Trigeminal neuralgia induced by odontogenic keratocyst associated with impacted supernumerary teeth: A rare case report

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Abstract
Odontogenic keratocyst(OKC)is a cyst of tooth origin with an aggressive behavior including a high recurrence rate, it has been rechristened to keratocystic odontogenic tumor(KCOT) as it be the reflects its neoplastic nature. We report a case of KCOT in association with an impacted supernumerary tooth along with Trigeminal Neuralgia, that subsided by itself after removal of the cyst.

Keywords: Odontogenic Keratocyst, Supernumerary tooth, Trigeminal Neuralgia

INTRODUCTION
Trigeminal neuralgia (TN or TGN), also known as prosopalgia, or Fothergill’s disease is a neuropathic disorder characterized by episodes of intense pain in the face, originating from the trigeminal nerve. The clinical association between TN and hemifacial spasm is called tic douloureux.[1]

TN affects the trigeminal nerve, one of the largest nerves in the head. The trigeminal nerve sends impulses of touch, pain, pressure and temperature to the brain from the face, jaw, gums, forehead and around the eyes.

Except for a few identified organic causes, its etiology for long remained uncertain, so it was called “idiopathic” neuralgia. Even now, with the sound hypothesis of neurovascular conflict (NVC), the pathophysiology of this disease still has obscure corners.[2]

It is a well-recognized disorder that most commonly occurs in people over 50 years of age and slightly more common in women than in men.[3]

Low-intensity mechanical stimulation of the trigger zones by speaking, washing or eating can provoke pain. The condition is usually treated with carbamazepine or other anticonvulsant medications although surgical management has also been used in recalcitrant.[3-5]

Diagnosis of TN is based primarily on a history of characteristic pain attacks that are consistent with specific, widely accepted research and clinical criteria.

The diagnostic criteria of the International Headache Society (1988) are as follows:[8]
• Paroxysmal attacks of facial pain which last for few seconds to <2 min

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- Pain has at least 4 of the following characteristics
  - Distribution along one or more divisions of the trigeminal nerve,
  - Sudden, intense, sharp, superficial, stabbing or burning in quality,
  - Pain intensity is severe,
  - Precipitation from trigger areas or by certain activities such as eating, talking, washing the teeth or cleaning the face,
  - Between paroxysms, the patient is entirely asymptomatic.
- Attacks are stereotyped in the individual patient
- No neurological deficit and exclusion of other causes.

For most patients with TN, the results of their clinical examination, imaging studies and laboratory tests are unremarkable. This type of TN is called “classical TN,” and its cause is unknown although vascular compression of the trigeminal nerve roots is often suggested.[5]

In a smaller group, the signs and symptoms of TN are secondary to those of another disease process that affects the trigeminal system, such as multiple sclerosis or a cerebellopontine angle tumor or any odontogenic cysts or tumors. This type of TN is called “symptomatic trigeminal neuralgia.”[5]

The case report presented here describes an odontogenic keratocyst in relation to an impacted supernumerary tooth with symptomatic unilateral trigeminal neuralgia.

**CASE REPORT**

An 82-year-old female patient, was reported to the department of Oral Medicine having history of facial pain which was right sided, electric shock like, and confined to the right half of the lower lip and also lower half of the face on same side with duration of 10 months [Figure 1]. The patient had previously visited 3 dentists who were unable to relieve patients pain later for which she underwent extraction of decayed lower right back molar thinking it to be the root of cause [Figures 2 and 3]. Following the extraction, pain was still not relieved in the area. Then various pharmacological remedies were given like Tergretol 100mg, TID along with Gabapentin, where the pain was temporarily relieved, but patient persisted to have the shock like facial pain. The patient had a medical history of hypertensive since 15yrs and was on medication.

At the outset of the consultation and history, the patient described her pain as a dull aching pain in the area of 46, 47 and 48 (which the patient had got it removed) that was exacerbated with chewing. Clinical evaluation demonstrated an edentulous arch in relation to 46, 47 and 48 regions.
with attrition and cervical abrasion in relation to tooth 44 and 45. The tooth responded normally to temperature. Infrequently, pain of short duration radiated through the right lower half of the face. The patient stated that the pain occurred spontaneously at times – while speaking, laughing and when exposed to cold wind (especially while traveling on a 2 wheeler) and described it as electric shock like, which lasted for 2–3 s, and was so severe that the patient had tears in her eye during the attack phase.

In light of the patient’s history and clinical examination, a provisional diagnosis of residual infection-induced neuropathic pain was given.

The patient was advised to get an OPG done to rule out if any residual infection was present within the alveolus, but to our surprise, the OPG revealed an impacted supernumerary tooth within the body of the mandible, that was surrounded by an well-defined unilocular radiolucency, with a sclerotic border, that was measuring approximately 2.5 cm × 1.5 cm in size and seemed to be encroaching on the mandibular canal; supernumeraries were also present in the 1st and the 3rd quadrants [Figure 4]. To rule out any cortical plate expansion, a mandibular cross-sectional occlusal radiograph was taken, which did not reveal any expansion [Figure 5].

Based on the radiological appearance a diagnosis of dentigerous cyst was made, with a differential diagnosis – Odontogenic Keratocyst & cystic Ameloblastoma was given.

She was further investigated with CBCT to rule out, if the lesion was impinging on the mandibular canal. The CBCT reports revealed the involvement of the nerve [Figures 6-8 and Table 1].

Following the radiographic findings including CBCT reports, the patient was advised excision of the lesion. She underwent medical workup with complete hemogram, liver function test and peripheral smear, which were within normal limits and a physician consent was obtained.

The surgical procedure ENUCLEATION was done in toto under local anesthesia. The patient was given antibiotics and anti-inflammatory drugs for 5 days [Figures 9-14].

Postoperative healing was uneventful, but the patient had paresthesia of the right half of the lower lip and the patient was advised to take neurobion forte daily for 30 days.

The excised specimen [Figure 13] was sent for histopathologic examination which revealed fibrocollagenous tissue lined by stratified squamous epithelium exhibiting palisading of basal epithelium and luminal surface containing wavy parakeratotic epithelial cells. Subepithelium shows mixed inflammatory cell infiltrate along with congested blood vessels [Figure 15].

During follow-up, there was a drastic improvement in her symptoms; hence, carbamazepine was tapered and stopped after a month.

DISCUSSION

Trigeminal nerve neuropathy can underlie symptomatic TN and can involve any segment of the nerve, from its central origins to its peripheral branches. Multiple sclerosis
and infarct are the most common abnormalities of the brainstem that cause TN. It is rarely attributed to an dentoalveolar disease like odontogenic cyst & tumor, which are the conditions that makes diagnosis of the disorder so difficult.

Furthermore, TN can mimic dentoalveolar disease, as it did in this patient’s case. Most of the tumor’s that are known to lead to the development of trigeminal neuralgia (acoustic and trigeminal schwannomas, meningiomas, epidermoid cysts, lipomas and metastases) are generally located in the posterior fossa.7,8

A keratocystic odontogenic tumor (KCOT) is a rare, benign but locally aggressive developmental cystic neoplasm. It most often affects the posterior mandible. It was previously called OKC.9

Figure 7: Axial view on cone-beam computed tomography

Figure 8: Cross-section view on cone-beam computed tomography

Figure 9: Soft-tissue incision placed distal to 45

Figure 10: A bony trough was placed on the alveolus

Figure 11: The lesion was retracted and here we can see the attachment of the lesion to the mandibular nerve

Figure 12: Complete removal of the lesion and the impacted tooth with thorough irrigation
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KCOTs are thought to arise from the dental lamina and associated with impacted teeth. Multiple odontogenic Keratocysts are a feature of nevoid basal cell carcinoma syndrome. Odontogenic Keratocysts are derived from the remnants of the Dental Lamina. And is known for its high recurrence rate, aggressive behavior.\textsuperscript{[10,11]}

KCOT can be found in the mandible and the maxilla but are twice as common in the mandible, with a predilection for the angle and ascending ramus. KCOT can cause TN when the cyst compresses on the nerve by eliciting a pressure effect.

The clinical findings of TN do not differentiate classical (basically idiopathic) trigeminal neuralgia from trigeminal neuralgia induced by other conditions like neoplastic disease / odontogenic cyst or tumors or foreign body induced. Diagnostic imaging studies like plain radiographs, OPG, CBCT in patients with trigeminal neuralgia and dental disease should be a part of evaluation of patients with symptoms of trigeminal neuralgia mimicking dental pain. Our case represents a dental disease–odontogenic cyst as a cause of trigeminal neuralgia with peripheral nerve compression.\textsuperscript{[12,13]}

This patient’s experience underscores the importance of a multidisciplinary approach to the diagnosis and management of orofacial pain. Because several forms of dentoalveolar disease, including musculo ligamentous conditions can cause dental pain, these diseases must first be ruled out with a thorough history and clinical examination. Then investigations for other conditions

| Table 1: CBCT Report |
|---------------------|
| **Content of the CBCT report** | **Description of the lesion** |
| Location of the lesion | Well-defined unilocular radiolucency seen in the right body of the mandible around an impacted supernumerary tooth, extending from 45 region distally up to 48 region Superoinferiorly: Crest to the lower border of the mandible Buccolingually: From buccal to lingual cortical plate |
| Size of the lesion | Roughly oval in shape, measures about 27 mm mesiodistally, 12 mm bucckolingually, 16 mm superoinferiorly |
| Margin of the lesion | The lesion is bordered by thin sclerotic margin, throughout; upper margin gets attached to the CEJ portion of the impacted tooth |
| Effects on adjacent structures | Thinning of the buccal and lingual cortical plates, displacement of the impacted teeth superiorly close to the crest, displaced the mandibular canal inferior close to the lower border of the mandible and also has perforated the upper wall of the canal in middle 3rd of the lesion |
| Radiologic impression | Large benign osteolytic lesion in the right mandible |
| D/D | Dentigerous cyst Unicystic ameloblastoma OKC |

OKC: Odontogenic keratocyst, CEJ: Cementoenamel junction, D/D: Differential diagnosis
known to cause orofacial pain, such as brain imaging etc., should be done. Finally, precise and open communication is necessary not only with the patient and health care givers, but also with the health care givers themselves. The importance of this requirement simply cannot be overstated.\textsuperscript{[9,12,13]}

CONCLUSION

The pathophysiology of TN is now better understood. Neurovascular compression is recognized as the main cause of “idiopathic” TN. Other uncommon causes are recognized as well.

There have been some dental causes also that are responsible for triggering of paroxysms of pain such as odontogenic cysts and tumors, foreign bodies such as broken RCT files and residual infections of odontogenic origin; all these can also trigger TN.

Medical treatment remains the first step and we can rely on very efficient drugs. However, these drugs are not devoid of side effects and often lose their efficacy with time. In situations where poor response to drugs and with dental disease, prompt and thorough evaluation for dental causes should be done, as surgical treatment of dental disease cures TN as seen in our case.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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