Atypical Facial Pain: An Enigma in Diagnosis and Treatment- A case report

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ABSTRACT

Atypical odontalgia (AO) is chronic orofacial pain condition characterized by the persistent orofacial pain that affects teeth or tooth socket after extraction in absence of any identifiable cause on clinical or radiographic examination. This causes a diagnostic dilemma for the clinician and is very frustrating to the patients. Patients demand treatments in a hope of pain relief and visit multiple clinics for the same. This leads to multiple unnecessary treatments without remission of the symptoms. These conditions are usually related to psychiatric, vascular or neurological problems. Hence, antidepressants like amitriptyline or serotonin-norepinephrine reuptake inhibitors (SNRIs) like duloxetine and antianxiety like ethyl loflazepate are reported to be effective in the treatment of AO. However, efficacy of such drugs varies among individuals and their symptoms. AO is a condition which carries sensory as well as complex psychological problems. Thus, while prescribing the drugs all the aspects including the variation in pharmaco-therapeutic responses should be considered. In this case report, we have reported a case of AO and have discussed its diagnostic workout, characteristic and the management aspect with combination of various medicines.

Key words: Atypical odontalgia, Antidepressants, Amitriptyline, Aripiprazole, Atypical facial pain, Sensitization.

INTRODUCTION

Orofacial pain can be a debilitating condition affecting the patient's quality of life. It is often misdiagnosed, leading the patients to suffer with prolonged, unnecessary and costly treatment. Up to 26% of population has suffered from the oro-facial pain, at some point of time, in their lives[1]. Atypical odontalgia (AO), also known as atypical facial pain, phantom tooth pain, persistent idiopathic pain or neuropathic orofacial pain, is a chronic orofacial pain condition, characterized by continuous pain affecting the teeth or teeth sockets after extraction in absence of any identifiable cause on clinical or radiographic examination[2]. AO although is a specific problem in dentistry but it is surprisingly complex as pain persists in tooth or teeth, or in a site where teeth have been extracted or following endodontic treatment, without any recognizable cause. Over time, the pain spreads or radiate to other areas leading to

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multiple unnecessary treatments, which does not resolve patient's symptoms. These disorders have been found to be associated with vascular, neuropathic or psychiatric conditions with some comorbid factors as well. Neural sensitization has also been thought to be a likely cause of atypical facial pain in root treated teeth or tooth socket[3]. The conditions like burning mouth syndrome (BMS) and atypical odontalgia (AO) are two common chronic orofacial pain conditions that are considered as somatoform disorders or psychogenic conditions as both lack organic cause[4]. Study also showed psychological stress, can cause transcriptomatic alteration in salivary composition and flow in submandibular glands and can cause stress related disorders like AO or BMS[5]. Dental migrane or facial pain of vascular origin has also been considered as a cause of AO[6]. Thus medicines like antidepressants, such as amitriptyline, antianxiety, such as benzodiazepine or anticonvulsants, such as carbamazepine or gabapentin are reported to have been prescribed to the patients suffering from AO[2-3]. However, the efficacy of such drugs varies depending on the symptoms and the case. In this article we present a case report of a patient suffering from AO and discuss its diverse characteristics and management.

CASE REPORT

A 50-year-old female, a front desk officer in a private office who was living with her husband and son (medical doctor) was referred to our center with chief complaint of heavy, piercing pain in right maxillary posterior area, since last 2 years. Her medical history was not significant and she was not under any medication for any other illness except, carbamazepine 100 mg in the morning and 200 mg in the evening for persistent orofacial pain. She had no significant family history and no psychiatric or psychological family history was identified.

Two and half years ago, she started to feel dull aching pain and discomfort in right maxillary posterior area. She had a history of root canal treatment of the right maxillary first molar at a private dental clinic few months back due to similar kind of symptoms. Initially there was a remission of the symptoms for few weeks but pain reappeared with more severe characteristics. Patient felt piercing kind of pain that radiated towards the right side of the jaw and sometimes under the eye and temporal region. Several radiographs were taken, clinical examination was conducted and antibiotics were prescribed, although no abnormalities were detected. Hence, Re root canal was performed without remission of symptoms and eventually the tooth was extracted. However, there was still no remission of symptoms. Consequently, maxillary right second premolar and third molar were also extracted when symptoms got worse, on patient's demand, although the tooth had no detectable abnormalities. With this treatment also, there was no remission of pain but her symptoms got worse. Then she was prescribed carbamazepine by her dentist. Carbamazepine appeared to be effective in relieving her pain. However, due to dizziness at the dosages prescribed, her son, a medical doctor reduced the dose to 100 mg twice after few months. The symptoms reappeared with decrease dose of drugs and then she was referred to our center by some other doctors.

After receiving the patient and taking her detailed history, we did both extra and intra oral clinical examination and took orthopantomograph (OPG) (Figure 1, 2 and 3). While examining the patient, we followed the diagnostic work out given by Glen T Clark[7]. No significant organic cause was found during examination.

The psychological assessment was also done by psychiatry assessment form, converted from Japanese to English and answers were evaluated. Although no significant findings were uncovered during the assessment, we found her very anxious about the unexplained pain for so

long, medication that made her dizzy and about reduced performance at work, which overall affected the quality of her life significantly. Obvious signs of depression were absent. The score of the Zung self-rating depression scale of the patient was 52, although no depressive mood, emotional disturbances, lack of energy or enthusiasm or suicidal thought were noted[8]. Based on the clinical examination, radiographs, medical history, results of the diagnostic work out and the psychological assessments, a diagnosis of atypical facial main (AO) was made.

Thus, we decided to explore alternative medications such as antidepressants or anti-anxiety even though those medicines may or may not cause dizziness. Initially, we planned to prescribe her with serotonin-norepinephrine reuptake inhibitors (SNRIs) such as Duloxetine or amitriptyline, while ethyl loflazepate as a potential anti-anxiety option. Given the efficacy of carbamazepine, which suggests neuronal over-activity, aripiprazole, an antipsychotic drug, was also considered alongside antidepressants and anti-anxiety medications. We started with ethyl loflazepate 0.5mg/day, along with aripiprazole 2mg/day and amitriptyline 10mg /day (SNRIs was not available in Nepalese market at that time), with follow-up every 2 weeks. As the symptoms of the patient was more or less same after 2 weeks, the dose of ethyl loflazepate was increased to 1 mg/day and amitriptyline to 20 mg/day with side effects being observed to withdraw if necessary. Patient was responding well to the medication in a month, with dizziness gone, we continued with the same medication for another 3 months.

Hospital anxiety and depression scale (HADs) was also used to access her anxiety level a week before (score: 9) we started the treatment, and after 4 months (score: 4), when her symptoms subsided. We kept a pain diary via visual analog survey scale every month for 4 months, with gradual decrease in score. In 4 months', time, since her first visit, her symptoms were cured with pain score recorded to be 1. Although, patient was extremely happy to feel normal after almost 2 and ½ years, she continued to take the same medicine with tapering dose (0.5mg/day ethyl loflazepate and 10 mg/day of amitriptyline) for another 4 months and finally ceased after 8 and 1/2 months after her first visit. She experiences complete remission of her symptoms.

DISCUSSION

AO is identified as a subgroup of persistent idiopathic orofacial pain disorder, by international headache society[9]. Recently, persistent dento-alveolar pain disorder (PDAP) is a new terminology for AO that has been put forward with new diagnostic criteria to address the limitations of existing terminology. In order to arrive at accurate diagnosis, one has to exclude all possible etiologies and it can become an interdisciplinary approach. PDAP is found to exist on around 1.6% of the population after root canal treatment, with unknown risk factors and etiology but is similar to post-surgical chronic pain conditions[10]. AO is predominant in

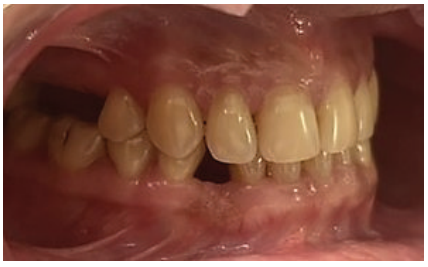

Figure 1

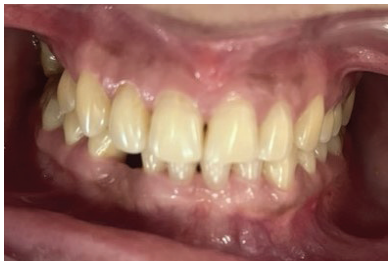

Figure 2

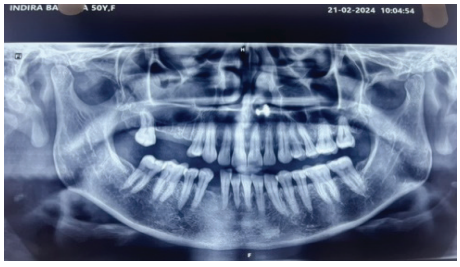

Figure 3

middle aged women[11]. Hence, the role of female hormones has been thought to be associated with these pain conditions, as there has been a therapeutic and physiological modification of estrogen levels in such patients[12]. In our case, the patient was in her early 50s and the onset of symptoms was although before the initiation of dental treatment, the symptoms got worse with each dental treatment. Literature suggests the possible occurrence of post traumatic peripheral pain neuropathies after dental treatment[13]. AO can occur in 3-6% of patients that underwent root canal treatment[14]. Hence, dental treatment can be a possible triggering or exaggerating factor for AO.

As for the characteristics of pain, our patient had a dull aching pain, which got severe lancinating type after root canal treatment that started to radiate to the temporal area. Even after extraction the severity and the nature of the pain remained more or less same. The location and the nature of the pain was similar to odontogenic pain which led to multiple dental treatments without remission rather exaggeration of the symptoms. These made such kind of conditions diagnostic dilemma. In this case following diagnostic work out was used to rule out the odontogenic cause[7]:

Phase I: If pain is present in vital tooth, then

Step 1: Perform following tests

a. Cold Test: To rule out non vitality
b. Periapical Radiographs to check apical pathology
c. A panoramic radiograph to rule out other maxillofacial diseases
d. A through head and neck examination to identify other causative factors
e. A cranial nerve examination which might be the cause of any sensory alterations

As there were no evidences of any of the disease, abnormalities or alterations, we proceeded to phase II.

Phase II: If pain lingers for more than 3 weeks in a vital tooth with no Periapical lesion

Step 2: Remove all the restoration and inspect under magnification for cracks

Not Applicable in our case as the tooth was already extracted.

Step 2.1: If there are no evidences of cracks then restore the tooth, keep the tooth slightly out of occlusion and make an orthotic appliance like occlusal splint to monitor and control the excessive tooth loading during sleep. However, if occlusal splint does not help and there are no signs of bruxism, premature contacts or sustained clenching during sleep and pain continues, stop the use of the appliance.

Step 2.2: If patients still complain of persistent pain with or without root canal treatment or in an extracted tooth site and clinicians see no evidence of cracks in a vital tooth, no periapical radiolucency, normal response to cold tests, no signs of bruxism, clenching or premature contacts, anesthetic tests protocols for topical, infiltration and get pain diary (one week) is suggested.

Topical anesthetics were applied and the site was infiltrated with local anesthetics. Pain diary was kept which was filled every week but there was no significant reduction in pain in a week.

Step 2.3: If topical anesthetic stops pain, then neurosensory stent with topical anesthetic is advised as long as required. However, if it does not reduce pain, then the medication protocol is adopted. Reexamine, repeat radiographs and pulp tests at intervals.

Medications protocols were adopted after a week.

As our examinations, radiographs and tests revealed no significant organic cause, we opted for medicines to treat her chronic pain condition.

Literature suggests AO as a psychogenic condition though the association between AO and psychogenic factors are not very clear[15-17]. In our case, we did her psychological assessment using psychiatry assessment form (Converted from Japanese to English) and Zung self-rating depression scale. As was patient being anxious when we received her, Hospital anxiety and depression scale (HADs) was used to asses her anxiety level before and after treatment. Pain diary was kept and pain was measured using Visual analog survey scale throughout the treatment. Although no significant score was seen in any of the assessment and scales patients' anxiety and pain level was recorded high before and in between the treatment.

While treating the AO, antidepressants like amitriptyline, has been found to be effective[17-18]. Amitriptyline works by activating serotonin and noradrenaline in the nervous system. This affects the descending pain inhibitory system of the neurotransmission pathway. Similarly, serotonin-norepinephrine reuptake inhibitors (SNRIs), block the reuptake of serotonin and norepinephrine in the brain and increases its availability and activity in the synaptic cleft, thereby improving the mood of the patient. However, all patients suffering from AO do not respond well to these medicines. Drugs like aripiprazole modulate dopamine and serotonin activity in brain. It acts a partial agonist at dopamine D2 and 5-HT1A receptors and an antagonist at 5HT2A receptors, rebalancing neurotransmitter levels to impact mood, behavior and thinking of a patient. Similarly, ethyl loflazepate binds to benzodiazepine site on GABA-A receptors and enhances GABA's inhibitory effect, in order to reduce neuronal excitability.

Hence, in this case we decided to use amitriptyline along with aripiprazole and ethyl loflazepate to treat chronic pain condition, anxiety and neuronal over activity due to intake of carbamazepine. This medication seemed to work well with the patient. From these findings, we could assume, serotonin, noradrenaline and dopaminergic system, all were involved in the pathophysiology of AO, similar to other case reports[2].

Pain is an unpleasant sensory and emotional experience due to actual or potential tissue damage[19]. Patients suffering from chronic pain conditions can undergo various biological and psychological changes, resulting in complex symptoms that are very difficult to diagnose and treat. Peripheral and central sensitization of trigeminal nerve pathways has also been reported, as a potential pathophysiology of AO[3,20]. Moreover emotional aspects, anxiety due to chronic pain conditions, multiple consultations, treatments and exacerbation of the symptoms despite all, can develop psychological components in patient. Hence, one should do a proper diagnostic workout to rule out all odontogenic cause of the pain and assess the psychological status of the patient before opting for its treatment.

CONCLUSION

AO shows various features and responses to drugs. It is considered not only a purely sensory problem, but also a considerably complex psychological problem, such as rumination about the pain. Investigating the difference in pharmaco-therapeutic responses might help to advance the treatment of AO. It is hard to diagnose AO precisely and we need an appropriate consensus about AO to prevent overtreatment. Further studies are needed to improve the diagnosis and treatment of AO.

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