Intraoral Sensory Abnormalities in A Patient with Juvenile Fibromyalgia

Takuya Naganawa1*, Abhishek Kumar2, Hitoshi Sato1, Toshihiro Okamoto1 and Tomohiro Ando1

1Department of Oral and Maxillofacial Surgery, Tokyo Women’s Medical University, Japan
2Department of Dental Medicine, Karolinska Institutet, Sweden

Submission: January 30, 2018; Published: August 24, 2018

*Corresponding author: Takuya Naganawa, Department of Oral and Maxillofacial Surgery, Tokyo Women’s Medical University School of Medicine, 8-1 Kawada-cho, Shinjuku-ku, Tokyo 162-8666, Japan; Email: tanaganawa@gmail.com

Abstract

A 9-year-old Japanese girl was referred to our institution with symptoms of intraoral burning pain. The patient had been diagnosed with juvenile fibromyalgia 6 month earlier. Intraoral qualitative sensory testing showed mechanical allodynia and heat hyperalgesia on the intraoral mucosa (tongue and cheek). Because of the lack of evidence for peripheral, organic changes in the oral mucosa, the sensory changes may be best explained by peripheral disturbance. According to the above findings, we diagnosed the patient with burning mouth syndrome (neuropathy), and we consider an association may exist between these reported somatosensory changes and JFM. The psychological managements by a child psychiatry specialist and hospitalization for changing their environments were performed. Moreover, the tooth brushing technique with avoiding the sites of allodynia was instructed carefully. After the 6 months of the management, the body pain and intraoral burning pain were reduced 7 to 2 on a NRS. The continuous dental managements may be important because of the difficulties of dental treatments in a condition of intraoral burning pain with allodynia [1-5].

Keywords: Juvenile fibromyalgia, Allodynia, Sensory changes, Intraoral qualitative sensory testing; Peripheral; Allodynia; Parts of body; Neuropathic pain; Brushing technique; Dental treatments; Patients; Tongue; Cheek; Similar Drugs; Hyperacusis; Knowledge

Abbreviations: JFM: Juvenile Fibro Myalgia; QST: Quantitative Sensory Testing; ACR: American College of Rheumatology

Introduction

Juvenile fibromyalgia (JFM) is a non-inflammatory chronic pain condition that most frequently affects the children at approximately 9-15 years of age. The male: female occurrence ratio of JFM is about 1: 4-8. It is a common, multifactorial disease, diagnosed on the basis of the total sum of painful illness, chronic fatigue, hypothermia, and many other autonomic signs / symptoms. JFM is characterized by constant widespread pain in different parts of body, poor sleep quality, daytime sleepiness and an altered mood. Various clinical forms of JFM are reported previously, however, to the best of our knowledge, there is no information about intraoral manifestations and somatosensory changes (neuropathy etc.) associated with JFM. Here, we report the case of a patient with JFM who exhibited intraoral somatosensory abnormalities [6].

Case report

A 9-year-old Japanese girl was referred to the Department of Oral and Maxillofacial Surgery, Tokyo Women’s Medical University Hospital, with symptoms of intraoral burning pain 7 on a numerical rating scale (NRS) ranging 0-10. The patient was previously diagnosed with JFM 6 month earlier. The patient was wearing sun glasses at all the time for protection of dazzling and reported hyperacusis. The patient reported difficulty in tooth brushing due to intraoral burning pain although intraoral examination revealed fair oral health status. An Intraoral Quantitative Sensory Testing (QST) with a Q-tip was performed to investigate mechanical allodynia and sensitivity to warm and cold perceptions were tested with a thermal device [7] The QST tests performed on the intraoral mucosa indicated mechanical allodynia on the tongue and cheeks, bilaterally. Although the NRS of heat stimuli on the tongue and cheek were high, there was no differences between right and left sides. According to the above findings, we diagnosed the patient with burning mouth syndrome (neuropathic pain).

The psychological managements by child psychiatry specialist and hospitalization for changing their environments were performed. Moreover, the results of intraoral sensory testing were identified with the patient, and then the tooth brushing technique with avoiding the sites of allodynia was instructed carefully. After the 6 months of the management, the body pain and intraoral burning pain were reduced 7 to 2 on...
In a trial with a small number of children, Pregabalin, tramadol, and gabapentin have been reported as effective. However, these agents are not proved to be efficacious. Moreover, these agents should be systematically tested if they are to be clinically used.

In this case, the psychological management with hospitalization was performed. Consultation (three times a week) by child psychiatry specialist and the temporarily isolation from her home environment were effective in this case. Following the results of the intraoral sensory testing, the diagnosed ‘burning mouth syndrome (neuropathic pain)’ was identified. The identified symptoms were shared with the patient, and then the tooth brushing technique was instructed carefully with avoiding the painful sites. The pain was relieved by child psychiatry specialist and the temporarily isolation from her home environment were effective in this case.

Discussion

JFM is characterized by constant widespread pain in different parts of the body, poor sleep quality, daytime sleepiness and an altered mood. The quality of life of individuals with chronic pain and their caregivers is severely restricted and the occurrence of symptoms of anxiety and depression is fairly common in this population. In this case, a NRS of 7 to 2 on a NRS without any medication treatments. Juvenile fibromyalgia has not been sufficiently appreciated thus far. Case collection is so sparse that specific diagnostic criteria have not been created. The most frequently used diagnostic criteria are those for adults that were set forth by the American College of Rheumatology (ACR) in 1990. These criteria include the following as central findings: pains persist for 3+ months over a wide area of the body and tender points are detected at 18 site. The tender point can be detected only when an appropriate site is pressed and it evades detection if a pressure site is distant from the appropriate site by as little as 5 mm. Physical examination of the joints as well as of the muscles of the entire body is crucial, including simultaneous detection of tender points, which is indispensable for making a diagnosis of juvenile fibromyalgia. The intraoral finding was not including these diagnostic criteria, moreover, there are no information of intraoral manifestation and sensory changes so far. Somatosensory changes were also found to be associated with the present case of JFM. The mechanisms of peripheral neurological changes of JFM have not yet been clarified, however, intraoral somatosensory changes related to JFM may be due to progressive lesions associated with this disease. Thus, continuous follow-up and sensory testing 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 JFM. Therefore, a greater accumulation of cases is needed to clarify intraoral abnormalities of JFM patients.

References

1. Naganawa T, Baad-Hansen L, Iida T, Ando T, Svensson P (2015) Assessment of human intraoral thermal sensitivity with simple devices in the clinic: implications for orofacial pain conditions. J Oral Facial Pain Headache 29(1): 93-99.
2. Yokota S, Kikuchi M, Miyamae T (2013) Juvenile fibromyalgia: Guidance for management. Pediatr Int 55(4): 403-409.
3. Wolfe F, Smythe HA, Yunus MB, Bennett RM, Bombardier C, et al. (1990) The American College of Rheumatology 1990 criteria for the classification of fibromyalgia: Report of the multicenter criteria committee. Arthritis Rheum 33(2):160-172.
4. Yunus MB, Maci AT (1985) Juvenile primary fibromyalgia syndrome. A clinical study of thirty-three patients and matched normal controls. Arthritis Rheum 28(2): 138-1345.
5. Kashikar-Zuck S, Graham TB, Huenefeld MD, Power SW (2000) Juvenile primary fibromyalgia syndrome. Arthritis Care Res 13: 388-397.
How to cite this article: Takuya N, Abhishek K, Hitoshi S, Toshihiro O, Tomohiro A. Intraoral Sensory Abnormalities in A Patient with Juvenile Fibromyalgia. Open Access J Neurol Neurosurg. 2018; 8(2): 555738. DOI: 10.19080/OAJNN.2018.08.555738.

6. Yokota S (2009) Juvenile fibromyalgia: Guidance for diagnosis and management. In: Nishioka K (Ed.) Guideline for Management of Fibromyalgia. Medical Review, Tokyo, pp.75-82.

7. Yokota S, Umebayashi H, Miyamae T (2007) Our experience of 3 cases with childhood fibromyalgia. J Jpn Pediatr Soc 111: 462-468.

8. Miyamae T, Yokota S (2008) The actual state and clinical features of fibromyalgia in our country. J Jpn Pediatr Soc 112: 1769-1777.

9. Naganawa T, Sato H, Kumar A, Iida T, Nagawa E, Okamoto T Ando T (2015) Clinical Presentation of Oral Manifestations and Intraoral Somatosensory Changes in Fahr's Disease. J Pain Relief 4: 214-216.

10. Naganawa T, Kumar A, Sato H, Uchida J, Okamoto T Ando T. Intraoral sensory abnormalities caused by tooth extraction in a patient with chronic inflammatory demyelinating polyneuropathy. J Clin Neurosci In Submission.

11. Naganawa T, Baad-Hansen L, Ando T, Svensson P (2015) Influence of topical application of capsaicin, menthol and local anesthetics on intraoral somatosensory sensitivity in healthy subjects-temporal and spatial aspects. Exp Brain Res 235(4): 1189-1199.

Your next submission with Juniper Publishers will reach you the below assets

- Quality Editorial service
- Swift Peer Review
- Reprints availability
- E-prints Service
- Manuscript Podcast for convenient understanding
- Global attainment for your research
- Manuscript accessibility in different formats (Pdf, E-pub, Full Text, Audio)
- Unceasing customer service

Track the below URL for one-step submission

https://juniperpublishers.com/online-submission.php

This work is licensed under Creative Commons Attribution 4.0 License
DOI: 10.19080/OAJNN.2018.08.555738