

# Idiopathic first bite syndrome – A rare case report with review of literature

Ramakant S Nayak<sup>1</sup>, S Rakendu<sup>1</sup>, Vijayalakshmi S Kotrashetti<sup>1</sup>, Ranganath Nayak<sup>2</sup>

Departments of <sup>1</sup>Oral Pathology and Microbiology and <sup>2</sup>Oral and Maxillofacial Surgery, Maratha Mandal's NGH Institute of Dental Sciences and Research Centre, Belgaum, Karnataka, India

## Abstract

First bite syndrome (FBS) is a condition that classically presents as severe pain in the preauricular region, initiated on the first bite of a meal. In most of the cases reported, it is associated with a history of upper neck surgery or tumor of the parotid salivary gland or parapharyngeal space (PPS). Some propose that FBS arises due to damage to the cervical sympathetic trunk leading to the loss of sympathetic innervations to the parotid salivary gland. Literature also showed occurrence of this syndrome in individuals who had no history of parotid tumor, PPS tumor or surgery of the upper neck, and such cases are referred to as idiopathic FBS (IFBS). There are very few case reports reported on IFBS. We report the one such rare case of IFBS in a 35-year-old male, referred to the outpatient department, with a 5-month history of severe, sharp pain and bilateral swelling in the parotid region occurring only on the first bite of eating and would diminish over few minutes.

**Keywords:** First bite syndrome, parapharyngeal space, parotid pain, parotid swelling

**Address for correspondence:** Dr. Vijayalakshmi S Kotrashetti, Department of Oral Pathology and Microbiology, Maratha Mandal's NGH Institute of Dental Sciences and Research Centre, Belgaum - 590 010, Karnataka, India.  
E-mail: drviju18@yahoo.com

**Received:** 02-Apr-2020, **Revised:** 06-May-2020, **Accepted:** 23-May-2020, **Published:** 09-Sep-2020

## INTRODUCTION

A broad spectrum of pathological conditions can affect the parotid glands. Some of which can be diagnosed clinically while others can be diagnosed only on imaging.

First bite syndrome (FBS) is one such condition which does not show any kind of abnormality on imaging. It was first described by Netterville (1998) as severe parotid pain elicited on the first bite of a meal without any prodromal symptoms.<sup>[1,2]</sup> The pain subsides gradually by the subsequent bites, and the pain is worst with the first bite and first meal of the day. Netterville *et al.* have proposed that FBS is caused by the loss of sympathetic innervations

to the parotid gland, which remains the most accepted hypothesis regarding the pathogenesis of the syndrome.<sup>[1]</sup>

The probable etiology for this syndrome could be associated with surgery to the neck, tumor of parapharyngeal space (PPS) or parotid gland. FBS has also been reported postoperatively in cases of cervical lymph node dissection, temporomandibular joint (TMJ) replacement, carotid endarterectomy or ligation, resection of stylohyoid ligament in Eagle's syndrome, bimaxillary osteotomy, etc.<sup>[2]</sup> The reason for occurrence of pain in these patients is due to loss of sympathetic innervations to the parotid gland postsurgically.<sup>[1,3,4]</sup> Apart from these, there are cases

### Access this article online

#### Quick Response Code:



#### Website:

www.jomfp.in

#### DOI:

10.4103/jomfp.JOMFP\_46\_20

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

**For reprints contact:** reprints@medknow.com

**How to cite this article:** Nayak RS, Rakendu S, Kotrashetti VS, Nayak R. Idiopathic first bite syndrome – A rare case report with review of literature. J Oral Maxillofac Pathol 2020;24:403.

reported in the literature where the syndrome is not associated with any of the abovementioned causes and are referred to as idiopathic FBS (IFBS).<sup>[2,5-9]</sup> The etiology for the same still remains unknown.

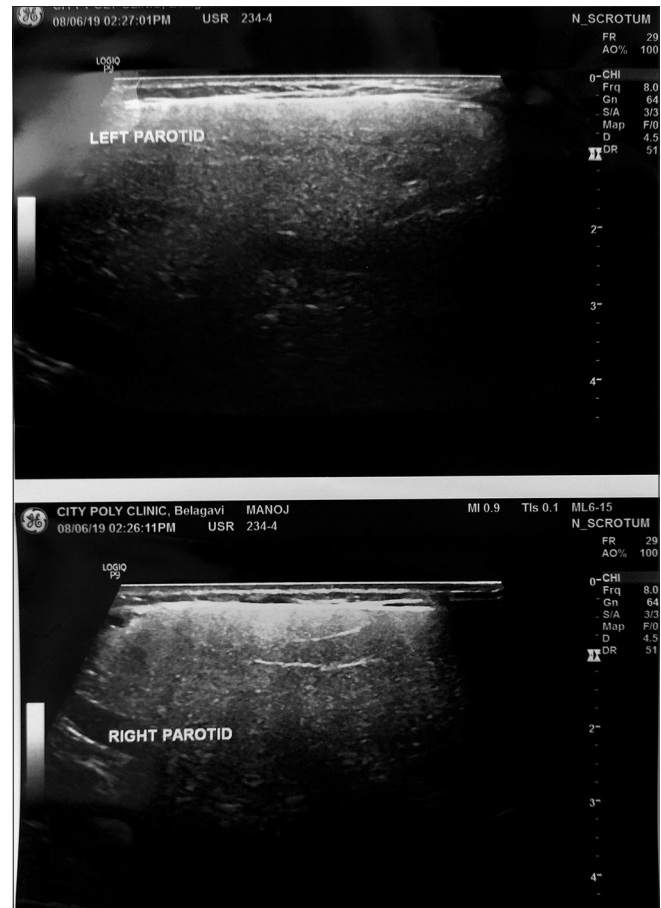
Till date, there have been only six published reports of IFBS which includes five case reports<sup>[2,6-9]</sup> and one case series,<sup>[5]</sup> and none are from India. We report a unique case of IFBS occurring in a 35-year-old male patient.

## CASE REPORT

A 35-year-old male patient reported to the outpatient department of our institute, Maratha Mandal's Nathajirao G. Halgekar Institute of Dental Sciences and Research Centre, Belagavi, on July 7, 2019, with a chief complaint of pain in the left and right lower jaw area for 5 months while having food. On detailed history, the patient informed that he experiences acute pain (10/10 on Numerical Rating Scale) on having the first bite of meal and gradually subsides on subsequent intake of food and subsides completely in few minutes after completion of meal. The pain was described as sharp and stabbing type. He experiences maximum pain while having the first bite of his food in the morning, and subsequently, with every first bite of the meal in a day, he experienced pain, but the intensity of pain reduced from morning to evening. During dinner, the intensity of pain experienced on the first bite of the meal was least. Because of pain during eating, the patient developed aversion for food which led to his weight loss.

Pertaining to medical history, the patient had met with an accident in 2014 and was hospitalized for 2 months and recovery was uneventful. Later in October 2018, he had a head injury following a fight where he was hit with a rod on the head, which was sutured, followed by which he was treated as a day-care patient. During this time, the patient was diagnosed with diabetes. Since then, he is on medication for diabetes (Glycomet 250 mg). The patient's blood glucose level was in normal limits with controlled level of glycosylated hemoglobin. Later in January 2019, he started experiencing pain in the parotid region while having food.

He initially visited an ENT specialist for the same, where he was diagnosed to have parotitis and was prescribed with medication which did not give any permanent relief. The patient is unaware of the medicine prescribed. Later, he visited another two ENT specialists. He was advised for scanning. Ultrasonographies of both right and left parotid glands were taken [Figure 1]. A computed tomography



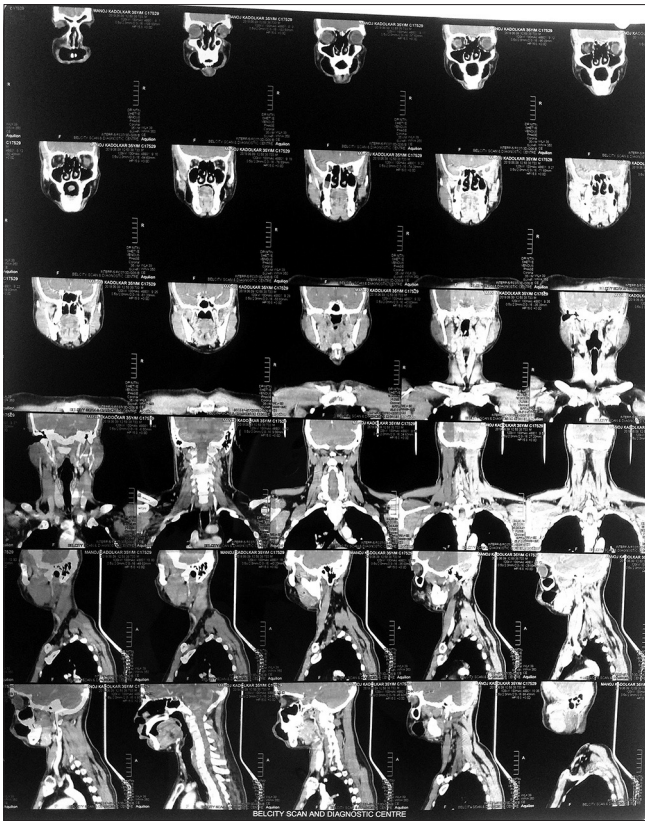
**Figure 1:** Photograph showing ultrasonography image with no abnormality detected except for a slight swelling in the parotid gland

scan was taken from the base of the skull to the lung apex. No relevant findings or abnormalities were noted except for a slight enlargement of the glands [Figure 2]. He was asked to continue with the same medication, which did not give him any permanent relief. Hence, he was referred to dental institute.

On examination, bilateral swelling was seen on the angle of mandible with raised earlobe [Figure 3]. On palpation, tenderness was elicited on the left side. On milking of parotid glands, clear but scanty fluid was noted. On intraoral and extraoral examination, no other abnormalities were detected.

Based on the history, symptoms observed and records of imaging, a final diagnosis of FBS was arrived at.

From July, the patient was prescribed with drugs for neuropathic pain that is Pregeb M 75, one tablet once daily, and Teedal, one tablet twice daily. The patient was called for weekly follow-up. After 5 months patient showed 80% reduction in pain and rated 2/10 on pain scale for pain. The patient is still under follow-up.



**Figure 2:** Photograph showing computed tomography scan with no abnormality detected, only slight swelling of the parotid gland observed

### Review of literature

On literature search, we found six published reports (one case series and five case reports) of IFBS, out of which two case reports were in Japanese language and abstract was available in English; hence, it was also included in review [Table 1].

The first case was reported in the year 2010 by Kurokawa *et al.* of a patient with a 6-month history of pain in the parotid region that occurred after the first bite of every meal. Pain decreased after 6 repeats of stellate ganglion blocks (SGBs) and completely disappeared after 16 repeats of SGB.<sup>[9]</sup> The patient had no history of trauma or surgery.

Wemyss *et al.* reported a case of the patient who had severe, sharp unilateral pain on the first bite of eating. No abnormalities were detected with respect to TMJ, PPS, trigeminal nerve or parotid gland. FBS was suspected, and the patient was immediately started with carbamazepine 200 mg, three times daily. It was proved to be successful in treating, and in 4 months, symptoms resolved completely.<sup>[2]</sup>

A similar case with symptoms consistent with FBS was reported by Stoopler *et al.* Medical therapy of the patient included oxcarbazepine and gabapentin without benefit.



**Figure 3:** Extraoral photograph showing mild bilateral parotid gland swelling with slight increase in the left side

Later, patient was given botulinum toxin A injections to the parotid gland and patient reported improvement in pain with the treatment.<sup>[6]</sup>

Chiba *et al.* reported case series of 14 patients with idiopathic parotid pain (IPP), and they opined that clinical picture and pathogenesis of IFBS remains unknown and also only a few cases have been reported in the literature, where the difference in clinical presentation of IPP and IFBS has not been clearly described. Hence, they referred their cases as IPP instead of IFBS. Out of 14 cases analyzed retrospectively by checking the medical records, they considered only 9 cases that were diagnosed with diabetes as IPP and the rest 5 cases were not considered as they had no history of diabetes. Their main aim was to find if there was any association of diabetic neuropathy with FBS. In this study, the authors ruled out various pathogeneses reported earlier such as the loss of sympathetic innervations to the parotid gland and gustatory stimuli which evoked facial pain. Thus, the pathogenesis of IPP in the study remained to be elucidated. They believed that there might be an association between diabetic neuropathy and pathogenesis of IPP, but it is unlikely that the diabetic neuropathy alone contributed to the development of IPP. Thus, the relationship between IPP and diabetes was speculated and required further studies.<sup>[5]</sup>

Hayashi *et al.* reported a case of IFBS treated with rikkosan. Here, the treatment with carbamazepine was ineffective and was, in turn, treated with the Kampo medicine rikkosan gargles along with dietary strategy. In this case, in 1 year, IFBS improved with no recurrence.<sup>[8]</sup>

The literature review provided with total of five individual case reports of IFBS and one case series of five cases

**Table 1: Review of literature**

Year	Title	Authors	Findings	Treatment/follow-up
2010	IFBS treated with repeated SGB: A case report <sup>[9]</sup>	Kurokawa H, Takezaki T, Kawaguchi R, Nakao M, Sato N, Unetani H <i>et al</i>	A case of man in his thirties with a 6-month history of pain in the parotid region that occurred after the first bite of every meal	The pain completely disappeared after 16 repeats of SGB and did not recur for 2.5 years
2012	A case of IFBS with bilateral onset <sup>[7]</sup>	Chiba M, Hirotsani H, Echigo S	A case of 33-year-old man, with no prior surgical history, presented with bilateral orofacial pain consistent with FBS	Conservative therapy, including drug therapy and self-care. At 6 months, his symptoms had almost resolved
2016	Letter to the editor: IFBS <sup>[6]</sup>	Eric T. Stoopler, Sophia Elmuradi, Thomas P. Sollecito, Natasha Mirza	A case of 56-year-old African American man with severe, bilateral facial pain of 40-year duration	Oxcarbazepine, 150 mg twice daily, and gabapentin. Currently receiving botulinum toxin A injections to the parotid glands with self-reported improvements
2018	Clinical features of IPP triggered by the first bite in Japanese patients with Type 2 diabetes: A case study of nine patients <sup>[5]</sup>	Masatoshi Chiba, Hiroaki Hirotsani and Tetsu Takahashi	Case series of 14 patients with idiopathic parotid pain. Out of which, nine patients were included in the study since they had a history of diabetes, whereas five were excluded as they did not have diabetes or any other underlying pathology	Dietary modifications, to which one patient responded and pharmacological management consisting of pregabalin or neurotropin
2019	A case of IFBS responding to carbamazepine <sup>[2]</sup>	C. Wemyss, J. Gibson and K. Ryan	A case of 51-year-old male patient with severe, sharp, unilateral pain occurring only on the first bite of eating	200 mg carbamazepine, three times daily 4 months later, complete resolution of symptoms was noted
2019	IFBS treated with rikkosan: A case report <sup>[8]</sup>	Hayashi K, Onda T, Ogane S, Kamio T, Ohata H, Takano N, Shibahara T	A case of 81-year-old woman who developed sharp pain in the parotid region with the first bite of every meal	Rikkosan gargle and dietary modifications After 1 year, complete resolution of symptoms with no recurrence was noted
2020	IFBS	Present case	A 35-year-old male patient with chief complaint of sharp pain associated bilaterally with parotid on every first bite of the meal	Pregeb M 75 prescribed for 5 months showed 80% reduction in pain

FBS: First bite syndrome, IFBS: Idiopathic FBS, SGB: Stellate ganglion block; IPP: Idiopathic parotid pain

without diabetes diagnosed as IFBS and nine cases with diabetes. We report an additional case of IFBS with diabetes.

## DISCUSSION

FBS is a painful condition which classically presents as severe pain in the parotid region, initiated on the first bite of a meal. It is thought to be accentuated by particular gustatory stimuli (acids, sour, spicy or sweet foods or drinks) and salivation, and some even say the thought of food.<sup>[2]</sup> FBS is characterized by sharp pain in the parotid region at the first bite of a meal which gradually decreases as mastication continues and is thought to be caused by sympathetic nerve damage. When FBS occurs with no history suggestive of nerve damage, it is classified as IFBS.

With oral intake, the secreted parasympathetic neurotransmitter cross-stimulates the sympathetic receptors, causing an autonomic imbalance. This leads to supramaximal contraction of the myoepithelial cells due to supranormal response to parasympathetic stimulation of myoepithelial cells, and excessive contraction of these cells causes pain on the parotid region on the first bite.<sup>[2]</sup> It is usually associated with a history of upper neck surgery or tumor of the parotid salivary gland or PPS, or damage to, or resection of the cervical sympathetic trunk.

Netterville's 1998 case series presented 49 patients who were treated with surgery for vagal paragangliomas. FBS was reported postoperatively in eight of the nine patients

who had sympathetic trunk resection. Since then, FBS has also been implicated postoperatively.<sup>[1]</sup>

Tumors reported to have caused symptoms of FBS are tumors of the PPS (synovial sarcoma), deep poles of the parotid (adenoid cystic carcinoma, squamous cell carcinoma and mucoepidermoid carcinoma) and submandibular gland (adenoid cystic carcinoma).<sup>[2]</sup> With respect to the present case, neither the patient underwent any kind of surgery nor was he diagnosed with any tumor. On doing the literature survey, few cases of IPP have been reported. The findings of this case favored the diagnosis of IPP, first of its kind being reported in India. The term IPP is used interchangeably for IFBS.

In the present case, though the patient had diabetes, he was diagnosed with diabetes only a few months dated before he got the symptoms of FBS. Hence, the duration since when he has been suffering from diabetes cannot be ascertained. However, he had not developed any neurological changes associated with diabetes.

Whenever we discover a case of bilateral parotid pain with swelling, based on clinical examination and imaging, various possible diagnoses are to be ruled out. The following are the differential diagnosis to be ruled out.

Sialolithiasis presents as moderately severe pain while salivation, particularly just before, during and after meals

with palpable stones in the duct or gland. Here, the pain was not continuous in nature neither there was any calcification noted in the scan. Acute suppurative parotitis is an acute painful condition with rapid swelling of the parotid gland, and a purulent discharge may be expressed with a low-grade fever. In this case, the pain was only during intake of food. Chronic sialadenitis is characterized by intermittent swelling of the gland which may lead to the development of fibrous masses. Viral parotitis presents as a firm, rubbery swelling of the salivary glands producing a continuous pain upon mastication. Sialadenosis is a bilateral persistent painless, noninflammatory condition.<sup>[10]</sup>

Literature review suggests different treatments being used for the management of FBS. The most common treatment carried out being pharmacological management. Nonsteroidal anti-inflammatories and codeine-based analgesia have proved ineffective in all cases. Other drugs used have included carbamazepine (with or without amitriptyline), rikkosan, pregabalin and neurotropic agents. Dietary modifications were also done.<sup>[2,5,8]</sup> Botulinum toxin A injections to the parotid glands and SGBs have also been used to successfully treat the idiopathic cases.<sup>[6,9]</sup>

For the present case one month use of pregabalin tablets showed significant improvement and pain almost reduced to 80% by the end of 5 months. Patient rated pain as 2/10 on the numerical pain scale.

## CONCLUSION

This case highlights the need of proper history taking by the dentist before arriving at the final diagnosis. FBS should not be confused with the various salivary gland disorders and parotid swellings which could lead to inappropriate management. Hence, all other conditions have to be selectively ruled out based on the various findings.

It is a rare case, only a few been reported yet in the literature. The present case is the first case reported in India. Clinicians might sometimes be misled due to its rarity. Hence, clinicians must be aware about the classic

presentation of the syndrome and its timely proper management.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

## Financial support and sponsorship

Nil.

## Conflicts of interest

There are no conflicts of interest.

## REFERENCES

1. Netteville JL, Jackson CG, Miller FR, Wanamaker JR, Glasscock ME. Vagal paraganglioma: A review of 46 patients treated during a 20-year period. *Arch Otolaryngol Head Neck Surg* 1998;124:1133-40.
2. Wemyss C, Gibson J, Ryan K. A case of idiopathic First Bite Syndrome responding to carbamazepine. *Oral Surgery ISSN* 2019;1752-2471.
3. Chiu AG, Cohen JI, Burningham AR, Andersen PE, Davidson BJ. First bite syndrome: A complication of surgery involving the parapharyngeal space. *Head Neck* 2002;24:996-9.
4. Linkov G, Morris LG, Shah JP, Kraus DH. First bite syndrome: Incidence, risk factors, treatment, and outcomes. *Laryngoscope* 2012;122:1773-8.
5. Chiba M, Hirotsu H, Takahashi T. Clinical features of idiopathic parotid pain triggered by the first bite in Japanese patients with type 2 diabetes: A case study of 9 patients. *Pain Res Treat* 2018;2018:1-6.
6. Stoopler ET, Elmuradi S, Sollecito TP, Mirza N. Idiopathic first bite syndrome. *J Oral Maxillofac Surg* 2016;74:872.
7. Chiba M, Hirotsu H, Echigo S. Bilateral idiopathic first bite syndrome: A case report. *J Japanese Stomatol Soc* 2012;61:327-30.
8. Hayashi K, Onda T, Ogane S, Kamio T, Ohata H, Takano N, Shibahara T. Idiopathic first bite syndrome treated with Rikkosan: A case report. *J Oral Maxillofac Surg* 2019;31:350-5.
9. Kurokawa H, Takezaki T, Kawaguchi R, Nakao M, Sato N, Umetani H, *et al.* Idiopathic first bite syndrome treated with repeated stellate ganglion blocks: A case report. *J Japan Soc Pain Clin* 2010;17:17-20.
10. Shafer WG, Hine MK, Levy BM. *A Textbook of Oral Pathology*. 6<sup>th</sup> ed. Philadelphia, USA: Saunders; 2009.