

CASE REPORT

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Traumatic neuroma on the tongue tip: A case report and review of the literature

Yosuke Jinno, Mitsuhiro Aoki, Hideki Mori

ABSTRACT

Introduction: Traumatic neuroma is a non-neoplastic proliferation of a nerve occurring in response to an injury or surgery. Traumatic neuroma on the tongue tip is a very rare proliferating lesion in the oral cavity. We present a case of traumatic neuroma on the tongue tip. This case may be valuable for understanding the pathogenesis of the neuroma.

Case Report: A 79-year-old man developed a painful nodule on the tongue tip. Histologically, the nodule consisted of a tortuous proliferation of nerve fascicles, and diagnosed as a traumatic neuroma. The bundles of the neuroma expressed a strong activity of the neural marker S-100. Expression of alpha smooth muscle actin (α -SMA) being a phenotypic marker of myoblast activity was also positive in the surrounding connective tissues of the nodule suggesting a relation to the painful natures of the neuroma. Cross-sectioned nerve bundles of the neuromas seemed to be 5–7 times larger than of the normal bundles suggesting intensity of proliferation the nerve bundles. The neuroma was considered to occur by repeated stimuli of the tongue tip to the front teeth.

Conclusion: Traumatic neuroma on the tongue tip is quite rare. Onset of the neuroma was suggested repeated stimuli of the tongue tip to the front teeth. Expression of

α -SMA of the bundles of the neuroma seemed to accord with the painful natures of the neuroma.

Keywords: Pain, Tip, Tongue, Traumatic neuroma

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INTRODUCTION

Traumatic neuroma is a well-known disorder involving peripheral nerves, which usually occurs to an injury or surgery. The lesion is not a true neoplasm; rather it presents an exaggerated response consisting of reactive hyperplasia of the nerve tissue. Although they are not true neoplasms, the pain associated with traumatic neuromas can be usually untense and debilitating. The principal features of oral traumatic neuroma are similar to the traumatic neuromas elsewhere in the body. The most common oral sites are lower lip, tongue, and mental nerve area. Clinically, traumatic neuromas are divided into painful (symptomatic) type and painless (asymptomatic) type. As specific natures of the traumatic neuroma are present the recurrence in the same place [1] and spontaneous remission [2]. So far as we are concerned, few cases of traumatic neuroma of the tongue tip have been described. Following recent clinicopathological analysis of 157 cases of oral neural tumors in last 44 years period, 12 cases of traumatic neuromas of the tongue were diagnosed. Of them, cases on dorsal tongue were 7 (50.1 %), those on lateral tongue was 4 (16.7%), respectively. Cases on tip of tongue was only 1 (4.2%) [3]. We present here a case of painful traumatic neuroma on the tongue tip. This case of the

Yosuke Jinno¹, MD, PhD, Mitsuhiro Aoki², MD, PhD, Hideki Mori³, MD, PhD

Affiliations: ¹Department of Dental and Oral Surgery, Ogaki Tokushukai Hospital, 6-85, Hayashi-machi, Ogaki City, Gifu 503-0015, Japan; ²Department of Otolaryngology, Ogaki Tokushukai Hospital, 6-85, Hayashi-machi, Gifu 503-0015, Japan; ³Department of Diagnostic Pathology, Ogaki Tokushukai Hospital, 6-85-1, Hayashi-machi, Ogaki City, Gifu 503-0015, Japan.

Corresponding Author: Hideki Mori, Department of Diagnostic Pathology, Ogaki Tokushukai Hospital, 6-85-1, Hayashi-machi, Ogaki City, Gifu 503-0015, Japan; Email: hi4de7mo6ri1@gmail.com

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traumatic neuroma may be valuable for understanding the pathogenesis of the neuroma.

CASE REPORT

A 79-year-old man was referred to the department of dentistry and oral surgery of our hospital for a thorough examination. The patient was complaining a painful solitary nodule on the tongue tip for last four months. He had tobacco use 10–15 cigarettes per day during the age 30–50 and drank beer 3–4 bottles (360 mL)/week. He had no constitutional symptoms except hypertension and hyperlipidemia, or history of irradiation. His family history was unremarkable. Before the surgery, he had higher serum level of neural fat (342 mg/dL) and blood sugar (120 mg/mL). No abnormal evidence was present on the examination of urine. Physical examination was notable for a 3-mm mass with polyp shape on the tongue tip. Mucous membrane surrounding the nodule was hyperemic. Multiple tongue indentations were present on the area surrounding the nodule (Figure 1). No other palpable masses or lymph nodes were found in the oral cavity, head, or neck. The mass was excised under local anesthesia.

Histologically, mild infiltration of lymphocytes and proliferation of small vessels remained in the subepithelial area at the top of the lesion (Figure 2). The nerve nodules developed and occupied widely in the submucosal space consisted of a non-encapsulated haphazard, tortuous proliferation of nerve bundles of nerve fascicles including axons, Schwann cells, and fibroblasts embedded in the background of collagens (Figures 2 and 3). Strong positivity of the bundles for a neural marker, S-100, was present, particularly in the axons (Figure 4). Alpha smooth muscle actin (α -SMA) was also positive in the peripheral area of the neural



Figure 1: Macroscopy of the tongue of this patient a week after the excision.

⇒: Excision mark. ⇓⇓⇓: Indentations.

bundles of the nodule (Figure 5). Pathological diagnosis of the nodule was traumatic neuroma. In this case, nearby the hyperplastic nerve bundles were present some of the normal nerve bundles. Cross-sectioned area of the nerve bundles of the hyperplastic lesions were approximately 5–7 times larger than of the normal bundles (Figure 3 and Table 1).

After the removal of the lesion, the patient reported complete cessation of oral symptoms.

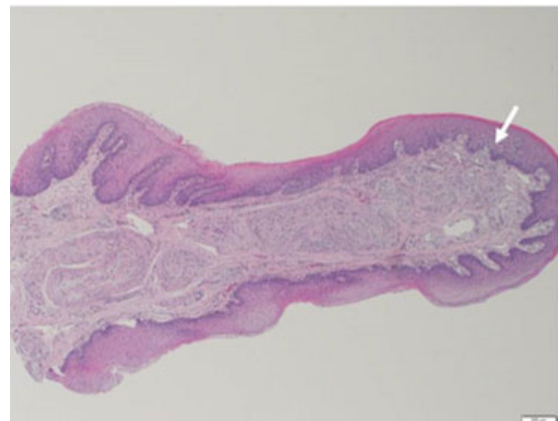


Figure 2: Excised lesion with polyp shape. Submucosal space of this polyp is occupied by the tortuous proliferation of nerve fascicles. ⇓: Chronic inflammation.

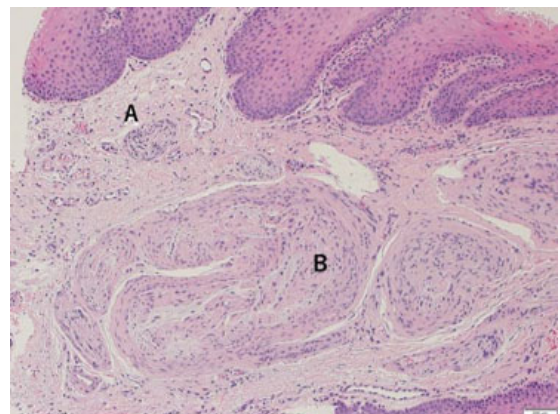


Figure 3: (A) Normal nerve bundle. (B) Hypertrophic nerve bundle.

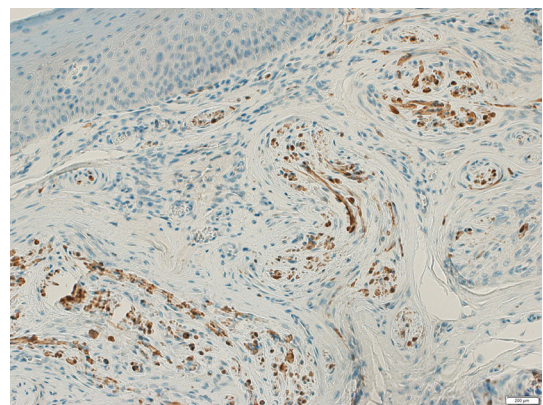


Figure 4: S-100 is expressed particularly in the axons of the bundles.

Table 1: Clinical and pathological characteristics of the traumatic neuroma on the tongue tip

Age (sex)	Type of the lesion	Size of lesion	Tongue indentation	Histological features		
				Routine HE	S-100	α -SMA
79 (male)	Painful	3 mm	+	Tortuous proliferation of nerve bundles	Particularly positive in the axons	Positive in the peripheral area of the nerve bundles

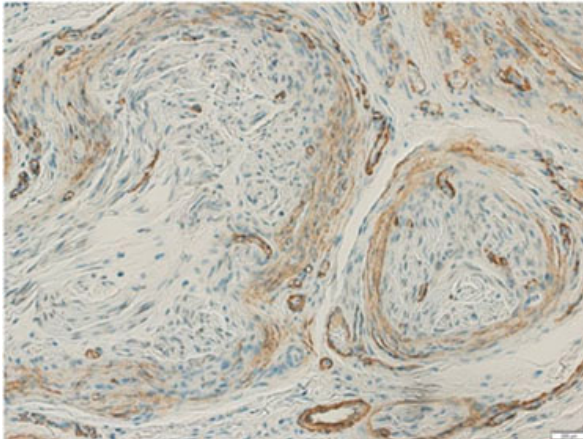


Figure 5: α -SMA is expressed in the peripheral area of the bundles.

DISCUSSION

Traumatic neuroma is regarded as a tumor-like proliferation of neural tissue that develops after a nerve has been severed or otherwise damaged. A follow-up study of the traumatic neuroma of the tongue was done by Touchette and Gessler [1]. In that study, the patient developed second traumatic neuroma (8 mm) at 18 months after the original surgery of the first neuroma (3 mm) in the same location. Etiological factors for this lesion include previous surgical procedures, crushing injuries, and lacerations. Stretching and bleeding to the surrounding tissue have also been considered [4]. However, signaling mechanisms and development pathways involved in the formation of the lesion are poorly understood [5, 6].

For the present case of the traumatic neuroma, initial phase is suggested an oral mucositis induced by repeated contact of the tongue tip to the front teeth. Similar case of the tongue in which development of neuroma was possibly related to pressing the tongue against a metal framework was reported [7]. On the first examination of the tongue, the tip area was blushing and multiple tongue indentations were recognized (Figure 1). Tongue indentation is regarded as a proof of repeated contact of the tongue to the front teeth. Thus, occurrence of the lesion will be importantly related to such extrinsic stimuli. Histological signs of chronic inflammation have been shown to be associated with painful neuromas [8]. However, pain and abnormal sensitivity can also occur in the absence of inflammation.

Weng et al. [9] identified the expression of α -SMA in painful neuromas. α -SMA is a phenotypic marker for myofibroblast activity, which contributes to increased contractile activity of myofibroblasts [10]. In this study, histological expression of α -SMA and S-100 was clear respectively in the nerve bundles. The expression of α -SMA is agreement with the painful natures of the present traumatic neuroma.

In this case, comparison of small normal nerve and hypertrophic nerves in the traumatic neuroma was possible. Results suggested that hypertrophic nerve bundles of the neuroma started up from of the small normal nerves. Furthermore, such evidences gave form to the exaggerated response to injury of nerve tissues consisting of reactive hyperplasia. It appears that common patients including this case start to be aware of abnormal feeling or pain for the nodule for past several months. Doubling time for the growth of the mass of traumatic neuroma (reactive hyperplasia) seems to be quite short. Oliveira et al. [6] documented the progression of nerve regrowth and neuroma development from the time of injury for only 90 days after amputation, in the experimental studies.

Accordingly, present case of painful traumatic neuroma in the tongue tip may be valuable for understanding of the pathogenesis of traumatic neuroma.

CONCLUSION

Traumatic neuroma on the tongue tip is a very rare proliferating lesion in the oral cavity. Onset of our case was suggested a repeated contact to the front teeth. Histologically, the neural bundle of the neuroma was 5–7 times larger than of the normal neural bundle. α -SMA was expressed in the neural bundles of the neuroma. This seems to coincide with painful nature of the neuroma.

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Author Contributions

Yosuke Jinno – Design of the work, Acquisition of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Mitsuhiro Aoki – Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published,

Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Hideki Mori – Conception of the work, Design of the work, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

Source of Support

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Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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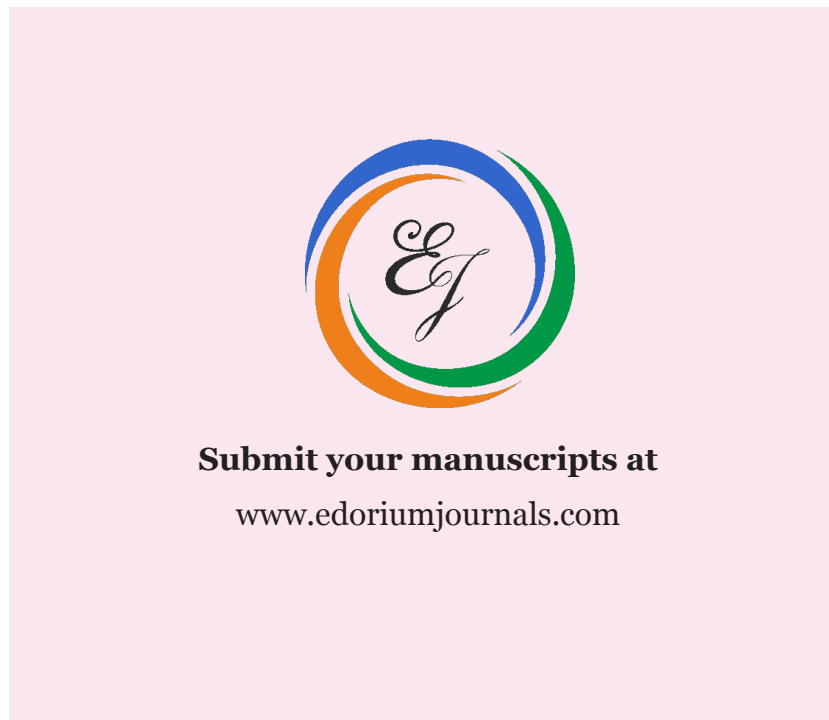
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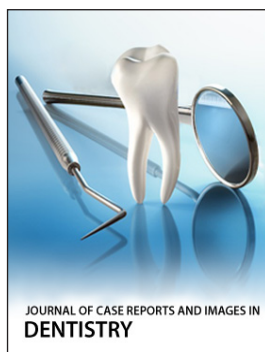
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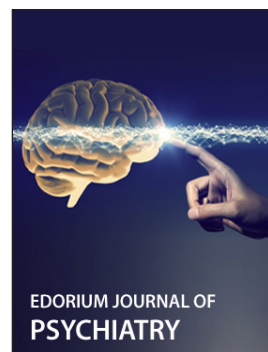
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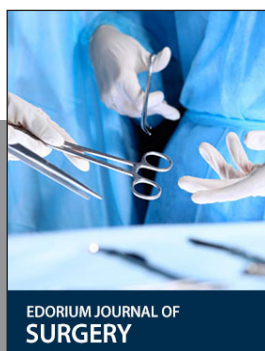
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