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Case Report Open Access

Intraoral Sensory Abnormalities Caused by Tooth Extraction in a Patient with Chronic Inflammatory Demyelinating Polyneuropathy

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Abstract

Background: Chronic inflammatory demyelinating polyneuropathy (CIDP) is considered to be an immune-mediated, acquired disease of the peripheral nerves. CIDP patients infrequently develop neuropathy with various clinical forms, such as blink reflex and trigeminal neuropathy. However, to our knowledge, there is no information about intraoral somatosensory changes associated with CIDP. Here, we report the case of a patient with CIDP who exhibited somatosensory changes investigated by quantitative sensory testing (QST).

Case Report: A 77-year-old Japanese man was referred to our institution with symptoms of intraoral pain and swollen gingiva. The patient had been diagnosed with CIDP 8 years earlier. The second premolar of the right mandible was diagnosed with apical periodontitis, and tooth extraction was performed to control his pain and inflammation. Following the tooth extraction, local inflammation and pain subsided, however, the patient reported spontaneous dysesthesia on the adjacent gingiva of the extracted tooth. Intraoral qualitative sensory testing of the gingiva showed gingival mechanical allodynia and cold hypoalgesia at the extraction site. We diagnosed the patient with painful post-traumatic trigeminal neuropathy related to tooth extraction.

Conclusion: Because of the lack of evidence for peripheral, organic changes in the oral mucosa, the sensory changes may be best explained by peripheral disturbance, an association may exist between these reported somatosensory changes and CIDP.

Keywords: Hyperalgesia; Neuralgia; Pain; Tooth extraction

Introduction

Chronic inflammatory demyelinating polyneuropathy (CIDP) is considered to be an immune-mediated, acquired disease of the peripheral nerves. CIDP is a rare yet underestimated condition, with an estimated incidence of 1-7.7/100,000 persons [1,2]. CIDP patients infrequently develop neuropathy [3,4] with various clinical forms, such as blink reflex and trigeminal neuropathy [5,6]. However, to our knowledge, there is no information about intraoral somatosensory changes associated with CIDP. Here, we report the case of a patient with CIDP who exhibited somatosensory changes investigated by quantitative sensory testing (QST).

Case Report

A 77-year-old Japanese man with poor oral hygiene was referred to the Department of Oral and Maxillofacial Surgery, Tokyo Women's Medical University Hospital, with symptoms of intraoral pain and swollen gingiva. The patient was previously diagnosed with CIDP 8 years earlier and had been receiving treatment for chronic renal failure caused by glomerulonephritis for 12 years. The patient reported difficulty in tooth brushing due to muscle weakness and loss of upper limb strength associated with CIDP. The right mandibular second premolar was diagnosed with apical periodontitis and the tooth was subsequently extracted to control pain and inflammation. Following tooth extraction, local inflammation and pain subsided, however, the patient reported spontaneous pain (3 on a numerical rating scale ranging 0-10) and perceptual distortion at the gingiva adjacent to the extracted tooth. Intraoral QST with a Q-tip to investigate mechanical allodynia and testing with a thermal device to investigate warm and cold perceptions [7] at the affected site indicated mechanical allodynia and cold hypoalgesia compared with the contralateral side. According to the above findings, we diagnosed the patient with painful post-traumatic trigeminal neuropathy related to tooth extraction. Interestingly, mechanical allodynia was observed in the molar region of bilateral mandibles and first molar regions of the right maxilla where the tooth extraction was performed.

Discussion

CIDP is an acquired disease of the peripheral nerves with a hallmark presentation of multifocal demyelinating lesions. Since its first description as a clinically well-defined entity, it has been mostly characterized as a predominantly motor, chronic progressive or relapsing diffuse polyneuropathy [8]. Previously, Cruccu reported facial nerve abnormalities [9], and Kimura presented a significantly higher frequency of facial nerve conditions in a CIDP patient compared with diabetic polyneuropathy patients [6]. However, intraoral somatosensory changes were not described because of difficulties in sensory testing in the intraoral region due to the specific structure of the oral cavity. In the present case, we used intraoral thermal devices specifically developed for intraoral QST, which we

previously demonstrated had sufficient reliability [7]. Interestingly, in the present case report we identified gingival mechanical allodynia and cold hypoalgesia at all extraction sites. However, to our knowledge, no previous reports have described abnormalities of intraoral sensory loss or gain in CIDP patients. Because of the lack of evidence for peripheral, organic changes in the oral mucosa, sensory changes may be best explained by peripheral disturbance.

The findings of the present case suggested that tooth extraction could be a possible cause of small nerve fiber injury [10] and associated tooth extraction evoked intraoral sensory abnormalities such as allodynia and perceptual distortion. Somatosensory changes were also found to be associated with the present case of CIDP. The mechanisms of peripheral neurological changes of CIDP have not yet been clarified, however, intraoral somatosensory changes related to CIDP may be due to progressive lesions associated with this disease. Thus, continuous follow-up and QST to assess disease progression may be helpful to clarify these issues. To our knowledge, this report is the first to describe intraoral manifestations and somatosensory changes in a patient with CIDP. However, a greater accumulation of cases is needed to clarify intraoral abnormalities of CIDP patients.

Conclusion

Because of the lack of evidence for peripheral, organic changes in the oral mucosa, the sensory changes may be best explained by peripheral disturbance, an association may exist between these reported somatosensory changes and CIDP.

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