Research Article

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ATYPICAL ODONTALGIA: A NON-ODONTOGENIC TOOTHACHE OF NEUROPATHIC ORIGIN

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ABSTRACT

Pain as a presenting symptom often has multidimensional qualities including sensory, cognitive, discriminate and affective qualities. Pain in the orofacial region represents a diagnostic challenge for even an astute practitioner due to the complex mechanism in which odontogenic and nonodontogenic pain may simulate each other. Patients presenting with atypical odontalgia (AO) complain of toothache often in absence of any relevant clinical or radiographic findings. No definite diagnostic criteria or treatment guidelines are yet available for management of such a condition. Among the current hypothesis available regarding the pathophysiology of AO, the most relevant one describes it as a neuropathic pain condition. Another important related factor that should be taken into consideration in such cases is the psychogenic background of the patient. This review describes the clinical presentations of atypical odontogenic pain and suggests the need to establish the accurate diagnostic criteria for identification and outline the treatment algorithm which focuses more on patient counselling and avoidance of invasive approach

Keywords: Atypical odontalgia, Phantom tooth pain, Neuropathic pain, Oro-facial.

INTRODUCTION

Most of the patients approaching clinician for pain relief, present with symptoms in orofacial region.[1,2] Although the International Headache Society (IHS) includes Atypical odontalgia (AO) as a subcategory along with persistent idiopathic facial pain disorder, it still is mostly an unrecognised diagnosis, since this term is often attributed to symptom complex which do not fit into the set of any established diagnostic criteria or classification of dental pathologies.[3] First described in literature, McElin and Horton in 1947, such a condition has often been reported in cases of endodontic therapy.[4]

The presence of unexplained toothache in absence of relevant clinical findings often prompts the dentist to perform dental treatment in absence of any pathology. Such uncalled for interventions often exacerbate the existing pain, thus directing the practitioner afterwards to diagnose the condition as atypical toothache.[5] IHS defines both AO and atypical facial pain as "persistent facial pain that does not have the characteristics of the cranial neuralgias and is not attributed to another disorder".[6] IHS also describes AO as a chronic pain condition where persistent or continuous pain is present for over 6 months after tooth extraction in the absence of any pathology.[7] Hence AO has also been termed as phantom tooth pain.[8,9]

Pathophysiology

There is scarce knowledge and understanding of the underlying mechanisms that mitigate pain in AO. The possible theories and hypothesis that have been put forward to explain the pathophysiology are based on neuropathogenic, vascular, psychogenic or idiopathic origins.[10] Since the clinical presentation follows a varied course in AO patients, it is possible that each case may have a different etiology. The following mechanisms have been described in literature to explain the origin of AO.

Psychogenic origin

Emotional component of the pain assumes the primary role in pain conditions that have a psychological etiology.[11] Although the psychological problems may act as a predisposing or a secondary factor, they cannot be attributed as the sole cause of pain in AO. This was supported by Brooke and Merskey who described the role of depression or emotional alteration only as a possible contributing factor instead of being the root cause of AO.[12]

Vascular origin

The role of vascular events in pathophysiology of AO was first suggested by Rees and Harris in 1979.[13] They observed the presence of migraine attacks in 30% of the patients reporting AO symptoms. The vascular pain hypothesis however is not considered significant in the genesis of AO by various authors due to absence of scientific evidence in its support.[11,14]

Neuropathic origin

The most accredited hypothesis available in literature regards AO as a neuropathic pathologic condition. The International Association for the Study of Pain defines neuropathic pain as a "pain initiated or caused by a primary lesion or dysfunction in the nervous system".[15] The chief characteristic of neuropathic pain is the partial of complete change in innervations area of a particular part of nervous system resulting in paradoxical presence of pain and hypersensitivity.[16] AO has been proposed to be a neuropathic disease state primarily due to the phenomena of deafferentation. Deafferentation of nerve fibres often occurs following a traumatic injury and results in persistent pain, paresthesia, dysesthesia even after complete healing of wound.[6] Deafferentation of primary afferent trigeminal nerve fibres may

occur following invasive dental procedures including endodontic therapy, tooth extraction, periapical and periodontal surgery, nerve block injection.[10] This was supported by List et al who observed in their study that 83% of the patients suffering from AO reported the onset of pain following an invasive dental procedure.[17] Central sensitization of primary afferent fibres following neural injury is characterised by spontaneous pain and hypersensitivity phenomena. The sensitization of nociceptive fibres occurs due to up-regulation of sodium channels resulting in reduced activation threshold, spontaneous ectopic activity.[18]
Exaggerated response to stimuli and continuous chronic pain is also attributed to activation of N-methyl-D-aspartate (NMDA) receptors and metabotropic glutamate receptors on second order neurons and binding of glutamate neurotransmitter.[18] Traumatic neuronal injury may also result in sprouting of nerve fibres or formation of nerve collaterals. If such nerve collaterals from A β fibres reach the second order neurons in dorsal horn of spinal cord, then even the non-noxious stimuli like thermal changes and pressure application on tooth is also experienced as pain, termed as allodynia.[6] Increased excitability of primary neurons may also occur in the event of loss of inhibitory pathways of pain control, including damage to inhibitory interneuron or down-regulation of presynaptic inhibitory receptors.[19]

Clinical Features

The most characteristic symptom of AO is pain which is localised to a particular tooth or region mostly as a dull ache and less commonly having a burning or throbbing quality.[6] Pain is non-paroxysmal in nature and is present mostly throughout the day, often chronic in nature (over 6 months). There is almost always a lack of clinical or radiologic signs of any pathosis.[13] AO is mostly prevalent in females who are in their mid 40s.[20,21] 3-6% of the patients present with a history of having undergone endodontic treatment.[22] Maxillary arch is involved more frequently compared to Mandibular arch, with a preponderance for posterior teeth. Another important feature is the absence of pain while sleeping, although it starts soon after awakening.[6] Pain aggravating factors may include touch, thermal changes, percussion.[6]

Diagnosis

The diagnosis of AO remains a controversial topic since there is absence of any specific diagnostic criteria or guidelines till date. The diagnosis is mainly established after exclusion of other pathologies originating from dental tissues. International headache society have included AO in their "Classification and Diagnostic Criteria for Headache Disorders, Cranial Neuralgias and Facial Pain" as diagnosis 12.8 "facial pain not fulfilling criteria in groups 11 and 12" (11: Headache or facial pain associated with disorder of cranium, neck, eyes, ears, nose, sinuses, teeth, mouth or other facial or cranial structures," 12:cranial neuralgias, nerve trunk pain and deafferentation pain")[3]. The diagnostic guidelines described are mentioned in table 1.
Later several other authors suggested further modified set of criteria to identify and diagnose AO. First among these were mentioned by Graff-Radford and Solberg who suggested inclusion of AO in IHS Classification under the diagnosis 11.6, headache or facial pain associated with "disorder of teeth, jaws and related structures" and called it as "idiopathic toothache" (table 2).[9]
Another set of criteria based on the clinical features of pain in AO were put forward by Marbach in 1993 (Table 3). He proposed the term "phantom tooth pain" for AO.[23] Later in a review, they further revised these criteria in order to make the diagnosis of AO more definite.[24]

Differential Diagnosis

Since AO is primarily a diagnosis of exclusion, various orofacial pathologies have to be ruled in a patient suffering from chronic facial pain in order to arrive at a definitive diagnosis. Conditions causing chronic pain similar to AO include trigeminal neuralgia,[25,23] toothache of pulpal origin,[26] maxillary sinusitis,[26] temporomandibular joint disorders,[27] cracked tooth syndrome,[27] pain related to masticatory muscles,[27] migraines,[27] postherpetic neuralgia,[27] ear and eye diseases.[27]
The most relevant and tough situation is the differentiation between AO and pain of endodontic origin or pulpal tooth pain. Melis et al have mentioned five features that are characteristic of AO pain but are absent in pulpal toothache: (1) continuous toothache in the absence of any pathology. (2) percussion of the involved tooth gives inconsistent response and the pain is not affected by thermal or mechanical stimulation; (3) the quality of pain remains consistent over the course of time whereas the pulpal toothache tends to change in severity with time; (4) pain is not relieved by dental treatment; (5) local anesthetics produce inconsistent effect in relieving pain.[6]
Another significant neurological disease causing chronic orofacial pain similar to AO is trigeminal neuralgia. In order to aid the clinicians in distinguishing between the two conditions, few clinical features characteristic of trigeminal neuralgia were listed by Marbach et al including the presence of trigger points; the quality of pain which is sharp, sudden, stabbing and electrical pain where as the pain in AO is dull and continuous.[27]
In examining patients suffering from migraines headache, who are suspected of having AO, it is important to remember that the pain is relieved by taking medications which are prescribed for treating migraine whereas the AO pain is unaffected by such medications.[20,28,29] Among other conditions that are confused with AO, pain originating from masticatory muscles or temporomandibular joint disorders often radiates to other areas of face, neck and shoulder along with exacerbation on movements of jaw.[23] Myofascial pain is commonly characterised by the presence of trigger points which elicit pain on palpation.[30] Diseases of ear, eye and sinus have typical accompanying symptoms that aid in establishing a definitive diagnosis.[31]

Treatment

Since the underlying pathophysiologic mechanism of AO is still not fully understood till date, it is impossible to achieve a complete cure. Therefore, instead of treating the root cause, a preferable approach has been directed toward managing physical and psychologic symptoms associated with the condition. The first and perhaps the most important step during management of AO is patient counselling. Patients suffering from chronic orofacial pain have already consulted several clinicians and undergone multiple invasive treatment procedures with little relief in pain or even worsening of symptoms. Therefore, it is of utmost importance to reassure the patients and provide detailed explanation regarding the nature of their disease and the possible treatment options available.[32] Additionally, the management of these cases is complicated by the co-existence of other chronic diseases including psychiatric illness. Therefore, an interdisciplinary treatment plan involving medical, dental as well as psychiatric specialists is the most effective approach in managing these patients.[10]
Since AO has been largely accepted to be a neuropathic disorder, the pharmacologic strategies for pain management rely on the medications that are prescribed in other neuropathic diseases. Jensen et al have stated that several medical experts suggest the treatment plan of neuropathic pathology to be dictated by the specific clinical symptom like allodynia or hyperalgesia based on the rationale that a specific pain symptom involves a specific receptor pathway.[33] This however may not be

a successful approach since neuropathic pain mechanisms involves multiple pathways and receptors.[33] Most of the cases of AO reported in literature have demonstrated best results with prescription of tricyclic antidepressants (TCA) alone or in combination with phenothiazines which are among the most frequently used neuroleptic agents.[20,34,35,36] The analgesic effect of TCA is produced by blocking the reuptake of neurotransmitters noradrenaline and serotonin in the endogenous pain inhibitory system.[37] Phenothiazines are employed to aid in potentiating the analgesic effect of TCA however, they should be prescribed with caution due to the risk of irreversible adverse effects in the nervous system causing tardive dyskinesia.[6,10] Most commonly prescribed TCA is Amitriptyline in the starting dose of 25 mg and ranging up to 100 mg.[35,36,38,39] Other drugs of same class that have been employed include imipramine, nortriptyline and dothiepen.[6] However the use of tricyclic drugs is limited by their side effects: dry mouth, weight gain, constipation, urinary retention.[6]

Other medications that have been tried with varying success include gabapentin, phenytoin, clonazepam, baclofen, aspirin, MAO inhibitors.[23,24] Opioid analgesics including oxycodone, ketamine, controlled release morphine, methadone have also been tried with moderate pain relief.[23,24] Injection of local anaesthetic agents and corticosteroids as well as giving nerve block via stellate or sphenopalatine ganglia to modulate sympathetic and parasympathetic pathways has also been found to be effective to some extent in alleviating pain.[23,24,40] Topical application of pain relieving medications has also been recommended in few reports. Topical capsaicin at the concentration of 0.025% for 4 weeks or topical anaesthetic and eutectic mixture of lidocaine and prilocaine bases (EMLA) cream at the concentration of 5% sometimes results in effective pain relief.[41]

An algorithm for treatment of neuropathic pain was proposed by Finnerup et al.[18] Based on this algorithm the first and foremost strategy involves patient counselling and avoiding any invasive or surgical treatments. Keeping in mind the safety profile and minimising the risk of adverse effects, the first line of pharmacologic management employs topical analgesics including anaesthetic agents like lidocaine or topical capsaicin. In case of ineffectiveness of these agents, TCA are prescribed unless contraindicated. However, if the pain still persists, then anticonvulsants like gabapentin or pregabalin are prescribed. As a last resort in some cases, opioid analgesics like tramadol and oxycodone are given to relieve pain.

Table 1: Diagnostic criteria given by headache classification committee of the international headache society for atypical odontalgia 12.8 Facial pain not fulfilling criteria in groups 11 and 12

(a)	Pain occurs daily for most or all of the day.
(b)	Initially at the time of onset, pain is limited to a specific region on unilateral side of face and may later spread to wider region. Pain is present as a deep ache which is difficult to localize.
(c)	No sensory loss is associated with pain.
(d)	Diagnostic investigations including radiographs fail to reveal any abnormality.

Table 2: Diagnostic criteria proposed by Graf-Radford and Solberg for atypical odontalgia (idiopathic tooth pain) 11.6.1 idiopathic toothache (atypical odontalgia)

(a)	Pain in a tooth or a tooth site.
(b)	Continuous or almost continuous pain.
(c)	Pain persisting more than 4 months.
(d)	No sign of local or referred pain.
(e)	Equivocal somatic nerve block.

Table 3: Diagnostic criteria proposed by Marbach for atypical odontalgia (phantom tooth pain)

1.	The onset of pain is associated with a nerve injury caused by dental, medical procedure or facial trauma.
2.	The pain may persist after healing of injured tissues.
3.	Pain occurs more commonly in patients having suffered dental or facial pain during or immediately before dental treatment or nerve injury.
4.	Pain is present as a continuous dull ache with difficulty in localising the exact site of pain or exact tooth. Patient's sleep is unaffected by pain.
5.	Clinical and radiographic investigations reveal absence of any pathology.
6.	Both genders are affected.
7.	There is limited data in literature relating depressive mental illness as an etiology in phantom tooth pain. Therefore personality disorders cannot be considered as a contributing factor or a consequence of phantom tooth pain.

CONCLUSION

The scarcity of knowledge regarding the exact etiology and pain mediating mechanism involved in AO is responsible for making its diagnosis and treatment a difficult task for even an astute clinician. On the basis of current information, AO appears to be primarily a neuropathic condition. Psychological disturbances are mostly present as a consequence instead of being a contributing factor, since the patient has often undergone multiple treatment procedures with little relief in pain. Therefore, patient counselling and avoidance of invasive treatment interventions are perhaps the two most important aspects of treatment or pain management. Majority of pharmacologic strategies are derived from cases of AO or chronic oro-facial pain that have been reported in literature. Additional research pertaining to specific pain mechanism is required to establish an evidence based treatment protocol that is beneficial for both the patient as well the clinician.

REFERENCES

1. Sarlani E, Balciunas BA, Grace EG. Orofacial pain – Part I: assessment and management of musculoskeletal and neuropathic causes. AACN Clin Issues 2005; 16:333–46. http://dx.doi.org/10.1097/00044067-200507000-00007
2. Annino DJ, Goguen LA. Pain from the oral cavity. Otolaryngologic Clinics of North America. 2003; 36:1127-35. http://dx.doi.org/10.1016/S0030-6665(03)00132-4
3. Headache Classification Committee of the International Headache Society, "Classification and diagnostic criteria for headache

disorders, cranial neuralgias and facial pain". Cephalalgia 1988; 8 Suppl 7:1–64.
4. McElin TW, Horton DT. Atypical facial pain: a statistical consideration of 65 cases. Ann Intern Med.1947; 27:749-53. http://dx.doi.org/10.7326/0003-4819-27-5-749
5. Abiko Y, Matsuoka H, Chiba I, Toyofuku A. Current evidence on atypical odontalgia: diagnosis and clinical management. Int J Dent.[10.1155/2012/518548] 2012 Jul [cited 2002 July 9].
6. Melis M, Lobo SL, Ceneviz C, Zawawi K, Al Badawi E, Maloney G et al. Atypical odontalgia: a review of the literature. Headache 2003; 43:1060–74. http://dx.doi.org/ 10.1046/j.1526-4610.2003.03207.x
7. Woda A, Pionchon P. A unified concept of idiopathic orofacial pain: clinical features. J Orofac Pain. 1999; 13:172–84.
8. Marbach JJ. Phantom tooth pain: deafferentation pain. Pa Dent J (Harrisb). 1984; 51:32–33.
9. Graff-Radford SB, Solberg WK. Atypical odontalgia. J Craniomandib Disord. 1992; 6:260–65.
10. Baad-Hansen L. Atypical odontalgia - pathophysiology and clinical management. J Oral Rehabil 2008; 35:1-11. http://dx.doi.org /10.1111/j.1365-2842.2007.01813.x
11. Graff-Radford SB, Solberg WK. Is atypical odontalgia a psychological problem? Oral Surg Oral Med Oral Pathol 1993; 75:579–82. http://dx.doi.org/10.1016/0030-4220(93)90228-V
12. Brooke RI, Merskey H. Is atypical odontalgia a psychological problem? Oral Surg Oral Med Oral Pathol 1994; 77:2–3. http://dx.doi.org/10.1016/S0030-4220(06)80094-0
13. Rees RT, Harris M. Atypical odontalgia. Br J Oral Surg. 1979; 16:212–18. http://dx.doi.org/10.1016/0007-117X(79) 90027-1
14. Bussone G, Tullo V. Reflections on the nosology of craniofacial pain syndromes. Neurol Sci. 2005; 26 Suppl 2:61–64. http://dx.doi.org/10.1007/s10072-005-0409-6
15. Merskey H, Bogduk N. Classification of chronic pain. Descriptions of chronic pain syndromes and definitions of pain terms. 2nd ed. Seattle: IASP Press; 1994.
16. Finnerup NB, Otto M, McQuay HJ, Jensen TS, Sindrup SH. Algorithm for neuropathic pain treatment: an evidence based proposal. Pain 2005; 118:289–305. http://dx.doi.org/ 10.1016/j.pain.2005.08.013
17. List T, Leijon G, Helkimo M, Oster A, Dworkin SF, Svensson P. Clinical findings and psychosocial factors in patients with atypical odontalgia: a case–control study. J Orofac Pain 2007; 21:89–98.
18. Finnerup NB, Jensen TS. Mechanisms of disease: mechanism based classification of neuropathic pain – a critical analysis. Nat Clin Pract Neurol 2006; 2:107–15. http://dx.doi.org/10.1038 /ncpneuro0118
19. Costigan M, Woolf CJ. Pain: molecular mechanisms. J Pain 2000; 1 Suppl 1:35-44. http://dx.doi.org/10.1054/ jpai.2000.9818
20. Reik L Jr. Atypical Odontalgia: a localized form of atypical facial pain. Headache 1984; 24:222-24. http://dx.doi.org /10.1111/j.1526-4610.1984.hed2404222.x
21. Schnurr RF, Brooke RI. Atypical Odontalgia. Update and comment on long-term follow up. Oral Surg Oral Med Oral Pathol 1992; 73:445-48. http://dx.doi.org/10.1016/0030-4220(92)90322-H
22. Campbell RL, Parks KW, Dodds RN. Chronic facial pain associated with endodontic therapy. Oral Surg Oral Med Oral Pathol 1990; 69:287-90. http://dx.doi.org/10.1016/ 0030-4220(90)90288-4
23. 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. http://dx.doi.org/10.1016/0030-4220(93)90413-X
24. Marbach JJ, Raphael KG. Phantom tooth pain: a new look at an old dilemma. Pain Med. 2001; 1:68-77. http://dx.doi.org/ 10.1046/j.1526-4637.2000.00012.x
25. Pertes RA, Bailey DR, Milone AS. Atypical odontalgia-a nondental toothache. J N J Dent Assoc 1995;66:29-33.
26. Marbach JJ. Orofacial phantom pain: theory and phenomenology. J Am Dent Assoc 1996; 127:221-29. http://dx.doi.org/10.14219/ jada.archive.1996.0172
27. Marbach JJ. Phantom tooth pain: differential diagnosis and treatment. N Y State Dent J 1993; 59:28-33.
28. Nicolodi M, Sicuteri F. Phantom tooth diagnosis and an amnestic focus on headache. N Y State Dent J 1993; 59:35-37.
29. Sicuteri F, Nicolodi M, Fusco BM, Orlando S. Idiopathic headache as a possible risk factor for phantom tooth pain. Headache 1991 ;31: 577-81. http://dx.doi.org/10.1111/ j.1526-4610.1991.hed3109577.x
30. Travell JG, Simon DG. Apropos of all muscles. Chapter 3. In: Travell JG, Simons DG, editors. Myofascial Pain and Dysfunction. The Trigger Point Manual. The Upper Extremeties. Baltimore, Md: Williams & Wilkins; 1983:45-102.
31. Brooke RI. Atypical Odontalgia. A report of twenty-two cases. Oral Surg Oral Med Oral Pathol 1980;49:196-99. http://dx.doi.org/10.1016/0030-4220(80)90044-4
32. Brooke RI, Schnurr RF. Diagnosing and treating atypical odontalgia . J Mass Dent Soc 1995;44:30-32.
33. Jensen TS, Baron R. Translation of symptoms and signs into mechanisms in neuropathic pain. Pain 2003; 102:1–8. http://dx.doi.org/10.1016/s0304-3959(03)00006-x
34. Sinha S, Pandeya S.N., Verma A, Yadav D. Synthesis and biological activity of phenothiazine derivatives. Int J Res Ayurveda Pharm 2011; 2:1130-37.
35. Kreisberg MK. Atypical odontalgia: differential diagnosis and treatment. J Am Dent Assoc 1982; 104:852–54. http://dx.doi.org/ 10.14219/jada.archive.1982.0297
36. Kaspo GA. Atypical Odontalgia in the dental office. J Mich Dent Assoc 1995; 77:34-36.
37. Clark GT. Persistent orodental pain, atypical odontalgia, and phantom tooth pain: when are they neuropathic disorders? J Calif Dent Assoc 2006;34:599–609.
38. Marbach JJ. Phantom tooth pain. J Endod 1978; 4:362-72. http://dx.doi.org/10.1016/S0099-2399(78)80211-8
39. Lilly JP, Law AS. Atypical odontalgia misdiagnosed as odontogenic pain: a case report and discussion of treatment. J Endod 1997; 23:337-39 http://dx.doi.org/10.1016/S0099-2399(97)80419-0
40. Gross SG. Atypical odontalgia: a cause for dental failure. J Conn State Dent Assoc 1991; 67:36-37.
41. Vickers ER, Cousins MJ, Walker S, Chisholm K. Analysis of 50 patients with atypical odontalgia. A preliminary report on pharmacological procedures for diagnosis and treatment. Oral Surg Oral Med Oral Pathol Endod 1998; 85:24-32. http://dx.doi. org/10.1016/S1079-2104(98)90393-6

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