ABSTRACT
Atypical odontalgia is a pain disorder featured by constant pain without any obvious dental pathology. We report a 38-year-old man having chronic orofacial pain in the past two years. The pain was first located in the left maxillary premolar-molar region, but spread to the contra lateral mandible, occipital region, head and shoulders over time. The pain was described as dull and continuous with the intensity of #6 (VAS), and not relieved even after root canal therapy of all teeth and extraction of wisdom teeth. Some comorbid conditions such as TMJ problems and involvement of masticatory muscles were also reported. Finally, the diagnosis of atypical odontalgia was established due to lack of evidence for any organic pathology on clinical and paraclinical investigations, and a combination therapy including fluoxetine and clonazepam was started. Dental practitioners should consider AO when confronting toothache without any reasonable organic cause and avoid unnecessary dental procedures.

KEYWORDS
Atypical toothache; Malpractice; Endodontics.

RESUMO
A odontalgia atípica (OA) é um distúrbio doloroso caracterizado por uma dor constante sem qualquer patologia dentária óbvia. Relatamos um homem de 38 anos com dor orofacial crônica nos últimos dois anos. A dor foi inicialmente localizada na região pré-molar-molar superior esquerdo, mas se espalhou para o lado oposto da mandíbula, região occipital, cabeça e ombros ao longo do tempo. A dor foi descrita como maçante e contínua com a intensidade de # 6 (EVA), e não aliviada mesmo após tratamento de canal de todos os dentes e extração dos dentes do siso. Algumas comorbidades como problemas de ATM e envolvimento dos músculos mastigatórios também foram relatadas. Finalmente, o diagnóstico de odontalgia atípica foi estabelecido devido à falta de evidência para qualquer patologia sistêmica nas investigações clínicas e exames complementares, e uma terapia combinada incluindo fluoxetina e clonazepam foi iniciada. Os dentistas devem considerar OA ao enfrentar uma dor de dente sem qualquer causa sistêmica razoável e evitar procedimentos odontológicos desnecessários.

PALAVRAS-CHAVE
Odontalgia atípica; Imperícia; Endodontia.
INTRODUCTION

A typical odontalgia (AO), also known as phantom tooth pain, non-odontogenic tooth pain, persistent orodental pain, persistent dento-alveolar pain disorder, and idiopathic periodontalgia, is a disorder characterized by continuous pain without any pathological changes and/or stronger pain than would be expected from the clinical features [1-3]. This entity was first described by Mc Elin and Horton in 1942 [4]. AO, according to the International Association for the Study of Pain, is defined as a “severe throbbing pain in the tooth without major pathology” and “persistent (chronic) continuous pain symptom located in the dento-alveolar region, which cannot be explained within the context of other disorders” [1]. In addition, on the basis of International Headache Classification, it is classified as a persistent facial/oral pain, recurring daily for more than 2 hours per day over more than 3 months in the absence of a clinical neurological deficit [5]. This condition can be occurred in all ages with a sex predilection in women; 80% -90% of all cases are females [2,3]. The main pathophysiology of AO is unclear. However, possible mechanisms include those of neuropathic, vascular, or psychogenic origin [2]. Miura et al., showed a dental trigger of AO in 56.7% of dental patients [6]. It is also estimated that about 96.000 of new cases of persistent pain occur following relatively routine dental procedures per year, with 61.000 of these cases attributed to AO [7,8]. On the other hand, Polycarpoue et al., reported that the prevalence of AO after a successful root canal therapy is 12% [9]. Unfortunately, in many cases, the chronic and persistent nature of pain prompt dental practitioner to treat teeth in the absence of any pathological findings. The treatments often exacerbate the pain instead of relieving it and dentists usually make a diagnosis of AO after the failure of various invasive therapies [3]. The aim of this paper was to report invasive dental malpractice in a patient with AO.

CASE REPORT

A 38-year-old male patient presented with the history of a maxillofacial chronic pain in the past 2 years. The pain were first located in the left maxillary premolar-molar region referred from the occipital and neck regions, and contralateral side as well. The pain was described as dull and continuous(constant), and not relieved even after root canal therapy of teeth #12, #13, #14. Soon after, the pain was perceived in the right mandibular premolar-molar area prompting the dentist to perform endodontic treatment on #30, which was previously restored with a deep amalgam restoration. This procedure neither discontinued the pain nor stopped its spreading to both upper and lower jaws. In addition, the patient’s sleep and feeding patterns was disturbed due to diffuse pain and anxiety. Following deterioration of the patient’s condition wisdom teeth were extracted, all of minimally decayed teeth were restored, and then endodontically treated one after another. In spite of having 28 restorations and 4 teeth extractions the pain was persistent till the patient was referred to oral and maxillofacial medicine specialist. The patient had a recent history of a dull aching pain continuing all day long with the intensity of #6 according to visual analogue scale (VAS), which impaired the quality of life severely so that the patient could sleep no more than 4 hours a day. Moreover, the patient was suffering from cancer phobia, because the cause of pain was not elucidated. Previous medical records showed nothing remarkable in neurologic and ENT examinations as well as computed tomography (CT), magnetic resonance imaging (MRI), audiometry, and laboratory investigations. Extra oral examination revealed temporomandibular disorder with severe involvement of masticatory muscles especially in masseter and lateral pterygoid bilaterally. Although intra oral examination showed no abnormalities two serial panoramic views (Figure 1a,b) demonstrated numerous unnecessary tooth fillings and endodontic therapies. Regarding social impact of pain, the patient experienced such inability that...
was about to lose his job and also unable to accomplish daily routines. On the other hand, occupational stress aggravated the severity of pain. Noticeably, there were some comorbid conditions such as GI disorders and feet pain, which were attributed to orofacial pain by the patient with pointing to pain referral route from jaws to head, neck, and shoulders as well as the above-mentioned regions. According to all information and lack of evidence for any organic pathological process the diagnosis of atypical odontalgia (AO) was established, and the patient underwent pharmacologic treatment according to a combination therapy with fluoxetine and clonazepam suggested by Mortazavi et al. [2].

Psychiatric disease [11]. It is also noted that AO patients may have history of depression or depressive symptoms in 66% and 41% of cases, respectively [3]. In an extensive retrospective study, the most common psychiatric problem of AO patients was depression followed by anxiety, somatic symptom disorders, insomnia disorders, obsessive-compulsive disorders, eating disorders, and personality disorders [6]. Continuous pain can affect various body and brain processes. It has been recently demonstrated that there is involvement of peripheral and central sensitization of trigeminal pathways in AO as well as a relationship between chronic pain and central sensitization [1]. In addition, emotional aspects such as rumination about the pain might be related to AO [1,3]. Many of these findings were in accordance to our patient's history such as anxiety, sleep disorder, job stress, and cancer phobia.

According to Miura et al., the age range of AO patients was between 18 and 86 years, with the mean age of onset being 53.62 years. The median duration of AO was 24 months, and women affected more frequently. A dental trigger was found in more than half of patients, 14.4% developed AO after root canal therapy, 12.3% after dental extraction, and 12% after prosthesis treatments [6]. There are a similarity between our case and Miura's in terms of age, disease duration, and type of dental procedures. Polycarpou et al., described that the risk factors leading to AO after endodontic therapy are included as preoperative pain from tooth, preoperative tenderness to percussion of the tooth, previous painful dental treatment, female gender, previous chronic pain disorders, and whether surgical treatment was received [2,3].

The pain is usually dull and can be presented localized, generalized, unilaterally, or bilaterally.

**DISCUSSION**

Although the relationship between AO and psychological problems is still unclear, because of the absence of any organic cause AO is often considered as a psychogenic condition [1,10]. Takenoshida et al., reported that 60% of AO patients had been diagnosed with a psychiatric disease [11].
As antidepressants are reported to be effective in the treatment of AO [1,3,2], the patient underwent medical treatment according to a combination therapy with fluoxetine and clonazepam suggested by Mortazavi et al., [2] Fluoxetine is a selective serotonin reuptake inhibitor (SSRI) antidepressant agent and clonazepam is a benzodiazepine derivative with anticonvulsant, muscle relaxant, analgesic, and anxiolytic properties [2]. Some other medications used for AO include gabapentin, tricyclic anti-depressants (TCAs), topical anesthetics, and opioids [13,14].

CONCLUSION

Atypical odontalgia should be considered as a non-odontogenic cause of toothache by dental practitioners when clinical and radiographical findings are normal and be a referred pain from a musculature. This case report shows the importance of establishing an accurate diagnosis in order not to generate inappropriate dental treatments.

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REFERENCES