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CLINICAL PAPER

Lingual Hypoglossal Reflex: An Unusual Reflex of Head and Neck

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Abstract

Introduction Oral-pharyngeal reflexes demonstrate a wide range of complexities due to their intricate synaptic pattern. Most of the reflexes are protective in nature such as preventing aspiration. These reflexes in oral cavity affect the muscles and can be evoked either in isolation or in combination in order to achieve a specific response. Certain sensory inputs induce an entire motor behavior pattern as seen in lingual hypoglossal reflex.

Materials and Methods A case report depicting this reflex have been presented. To our knowledge, this is the first case documented in a living human. Along with this, cadaveric studies have been also done in fifteen specimens.

Results Extralingual anastomosis between lingual nerve and hypoglossal nerve were found in six specimens.

Conclusion Hereby, we present a clinical paper of this unusual phenomenon which has not been documented in the literature.

Keywords Reflex · Lingual nerve · Hypoglossal nerve · Synapses

Introduction

Reflexes are actions performed in response to a stimulus without conscious thought. They occur before the brain is aware of what is happening. Reactions to a particular stimulus are processed in the spinal cord, bypassing the control of the brain [1]. When a movement occurs through a reflex action, the brain becomes aware of such movement naturally which in turn can regulate it somewhat, but cannot control it.

According to Charles Sherrington, reflexes are specific sensory inputs that can induce motor responses subconsciously resulting in reciprocal effects on various motoneuron pools [1]. Humans have various reflexes all over the body with specific function. They are mainly protective in nature. Some of the notable reflexes in maxillofacial region include corneal reflex, pupillary reflex, gag reflex, respiratory reflex, vestibulo-ocular reflex, oculocardiac reflex which are all well addressed in the studies [2–6].

Lingual hypoglossal reflex is one among them which is not much popular as others mentioned above and its presence is not routinely noted in the clinics. Hereby, we report an interesting case of lingual hypoglossal reflex with its literature insights. Though some studies have been done in animals and cadavers, its presence has not been mentioned in an alive person [7–11]. This clinical paper also highlights study done in 15 cadaveric specimens, out of which six revealed lingual and hypoglossal nerve anastomosis. To our knowledge, this is the first case report to mention about lingual hypoglossal reflex in a living human.

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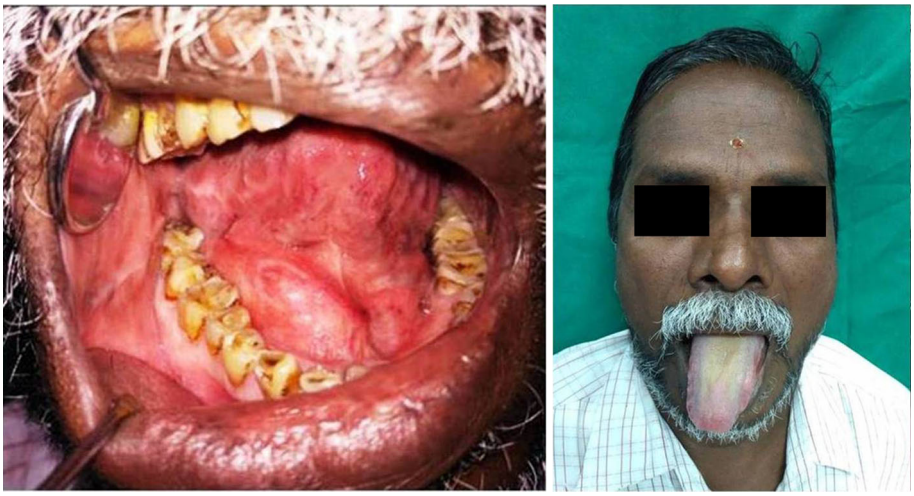

Fig. 1 Swelling in right floor of mouth and deviation of tongue toward right side

Case Report

A 53-year-old male patient reported to the department of Oral and Maxillofacial surgery with complaints of pain and swelling below the tongue of 6 months duration. On inspection, a well-defined swelling of size 5 × 3 cm was found on the floor of the mouth extending anteroposteriorly from teeth 42–47 region and mediolaterally from lingual sulcus to mid-line. Swelling was firm and tender on palpation with an erythematous overlying mucosa. No pus discharge or evidence of ulceration was noted intraorally in the floor of the mouth. There was a deviation of the tongue toward the lesion side (right side) with normal protrusive movements (Fig. 1). CT contrast revealed an intensely enhancing diffusely enlarged mass medial to the body of mandible (Fig. 2). Since this swelling under the tongue was present over a long period of time showing no signs of regression and signs of inflammation, a diagnosis of chronic sialadenitis was made and surgical removal of the sublingual gland was planned under general anesthesia.

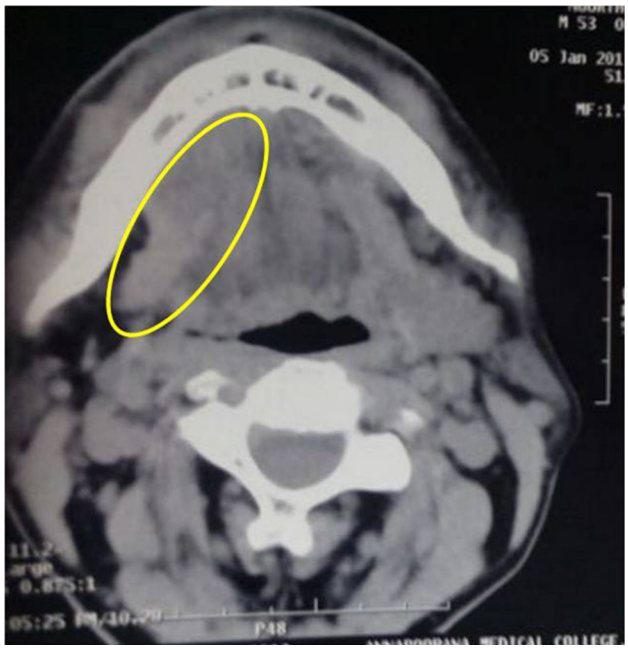

Fig. 2 Axial CT showing hyperdense mass medial to body of right mandible

Intraorally, incision was placed on the mucosa of the floor of the mouth in relation to teeth 43–47 region parallel to the Wharton's duct. Careful dissection was carried out to isolate the gland not injuring the Wharton's duct. The gland which was well encapsulated and showing no signs of infiltration was removed in toto, and surgical site was closed primarily with 3–0 vicryl. The gland was enlarged with a total dimension of about 2.5 × 2.5 cm (Fig. 3). Biopsy report was consistent with chronic sialadenitis.

On periodical follow-up, progressive exaggerated deviation of tongue to the right was noted with loss of protrusion only during first one and half months (Fig. 4). This complication has occurred after removal of sublingual gland in spite of the surgical plane being well away from the hypoglossal nerve. Hypoglossal nerve gives the motor supply to the tongue which usually lies below the mylohyoid muscle in the submandibular triangle. This intrigued us to search the literature and do cadaveric dissections on fifteen specimens in association with Department of Anatomy.

Out of fifteen cadaveric dissections, anastomosis between lingual nerve and hypoglossal nerve at various levels extralingually was seen in six specimens (Fig. 5). The literature review coupled with the cadaveric dissections helped us to understand the interesting concept of lingual hypoglossal reflex (video attached as Supplementary file).

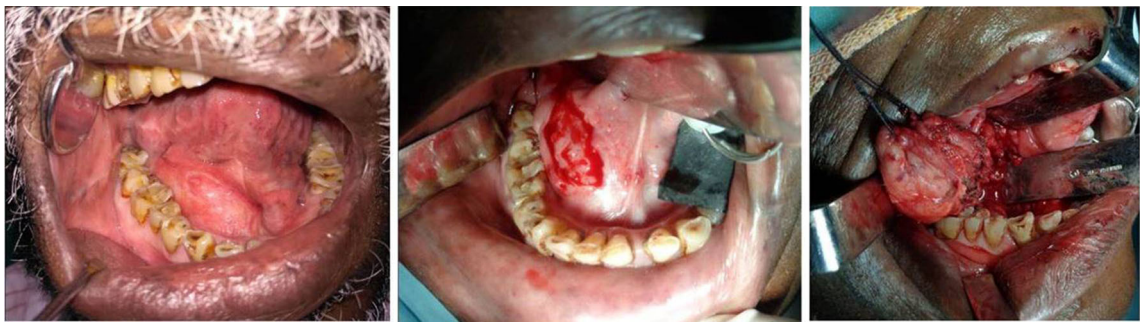

Fig. 3 Surgical excision of right sublingual gland

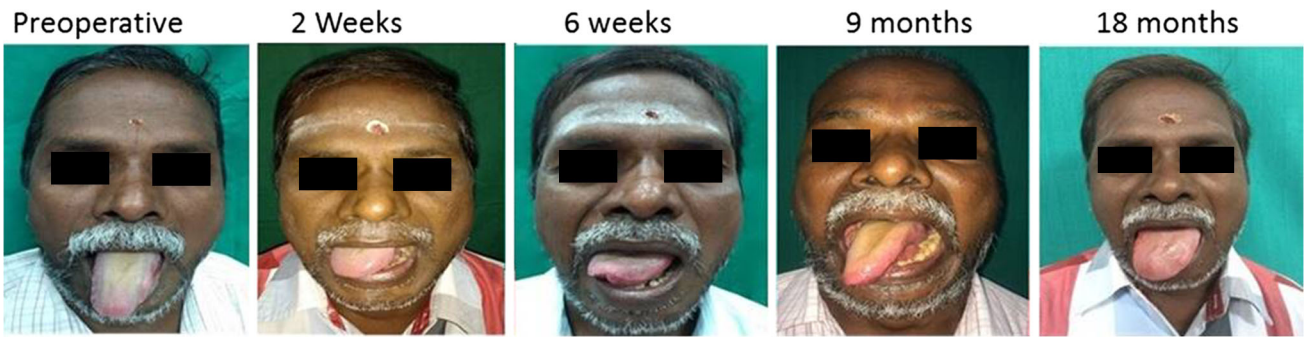

Fig. 4 Comparison of tongue protrusion

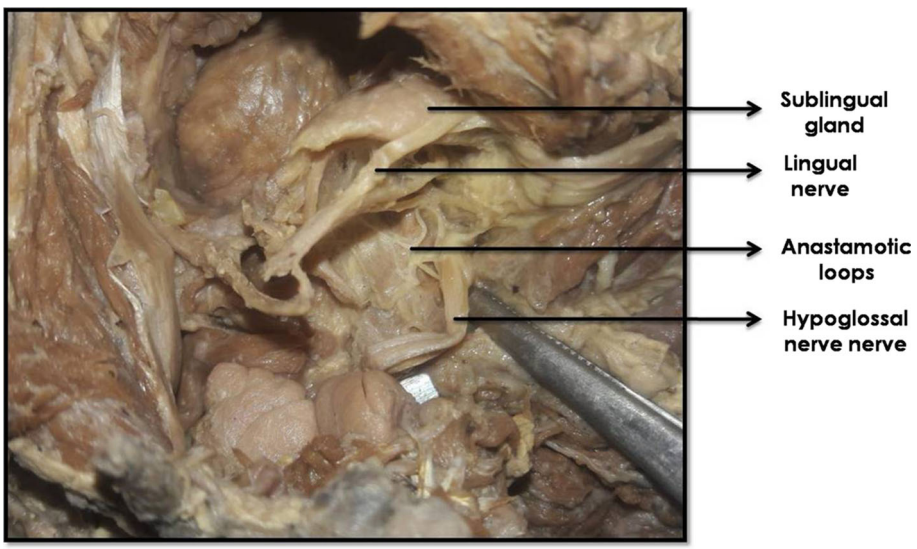

Fig. 5 Cadaveric specimen showing anastomotic loops between lingual and hypoglossal nerve

Discussion

A typical reflex arc comprises of: neural receptors located on sensory fibers, afferent pathway of sensory fibers, central synaptic connections that almost always uses interneurons, and the efferent pathway composed of the motoneurons, or autonomic post-ganglionic neurons innervating the effector organ [12].

In lingual hypoglossal reflex—afferent pathways are through lingual nerve, efferent being the hypoglossal nerve and reflex action is due to the synaptic connections between these two nerves [1] (Fig. 6). When a mechanically stimulation is given to filiform papillae, the tip of the tongue curves upward and on electrical stimuli the tongue hollows from side to side with the upward curvature of lateral edges and depression of back of the tongue as demonstrated by Miller and Sherrington which correlated with our patient [13].

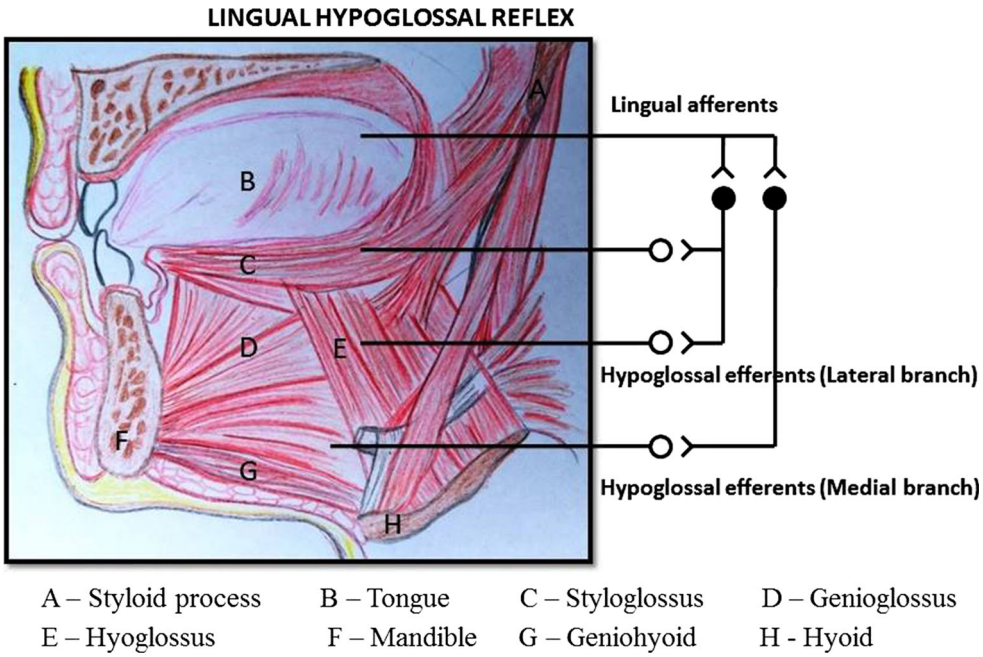

Fig. 6 Synaptic connections between lingual and hypoglossal nerve

Thus, the motor response in the hypoglossal nerve is induced due to specific sensory inputs given to lingual nerve subconsciously resulting in the above said movements. Stimulation of either lingual nerve produced bilateral but not symmetrical movement of the tongue.

Tongue has four intrinsic and extrinsic muscles which coordinates its various movements like protrusion, retrusion and side-to-side movement. Except palatoglossus, all are innervated by hypoglossal nerve. The protrusive and intrinsic muscles of tongue are innervated by medial branch, and the retrusive muscles are innervated by lateral branch of hypoglossal nerve [14, 15]. The elicited response on hypoglossal motor neurons depends on the site of sensory input and depending on the action of muscles in response to stimulation [16].

Lingual hypoglossal reflex occurs in the tongue due to the post-synaptic transmission of sensory inputs to hypoglossal nerve via lingual nerve stimulation. Eventually retrusive movements of tongue occur predominantly as stimulation of lingual nerve results in suppression of the protrusive muscles [1].

In order to assess whether the reflex was seen to involve the strap muscles of the neck, stimulation of neck was done, but contraction of infrahyoid muscles was not noted. This infers that the reflex arc to be apparently above the Pirogov's triangle of the neck.

Communication patterns between lingual and hypoglossal nerves on cadaveric dissections and autopsies have been reported in the literature. In 1958, Fitzgerald and Law studied about the interconnections between these two nerves in specimens of humans, pigs, cats, dogs and rabbits. Peripheral connections between the lingual nerve and the lateral and medial divisions of the hypoglossal nerve have been described by them in all the specimens except rabbits. Laterally located connections were found more commonly in humans, whereas medially located connections were seen in cats, dogs and pigs [7].

Rusu et al. [17] in his cadaveric dissection of 12 specimens demonstrated lingual hypoglossal nerve synapses in:

1. Anterior border of hyoglossus muscle where the first translingual branch of lingual nerve was connected either to main trunk of hypoglossal nerve or to the styloglossal or hyoglossal branch of hypoglossal nerve.
2. Six sides revealed a thin anastomosis between the hypoglossal nerve and the lingual nerve trunk which was consistent with our findings obtained through cadaveric dissections.

Zur et al. with his Sihler's staining confirmed the interconnections between the lingual and hypoglossal nerve in the tongue. His study on five autopsied tongue showed the medial and lateral branches of the lingual nerve innervating the middle and anterior thirds of tongue, respectively, which were restricted to the ventrolateral tongue mucosa, and both had anastomotic branches with the hypoglossal nerve in the body of the tongue. They also noted such anastomotic branches to be 0.25 mm in diameter. They reported that the presence of such communications leads to the stimulation of hypoglossal nerve while passing a probe or mechanical stimuli to the lingual nerve leading to what is known as "lingual hypoglossal reflex" [15].

Blom and Skoglund [18] found reflex discharges on the twelfth cranial nerve upon stimulation of lingual nerve.

Nakahara et al. [11] were the first to record reflex discharge in the hypoglossal nerve caused by gustatory stimulation of the tongue in frogs. INOUE investigated the relation between stimulating portions of the tongue and the reflex discharges in the branch to the hyoglossal and genioglossal branch of hypoglossal nerve. He found that both branches generated reflex discharges in the tip of the tongue in response to the given stimulations [10].

Tongue deviation which occurs as part of this reflex may also be associated with an undiagnosed medullary lesion close to the hypoglossal nucleus. In our patient, MRI brain was done which ruled out lesion in the medulla oblongata. The other implication of this reflex is that this may explain traumatic ulcers of lateral border of tongue of unknown etiology.

Conclusion

Lingual hypoglossal reflex is a rare phenomenon in humans. Examination of this reflex is not done routinely in clinics. It is a proven fact that manifestations of oral cavity act as mirror for the underlying systemic lesions/conditions. Hereby, the authors emphasize the importance of its recognition and to include stimulation of tongue with a probe as a part of their routine chair side examination.

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Compliance with Ethical Standards

Conflict of interest All authors declare that they have no conflict of interest.

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