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Case Report

Twitchy Pain of Left Face: A Rare Case of Idiopathic Trigeminal Neuralgia after Tooth Extraction in a Young Child

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ABSTRACT

Introduction: *Idiopathic Trigeminal neuralgia in children is a rare case presentation that poses a challenge in dental practice. This case involved a sequence of multi-management by several departments including Paediatric Dentistry, Oral Maxillofacial Surgery (OMFS), Otorhinolaryngology (ORL) and Paediatric Neurology. Case description: A 10-year-old girl was referred initially to the Paediatric Dentistry Specialist Clinic, School of Dental Sciences, Universiti Sains Malaysia (USM), Malaysia with the complaint of left facial pain associated with muscle twitching, weakness and swelling for five days prior to this first visit. There was a history of tooth extraction of her lower left second primary molar on the day before the episode of left facial pain and twitching. Clinical examination showed left facial involuntary twitching with visual analogue scale (VAS) pain score of 8, some evidence of left facial weakness, numbness but no facial swelling. No significant finding on the intraoral examination, including good healing signs at the exodontia site. The investigations*

included orthopantomography, Cranial Nerves II-XII examination and tests, blood investigations, Blood Urea and Serum Electrolyte (BUSE), electromyography (EMG) and electroencephalogram (EEG). The final diagnosis was left Idiopathic Trigeminal Neuralgia that is associated with tooth extraction. After a period of a month, the symptoms and signs were gradually reduced and ceased by taking oral medications such as Baclofen, Neurobion and Gabapentin. Joint management from multi-departments with close follow ups help to provide the conservative and safe treatment for this patient.

Keywords: *child, trigeminal neuralgia, extraction, muscle twitching, spasm*

INTRODUCTION

A chronic orofacial pain involving one or more divisions of the fifth cranial nerve is known as trigeminal neuralgia (TN). TN involves one or more branches of the trigeminal nerve. It is a prevalent cause of face pain in adults, primarily affecting those in their fourth and fifth decades of life, however it is uncommon in children (Ivanes & Moreno, 1999). The prevalence of TN among children was reported as 0.5% out of 1040 at the referred-based paediatric headache clinic within a 5 years period (Brameli et al., 2020). According to International Classification of Orofacial Pain 1st edition, classical TN is characterized by "recurrent unilateral brief electric shock-like pains, abrupt in onset and termination, limited to the distribution of one or more divisions of the trigeminal nerve and triggered by innocuous stimuli"(ICOP, 2020). The causes might be a result of another disorder such as tumour, vascular malformation, and multiple sclerosis; while most of the cases are idiopathic (Childs et al., 2000).

Idiopathic Trigeminal Neuralgia (ITN) is a recurrent paroxysms of unilateral facial pain fulfilling criteria of classical TN with neither electrophysiological tests nor MRI showing significant diagnostic findings (ICHD-3, 2018). Pharmacological first-line treatment for TN is with carbamazepine and oxcarbazepine. Other treatment choices include lamotrigine, phenytoin, clonazepam, gabapentin, pregabalin, topiramate, levetiracetam, tocainide, and surgery (Obermann, 2010).

Majority data regarding TN and pain, including headache are from the adult population. Data from children and adolescents is generally sporadic in the form of case reports. The recent study survey among children in a headache clinic reported all the cases involved were ITN with continuous concomitant pain with different history and presentation (Brameli et al., 2020). Here, we present a case of Idiopathic TN with muscle spasm on the left face, which occurred after lower left posterior tooth extraction in a child patient.

CASE DESCRIPTION

A 10-year-old girl was referred from Klinik Rawatan Keluarga (Pergigian) to Paediatric Dentistry Specialist Clinic, USM with a chief complaint of left facial muscle pain associated with continuous twitching, swelling and numbness for 5 days prior the first visit. She also complained of saliva drooling on the left corner of the mouth. The pain on the left face had sudden onset, intermittent, shock-wave in nature, aggravated by skin touching and associated with prickling and giddiness sensation, particularly on the left peri-orbital region. It lasted for about 2 minutes for each episode of pain. Patient claimed the pain score was 8 based on visual analog scale (VAS) as in Figure 1(A). The middle third of left facial swelling was reduced after taking the oral amoxicillin and metronidazole prescribed by the previous dental practitioner.

Further history showed history of tooth extraction, that was lower left primary molar tooth (#75) by the general dental practitioner (GDP) on 2nd December 2021. According to the patient and her father, the tooth extraction treatment was uneventful, and she was prescribed paracetamol. The left facial pain, muscle twitching and swelling started on the day after tooth extraction (3rd December 2021). She went to the same GDP, who was then prescribed oral amoxicillin and metronidazole. Even after completing the antibiotic, the left facial pain and twitching still did not resolve, even though no increment in severity. Otherwise, the left facial swelling

reduced after completing the course of oral antibiotics. She denied any episode of the same complaint before, and no history of prolonged headache, dizziness or blurring vision.

Her past medical history revealed previous diagnosis of left aural polyp with otitis externa secondary to trauma under Otorhinolaryngology (ORL) unit, Hospital USM. Currently no follow up needed for the latter medical problem. She was also seen by the Psychiatric team for management of mild anxiety, but not on medication. Her last visit at Psychiatric clinic was in May 2021 with no further follow up recorded.

On general examination, she was well and alert to time, place and person. No other physical findings on other parts of the body except for the patient's face.

EXAMINATION	FINDING(S)
Extra-oral examination	Pain and tenderness at dermatome area of maxillary (V2) and mandibular (V3) innervations Left facial muscle twitching Able to raised left eyebrow (weak)
Intra-oral examination	Oral hygiene was fair Extraction site #75: - Surrounding gingiva healed good, no tenderness - No pus discharge, no tenderness upon palpation
Cranial Nerves II-XII examination and tests	CN II (No abnormalities detected) CN VII (affected) CN VIII (affected) CN IX-XII (No abnormalities detected)

INVESTIGATION	FINDING(S)
Orthopantomography (Figure 2)	*No abnormalities detected*
Blood investigations: Full blood count and blood urea serum electrolyte (BUSE)	*Within normal range*

Based on the initial impression by Paediatric Dentistry and Oral Maxillofacial Surgery (OMFS) specialists, the patient had been diagnosed with Bell's palsy. Another differential diagnosis was Left TN. She was prescribed Tablet Baclofen 5mg 3 times a day and 1 Tablet Neurobion 3 times a day; both taken for 5 days. At the same time, the Otorhinolaryngology (ORL) also saw this patient as to rule out any possible relation to her previous diagnosis that might involve the ear part. The ORL team had further diagnosed her as having Left Hemi Facial Spasm.

The patient was reviewed 5 days later, with slight improvement in terms of reduced left muscle spasm. However, the left facial pain still lingered, but no increment in severity. She was then referred to the Paediatric Neurology Department in Hospital USM for further neurological assessment. Further investigations such as electromyography (EMG) and electroencephalography (EEG) were done on the face region, and the results

were normal. After thorough discussion between Paediatric Dentistry and Paediatric Neurology teams, the final diagnosis was Left ITN. Her medication was changed to Tablet Gabapentin 80mg (7 mg/kg) once daily.

Upon follow up of a month review at Paediatric Dentistry clinic, there was no facial twitching as in Figure 1(B). However, patient complaints of slight tenderness on palpation at the left inner cheek region, with VAS pain score 2 of out of 10. Few days prior to the review, the patient was seen by the Paediatric Neurology department, and she was instructed to cease Gabapentin consumption. Another 2 months review was given, but the patient failed to attend.

In this case report, we share the management of ITN in a child with additional and rare symptoms of left hemifacial muscle spasm, using pharmacological intervention as the first line treatment.

Figure 1: Preoperative (A) and postoperative one month follow-up (B)

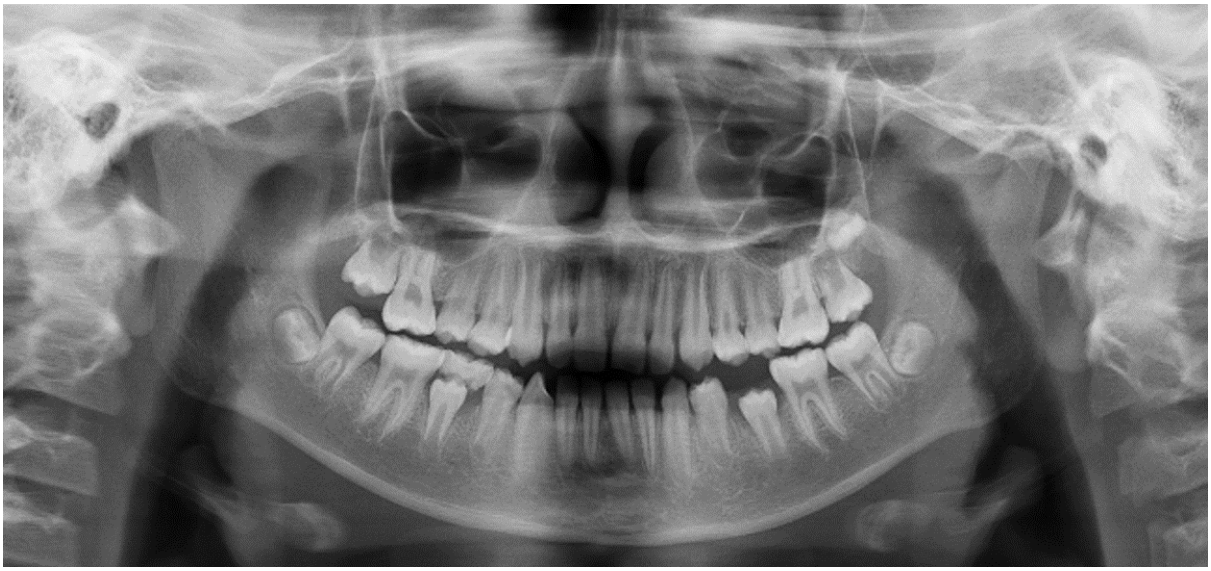

Figure 2: Orthopantomography taken 12 December 2021

DISCUSSION

ITN does not commonly happen in children. The first case of TN involving a child patient was reported in a 10-year-old boy in 1921 (Barclay, 1921). Over time, the diagnosis of TN proved to be a challenge because some clinical features might represent differently, apart from the child being less capable describing important symptoms. Thus, it is vital to focus on clinical features, etiology and treatment of craniofacial pain in children in order to avoid misdiagnosis and start up an appropriate treatment without delay (Raieli, 2019). In this case, the associated symptom of muscle twitching or so called 'spasm' that happens concurrently with the facial pain is somewhat alarming to get to the correct diagnosis.

Hemifacial spasm is a chronic condition characterised by involuntary contractions of muscles innervated by facial nerve, on one side of the face. It usually appears in the fourth to seventh decade of life, and rarely occurs in children. A case report of 61-year-old revealed that the patient had suffered the hemifacial spasm for almost 20 years, and the facial pain for 5 years due to a dolichoectatic left vertebral artery and displaced both the facial and trigeminal nerves (Crevier-Sorbo et al., 2019). The combination of hemifacial spasm and TN is also known as painful tic convulsif syndrome. The latter case was managed by surgical microvascular decompression due to long standing symptoms. The child patient in this case presented with left hemifacial muscle spasm that lasted for almost 2 weeks, which gradually decreased and responded to the treatment.

Other aetiologies of TN to be considered are the dental and other facial pain issues including dental caries, pulpitis, dental sensitivity, pericoronitis, cracked tooth, alveolar osteitis, salivary stone, temporomandibular disorders, glossopharyngeal neuralgia, post-herpetic neuralgia, burning mouth syndrome and many more (Lambru et al., 2021). A case report shows an 11-year-old TN child with severe Epstein-Barr virus infection who developed mild meningoencephalitis that linked to the pathogenesis of TN (Solth et al., 2008).

In this present case, the justification of not prescribing magnetic resonance imaging (MRI) is that apart from no significant findings through history and examination, the electromyography (EMG) and electroencephalography (EEG) results were within normal findings. This aligns with the recommendation by ICOP (2020). There was also no sign of neurological pathology as to rule out any brain disorder. It is interesting

that this present case depicted several signs and symptoms that interchangeably lead to few diagnoses such as ITN, Hemifacial spasm and Bell's palsy.

Approximately 22 out of 117 adult TN patients treated with Gamma Knife radiosurgery reported that the orofacial pain had actually either started or aggravated after their dental procedure (Tripathy et al., 2020). This again raises the doubt whether the primary diagnosis was TN or otherwise. There is a chance of some iatrogenic injury to the inferior alveolar nerve/lingual nerve, leading to sensory neuropathy which includes TN. The neuropathic pain on the orofacial region has a very complex pathophysiology and can be initiated by dental treatments such as third molar or implant surgery, endodontic treatments and local anaesthesia injections (Tinastepe & Oral, 2013). Similarly in this present case, the local anaesthesia or traumatic injury to the relevant nerve plexus during tooth extraction may instigate the TN and muscle spasm.

Treatment Options

1. Conservative Approach

Treatment is critical whereas a lack of pain-free episodes can lead to outcome decrease in key functions. In patients with TN, pharmacological intervention is usually the first line of treatment. Carbamazepine is the first-line therapy for TN; however, other medicines such as baclofen, gabapentin, lidocaine, and misoprostol have shown benefit in refractory patients (Jones et al., 2019). Several case studies that involved children prescribed carbamazepine with different dosages, from 15 mg/kg/day to 800 mg/day, and some combined with gabapentin 1200 mg/day to treat TN (Raieli et al., 2001; Lopez et al., 2004).

A cochrane review concludes that it is uncertain regarding the clinical effectiveness of botulinum toxin decays over time, including with repeated treatment sessions for treatment of hemifacial spasm (Duarte et al., 2020). In this present case, since the hemifacial spasm is not a chronic condition and no investigation finding including via MRI, further invasive treatment like botulinum injection is not indicated.

2. Surgical intervention

Surgical intervention is an option whenever medication therapy alone does not succeed. One-third of patients do not receive adequate symptom relief with medication therapy alone (Brendtsen et al., 2012). Prolonged TN symptoms can eventually lead to disinhibition of pain pathways in the spinal trigeminal nucleus, creating chronic background pain and probable progression of the disease (Chicoine et al., 2019). Microvascular decompression (MVD), stereotactic radiosurgery (SRS) or rhizotomy with radiofrequency thermocoagulation, mechanical balloon compression, or glycerol injection are surgical approaches used to treat TN (Bahgat et al., 2011; Bender et al., 2011).

An earlier choice to perform surgery is even more crucial in the paediatric population, where a lack of suspicion of the condition might lead to delays in diagnosis and treatment. The MVD was deemed a success in a child whose symptoms were caused by nerve compression via the superior cerebellar artery and anterior inferior cerebellar artery (Solth et al., 2008). Majority or 71% patients were satisfied with their post-operative condition after Gamma Knife radiosurgery, regardless of the presence of preceding dental pain that some had required multiple tooth extractions even without any significant indications (Tinastepe & Oral, 2013). Thus it requires multiple team approaches to rule out either dental cause or others before finalizing the diagnosis.

Limitation for this case is that the patient failed to follow up in 3 months, 6 months and 9 months review. We believe that the symptoms remain resolved as the patient defaulted on the follow up appointment.

CONCLUSION

ITN is a rare case reported particularly in paediatric patients. Although there is still a lack of complete understanding of the pathophysiology of TN generally, and specifically ITN, neurovascular conflict is currently the most widely recognised explanation. Proper examination in order to reduce the chances of misdiagnosis and postoperative complications, which could have long-term consequences for young patients. Non-invasive

treatment such as using medication should be the first line of treatment. Joint management from multi-departments with close follow-ups help to provide the conservative and safe treatment for this patient. Long-term follow- up and a better understanding of the pathophysiology of ITN may help to provide a solution in the future and to prevent relapse.

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CONFLICTS OF INTEREST

The authors declare no conflict of interest.

ETHICAL APPROVAL

The patient's anonymity is carefully protected. We have obtained written consent of the patient and her guardian for the use of clinical information and photos for publication.

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