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REVIEW

First bite syndrome

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Summary Based on a review of the indexed medical literature (PubMed database), the authors describe the clinical features leading to the diagnosis of first bite syndrome, the pathophysiology of this syndrome and analyse the various treatment options available to otorhinolaryngologists to manage this syndrome.

Introduction

A Google search using the terms ''first bite syndrome'' retrieved several tens of thousands of responses, about two-thirds of which corresponded to questions from patients and/or practitioners confused by this disease. This large number of search results contrasts with the small number of articles published in the medical literature, as less than 25 articles were found in the PubMed database [1—22]. In view of this discrepancy between the high demand and the poor supply, we conducted a review of the literature to define the clinical features suggestive of the diagnosis of first bite syndrome, describe the pathophysiological hypotheses proposed to explain the development of this syndrome and analyse the treatment options currently available to oto-rhinolaryngologists to manage this syndrome.

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Diagnosis

The term ''first bite syndrome'' was first used in the indexed medical literature by a North American gastroenterologist Dr W.S. Haubrich in 1986 [1] in an article published in the *Henry Ford Hospital Medical Journal* describing 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 [1]. Intrigued by this painful dysphagia, which, according to him had not been previously described in medical textbooks of the time devoted to oesophageal diseases, Haubrich retrospectively reviewed the cohort of 949 patients seen between 1983 and 1985. He then discovered that all patients experiencing these symptoms presented an associated oesophageal disease (sliding hiatal hernia in 17 cases, Schatzki B rings in three cases, cancer of the oesophagogastric junction in three cases and tertiary contractions of the distal oesophagus in one case). In the light of these findings, Haubrich proposed transient oesophageal spasm as the pathophysiological basis for this syndrome [1].

This paper was the only article on this subject until 1998, when a North American otorhinolaryngologist, Dr. James

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L. Netterville [2] again used this term. In an article devoted to paragangliomas of the vagus nerve and their treatment, Netterville described an early postoperative pain syndrome (totally unrelated to the clinical features described by Haubrich [1]), characterized by sudden onset without prodromal symptoms, at the first bite, of pain or severe cramping in the ipsilateral parotid region. The patient may even avoid eating due to the severity of this syndrome. The pain, which gradually resolves with the following mouthfuls, recurs at each meal and is accentuated by the use of sialogogues. In all cases of the series reported by Netterville et al. [2], this syndrome was associated with a lesion of the ipsilateral cervical sympathetic trunk lesion. However, well before this publication, Gardner and Abdullah [23], in 1955, had already clearly described these symptoms in a study that analysed the differences between pre- and postganglionic cervical sympathectomy (superior cervical ganglion) in the treatment of certain vascular or atrophic brain lesions.

Following this publication by Netterville et al. [2], several papers concerning first bite syndrome [3—22] (Table 1) reported sudden onset of intense head and neck pain in the parotid region, with no prodromal symptoms. Pain was triggered by the first bite and then gradually resolved with subsequent bites. However, several clinical variants were reported. For example, the syndrome can occur several months rather than several days after upper neck surgery [4] and the pain can also be located in the mandibular region or oral cavity and radiate to the ear [12]. Some authors [11,13,14,18,20] have reported that pain can also be triggered by salivation, simply thinking about a meal or by simple contact with various foods even in the absence of chewing. An associated cervical sympathetic trunk lesion was present in only 42.8% of the 112 cases published (Table 1) since the article by Netterville et al. [2]. Two main aetiological classes can be distinguished: postoperative syndromes and tumours. More than 95% of published cases correspond to postoperative syndromes [1—14,18—21] (Table 1) following upper neck surgery (resection of mixed and/or cervical sympathetic nerve tumours, deep cervical lymph node dissection, parotid gland surgery, particularly involving the deep lobe of the parotid gland, parapharyngeal and infratemporal fossa surgery, carotid bifurcation and/or internal carotid artery surgery, resection of the styloid process). Tumours responsible for first bite syndrome were malignant in all cases arising from the deep lobe of the parotid gland, parapharyngeal space or ipsilateral submandibular gland and, in a few cases, the tumour was detected on conventional imaging only several months after onset of the pain [15—17,22].

All authors agree that the positive diagnosis is exclusively clinical, based on the findings of clinical interview and a normal physical examination [2—20]. However, several differential diagnoses must be eliminated. The leading differential diagnosis is temporomandibular joint dysfunction, which can be easily distinguished, as first bite syndrome is not associated with pain or discomfort on mobilization and palpation of the ipsilateral temporomandibular joint. The second differential diagnosis is gastro-oesophageal reflux, which can also be easily excluded in the absence of pain in the parotid region and the presence of associated digestive and pharyngeal signs. Glossopharyngeal neuralgia, triggered by ipsilateral intraoral palpation of the subtonsillar region,

Table 1 Aetiologies and symptoms associated with first bite syndrome (PubMed meta-analysis).

Authors	Year	N	Aetiology	Associated symptoms
Series				
Netterville et al. [2]	1998	9	PO	9 cases of cervical sympathetic trunk injury
Chiu et al. [3]	2002	12	PO	6 cases of cervical sympathetic trunk injury
Kawashima et al. [7]	2009	9	PO	3 cases of cervical sympathetic trunk injury
Linkow et al. [19]	2012	45	PO	10 cases of cervical sympathetic trunk injury
Abdeldaoui et al. [20]	2013	17	PO	12 cases of cervical sympathetic trunk injury
Case reports				
Cernea [4]	2006	1	PO	—
Kamal et al. [5]	2007	1	PO	Cervical sympathetic trunk injury
Ali et al. [6]	2008	1	PO	Cervical sympathetic trunk injury
Mandel and Syrop [8]	2008	1	PO	Cervical sympathetic trunk injury
Borras Pereira et al. [9]	2009	1	PO	Cervical sympathetic trunk injury
Casserly et al. [10]	2009	1	PO	Cervical sympathetic trunk injury
Lee et al. [11]	2009	5	PO	2 cases of cervical sympathetic trunk injury
Philips and Farquhar-Smith [12]	2009	1	PO	—
Albasri et al. [13]	2009	2	PO	—
Costa et al. [14]	2011	2	PO	—
Deganello et al. [15]	2011	1	T	—
Dierk et al. [16]	2011	1	T	—
Liberman and Har-El [17]	2011	1	T	Cervical sympathetic trunk injury
Wong et al. [18]	2011	1	PO	—
Simms et al. [21]	2012	3	PO	—
Guss et al. [22]	2012	1	T	—

N: number; PO: postoperative; T: tumour.