



ISSN Print: 2394-7489
ISSN Online: 2394-7497
IJADS 2021; 7(1): 183-186
© 2021 IJADS

www.oraljournal.com

Received: 17-11-2020

Accepted: 25-12-2020

Joel Eduardo Sanchez Nuñez

Posgrado de Odontología Avanzada,
Master in Science Student,
Universidad Autónoma de Nuevo
Leon, Facultad de Odontología
Monterrey, Nuevo Leon, Mexico

Myrna Delia Salinas Quiroga

Cuerpo Academico UANL-CA-395,
Universidad Autónoma de Nuevo
Leon, Facultad de Odontología
Monterrey, Nuevo Leon, Mexico

Valentin Zaragoza Magaña

Cuerpo Academico UANL-CA-395,
Universidad Autónoma de Nuevo
Leon, Facultad de Odontología
Monterrey, Nuevo Leon, Mexico

Rosa Isela Sanchez Najera

Departamento de Histología. Doctor
en Investigación Universidad
Autónoma de Nuevo Leon, Facultad
de Odontología Monterrey, Nuevo
Leon, Mexico

Lizeth Edith Rodriguez Quintanilla

Cuerpo Academico UANL-CA-363,
Universidad Autónoma de Nuevo
Leon, Facultad de Odontología
Monterrey, Nuevo Leon, Mexico

Sara Saenz Rangel

Cuerpo Academico UANL-CA-363,
Universidad Autónoma de Nuevo
Leon, Facultad de Odontología
Monterrey, Nuevo Leon, Mexico

Maria Fernanda De Leon Gomez

Departamento de Fisiología.
Dentistry student, Universidad
Autónoma de Nuevo Leon, Facultad
de Odontología Monterrey, Nuevo
Leon, Mexico

Juan Manuel Solis Soto

Departamento de Fisiología. Doctor
en Ciencias, Universidad Autónoma
de Nuevo Leon, Facultad de
Odontología Monterrey, Nuevo Leon,
Mexico

Corresponding Author:

Joel Eduardo Sanchez Nuñez

Posgrado de Odontología Avanzada,
Master in Science Student,
Universidad Autónoma de Nuevo
Leon, Facultad de Odontología
Monterrey, Nuevo Leon, Mexico

Atypical odontalgia: An update

Joel Eduardo Sanchez Nuñez, Myrna Delia Salinas Quiroga, Valentin Zaragoza Magaña, Rosa Isela Sanchez Najera, Lizeth Edith Rodriguez Quintanilla, Sara Saenz Rangel, Maria Fernanda De Leon Gomez and Juan Manuel Solis Soto

DOI: <https://doi.org/10.22271/oral.2021.v7.i1c.1134>

Abstract

Introduction: Atypical odontalgia (AO) is one of the most complicated challenges that a dentist can face for an adequate diagnosis and treatment plan. It can cause prolonged visits and costly overtreatments for the patient, increasing the time of resolution of the condition.

Objective: To analyze the literature about AO, as well as its epidemiology, pathophysiology, predisposing factors, associated pathologies and treatment.

Methodology: Information was searched in PubMed, SCOPUS and Google Scholar. Key words were used to search for information such as: atypical odontology, epidemiology, pathophysiology, related disease, treatment, among other relevant articles.

Results: AO occurs in women (85%) and men (20%) between 50 - 60 years old, hormonal or psychiatric disorders, related to local neuropathies due to problems in electrical conduction, and present or largely confused with trigeminal neuralgia and burning mouth syndrome mainly, sometimes it is possible to identify duct treatment or a history of aggressive therapy in the area, and can be treated with Amitriptyline, Imipramine, Aripiprazole, and locally with Botulinum Toxin A with favorable results found at 4 weeks.

Conclusion: AO does not have specific characteristic criteria for its diagnosis, leaving a wide spectrum of possibilities that represent a complex pathology that needs further study.

Keywords: Atypical odontalgia, epidemiology, related disease, treatment

1. Introduction

Atypical odontalgia is one of the most frustrating conditions in dentistry, which not only causes frequent and possible overtreatment visits, but also problems and comorbidities beyond those found in the clinic, which put the dentist at a disadvantage [1].

First reported in 1947 [2], atypical dentistry is described as a constant throbbing sensation or pain in a tooth, teeth or extraction site that is persistent and unrelenting, and not significantly affected by exposure to hot or cold food or drink, either by chewing or biting. The pain may or may not be relieved by injection of local anesthetic. The intensity of the pain can range from very mild to very severe and negatively impacts life [3]. There is usually no identifiable cause to explain the pain and it often follows or is associated with a history of some type of dental procedure, such as having root canal treatment or a tooth extraction. Occasionally, pain can occur for no reason [4]. The pain is felt in a tooth or teeth and persists despite treatment to relieve the pain. This often presents a frustrating and confusing situation for both patient and dentist, and can lead to more and more dental treatment, neither of which is effective in relieving pain [5]. In 1993, a new term was introduced for this condition called phantom tooth pain [6]. AO has been referred to by different names such as phantom tooth, idiopathic tooth pain, chronic continuous dental-alveolar pain, and persistent dental-alveolar pain disorder [7, 8, 9, 10]. Since 2004, the International Headache Society first recognized the term Idiopathic Persistent Orofacial Pain, replacing that of AO and maintained it to date [11]. Conditions representing chronic orofacial pain such as persistent idiopathic facial pain, atypical dentistry, and burning mouth syndrome are usually grouped together under the concept of orofacial pain, and which represent a diagnostic and therapeutic challenge due to their different hypotheses as responsible for the pain, as well as the pathophysiological mechanisms [4].

AO is a characteristic part of the endodontal clinic, can be nonspecific, radiated or referred and even be associated with endodontically treated dental organs, when there is no radiographic identification of the causal motive, the patient's psychosocial history should be considered as stress due to altered circulation of catecholamines and sympathetic modification of pain maintained, unrealistic expectations of eliminating pain, denying or accepting advice from the dentist; environmental factors such as marital problems, economic difficulties, the identification of these factors helps the clinician to refrain from performing irreversible treatments such as the extraction of a tooth^[12].

Currently in the literature there is no adequate and concise review on atypical odontalgia therefore its epidemiology, pathophysiology, associated factors and treatment were particularly analyzed.

2. Materials and methods

Articles on the subject published through the PubMed, SCOPUS and Google Scholar databases were analyzed, with emphasis on the last 5 years. The quality of the articles was evaluated using PRISMA guidelines, i.e., identification, review, choice and inclusion. The quality of the reviews was assessed using the measurement tool for evaluating systematic reviews (AMSTAR-2). The search was performed using Boolean logical operators AND, OR and NOT. It was realized with the words atypical odontalgia, epidemiology, pathophysiology, related disease, treatment. The keywords were used individually, as well as each of them related to each other.

3. Results & Discussion

3.1 Epidemiology

Epidemiology in one study estimated the incidence of persistent idiopathic facial pain at 4.4 per 100,000 people per year^[13]. Prevalence rates are estimated at 0.03%^[10]. Studies confirm a high proportion of female patients with AO of up to 85% in previous studies^[14]. Although other studies mention different age ranges, it may be consistent and similar, with the average age of 56.9 years, and 55.4 years respectively^[15, 16]. This may be sufficient to explain that there is a significant statistical difference in gender and average age distribution in the population of AO patients as well, possibly because women are more likely to have recurrent variable pain and to express it more intensely than men due to hormonal mechanisms that are not well understood^[17]. On finding it some report approximate estimates of 1.03%, indicating that it is a rare disorder, approximately 10-21% of the population in pain clinics, 21-27% with idiopathic facial pain and 3% unilateral pain in third level neurological clinics^[18]. Results comparable to other studies where 2.1% of patients presented facial pain without indicating the region of the face and could be related to myofascial origin. This is due to the side effects of the treatment and its evolution that resolve spontaneously^[19].

AO occurs in women (85%) and men (20%) between 50 and 60 years of age or in ages close to hormonal decompensation related to menopause. It is also more common in those who suffer from psychiatric diseases and with a history of some type of treatment or exhaustive manipulation of the hard and soft tissues of the face.

3.2 Pathophysiology

Currently the pathophysiology is controversial, but the hypotheses are more related to neuropathic, psychogenic,

vascular or even idiopathic causes^[20, 21]. It is mentioned that, in the absence of causes related to AO, most researchers associate it with one or two prevailing theories, one emphasizing that AO is primarily a manifestation of a psychiatric disorder, such as depression, hysteria or post-traumatic stress; the other theory relates it to a form of neuropathic pain such as phantom tooth or sympathetically maintained pain^[22, 3]. Another possible hypothesis could be based on Melzack's theory of neuromatrix, a neural connection in which the composition and connections are genetically determined and then influenced by multiple signals coming from different parts of the body^[23]. A hypothesis was also made about the hyperreactivity of the trigeminal nucleus in response to the electrical activity of the peripheral trigeminal nerve, which resulted in symptoms of trigeminal neuropathy^[24]. It has been mentioned a multineural reflex of the trigeminal system in the brain stem which produces the symptoms^[25]. Some other studies have established that there is no pathophysiological evidence among the role of trigeminal dorsal root neurovascular compression in persistent idiopathic facial pain^[26, 27, 28]. Many of these patients have a history of mild trauma and sensory changes that have initiated the suggestion that idiopathic persistent facial pain may represent the extremes of a spectrum of clinical presentations^[29]. The pathophysiology of persistent idiopathic facial pain is still enigmatic, however, neuropathic mechanisms may be relevant, interdisciplinary collaboration is needed to rule out or manage secondary causes, psychiatric co-morbidities assessments are recommended in the early stages of the disease, and should be addressed in the treatment plan, however research is still needed to establish clear diagnostic criteria and treatment strategies based on clinical findings and individual physiology and event management^[18]. Misdiagnosis of pain can lead to frustrating results and unnecessary treatment, with root canal therapy sometimes performed on all teeth^[30]. Pain management in patients with AO is not encouraging; medications and surgical therapies have been used with inconclusive results. For the time being, AO is considered to be neuropathic in origin and new techniques will be attempted in conjunction with those that demonstrate similar efficacy in neuropathies^[31].

Most of the AOs are neuropathic in nature and therefore we can conclude that, although the concept of pain is controversial, it evidently affects the nerve endings and can even be evident when the origin is specific to the location or relationship to its sensory or motor root. Further studies are needed to understand the different methods.

3.3 Predisposing factors and associated pathologies

Odontogenic pain such as pulpitis, periapical periodontitis and fissure tooth syndrome should be ruled out after a thorough examination and necessary complementary tests such as x-rays, anesthetic blocks, ruling out sinus pain, myofascial pain and symptoms related to pain from temporomandibular joint dysfunction, as well as trigeminal neuralgia^[32]. Trigeminal neuralgia has been frequently documented as a major cause of atypical dentistry, however, pontine stroke as a cause of AO is limited to few cases, therefore, prevalence is not established, so neuroimaging; brain MRI could show areas of infarction in the right bridge near the entrance area of the trigeminal root^[33]. Others criticized the hypothesis and questioned the relevance of the psychological component in pain by evaluating and comparing 19 patients with AO using the Minnesota Multiphase Personality Inventory with 19 patients

with headache, they found no difference in the scale suggesting that the psychological role does not play a significant factor^[34]. Authors studied the association of AO and burning mouth syndrome (BMS), however, they found different epidemiological characteristics, sleep quality, and pain experiences among those with AO and burning mouth syndrome than did AO alone, suggesting BMS as a comorbidity disorder in AO patients that contributes to a more painful experience, as the presence of psychiatric comorbidities in both groups aggravated sleep quality, with little impact on the pain experience^[35]. Sometimes evidence of past aggressive treatment, local disease, or an uncertain history can be dismissed because of its low magnitude of sensory relevance, and that is when the AO becomes completely nonspecific and demonstrates a psychiatric component (60%), and where amitriptyline and aripiprazole gave favorable results, although not in all patients with psychiatric disorders but with burning mouth syndrome^[36]. Similar conclusions are reported, where the total of their 383 patients with AO, 177(46.2%) had psychiatric comorbidities, the most common being depression (15%) anxiety (10.1%), bipolar disorder (3%), schizophrenia (1.8%), and reporting that half of their participants had no dental correlation with OA making the hypothesis of a dental-cause relationship with AO controversial if not refuted^[37].

AO is related in order of frequency to trigeminal neuralgia, burning mouth syndrome, endodontic treatment, trauma or aggressive tissue manipulation. It can be considered not only a sensory problem, but more complex, of psychological nature, such as mental rumination, among them the most common being depression and anxiety. Researching how to treat the different related diseases and their pharmacological responses could help advance the treatment of AO.

3.4 Treatment

Of the different treatment modalities, we find those directed to control in the component of psychiatric disorders and those that need some type of accepted or experimental physical intervention. Although AO is a little-known condition, the understanding that it is similar to conditions that cause neuropathic pain is accepted, so it is necessary to obtain a diagnosis before intervening surgically or unnecessarily and should be treated similarly to other painful facial neuropathies^[38]. Most reports recommend the use of tricyclic antidepressants, amitriptyline as the main agent for managing AOs, imipramine, or aripiprazole, a partial dopamine agonist, or, if contraindicated, a specific serotonergic noradrenergic antidepressant such as mirtazapine or sodium valproate alone or in combination with the first two^[36]. These drugs have been beneficial in reducing pain, however the use of a partial dopamine agonist alone or in combination with tricyclic antidepressants resulted in fewer side effects, so it was better accepted by patients. Favorable results ranged from 4 (46.3%) to 16 weeks (65.9%) of use for 165 patients^[3].

Among the most accepted clinical interventions is the use of Botulinum Neurotoxin Type A injections. Where several authors used it locally when finding unsatisfactory results after the administration of antidepressants and antiepileptic drugs, the patient presented significant reduction in pain intensity and proposed the creation of trial designs to use it in a safe and effective way^[39], were used in areas of the gum, hard palate and upper lip, using 15-30U, with a total number of injections of 6-12 and a follow-up period of 6-20 months, the 4 patients obtained improvement with almost complete elimination of pain, lasting up to 6 months^[40]. Similar data to

another study in which its 9 patients presented significant 2% reduction in pain intensity, a response latency of 1-15 days with an effect duration of 2-6 months without adverse effects and concluded that it is a safe and effective option, but with insufficient evidence to establish it as a treatment guide^[41].

Pharmacological treatments such as amitriptyline, imipramine, aripiprazole, mirtazapine, or sodium valproate may have adequate long-term results, however, they involve prolonged use, which can result in alterations and undesirable side effects, so it is proposed to conduct studies where they are used in combination with local techniques such as infiltration of botulinum toxin A.

4. Conclusions

Atypical odontalgia is a rare condition, approximately 4.4 per 100,000 people, found in a higher percentage of women in the 50 to 60 age range. The pathophysiology is controversial and may be due to hyperreactivity of the trigeminal nucleus or poor electrical conduction causing local neuropathy. These patients usually present psychological and psychiatric disorders such as depression, anxiety, and symptoms related to trigeminal neuralgia, burning mouth syndrome, trauma and aggressive oral manipulation. Treatment modalities have been proposed, pharmacological in minimal doses, and those of intervention such as local injection of Botulinum Toxin A, both modalities have proved favorable.

5. References

1. Toyofuku A. Psychosomatic problems in dentistry. *Biopsychosoc Med* 2016;10:14.
2. McElin T, Horton D. Atypical facial pain: a statistical consideration of 66 cases, *Ann Intern Med* 1947;27(5):749-768.
3. Tu TTH, Miura A, Shinohara Y, Mikuzuki L, Kawasaki K, Sugawara S, *et al.* Pharmacotherapeutic outcomes in atypical odontalgia: determinants of pain relief. *J Pain Res* 2019;12:831-839.
4. Forssell H, Jääskeläinen SK, List T, Svensson P, Baad-Hansen L. An update on pathophysiological mechanisms related to idiopathic oro-facial pain conditions with implications for management. *J Oral Rehabil* 2015;42(4):300-322.
5. Durham J, Stone SJ, Robinson LJ, Ohrbach R, Nixdorf DR. *Int Endod J* 2019;52(3):279-287.
6. Marbach JJ. Is phantom tooth pain a deafferentation (neuropathic) syndrome? Part I: Evidence derived from pathophysiology and treatment. *Oral Surg Oral Med Oral Pathol* 1993;75:95-105.
7. Marbach JJ. Phantom tooth pain. *J Endod* 1978;4:362-72.
8. Graff-Radford SB, Solberg WK. Atypical odontalgia. *J Craniomandib Disord* 1992;6(4):260-5.
9. Headache Classification Subcommittee of the International Headache Society: The International Classification of Headache Disorders, 2nd ed. *Cephal* 2004;24(1):9-160.
10. Mueller D, Obermann M, Yoon MS, Poitz F, Hansen N, Slomke MA, *et al.* Prevalence of trigeminal neuralgia and persistent idiopathic facial pain: a population-based study. *Cephal* 2011;31(15):1542-1548.
11. The international classification of headache disorders. headache classification committee of the International Headache Society. 3rd ed. *Cephal* 2018;38:1-211.
12. Parashos P, Vickers ER. Atypical Odontalgia. *Aust Endod J* 2000;26(3):121-123.

13. Koopman JS, Dieleman JP, Huygen FJ, de Mos M, Martin CG, Sturkenboom MC, *et al.* Incidence of facial pain in the general population. *Pain* 2009;147(1-3):122-7.
14. Baad-Hansen L. Atypical odontalgia-Pathophysiology and clinical management. *J Oral Rehabil* 2008;35:1-11.
15. Pigg M, Svensson P, Drangsholt M, List T. Seven-year follow-up of patients diagnosed with atypical odontalgia: a prospective study. *J Orofac Pain* 2013;27(2):151-164.
16. Ram S, Teruel A, Kumar SK, Clark G. Clinical characteristics and diagnosis of atypical odontalgia: implications for dentists. *J Am Dent Assoc* 2009;140(2):223-228.
17. Porporatti AL, Costa YM, Stuginski-Barbosa J, Bonjardim LR, Conti PC, Svensson P, *et al.* Quantitative methods for somatosensory evaluation in atypical odontalgia. *Braz Oral Res* 2015;29(1):1-7.
18. Benoliel R, Gaul C. Persistent idiopathic facial pain. *Cephal* 2017;37(7):680-691.
19. Malacarne A, Spierings ELH, Lu C, Maloney GE. Persistent Dentoalveolar Pain Disorder: A Comprehensive Review. *J Endod* 2018;44(2):206-211.
20. Tarce M, Barbieri C, Sardella A. Atypical odontalgia: An up-to-date view. *Minerva Stomatol* 2013;62(5):163-181.
21. Rees RT, Harris M. Atypical odontalgia. *Br J Oral Surg* 1979;16(3):212-218.
22. Vickers ER. Atypical odontalgia. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1998;85(6):628-629.
23. Melzack R. From the gate to the neuromatrix. *Pain* 1999;(6):121-126.
24. King RB. Evidence for a central etiology of tic douloureux. 1967. *J Neurosurg* 2007;107(1):232-237.
25. List CF, Williams JR. Pathogenesis of trigeminal neuralgia; a review *AMA Arch Neurol Psychiatry* 1957;77(1):36-43.
26. Maarbjerg S, Wolfram F, Heinskou TB, Rochat P, Gozalov A, Brennum J, Olesen J, Bendtsen L. Persistent idiopathic facial pain - a prospective systematic study of clinical characteristics and neuroanatomical findings at 3.0 Tesla MRI. *Cephal* 2017;37(13):1231-1240.
27. Kuncz A, Vörös E, Barzó P, Tajti J, Milassin P, Mucsi Z, *et al.* Comparison of clinical symptoms and magnetic resonance angiographic (MRA) results in patients with trigeminal neuralgia and persistent idiopathic facial pain. Medium-term outcome after microvascular decompression of cases with positive MRA findings. *Cephal* 2006;26(3):266-276.
28. Lang E, Naraghi R, Tanrikulu L, Hastreiter P, Fahlbusch R, Neundorfer B, *et al.* Neurovascular relationship at the trigeminal root entry zone in persistent idiopathic facial pain: Findings from MRI 3D visualisation. *J Neurol Neurosurg Psych* 2005;76(11):1506-1509.
29. Forssell H, Tenovuo O, Silvioniemi P, Jääskeläinen SK. Differences and similarities between atypical facial pain and trigeminal neuropathic pain. *Neurology* 2007;2;69(14):1451-1459.
30. Marbach JJ, Hulbrock J, Hohn C, Segal AG. Incidence of phantom tooth pain: an atypical facial neuralgia. *Oral Surg Oral Med Oral Pathol* 1982;53(2):190-193.
31. Melis M, Lobo SL, Ceneviz C, Zawawi K, Al-Badawi E, Maloney G, *et al.* Atypical odontalgia: A review of the literature. *Headache* 2003;43(10):1060-1074.
32. Bosch-Aranda ML, Vázquez-Delgado E, Gay-Escoda C. Atypical Odontalgia: A Systematic Review Following the Evidence-Based Principles of Dentistry. *CRANIO* 2011;29(3):219-226.
33. Rajat Goel, Sanjeev Kmar, Ajay Panwar, Abhishek B Singh. Pontine Infarct Presenting with Atypical Dental Pain: A Case Report. *Open Dent J* 2015;9(2):337-339.
34. Graff-Radford SB, Solberg WK. Is atypical odontalgia a psychological problem?. *Oral Surg Oral Med Oral Pathol* 1993;75(5):579-82.
35. Tu TTH, Miura A, Shinohara Y, Mikuzuki L, Kawasaki K, Sugawara S, *et al.* Evaluating Burning Mouth Syndrome as a Comorbidity of Atypical Odontalgia: The Impact on Pain Experiences. *Pain Pract* 2017;18(5):580-586.
36. Takenoshita M, Miura A, Shinohara Y, Mikuzuki R, Sugawara S, Tu TTH, Kawasaki K, *et al.* Clinical features of atypical odontalgia; three cases and literature reviews. *Biopsychosoc Med* 2017;11:21.
37. Miura A, Tu TTH, Shinohara Y, Mikuzuki L, Kawasaki K, Sugawara S, *et al.* Psychiatric comorbidities in patients with Atypical Odontalgia. *J Psychosom Res* 2018;104:35-40.
38. Ghurye S, McMillan R. Orofacial pain - an update on diagnosis and management. *Br Dent J* 2017;223(9):639-647.
39. López-Bravo A, Jarauta-Salvador F, Lecina-Monge J, Oliveros-Cid A, Marín-Gracia M, Santos-Lasaosa S, *et al.* OnabotulinumtoxinA in the treatment of atypical odontalgia: description of a clinical case. *An Sist Sanit Navar* 2019;42(2):209-213.
40. Cuadrado ML, García-Moreno H, Arias JA, Pareja JA. Botulinum Neurotoxin Type-A for the Treatment of Atypical Odontalgia. *Pain Med* 2016;17(9):1717-21.
41. García-Sáez R, Gutiérrez-Viedma Á, González-García N, Gómez-Mayordomo V, Porta-Etessam J, Cuadrado ML, *et al.* Onabotulinumtoxin A injections for atypical odontalgia: an open-label study on nine patients. *J Pain Res* 2018;11:1583-1588.