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CASE REPORT

Gustatory dysfunction after mandibular zoster

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Abstract Varicella zoster, limited to the mandibular nerve, is rare. Classical symptoms are pain, hypesthesia and vesicular eruption restricted to the third trigeminal segment (V3). Little is known on taste affection after mandibular nerve zoster. We report two cases of patients suffering from mandibular zoster associated with subjective taste disorder. In both cases, gustatory measures confirmed ipsilateral hemiageusia of the anterior two-thirds of the tongue. After 2 months, the symptoms regressed and psychophysical measures came back to normal values, whereas post-zoster neuralgia lasted for more than 1 year. Gustatory dysfunction is a possible symptom after mandibular nerve zoster. In contrast to post-zoster neuralgia, taste function seems to recover quickly.

Keywords Zoster · Gustatory · Taste · Trigeminal · Mandibular · Dysgeusia · Tooth extraction

Introduction

Zoster limited to the mandibular nerve is rare. Emotional stress and compromised immune system are favoring factors. It has also been reported to occur several days after dental treatments [1, 2]. The clinical course is mostly self-limiting, but severe complications such as neuralgia, tooth loss, mandibular necrosis or tooth growth disorders, have been reported [3–5]. Despite the expected affection of the anterior two-third of the hemi-tongue (lingual nerve), taste disorders after mandibular zoster and their clinical course have almost never been reported [6]. The aim of the present study was to assess and follow up gustatory function in patients with zoster limited to the mandibular branch.

Materials and methods

Case 1

An otherwise healthy 73-year-old woman underwent root canal treatment of the first left upper premolar tooth (tooth 25) 5 days before visiting our outpatient clinic. Two days later, she noticed a burning pain on the left inferior half side of her face and the left anterior hemi-tongue. The day before coming to the clinic, she also developed left ear numbness, left ear tinnitus and vesicular eruption from the left parotid region and cheek to the chin (V3 segment), as well as on the left anterior two-thirds of the tongue. Pulling out the tongue was painful. She was complaining of total hemiageusia without dysgeusia (e.g., bad or metallic taste) on the left tongue side.

The examination showed a left facial vesicular scabby exanthema of the skin of the third trigeminal root (V3), extending from the tragus to the chin including the mandibular skin, as well as similar lesions on the posterior wall of the external auditory duct (EAD) and the tympanic membrane. Within the oral cavity, vesicular lesions were spread from the left cheek mucosa onto the inferior left part of the tongue. A lateralized Weber tuning fork test to the

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left side and a positive Rinné test on both sides were found upon formal audiometry. Pure tone audiometry revealed slight asymmetric thresholds, with the left side being 10 dB less sensible than the right side. Both thresholds were within normal ranges. There was no nystagmus or any facial weakness. The cutaneous lesions had the typical aspect of varicella zoster virus (VZV) reactivation.

Taste testing, performed with "Taste Strips" [7], showed 3 correctly identified tastants out of 16 presentations on the left anterior two-thirds of the tongue, against 13 on the right side: this reflects a severely impaired gustatory function of the left anterior two-thirds of the tongue, compared with a normal gustatory function of the right tongue side. Olfactory function was normal (*n*-butanol threshold of 7.75 points [8]).

We concluded on zoster of the V3 branch of the trigeminal nerve. The diagnosis was confirmed by direct immunofluorescence from the swab of oral vesicular lesions showing VZV antigen. We suspected that the lateralized tuning fork test and the tinnitus might be due to vesicular affection of the tympanic membrane and involvement of the tensor tympanic muscle, which is also innervated by branches of the mandibular nerve.

We decided to treat her with an oral antiviral treatment (Valtrex 1 g, 3×/day) and a topical anti-inflammatory cream (Tannosynt, 2×/day) for 10 days. On the next day, she came back because of an increasing erythema and we diagnosed a secondary infection with a cellulitis from the left inferior face to the upper thorax. She was then hospitalized for intravenous and topical antibiotic therapy (Floxapen 500 mg, 3×/day and Fucidine locally, 2×/day), continuing the antiviral therapy (Valtrex 1 g, 3×/day). After 7 days of treatment, the cellulitis regressed and she could be discharged.

Two weeks after the onset of her zoster, pain, taste loss, hypesthesia and allodynia of the left cheek region worsened. Physical examination, at that moment revealed a mottled aspect of the skin of the left V3 region with some residual scabs and an acute external otitis. The latter was successfully treated with topical instillation of a gentamicin and steroids cream. In addition to the acute external otitis, an astonishingly quick onset of post-zoster neuralgia probably contributed to the facial pain. At this time, the oral cavity lesions had disappeared and the mucosa was normal.

She noticed a completely recovered taste function within 2 months after the onset of the symptoms. Gustatory function of the left anterior two-thirds of the tongue returned gradually back to normal, with taste strip scores of 9 and 12 out of 16 points (on the left side), at 2 and 6 months, respectively. The results of the right side tests did not vary during this period. However, saliva seemed to be changed, with a more viscous and permanent "fatty" consistency in the oral cavity, and she was still complaining of hypesthesia and paresthesia on the very anterior left side of the tongue. At her last visit, more than 1 year after the onset of her zoster, she was still suffering from persistent left V3 post-zoster neuralgia and left V3 hypesthesia.

Case 2

A 34-year-old HIV-positive female patient visited our outpatient clinic with a vesicular scabby exanthema of the right V3 region. She had noticed a deep pain sensation of this region several days before the cutaneous eruption occurred, associated with right temporo-mandibular joint pain, right-sided hypesthesia of the face (limited to V3), oral burning and bitter dysgeusia. Examination showed vesicular lesions on the V3 segment of the right face skin and on the right hemi-tongue. Diagnosis was made by direct immunofluorescence from swabs of the oral vesicular lesions, showing VZV antigen. We treated her with an oral antiviral treatment (Valtrex 1 g, 3×/day) and a topical anti-inflammatory cream (Tannosynt, 2×/day) for 1 week. The taste tests showed 8 out of 16 correctly identified taste strips on the right anterior two-thirds of the tongue, against 13 on the left side. This reflects a slightly diminished gustatory function of the right anterior two-thirds of the tongue, compared with a normal gustatory function of the left side.

Like the first patient, she recovered quickly from her taste affection and bitter dysgeusia disappeared. Taste measures came back to normal values 2 months after initial testing, but post-zoster neuralgia lasted for more than 6 months.

Discussion

The present two cases illustrate well that gustatory function may be affected in mandibular VZV reactivation. These two cases also suggest that gustatory function recovers quicker than somatosensory function after mandibular zoster. None of the present cases developed permanent dysgeusia, although both cases developed post-zoster neuralgia.

The third trigeminal branch, the mandibular nerve (V3), further divides into two main trunks. Before this division, several small branches leave the main trunk of the V3, namely the nervus spinosus (sensory innervation of parts of the meninges), the medial pterygoid nerve and motor nerves to the tensor tympani and soft palate tensor muscles. Then, the V3 divides into anterior and posterior branches: the anterior branch gives raise to the masseter, deep temporal, buccal and lateral pterygoid nerves, whereas the posterior branch gives rise to the inferior alveolar, lingual

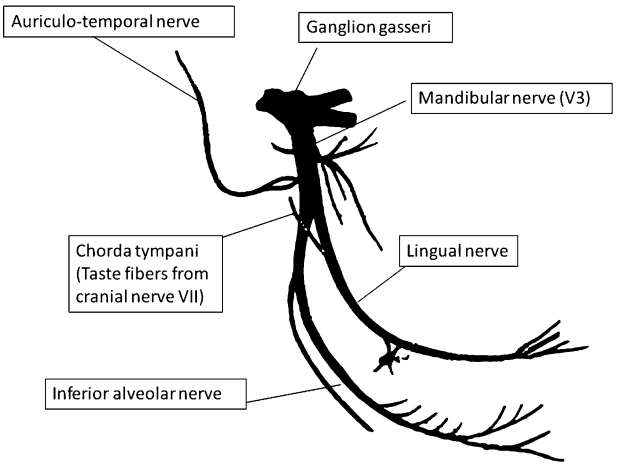

Fig. 1 Schematic representation of the mandibular nerve and its branches

and auriculotemporal nerves. While the lingual nerve carries sensory, parasympathetic and gustatory fibers for the anterior two-thirds of the tongue, the auriculotemporal nerve contains sensory fibers for parts of the external ear, EAD and tympanic membrane [9]. Further motor branches to the mylohyoid and anterior belly of the digastric muscle also split from this posterior branch. The mandibular nerve also gives off branches to the otic ganglion (Fig. 1).

The clinical presentation of mandibular zoster follows this anatomy. Besides the classical skin lesions, otological symptoms may occur due to affection of nerves giving sensory innervation to the eardrum and EAD, as well as nerves innervating muscles involved in the middle ear mechanics (tensor tympani) and Eustachian tube opening (soft palate tensor). Deep pain, associated with eating discomfort, may be due to the involvement of the numerous nerves innervating a broad range of mastication muscles. Finally, gustatory dysfunction might very likely be related to the involvement of the lingual nerve. This latter nerve gets its gustatory fibers from the chorda tympani, a branch from the facial nerve carrying taste and parasympathetic fibers, to supply the anterior two-thirds of the ipsilateral tongue with taste fibers. It can be considered as an anastomosis between the facial and mandibular nerves. Thus, an affection of the mandibular nerve may lead to taste disorders throughout the lingual branch.

It is all the more surprising that taste disorders have not yet been regularly reported or measured in mandibular nerve zoster. Apart from a single report about one patient suffering from subjective taste impairment [6], we did not find any further information on taste disorders after mandibular nerve zoster. Not surprisingly, both the patients were complaining about taste disorders, which we assessed by measured ipsilateral gustatory dysfunction. Interestingly, the first case also confirms possible VZV reactivation by dental treatment, as already described in literature [1, 2].

This is in contrast to Ramsay–Hunt syndrome, where taste disorders have already been described earlier [10]. This is probably due to the mandatory clinical feature of facial palsy in Ramsay–Hunt syndrome [11]. The gustatory fibers from the anterior hemi-tongue traveling with the chorda tympani join the facial nerve within the inner auditory canal before entering the brain stem with the intermedius nerve. This close anatomical relation between facial nerve and intermedius nerve makes patients with facial palsy probably more prone to experiencing taste disorders [12]. However, it is clinically sometimes difficult to distinguish the different presentations of zoster reactivation, especially since there is possible overlap with Ramsay–Hunt syndrome. Whereas the palsy can occur with a delay, zoster vesicles can be present or not (zoster sine herpete) and cranial nerves (CN) other than the VII and VIII can be affected concomitantly (e.g., V, IX, X) [6]. Since the present cases have never had any facial weakness or palsy along their clinical course, they are to be considered as Ramsay–Hunt syndrome.

The concomitant involvement of the external auditory meatus and eardrum with the mandibular region could be explained by viral spread through the auriculotemporal branch of the V3 or the chorda tympani. Nevertheless, the exact sensory innervation of the ear and particularly the external auditory meatus is still debated. It is generally accepted that the sensitive innervation of the concha, eardrum and EAD is supplied by the sensory fibers of the facial (Wrisberg), vagus and auriculotemporal nerves. However, the exact territories vary among authors. Moreover, recent anatomical papers on CN bring out several formerly not described distal anastomoses, such as those between the V and VII CN, the VIII and VII, as the already known tympanic plexus (CN VII and IX): this gives other possible pathways for viral spread. More macroscopic and microscopic anatomy research is needed to document these findings.

It is an interesting finding that our patients recovered quickly from their taste disorders, which was confirmed by normal taste strip scores measured 2 months after onset. The quick recovery time, after which taste disorders regressed in mandibular nerve zoster, stands in line with previously reported observations on taste dysfunction after ear or skull base surgery [13], microlaryngoscopy and tonsillectomy [14, 15]. This suggests a high regeneration potential of the gustatory system. In contrast, post-zoster neuralgia tends to last very long to disappear, from several months to even years. In light of this neuralgia, it is also remarkable that no distorted gustatory symptoms such as dysgeusia develop. Gustatory fibers may regenerate quicker than somatosensory

fibers, but, to our knowledge, little is known about the physiopathology of neural involvement by VZV.

Taken together, taste disorders may occur in mandibular nerve zoster. Based on two observations, it is not possible to claim dysgeusia as a regular symptom. However, taste dysfunction seems also to be transient, as seen in the present cases. We hope further reports will confirm and extend our surprising findings.

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Conflict of Interest None.

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