

First Bite Syndrome Presenting as Initial Symptom of Soft Tissue Mass in Base of Tongue and Pharyngeal Space Lead to Diagnosis of Squamous Cell Carcinoma Invading Trachea and Esophagus: A Rare Case Report

Satnam S Jolly¹, Vidya Rattan², Sachin Rai³

ABSTRACT

First bite syndrome (FBS) is considered as a development of pain in the preauricular region triggered by gustatory stimuli. It has been mostly associated with surgery of the parapharyngeal space, parotid gland, and upper neck. This hypersensitivity is thought to elicit a supramaximal contraction of myoepithelial cells during the first bite of a meal that subsides with continued masticatory action. We are reporting a rare case of preauricular pain after first bite of meal that leads to the diagnosis of squamous cell carcinoma invading the trachea and esophagus.

Keywords: Carcinoma, First bite, Hypopharyngeal cancer, Neck mass, No pathology, Pain.

International Journal of Head and Neck Surgery (2022): 10.5005/jp-journals-10001-1412

INTRODUCTION

First bite syndrome (FBS) is considered as a development of pain in the preauricular region triggered by gustatory stimuli.¹ The pain is typically worst with the first bite during the first meal of the day and gradually subsides with successive masticatory movements. It is recurring in nature and occurs before every meal. It has been mostly associated with surgery of the parapharyngeal space, parotid gland, and upper neck. Most theories on the pathology of FBS revolve around the concept of sympathetic denervation of the parotid gland with subsequent hypersensitivity of myoepithelial cells to parasympathetic neurotransmitters. This hypersensitivity is thought to elicit a supramaximal contraction of myoepithelial cells during the first bite of a meal that subsides with continued masticatory action.² In the literature, most of the reported cases show occurrence of FBS few days after parapharyngeal surgery. The purpose of this paper is to report an interesting case in which presenting symptom of FBS leads to the diagnosis of squamous cell carcinoma invading to trachea and esophagus.

CASE DESCRIPTION

A 46-year-old male reported to the unit of oral and maxillofacial surgery with the chief complaint of severe pain in the right preauricular region after taking first bite of each meal, which lasted for a moment and subsided with subsequent masticatory movements from last 6 months. The pain radiated toward parotid, temporal, and neck regions. On clinical examination, there was no abnormality detected in the oral cavity, face, and neck and no any weakness of the facial nerve was there (Fig. 1). The patient was a chronic cigarette smoker for the last 20 years and there was no history of previous neck pathology and surgery. Computerized tomographic scan of the neck showed a heterogeneous soft tissue mass in the region of base of tongue on left side and extending toward midline. Posterolaterally, the soft tissue mass

¹⁻³Department of Oral and Maxillofacial Surgery, Postgraduate Institute of Medical Education and Research, Chandigarh, India

Corresponding Author: Satnam S Jolly, Department of Oral and Maxillofacial Surgery, Postgraduate Institute of Medical Education and Research, Chandigarh, India, Phone: +91 9914287909, e-mail: satnamsurgeon@yahoo.co.in

How to cite this article: Jolly SS, Rattan V, Rai S. First Bite Syndrome Presenting as Initial Symptom of Soft Tissue Mass in Base of Tongue and Pharyngeal Space Lead to Diagnosis of Squamous Cell Carcinoma Invading Trachea and Esophagus: A Rare Case Report. *Int J Head Neck Surg* 2022;xx(x):1–3.

Source of support: Nil

Conflict of interest: None

was extending along the left pharyngeal wall in the tonsillar fossa of approximately of 3.5 × 2 × 3 cm in diameter. Laterally, it caused lateral displacement of left parapharyngeal space fat (Fig. 2). There was paratracheal lymphadenopathy with tracheal and esophageal invasion by the mass without involvement of major neck vessels. Full-body scan was done to rule out the primary or metastatic site. No evidence of tumor was found in other parts of body on radiological examination.

The biopsy of the right upper paratracheal mass was taken by a pulmonary medicine specialist via the endoscopic approach. Histopathological examination revealed a diagnosis of moderately differentiated squamous cell carcinoma, which on immunohistochemistry was CK5/6 positive. The patient received radiotherapy 30G/15 fraction for 2½ weeks and chemotherapy with paclitaxel 280 mg, cisplatin 80 mg, dexamethasone 16 mg, granisetron, and mannitol 20% intravenous for six cycles. After 6 months of this regime, the patient was totally free from pain in his jaw. The patient has been followed for 1 year with no recurrence till date.



Fig. 1: Clinical photograph of neck without any abnormality and swelling

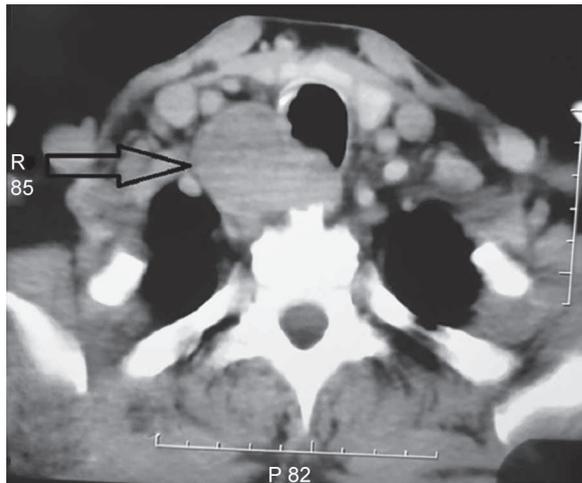


Fig. 2: Axial computerized tomographic scan showing soft tissue mass compressing the trachea and enlarged paratracheal lymph nodes (arrow showing a large lesion)

3 cm in length, which lies posterior to the carotid sheath at the level of the second and third cervical vertebrae. The symptoms of FBS develop when there is damage and irritation to superior cervical ganglion and postganglionic sympathetic plexus that are associated with external carotid artery, as this artery courses the parotid gland.^{7,8} Damage to plexus of superior cervical ganglion around the internal carotid artery can lead to Horner syndrome.⁹ Deganello et al.¹⁰ described a case of FBS, which occurred as a presenting symptom of adenoid cystic carcinoma of deep lobe of the parotid gland which involved the carotid artery plexus.

In our case, the lesion was in the base of the tongue involving the parapharyngeal space without involvement of carotid arteries. The reason in our case may be due to disruption of sympathetic pathways of neurosensory supply of the parotid gland by invasion of tumor to superior cervical ganglion plexus. The cause exactly is not known in our case. The patient responded very well to radiotherapy without any surgery and there is no recurrence of symptoms after 1 year of follow-up after chemotherapy too. This is the first case report in which alone symptoms of FBS lead to the diagnosis of squamous cell carcinoma of base of tongue invading to trachea and esophagus.

In many cases of FBS, no cause can be elicited and these are idiopathic in nature making the treatment of FBS difficult, as the pathophysiology is not very well understood in these cases. In most of the reported cases, the symptoms of FBS resolved spontaneously with time. The management is therefore directed as symptomatic relief with many drugs until natural resolution occurs.^{11,12} Ali et al.¹³ used botulinum toxin for symptomatic relief for many days until pain subsided by itself.¹³

This may be the first case in the literature in which a mass in the base of tongue and parapharyngeal region invading to trachea and esophagus lead to diagnosis of squamous cell carcinoma after the initial presenting symptoms of FBS in a healthy patient. All patients reporting with symptoms of FBS should undergo complete head and neck examination including computerized tomography and magnetic resonance scan to rule out underlying pathology. The dentist, otolaryngologist, and oral and maxillofacial surgeon should be aware of FBS so that these patients can be diagnosed and managed in early stages.

DISCUSSION

First bite syndrome has been recognized medically, but very few reports concerning it are available in the literature. Haubrich¹ in 1986 first time described it as a development of pain in parotid region after first bite of every meal. The pain is sharp in nature, excruciating in the parotid area and radiates toward the temporal region on the first bite of meal and is relieved with subsequent masticatory movements. Nettekville et al.² stated that due to damage to cervical sympathetic innervations during parapharyngeal surgery, there is a hypersensitivity of myoepithelial cells to parasympathetic neurotransmitters. This hypersensitivity elicits a supramaximal contraction of myoepithelial cells during the first bite of the meal that subsides with continued masticatory action. The second thought is considered that tumor irritation of the sympathetic system, together with concomitant tumor denervation of the parasympathetic system that can cause myoepithelial supersensitivity and hyperresponsiveness, can lead to FBS, as this might have happened in the present case.³⁻⁶ The sympathetic chain's superior cervical ganglion is approximately

REFERENCES

1. Haubrich WS. The first-bite syndrome. *Henry Ford Hosp Med J* 1986;34(4):275-278.
2. Nettekville JL, Jackson CG, Miller FR, et al. Vagal paraganglioma: a review of 46 patients treated during a 20-year period. *Arch Otolaryngol Head Neck Surg* 1998;124(10):1133-1140. DOI: 10.1001/archotol.124.10.1133
3. Kawashima Y, Sumi T, Sugimoto T, et al. First-bite syndrome: a review of 29 patients with parapharyngeal space tumor. *Auris Nasus Larynx* 2008;35(1):109-113. DOI: 10.1016/j.anl.2007.06.005.
4. Chiu AG, Cohen JI, Burningham AR, et al. First bite syndrome: a complication of surgery involving the parapharyngeal space. *Head Neck* 2002;24(11):996-999. DOI: 10.1002/hed.10162.
5. Reddy CE, Gupta AK. Clinical features of first bite syndrome. *Otolaryngol Head Neck Surg* 2009;141(3 Suppl 1):147-148.
6. Cernea CR, Nishio S, Hajaji FC, et al. First bite syndrome after resection of styloid process. *Otolaryngol Head Neck Surg* 2005;133(2):215.
7. Kamal A, Abd El-Fattah AM, Tawfik A, et al. Cervical sympathetic schwannoma with postoperative first bite syndrome. *Eur Arch Otorhinolaryngol* 2007;264(9):1109-1111. DOI: 10.1007/s00405-007-0308-y.

8. Mandel L, Syrop SB. First bite syndrome after parapharyngeal surgery for cervical schwannoma. *J Am Dent Assoc* 2008;139(11):1480–1483. DOI: 10.14219/jada.archive.2008.0073.
9. Albasri H, Eley KA, Saeed NR. Chronic pain related to first bite syndrome: report of two cases. *Br J Oral Maxillofac Surg* 2011;49(2):154–156. DOI: 10.1016/j.bjoms.2009.10.035
10. Degaenello A, Meccariello G, Busoni M, et al. First bite syndrome as presenting symptom of parapharyngeal adenoid cystic carcinoma. *J Laryngol Otol* 2011;125(4):428–431. DOI: 10.1017/S002221511000294X.
11. Phillips TJ, Farquhar-Smith WP. Pharmacological treatment of a patient with first-bite syndrome. *Anaesthesia* 2009;64(1):97–98. DOI: 10.1111/j.1365-2044.2008.05804.x.
12. Casserly P, Kiely P, Fenton JE. Cervical sympathetic chain schwannoma masquerading as carotid body tumour with a postoperative complication of first-bite syndrome. *Eur Arch Otorhinolaryngol* 2009;266(10):1659–1662. DOI: 10.1007/s00405-008-0902-7.
13. Ali MJ, Orloff LA, Lustig LR, et al. Botulinum toxin in the treatment of first bite syndrome. *Otolaryngol Head Neck Surg* 2008;139(5):742–743. DOI: 10.1016/j.otohns.2008.08.015.