Traumatic Neuroma of Bilateral Mental Nerve: A Case Report with Literature Review

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Abstract

Traumatic neuroma is not a true neoplasm but a neural reaction subsequent to damage of the peripheral nerve. We would like to present a rare case of traumatic neuroma arising from bilateral mental nerve with a history of extracted lower left first molar and lower right second molar more than 10 years ago. A 58-year-old female complained of sporadic pain and numbness sensation over lower face for 2 years. Two submucosal nodules were found at bilateral mandibular vestibule next to mental foramen with mild tenderness on palpation. The panoramic radiography showed two prominent clear radiolucent shadows adjacent to the right and left mental foramens with two residual imprints of extraction wounds at #36 and #47 areas. Excisional biopsy and ablation of bilateral mental nerve with cautery electrodes were performed. Histopathologically, it was consistent with traumatic neuroma. After 1-year follow up, the painful sensation was relieved and no recurrence was found.

Key words: traumatic neuroma, mental nerve.

Introduction

Traumatic neuroma is a reactive proliferation of neural tissue after damage to peripheral nerve. It is a firm, oval, slowly growing, palpable painful or asymptomatic nodule, no longer than 2 cm with normal coloration. It may be associated with paresthesia over the injury area¹. After damage, the proximal portion of the transected or crushed peripheral nerve will regenerate and reestablish innervations to distal stump through tube of proliferating Schwann cell. However, the interposition of wound-repairing tissue between the two severed segments prevents their re-establishment. Thus, the axons proliferating randomly tangle with connective tissue. A tumor-like mass may develop²,³. Clinically, neuralgic pain with trigger point in the area of neuroma or painful hypersensitivity to normal light tactile stimuli may be the prominent symptom. It commonly present as tenderness on slightly percussion or pressure, or distortion of surrounding tissue¹.
Microscopically, the definition of traumatic neuroma is a non-encapsulated, non-neoplastic tangled mass of axons, endoneurial cells, Schwann cells, and perineural cells proliferation in a dense collagenous matrix. In some lesions, inflammatory cells can be found in the neuroma matrixes.

In the head and neck region, traumatic neuroma has been reported to occur frequently subsequent to tooth extraction, parotidectomy, radical neck dissection surgery, ramus split surgery, and incision for drainage. The purpose of this report is to present a rare but interesting case of traumatic neuroma arising from bilateral mental foramen with previous extraction history more than 10 years ago. Besides, we also reviewed literatures and discussed with clinicopathological characters for differential diagnosis with therapeutic theorem.

Case report

A 58-year-old female complained of sustained sporadic radiating pain over lower face with chin numbness for 2 years. At the relapse of the pain, she may feel diffuse referred pain extending to the neck. She had experienced of cellulites over left mandible 20 years ago. Intraoral incision and drainage of the pus over lower mandible was performed, following endodontic treatment on left mandibular first premolar (#34) and extraction of lower left first molar (#36). According to her description, the extraction was under surgical procedures with flap reflection and trimming surrounding bone. The treatment outcome was not as satisfied as she expected because of the complication of swelling and painful sensation over surgical area. However, the painful sensation disappeared six months later. The right mandibular secondary molar (#47) was extracted 10 years ago without any complication. Recently, she found two growing masses over bilateral mandible vestibule areas with painful sensation elicited while slightly palpation. Then she came to our hospital for help.

On physical examination, two submucosal movable nodules about 5 mm × 5 mm on the right site and 3 mm × 2 mm on the left side in size are palpable at mandibular vestibule area next to bilateral mental foramen (Fig. 1). Painful sensation of lower face may be elicited while slightly palpating these areas. The panoramic radiograph showed two enlarged oval, radiolucent shadows over bilateral mental foramen with extraction imprints over #36 and #47 (Fig. 2). The computed tomography (CT) did not show any contribution (Fig. 3). According to the patient’s past dental history, traumatic neuroma was the first impression. Other soft tissue tumor such as neurofibroma or schwannoma should be ruled. Operation was performed two days after admission. The patient was put in supine position under general anesthesia. Mucoperiosteal flap was reflected. With slightly and carefully dissection, the mucoperiosteum was pushed down to expose mental foramen and nerve. Two bulbous nodules were dissected at the entrance of mental nerves with hemostat and were removed with cautery electrodes (Fig. 4). The surrounding nerve tissues were also removed to make a space for preventing recurrence. The flap was reposition and sutured with 5–0 silk.

Microscopically, numerous distinct neural bundles were trapped in a densely collagenous connective tissue matrix (Fig. 5, Fig. 6). One year latter, she was free from radiating pain over lower face and neck. Although the patient still felt numbness sensation at chin area, she felt free with the treatment outcome.
Fig. 1. Two movable submucous nodules about $5 \times 5$ mm in size (A) and $3 \times 2$ mm in size (B) are palpable at mandibular vestibule areas next to bilateral mental foramens.

Fig. 2. Panoramic radiograph revealed two enlarged oval radiolucent shadows at bilateral mental foramens (white arrow) with remaining extraction imprints over #36 and #47 area.
Fig. 3. Computed tomography (CT) of mandible shows unremarkable findings of the mandible.

Fig. 4. Operation dissected the nodular mass (A) with reactive proliferation of nerve tissue along the mental nerve was exposed (B).
Fig. 5. Microscopic Feature of the lesion: (A) Numerous neural bundles were trapped with densely collagenous connective tissue stroma. (H&E stain × 100); (B) Cross-sectioned nerve bundles within dense connective tissue. (H&E stain X250)

Fig. 6. Microscopic Feature of the lesion: (A) Haphazard arranging nerve bundles proliferate within in a fibrous stroma. (H&E stain × 125); (B) Cross-sectioned nerve bundle within connective tissue. (H&E stain X250)
Discussion

Amputation neuroma was a synonym to traumatic neuroma which was first used by Odier in 1811\(^1\). The symptoms were mainly painful swelling at the end of proximal nerve stumps following nerve transected although some were asymptomatic\(^{12,13}\). Cahn was the first one who reported the first traumatic neuroma of the mental foramen in 1939. He reported a middle-aged woman suffering with severe, radiating neuralgia of the lower face and suggested the unsuspected neuromas might be a result of oral surgery\(^{6}\). In the following years, several cases confirmed of traumatic neuroma have been contributed in dental literatures\(^{14-25}\). Sist and Greene confirmed the concept that traumatic neuromas in oral cavity may account for some cases of atypical facial pain and trigeminal neuralgia. In Sist’s research, the most frequent site of painful oral traumatic neuroma was located at the mental foramen. Most cases with painful sensation may be relieved after surgical intervention although in a few cases, removal of the lesions had limited effect on the pain pattern. This may be explained with that painful impulse from periphery induces the permanent discharge in ascending PGM (pattern generating mechanisms)\(^{26}\).

Recently, the development of the mechanism of symptomatic traumatic neuroma was discussed. Chronic irritation, mechanical destruction, complicated wound healing or irregular scar developed close to the sensory nerve injury may cause the development of symptomatic traumatic neuroma. When a nerve is transected, distal axons suffer Wallerian degeneration and then the axons and Schwann cells of the proximal stump proliferate. When a barrier between the distal and proximal stump is present or the distal stump is lost, nerve proliferation continues from the proximal stump, producing a disorganized cell tangle compose of neural fibers and connective tissue extending into the surrounding soft tissue. Cytokines (such as interleulin 1b, interleukin-10, transforming growth factors-\(\beta_1\)\(^{26}\) and other extracellular signals mingle together and the wound matrix and nerve mixed with a complicated, wound extracellular matrix. The irregular nerve fibers proliferated and immersed in scar tissue. Myofibroblasts contraction inside the wound and contracted the scar. The surrounding peri- and epinural tissue around the nerve fibers are attenuated to proliferate in order to defend neural fiber from contraction injury. The defensive reaction of the nerve makes tissue constriction. Then a balance between nerve regeneration (proliferation) and damage (contraction) developed.

Traumatic neuroma does not occur in motor nerve because it has no or very few potential for regeneration when the myeline sheath of the distal stump is not connected to the proximal stump. To avoid the development of traumatic neuroma, the injured or transected nerve should be placed out of the site of potential excessive fibroproduction site and/or to avoid the external factors leading to excessive fibroproduction\(^{27}\).

In the oral and maxillofacial region, traumatic neuroma has been reported subsequent to elective surgery approach such as tooth extraction\(^{28}\), neck dissection\(^9\), ramus split surgery\(^{10}\), incision and drainage\(^{11}\) and parotidectomy\(^{29}\). In several researches, extraction may be the most common etiologic factor in oral cavity\(^{15,16,26,30}\). In the mental region, the etiologies of traumatic neuromas included tooth extraction\(^{1,12}\), ill fitted denture wearing\(^{26}\), intraoral incision for drainage\(^{11}\). Detailed past
dental history taking may contribute to precise diagnosis. In our case, the etiology may be traced back to improper incision for drainage or during the procedure of complicate extraction of #36 and #47. When the nerve was damaged, the complicated healing (i.e. healing by secondary intension, or infection) may contribute to the development of traumatic neuroma\(^27\). Interesting finding from panoramic film, the remaining extraction imprints may be found although those were performed at least more than 10 years before. The healing of complicate healing (mainly infection or healing by secondary intension) may be the main etiology in our case.

Schwannoma or neurofibroma were also to be ruled out in our case. According to Batsakis, neurogenic tumors of the head and neck regions generally do not produce neurologic signs or symptoms\(^31\). Unilocular radiolucent shadow over mandible body should be ruled out with odontogenic cyst or tumors. Residual cyst, odontogenic keratocyst, primordial cyst, ameloblastoma and central giant cell lesions should be ruled out\(^32\). However, in our case, the bilateral radiolucent shadows of mandible were considered to have fewer incidences of “true” odontogenic tumors or cyst than normal anatomic structures or reactive lesions. And the histopathological report confirmed our diagnosis.

Considered to the treatment of traumatic neuroma, the best treatment of choice is simple excision with removal of the resembling proximal stump in an area away from old scar tissue\(^3,27\). Tay et al. found that diathermy may reduce the development of traumatic neuroma to 35%, and electrocoagulation of the proximal nerve stump seems to protect against the development of traumatic neuroma\(^33\). Other treatment options include steriotactic radiosurgery, local infiltration with steroid, sympathetic nerve block and ultrasonic therapy\(^28\). In our case, we removed the lesions and surrounding tissues with cautery electrodes to avoid the recurrence.

**Reference**

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雙側下顎頦神經外傷性神經瘤：病例報告與文獻回顧

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摘 要

外傷性神經瘤是周邊神經組織受到傷害後的一種增生反應。在臨床表徵上，它可能為痛或不痛的的腫塊，通常伴隨有外傷的病史。本報告提出一罕見雙側下顎頦神經外傷性神經瘤病例。患者為一位五十八歲女性，來院主訴為下顔面部疼痛與麻木兩年之久。在下顎雙側近頦孔前庭處可觸摸到兩顆結節腫塊，在觸診時會引起患者的下顏面疼痛感。根據理學與影像學診斷，而且患者側曾患蜂窩性組織炎有切開引流的病史，右側有拔牙的病史。初步診斷為外傷性神經瘤，但不能排除神經纖維瘤與神經鞘瘤。於是為患者安排手術，切除雙側腫瘤與頦神經。組織病理切片下證實為外傷性神經瘤。患者一年後追蹤，下顏面疼痛感已解除並無復發症狀。因此病例雙側的罕見性，並針對外傷性神經瘤的成因、臨床症狀與治療做文獻回顧，故提出此報告。

關鍵詞：外傷性神經瘤，顎神經。