomy was indicated for four patients for mucoepidermoid carcinoma. Two patients underwent excision of paraganglioma and branchial fistula respectively. The remaining patients who did not develop FBS underwent excision of the carotid body tumor (8), superficial parotidectomy (23), and total parotidectomy (36). The clinical data of the study are summarized in Table 1.

Sr No	Diagnosis	No	Surgery	FBS	Horner's syndrome	Recovery after one year
1	Vagal schwannoma	2	Excision	+	-	-

2.	Vagal schwannoma	5	Excision	+	+	+
3	Mucoepidermoid carcinoma parotid	4	Total parotidectomy	+	+	+
4	Paraganglioma neck	1	Excision	+	+	_
5	Branchial fistula	1	Excision	+	+	+
6	Carotid body tumor	8	Excision	-	-	
7	Pleomorphic adenoma	23	Superficial parotidectomy	-	-	
8	Other parotid lesions	36	Total parotidectomy	-	-	

The onset of FBS was immediately after the surgery. All the patients were given a trial of one month of carbamazepine, although there was no therapeutic benefit of the same. All patients were followed up for one year. Ten patients recovered completely at the end of one year while three patients did not show any sign of recovery even after one year.

DISCUSSION

'First bite syndrome' was first described in the indexed medical literature by North American Gastroenterologist Dr. W.S. Haubrich in 1986 in an article published in the *Henry Ford Hospital Medical Journal*[4]. *He* described the clinical features characterized by the occasional onset, without prodromal symptoms, at the first bite, of pharyngeal blockage of the food bolus, sometimes accompanied by retrosternal chest pain. Dr. Haubrich retrospectively studied a cohort of 949 patients between 1983 and 1985 and he proposed transient esophageal spasm as the pathophysiological basis of this disease. This was the only study describing this term till 1998 when Dr. Netterville again used this term in an article devoted to the paragangliomas of the vagus nerve and their treatment[5]. After the article by Netterville, several papers similarly described first bite syndrome. However several clinical variants were reported. One of the studies mentions that the syndrome can occur after several months rather than days after upper neck surgery[6]. Unlike the classical presentation of pain over the parotid region, the patient may also experience pain in the mandibular region or oral cavity, and may radiate to the ear[7].

Neck surgeries involving the infratemporal fossa, parapharyngeal space, and deep lobe of the parotid gland may present with many major and significant complications, particularly injury to ICA and IJV[8]. Also, there may be an injury to the cranial nerves like spinal accessory and sympathetic chain and traumatic chyle leak. First bite syndrome is reported as a minor complication of neck surgeries involving dissection along the carotid sheath and deep lobe of parotid gland. To this date, it remains under reported with a mention in a few case reports and series. It presents as severe pain in the parotid region which is of cramping nature. The pain usually starts with the first bite of the meal and is most severe during the first meal of the day. During the early post operative period, the pain is so intense that it may dissuade the patient from any oral intake. The symptoms thereafter improve gradually.

The sympathetic chain sacrifice in these patients may also lead to Horner's syndrome characterized by ipsilateral ptosis, miosis, and anhidrosis of the ipsilateral half of the face. In the study by Netterville et al nine out of 46 patients excised for vagal paraganglioma developed FBS along with Horner's syndrome. In our study, 13 patients out of 80 patients developed FBS. The incidence of this syndrome in our study is around nine percent which is in coherence with the international literature[3,11]. Two of them developed Horner's syndrome concurrently with FBS.

The cause of FBS is largely unknown, however, Netterville et al have proposed that FBS is caused by the loss of sympathetic innervation to the parotid gland[5]. Netterville et al proposed that FBS is due to the damage or removal of cervical sympathetic with loss of sympathetic innervations to the parotid gland. As a result of denervation, there is heightened sensitivity of the sympathetic receptors to the parasympathetic neurotransmitters. A supramaximal response by the myoepithelial cells is elicited on cross-stimulation by the release of parasympathetic neurotransmitters induced by chewing and biting[5]. This hypothesis of loss of the sympathetic innervation to the parotid gland is also supported by the studies done by Chiu et al[8] and Kwashima et al[9]. A direct injury to the cervical sympathetic trunk (cranial nerve surgery at the skull base and parapharyngeal space, deep cervical lymph node dissection, tumors), or postganglionic sympathetic efferents of the cervical sympathetic trunk, corresponding to the pericarotid sympathetic plexus (styloid process surgery, carotid surgery, parotid surgery with ligation of the external carotid artery, tumors) would therefore be the probable cause of this syndrome[4].

It is interesting to note that in our study out of the 40 patients who underwent total parotidectomy only four patients developed first bite syndrome. Given the extreme rarity of this syndrome compared to the relatively high rate of external carotid artery and/or parotid gland surgery with ligation or resection of the external carotid artery, sympathetic-parasympathetic conflict on myoepithelial cells of the parotid gland is probably not the only mechanism involved in the pathogenesis of this syndrome, whose pathophysiology has not yet been fully understood[4].

According to Gardener and Abdullah[9] and Kwashima et al[10], factors predisposing to the development of first bite syndrome are (a) preservation or injury to the superior cervical ganglion (in the case of cervical sympathetic trunk lesions), which would explain why all patients who underwent resection of the cervical sympathetic trunk without excision of this ganglion did not develop first bite syndrome or (b) ligation or resection of part of the external carotid artery and damage to the surrounding sympathetic plexus.Various treatment strategies have been proposed to relieve the pain of first bite syndrome. Non-steroidal anti-inflammatory drugs, acupuncture, anesthetic sprays or local anesthetic block and oral analgesics such as paracetamol, codeine, or narcotics, and tympanic neurectomy or auriculotemporal neurectomy have remained ineffective. Anticonvulsants (carbamazepine, pregabalin, gabapentin) used alone or in combination with tricyclic antidepressants (amitriptyline) can decrease the severity and/or duration of the pain[5,8,11,12]. In our study, we gave a trial of one month of Carbamazepine without any therapeutic benefit.

CONCLUSION

The first bite syndrome is a relatively rare condition occurring as a result of a sympathetic-parasympathetic imbalance in the innervations to the parotid gland. Although most of the studies support this hypothesis there may be other factors in play owing to the rarity of this syndrome as compared to the number of neck surgeries performed. Various treatment options have been tried but none has shown a significant therapeutic benefit. With a current incidence of around ten percent following upper neck surgery especially involving the deep lobe of the parotid gland or the cervical sympathetic chain, a surgeon needs to inform the patient preoperatively about this debilitating condition and the lack of any standard treatment.

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Conflicts of interest
There are no conflicts of interest.

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