

Atypical Facial Pain: A Mini-Review

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ABSTRACT

Persistent idiopathic facial pain (PIFP), previously called “atypical facial pain”, is a diagnostic entity that describes chronic facial pain without evidence of structural or other specific causes of pain. Most of the data clearly indicate that PIFP is a rare disorder. It is more common in women, and the mean age of onset is in the mid forties. Careful interdisciplinary collaboration is needed to establish the diagnosis and management of persistent idiopathic facial pain (PIFP). The diagnostic criteria for PIFP include the presence of daily or near daily pain that is initially confined but may subsequently spread, which cannot be attributed to any pathological process. So, the diagnosis of PIFP is mainly done by exclusion of other disorders.

Key words: persistent idiopathic facial pain; atypical facial pain; atypical odontalgia; trigeminal neuropathy

INTRODUCTION

Persistent idiopathic facial pain (PIFP), previously called “atypical facial pain”, is a diagnostic entity that describes chronic facial pain without evidence of structural or other specific causes of pain.^[1,2] The etiology of PIFP is unknown but surgery or injury in the trigeminal nerve distribution could be reported as a past precipitating event.

DEFINITION

The International Classification of Headache Disorders (ICHD, version 3) published by the International Headache Society (IHS) describes persistent idiopathic facial pain (PIFP) as ‘persistent facial and/or oral pain, with varying presentations but recurring daily for more than 2 hours per

day over more than 3 months, in the absence of clinical neurological deficit’. PIFP is the current diagnostic terminology that historically was considered under the name of atypical facial pain (AFP).^[2]

Diagnostic criteria for PIFP^[2]

Diagnostic criteria
A. Facial and/or oral pain fulfilling criteria B and C
B. Recurring daily for >2 hours per day for >3 months
C. Pain has both of the following characteristics: 1 Poorly localized, and not following the distribution of a peripheral nerve 2 Dull, aching or nagging quality
D. Clinical neurological examination is normal
E. A dental cause has been excluded by appropriate investigations
F. Not better accounted for by another ICHD-3 diagnosis

Comments from ICHD^[2]:

A wide variety of words are used to describe the character of Persistent idiopathic facial pain (PIFP) but it is most often depicted as dull, nagging or aching. It can have sharp exacerbations, and is aggravated by stress. Pain may be described as either deep or superficial. With time, it may spread to a wider area of the craniocervical region.

Persistent idiopathic facial pain (PIFP) may be comorbid with other pain conditions such as chronic widespread pain and irritable bowel syndrome. In addition, it presents with high levels of psychiatric comorbidity and psychosocial disability.

A continuum seems to exist from Persistent idiopathic facial pain (PIFP) induced by insignificant trauma to Painful post-traumatic trigeminal neuropathy caused obviously by significant insult to the peripheral nerves. Persistent idiopathic facial pain (PIFP) may originate from a minor operation or injury to the face, maxillae, teeth or gums but persist after healing of the initial noxious event and without any demonstrable local cause. However, psychophysical or neurophysiological tests may demonstrate sensory abnormalities.

The term *atypical odontalgia* has been applied to a continuous pain in one or more teeth or in a tooth socket after extraction, in the absence of any usual dental cause. This is thought to be a sub form of Persistent idiopathic facial pain (PIFP) although it is more localized, the mean age at onset is younger and genders are more balanced. Based on the history of trauma, atypical odontalgia may also be a sub form of Painful post-traumatic trigeminal neuropathy.

Etiology

Some risk factors have been suggested as etiologic factors but have not been proven. A role of female hormones has also been suggested as PIFP is more common in females. Interestingly, for many years, a psychogenic origin has been postulated for this condition,^[3] however it's not clear if psychological distress is the cause or the consequence of PIFP.

Prevalence

Most of the data clearly indicate that that PIFP is a rare disorder. The population prevalence of PIFP is estimated to be 0.03% and some studies have shown that it is more common in women.^[4,5] The mean age of onset is in the mid 40's.^[6]

Clinical features

Pain onset in PIFP is often associated with minor surgical or other invasive dental or otolaryngologic procedures; these may be reported as the initiating event or as an attempt to manage

the pain. However, many patients cannot reliably recall the sequence of events.

There are no clinically evident neurosensory deficits in PIFP.^[2] Clinical and imaging investigations do not reveal any relevant abnormalities of the face.

Pain in PIFP is usually deep but can be superficial as well. It is poorly localized, radiating and mostly unilateral, although up to 40% of cases may have bilateral pain. Pain is most often localized to the upper jaw, and may extend to the eyes, nose, cheek and temple. Severity of the pain may be aggravated by emotional stress. Most PIFP patients report persistent, long lasting (years) daily pain that tends to spread, in a non-dermatomal pattern, with time. Often PIFP may coexist with other chronic orofacial pain or headache syndromes. Psychiatric and psychosocial disorders have often been associated with PIFP.^[2,6]

Differential diagnosis

It is important for clinicians to clearly distinguish PIFP from other persistent orofacial pain disorders that may be confused with it, such as trigeminal neuralgia with persistent background pain, painful traumatic trigeminal neuropathies (PTTN), myofascial pain and others. The clinician must exclude other likely disorders by thorough clinical examination, follow up and imaging if there are indications.

In PTTN, pain is unilateral and may be precisely located to the distribution of the affected nerve with evident sensory dysfunction, particularly if a major nerve branch has been injured. In PTTN, pain intensity is moderate to severe and the pain character is usually burning or shooting, which is typical of a neuropathic pain syndrome. More often there is clinically severe allodynia, hyperalgesia or negative neurosensory signs, which should be absent in PIFP.^[7,8]

Contrary to trigeminal neuralgia, in PIFP there is a high prevalence of bilateral pain, depression and other chronic pain, and a low prevalence of stabbing pain, touch-evoked pain and remission periods. As opposed to trigeminal neuralgia, in PIFP

there is no association between the painful side and the presence of or degree of compression from a neurovascular contact in the root entry zone of the ipsilateral trigeminal nerve. In addition, TN is usually responsive to anticonvulsant therapy. [6]

In myofascial pain, patients will often describe dysfunction associated with chewing foods and a limited range of mandibular movements. Characteristically the pericranial, masticatory and cervical muscles are painful to moderate manual pressure and some may display the classical “trigger point” phenomenon in response to muscle manipulation. These findings should be absent in PIFP patients. [9-11]

Pathophysiology

Currently, the prevalent belief is that PIFP is a disproportionate reaction to a mild injury, but the exact pathophysiology is still unclear. [12] The large number of PIFP patients presenting with a history of mild trauma and subclinical sensory changes has led to the suggestion that PIFP and PTTN may represent extremes of a spectrum of clinical presentations. As such, PIFP would therefore be considered a neuropathic pain syndrome. [13]

How can GPs diagnose it with certainty?

The diagnostic criteria for PIFP include the presence of daily or near daily pain that is initially confined but may subsequently spread, [2] which cannot be attributed to any pathological process. So, the diagnosis of PIFP is mainly done by exclusion of other disorders.

Treatment

Careful interdisciplinary collaboration is needed to establish the diagnosis and management of persistent idiopathic facial pain (PIFP). Clinicians should be aware of the importance of listening to the patients and acknowledging the symptoms that patients are describing as a real condition. [14] Patient education is important to clarify the diagnosis, and certainly the patient should be discouraged from any further invasive interventions aimed at pain relief in the absence of clear associated pathology.

The use of medications, known to have an effect in painful neuropathies, i.e. antidepressants and antiepileptic drugs, could be of benefit in PIFP. Amitriptyline is often the first drug of choice in PIFP [15] as it reduces the nociceptive charges originating from the myofascial tissues and so helps in controlling the pain. Different medications have also been tried in clinical trials for management of PIFP as Selective serotonin reuptake inhibitors (SSRIs), Calcitonin, Sumatriptan, Topiramate and others. [16-18]

Other recent techniques used for relief of neuropathic pain as using high-frequency repetitive transcranial magnetic stimulation (rTMS) may be also promising for management of PIFP. [12]

Complementary and alternative medicine approaches as acupuncture, hypnosis, simple relaxation etc. [19,20] have also been suggested.

When to refer

Diagnosis of PIFP is difficult especially when the clinician cannot find an objective explanation to the patient’s subjective pain experience. So, there is a need for a multidisciplinary diagnostic approach for exclusion of relevant differential diagnoses.

REFERENCES

1. A. Woda, P. Pionchon, A unified concept of idiopathic orofacial pain: clinical features, *J Orofac Pain* 13(3) (1999) 172-84; discussion 185-95.
2. S. Headache Classification Committee of the International Headache, *The International Classification of Headache Disorders*, 3rd edition (beta version), *Cephalalgia* 33(9) (2013) 629-808.
3. C. Feinmann, M. Harris, R. Cawley, Psychogenic facial pain: presentation and treatment, *Br Med J (Clin Res Ed)* 288(6415) (1984) 436-8.
4. D. Mueller, M. Obermann, M.S. Yoon, F. Poitz, N. Hansen, M.A. Slomke, P. Dommers, E. Gizewski, H.C. Diener, Z. Katsarava, Prevalence of trigeminal neuralgia and persistent idiopathic facial pain: a population-based study, *Cephalalgia* 31(15) (2011) 1542-8.

5. V. Pfaffenrath, M. Rath, W. Pollmann, W. Keeser, Atypical facial pain--application of the IHS criteria in a clinical sample, *Cephalalgia* 13 Suppl 12 (1993) 84-8.
 6. S. Maarbjerg, F. Wolfram, T.B. Heinskou, P. Rochat, A. Gozalov, J. Brennum, J. Olesen, L. Bendtsen, Persistent idiopathic facial pain - a prospective systematic study of clinical characteristics and neuroanatomical findings at 3.0 Tesla MRI, *Cephalalgia* 27 (2016) 27.
 7. T. Renton, Z. Yilmaz, Profiling of patients presenting with posttraumatic neuropathy of the trigeminal nerve, *J. Orofac. Pain* 25(4) (2011) 333-44.
 8. M. Pigg, P. Svensson, M. Drangsholt, T. List, Seven-year follow-up of patients diagnosed with atypical odontalgia: a prospective study, *J Orofac Pain* 27(2) (2013) 151-64.
 9. E. Schiffman, R. Ohrbach, E. Truelove, J. Look, G. Anderson, J.P. Goulet, T. List, P. Svensson, Y. Gonzalez, F. Lobbezoo, A. Michelotti, S.L. Brooks, W. Ceusters, M. Drangsholt, D. Ettlin, C. Gaul, L.J. Goldberg, J.A. Haythornthwaite, L. Hollender, R. Jensen, M.T. John, A. De Laat, R. de Leeuw, W. Maixner, M. van der Meulen, G.M. Murray, D.R. Nixdorf, S. Palla, A. Petersson, P. Pionchon, B. Smith, C.M. Visscher, J. Zakrzewska, S.F. Dworkin, I.a.f.D.R. International Rdc/Tmd Consortium Network, I.A.f.t.S.o.P. Orofacial Pain Special Interest Group, Diagnostic Criteria for Temporomandibular Disorders (DC/TMD) for Clinical and Research Applications: recommendations of the International RDC/TMD Consortium Network and Orofacial Pain Special Interest Groupdagger, *J Oral Facial Pain Headache* 28(1) (2014) 6-27.
 10. C. Alonso-Blanco, C. Fernandez-de-las-Penas, A.I. de-la-Llave-Rincon, P. Zarco-Moreno, F. Galan-del-Rio, P. Svensson, Characteristics of referred muscle pain to the head from active trigger points in women with myofascial temporomandibular pain and fibromyalgia syndrome, *J Headache Pain* 13(8) (2012) 625-637.
 11. C.S. Stohler, C.J. Kowalski, J.P. Lund, Muscle pain inhibits cutaneous touch perception, *Pain* 92(3) (2001) 327-333.
 12. R. Benoliel, C. Gaul, Persistent idiopathic facial pain, *Cephalalgia* 37(7) (2017) 680-691.
 13. H. Forssell, O. Tenovuo, P. Silvoniemi, S.K. Jaaskelainen, Differences and similarities between atypical facial pain and trigeminal neuropathic pain, *Neurology* 69(14) (2007) 1451-9.
 14. A. Sardella, F. Demarosi, C. Barbieri, G. Lodi, An up-to-date view on persistent idiopathic facial pain, *Minerva Stomatol* 58(6) (2009) 289-99.
 15. Y. Sharav, E. Singer, E. Schmidt, R.A. Dionne, R. Dubner, The analgesic effect of amitriptyline on chronic facial pain, *Pain* 31(2) (1987) 199-209.
 16. S.D. Harrison, S.A. Balawi, C. Feinmann, M. Harris, Atypical facial pain: a double-blind placebo-controlled crossover pilot study of subcutaneous sumatriptan, *Eur Neuropsychopharmacol* 7(2) (1997) 83-8.
 17. G. Schwartz, M. Galonski, A. Gordon, M. Shandling, D. Mock, H.C. Tenenbaum, Effects of salmon calcitonin on patients with atypical (idiopathic) facial pain: a randomized controlled trial, *J Orofac Pain* 10(4) (1996) 306-15.
 18. M. Volcy, A.M. Rapoport, S.J. Tepper, F.D. Sheftell, M.E. Bigal, Persistent idiopathic facial pain responsive to topiramate, *Cephalalgia* 26(4) (2006) 489-91.
 19. G. Goddard, H. Karibe, C. McNeill, E. Villafuerte, Acupuncture and sham acupuncture reduce muscle pain in myofascial pain patients, *J Orofac Pain* 16(1) (2002) 71-6.
 20. R. Abrahamsen, L. Baad-Hansen, P. Svensson, Hypnosis in the management of persistent idiopathic orofacial pain--clinical and psychosocial findings, *Pain* 136(1-2) (2008) 44-52.
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