

REVIEW ARTICLE

Open Access

Multi-dimensionality of chronic pain of the oral cavity and face

Joanna M Zakrzewska

Abstract

Orofacial pain in its broadest definition can affect up to 7% of the population. Its diagnosis and initial management falls between dentists and doctors and in the secondary care sector among pain physicians, headache neurologists and oral physicians. Chronic facial pain is a long term condition and like all other chronic pain is associated with numerous co-morbidities and treatment outcomes are often related to the presenting co-morbidities such as depression, anxiety, catastrophising and presence of other chronic pain which must be addressed as part of management . The majority of orofacial pain is continuous so a history of episodic pain narrows down the differentials. There are specific oral conditions that rarely present extra orally such as atypical odontalgia and burning mouth syndrome whereas others will present in both areas. Musculoskeletal pain related to the muscles of mastication is very common and may also be associated with disc problems. Trigeminal neuralgia and the rarer glossopharyngeal neuralgia are specific diagnosis with defined care pathways. Other trigeminal neuropathic pain which can be associated with neuropathy is caused most frequently by trauma but secondary causes such as malignancy, infection and auto-immune causes need to be considered. Management is along the lines of other neuropathic pain using accepted pharmacotherapy with psychological support. If no other diagnostic criteria are fulfilled than a diagnosis of chronic or persistent idiopathic facial pain is made and often a combination of antidepressants and cognitive behaviour therapy is effective. Facial pain patients should be managed by a multidisciplinary team.

Keywords: Facial pain, Temporomandibular disorders, Trigeminal neuralgia, Burning mouth syndrome, Neuropathic pain, Persistent idiopathic facial pain, Cognitive behaviour therapy, Biopsychosocial

Introduction

This review will look at pain that predominantly presents in the lower part of the face and the mouth. The epidemiology and classification will be discussed and the diagnostic criteria presented together with a brief mention of management. The review will include a discussion about the multidimensionality of facial pain as there is increasing evidence throughout the field of chronic pain that psychosocial factors impact significantly not just on outcomes from management but also act as prognosticators and can even affect the way symptoms are reported. Many patients will have more than one pain diagnosis and there may also be an underlying psychiatric or personality disorder which

pre disposes to chronic pain and which will alter the presentation and significantly affect management [1].

When problems arise in this area patients become very confused as they are unsure as to whether they should consult a doctor or dentist. Equally health care professionals often struggle as it is rare for medical students to be taught in depth about the mouth and surrounding structures. On the other hand dentists do not have in depth knowledge of the biopsychosocial approach to head and neck pain, remain confused about management of non-dental pain and are very restricted in the types of drugs that they can prescribe [2,3]. Hence as Hals et al. [4] point out these patients often get stigmatized as "difficult" as few health care professionals feel capable of helping them single handed as they really need a multi-professional team. A recent study of the healthcare "journey" of chronic orofacial pain patients in the UK showed that 101 patients had attended a mean

Correspondence: j.zakrzewska@ucl.ac.uk Facial pain unit, Division of Diagnostic, Surgical and Medical Sciences, Eastman Dental Hospital, UCLH NHS Foundation Trust, 256 Gray's Inn Road, London WC1X 8LD, UK



of seven health care settings, seen a mean of three specialists and only 24% judged their treatment to be successful [5]. This is supported by a similar survey of German University dental schools in 2004 (poor response rate 45% of schools and based on 34,242 patients) that showed that patients were inadequately treated prior to referral but also indicated that many once in the dental schools were still not managed according to guidelines. Most were managed using only one modality despite 30% having psychological morbidity. Only 11% were referred for psychiatric or psychotherapy, 9% for pain therapy and 7% for neurological assessment whereas 30% were referred to oral and maxillofacial surgeons [6].

Review

Epidemiology

Aggarwal et al. [7] in a population based study in the UK using a postal questionnaire looked at frequently unexplained pains and found 7% of patients reported having a chronic orofacial pain, 15% reported chronic widespread pain, 9% irritable bowel syndrome and 8% chronic fatigue. Of these 9% reported more than one of these pains. A recent German population study using strictly defined criteria and face to face interviews by trained headache neurologists suggests that trigeminal neuralgia is commoner than persistent idiopathic facial pain but both are rare with a lifetime prevalence of trigeminal neuralgia of 0.3% versus 0.03% for persistent idiopathic facial pain [8]. Koopman et al. [9] using the research databases of all primary care physicians in the Netherlands searched for all cases of facial neuralgias and persistent idiopathic facial pain and found an incidence rate of 38.7 per 100,000 people years The diagnosis were validated by pain experts as they found up to 48% had been misdiagnosed by the primary care physicians. Trigeminal neuralgia and cluster headache were the most common types. On the other hand temporomandibular pain and muscle disorder-type pain (TMD) is common and population-based studies among adults report that approximately 10-15% have these disorders [10,11].

The pain "journey" and co-morbidity in chronic orofacial pain patients

It is crucial to remember that every long term condition including pain results in psychological morbidity and reduced quality of life. Increasingly it is being recognised that some patients have an increased risk of developing chronic facial pain [12] and recognising this group early may reduce multiple referrals and inappropriate management. It is known that predictors of poor outcome can be identified early [13,14]. A chronic pain patient who finally reaches a multi-disciplinary pain clinic will often have a long duration of pain, with significant

functional impairment and will have developed fixed ideas on cause, location and legitimacy of the pain and this will impact on the pain specialists approach [15]. Many as a result will have low expectations from their pain consultation [16]. Studies of chronic facial pain patients have shown similar breakdown in doctor patient relationships and patients express confusion about the varied views they had received on management of their pain and likely outcomes [17]. There is a general desire by orofacial pain patients to be understood, their pain to be acknowledged as real and to feel cared for as life has become hopeless and trust in the medical profession has been lost [18]. Patients expectations for outcomes from a pain clinic will vary and these need to be recognised so management plans are appropriate [19]. Illness beliefs have been found to affect outcomes in patients with orofacial pain [20].

Mental health status will affect the pain experience and conditions that are especially significant are depression and anxiety [21]. It is now known that there are neural markers for fear and anxiety which exacerbate chronic pain [22]. Mental defeat (a psychological construct which includes catastrophising) increases distress and disability from pain [23,24]. Patients with borderline personalities report higher pain levels than other pain patients [25]. A recent study by Taiminen et al. [1] of 63 patients with burning mouth syndrome or atypical facial pain supported these findings. They showed that over 50% of these patients had a lifetime mental health disorder especially depression and personality disorders were common. They demonstrated that the mental health problems predated the facial pain and they postulate that psychiatric conditions and these facial pains may be mediated by dysfunctional brain dopamine activity. The recent recommendations on rehabilitation of patients with temporomandibular disorders (TMD) also highlights the need to identify patients with mental health problems, termed red and yellow flags and it is suggested that this is done through a combination of questionnaires and clinical interview [26]. Another factor to take into account are the personality differences between doctors and patients as these can significantly affect a consultation [27] and patients anger and frustration with treatment can induce in clinicians emotions of anger/frustration which then hinders the consultation [28].

As in all consultations the history is crucial and clinicians must be prepared to listen to the patients narrative without interruption and with empathy [29]. Langewitz et al. [30] have shown that in a tertiary medical outpatient clinic the mean spontaneous talking time was 92 seconds with 78% of patients having finished their initial statement in two minutes yet physicians redirect patients' opening statement after a mean of 23 seconds. In those consultations were patients are allowed to complete

their opening statement patient satisfaction is improved and it leads to improved outcomes [31,32]. Cairns et al. [26] have suggest that dentists need additional training when taking histories from patients with TMD so they can identify broader issues which they suggest should include among others, chronicity, functional limitation, discrepancy between findings, overuse of medication, inappropriate behaviours, inappropriate expectations, inappropriate responsiveness to treatments, and risks of self-harm and suicide.

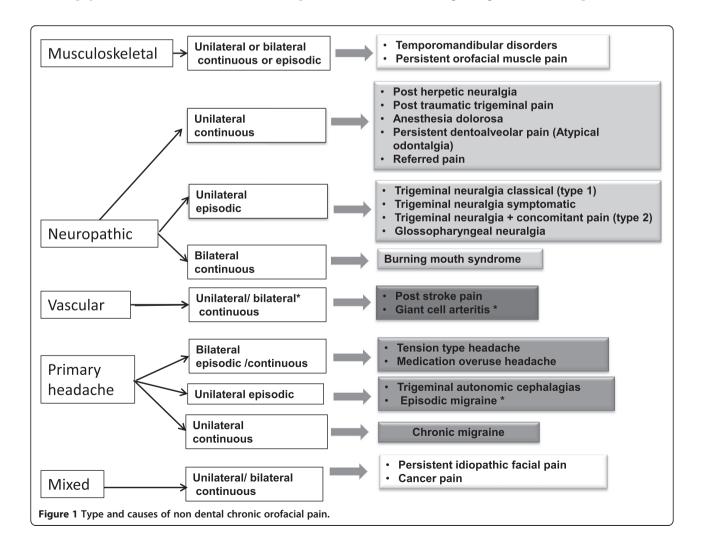
Much as clinicians like to lead their patients through a consultation it is often the uninterrupted opening statement that provides not only diagnostic markers but also details of impact on quality of life. Time is crucial, often restricted, but essential to make a diagnosis and establish a relationship with a patient as good communication is vital. Kenny [15] identified that there is often a struggle in complex pain consultations between the doctor and the patient as both want to function as speakers as both have their beliefs about the condition. This is further compounded by the fact physicians often do not understand their patients'

health beliefs [33]. Good patient doctor relationship will not only improve outcome expectations but will also decrease anxiety [34].

Classification and diagnosis

The International Headache Society (IHS) is in the process of updating its classification and most conditions covered here are to be found in chapter 13 [35]. Controversy remains about taxonomy and hence lack of international agreement with the result that there are a considerable number of labels being used for what may in fact be the same underlying conditions. An ontological approach would be of immense benefit especially if genotyping is to be done in the future as this would not necessitate any change in nomenclature when underlying mechanisms are identified [36].

When making a diagnosis it is useful to make the distinction whether this is a definite diagnosis, probable or even possible as this allows for a change in diagnosis once more facts come to light. Figure 1 shows a potential schema



based on possible causation and presentation of chronic facial pain.

Some chronic facial pain is strictly unilateral and follows clear neurological boundaries whereas others are very widespread in distribution. The majority of pain is continuous with flare ups but there are a few conditions which are very episodic and so it is worth eliciting these factors at the very beginning in order to narrow down the differential diagnosis. Benoliel et al. showed in their clinic population of 328 that chronic orofacial pain could be defined in the same way as chronic daily headache but stress that chronic orofacial pain includes a very heterogeneous group of symptoms [37]. There are a range of conditions that can result not only in pain but associated neuropathy which can be detected either by gross clinical examination or more detailed neurophysiological testing. Conditions that can result in neuropathy include trauma, inflammatory autoimmune disorders e.g. systemic scleroderma, Sjogren's syndrome, sarcoidosis, multiple sclerosis; rare vascular malformations, neoplasia anywhere along the trigeminal nerve and infections such as leprosy, viral, Lyme disease, syphilis [38]. Table 1 lists the main characteristics of the most common chronic non dental pains.

Dental pain

By far the commonest cause of pain in the lower face is dental i.e. pain related to the teeth and their surrounding structures. Few dental causes are chronic but given its high prevalence it needs to be considered in patients with other chronic pain who report a change in their symptoms which are not expected from the main condition [39,40]. Although some of the dental conditions are easy to diagnose with a careful examination using a good light others will need investigating with local imaging. Dentists are very good at diagnosis of most dental pain and so patients should be referred to them for an assessment. However, beware the dental practitioner who carries out irreversible dental treatment in a patient based on the patient's history alone with no clinical signs and X-ray validation. Many patients with trigeminal neuralgia have unnecessary teeth extractions and this has been documented by neurosurgeons. In the initial presentation this confusion is understandable as the pain is intermittent and sharp and can seem to the patient to be localised to the teeth. Equally those with neuropathic pain may undergo hours of complex dental treatment and find it does nothing to relieve their pain. The most challenging dental diagnosis is that of the cracked tooth as this can be very difficult to detect and so the symptoms can become chronic. More sophisticated imaging such as cone beam CT may be helpful. Dental pain needs to be treated mechanically together with analgesics and in some cases antibiotics.

Intraoral non dental pain

Within the mouth there are a variety of other causes of dental pain which are not related to the dental tissues.

Oral mucosal lesions such as recurrent oral ulceration, lichen planus, blistering conditions will cause chronic pain but there are very clear signs which make diagnosis easy. Salivary stones cause intermittent pain of relatively low intensity, pain most frequently occurs at the thought of food and when eating.

In some instances there is a clear history of nerve damage either due to dental procedures or to trauma whereas in other instances it currently remains impossible to determine the mechanism involved in causing the pain. This has led to some confusing nomenclature. Recently an international group of experts proposed the use of the term persistent dentoalveolar pain disorder [36] to encompass persistent pain without local disease (possible other pseudonyms include atypical odontalgia, phantom tooth pain). They propose that the symptoms could then be subdivided into those where there is a clear relationship to some form of trauma and others where the mechanism is unknown. Attempts have been made determine the patients' experience of this pain and some common themes emerge e.g. difficult to obtain a clear history, complex descriptors, and well localised, deep pain [41]. Terms such as post traumatic trigeminal neuropathy, peripheral painful traumatic trigeminal neuropathy PPTTN could then be used in those instances where there is a clear correlation between trauma and development of pain.

Atypical odontalgia

In atypical odontalgia the pain is very clearly localised to the dentoalveolar tissues either where teeth are still present or have been lost. There may or may not be a history of dental treatment prior to onset. The pain may move from one area to another. It is a dull throbbing continuous pain which can at times be sharp. It is often light touch provoked. Baad-Hansen postulates it is most likely to be a neuropathic pain and has different features from TMD [42,43]. Thus attempts have been made to characterise these patients but there is little evidence that pharmacological treatment along the lines of a neuropathic pain provide relief [43,44] but it probably is important to avoid more surgical interventions which can result in increased sensitisation.

Post traumatic trigeminal neuropathy

In some instances there is a clear history of nerve injury which can range from a dental extraction, root canal filling [45], local anaesthetic [46], implants [47], facial trauma [48]. Nerve injury, of varying degree, can be assumed to have been the cause and these conditions could be called a post traumatic trigeminal neuropathy

Table 1 The main characteristics of the most common chronic non dental pains and their managment

	Post traumatic trigeminal neuropathy	Buring mouth syndrome	Temporomandibular disorders	Trigeminal neuralgia	Persistent idiopathic facial pain	Trigeminal post herpetic neuralgia
Epidemiology	becoming increasingly common	rare	common	rare	rare	rare
Onset	3-6 months of traumatic event	slow	sometimes starts abruptly	memorable, sudden	slow	slow post herpes zoster
Duration	continuous with minor fluctuations, some have intermittent episodes	continuous	often constant	intermittent seconds to minutes	constant	constant
Periodicity	constant	can vary throughout the day	fluctuations often worse am/evening	refractory periods, many attacks a day periods of complete remission weeks, months	varies, can have periods of no pain	may be excacerbations
Site	distribution of a nerve branch, tooth or tooth bearing area		masseter, temporalis, around TMJ,ear, retromolar area	V2, V3 most common intraoral and extra oral	non anatomical, gradually gets larger	anatomic distribution, most common ophthalmic branch
Radiation	nil	all parts of the oral mucosa	may radiate to neck	only within trigeminal distribution	can spread over whole face, head, intra oral	little
Character	dull, burning, tingling, pins and needles at times sharp	burning, stinging, sore	aching,heavy, deep, can be sharp	sharp, shooting, lightening, may be a dull ache, burning after pain	dull, nagging, can be sharp	burning,, pins and needles
Severity	moderate to severe	mild to severe	variable moderate to severe	moderate to severe	moderate to severe	moderat to severe
Aggravating factors	touch	sometimes certain food,	prolonged chewing, opening wide, jaw movements	light touch, eating, some attacks are spontaneous	fatigue, stress	light touch,
Associated factors	may be altered sensation, reduced quality of life, history of trauma or dental procedure	altered taste, dry mouth, depression, anxiety, poor quality of life	clenching, bruxism, may have clicking of TMJ, locking, reduced opening, headaches, migraines	very rare autonomic features, fear of pain return, depression, poor quality of life	often other chronic pain, significant life events, vulnerable personalities,	may be altered sensation, skin changes
Examination	allodynia, hypoesthesia	nil, sometimes geographic tongue	palpation of muscles/joint induces same pain, unassisted reduced opening, clicking, intraorally evidence of frictional keratosis in cheeks, attrition of teeth	may trigger attack on touch, very rarely sensory changes	nil	allodynia, hypoaesthesia, hyperaesthesia
Management	drugs for neuropathic pain many benefit from CBT	neuropathic drugs, clonazepam, CBT	education, physiotherapy, psychology, anti- inflammatory drugs	carbamazepine/oxcarbazepine, neurosurgical procedures	CBT, antidepressant drugs	nortryptyline, pregablin, gapabentin, lidocaine patches

CBT cognitive behaviour therapy.

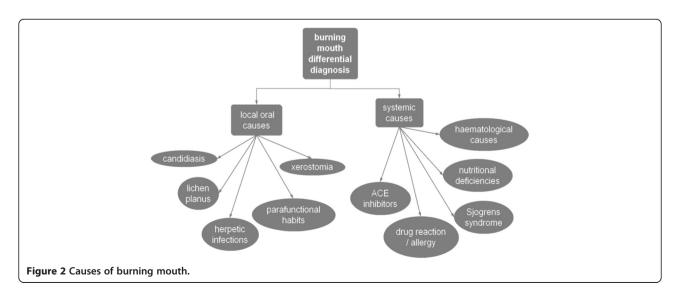
or peripheral painful traumatic trigeminal neuropathy PPTTN as recently proposed by Benoliel et al. [48]. Benoliel et al. compared 91 PPTN with 54 classical trigeminal neuralgia (TN) patients and showed that the temporal features of PPTTN were very varied with only 50% having continuous pain whereas others reported daily pain but which lasted less than 4 hours, some even had very short attacks that were similar to TN like pain. Pain is often described as burning, stabbing [43,48] but patients with definite nerve injury such as lingual and inferior alveolar nerve describe a feeling of pins and needles, fizzing and swollen sensations [47]. Pain in some circumstances is evoked in others it is spontaneous. Some sensory changes can often be detected on clinical testing and somatosensory testing will often show evidence of hypoaesthesia or allodynia, most imaging is negative. Meyer et al. have proposed a careful evaluation of peripheral nerve injuries that is both subjective (history, questionnaires and examination) and objective which includes quantitative sensory testing and imaging [49]. Renton et al. have shown how quality of life can be affected by trigeminal nerve injuries and the negative effects are more pronounced in patients with inferior alveolar nerve injury as opposed to lingual nerve injury [47]. Not all traumatic injuries result in pain some only present with sensory changes [47]. Although surgical repair is possible for inferior alveolar nerve most patients need to be managed according to guidelines for neuropathic pain.

Burning mouth syndrome glossodynia, stomatodynia

This strictly intraoral condition presents as a burning, discomfort of the oral mucosa especially the tongue for which local or systemic causes cannot be found. It is unusual in that it is especially common in postmenopausal women who have been found to have a high

level of anxiety and depression. The aetiology remains unknown and a variety of hypothesis have been put forward [50]. Neurophysiological testing and biopsies of the tongue have indicated that there are peripheral nerve changes with abnormal appreciation of temperature but central changes have also been noted on fMRI testing [51]. Not only do the patients report abnormal sensations but there are often other symptoms such as altered taste and disturbed salivary production [52]. The symptoms can be continuous but the intensity does vary throughout the day and some patients find eating mild food helpful. On examination the oral mucosa is normal although it is not unusual to see signs of a geographic tongue (erythema migrans) or fissured tongue and several patients have a habit of thrusting the tongue against their front teeth. Investigations are needed to exclude other causes of burning as indicated in Figure 2.

Management begins with acknowledging the symptoms as real and reassuring the patient that it is a recognised albeit rare condition. There have been a number of RCTs performed and a Cochrane systematic review [53]. Although a small study, not replicated again, cognitive behaviour therapy has been shown to be effective and it is the mainstay management in our facial pain unit. Clonazepam as a topical and systemic agent has been evaluated with mixed outcomes [54,55] but the significant side effect of drowsiness and the potential for addiction need to be taken into account. There are several RCTs on the use of alphalipoic acid, an antioxidant, but the outcomes are conflicting [53]. Given the evidence that this is potentially a neuropathic pain then drugs for neuropathic pain e.g. gabapentin, pregablin, tricyclic antidepressants can be tried. There is little data on prognosis. Sardella et al. in a survey of 53 patients showed that 3% had complete spontaneous resolution and less than 30% had a moderate improvement [56].



Extraoral pain with or without intra oral pain

These conditions often present with pain in both locations but equally can occur only extra-orally.

Temporomandibular disorders TMD

By far the commonest cause of non dental pain is a musculoskeletal pain related to the masticatory apparatus and it is principally an extra oral pain but pain is often felt in the retromolar area. TMD encompasses a variety of different conditions and most of the research in this area has been carried out by dentists with a specific interest in facial pain. The Research Diagnostic Criteria (RDC) for TMD where published in 1992 and have been used as a basis for research internationally [57]. The criteria include psychological distress and quality of life assessments which large studies in the US currently being done show are of great importance in prognosis [58,59]. Recently the RDC has been updated to make it more clinically relevant [60,61]. In the new proposals the commonest condition will be called myalgia and the criteria are: pain in the muscle affected by jaw movement and palpation of the masseter or temporalis induces the same pain. If the pain radiates to other local structures then it will be termed myofascial pain with referral. Disorders of the disc within the joint lead to clicking and crepitus and if the disc does not reduce then it will lead to limitation of opening and possible locking. On their own discs do not cause pain but they are often found in association with muscle pain. Degenerative disorders, joint dislocations as often found in hyper or hypomobility conditions do not cause significant pain. It is increasingly being recognised that TMD pain is related to other conditions such as fibromyalgia, back pain, migraine. It is also proposed that some headaches are due to TMD in that jaw movement induces the same headache [62]. Patients with TMDs do not cope as well with stress as general population [63] and oral health related quality of life is negatively affected by this condition becoming more pronounced in those with more symptoms [64].

A wide range of therapies are used but overall self-management through education, physiotherapy and with some cognitive behaviour therapy needs to be encouraged. Cairns et al. stress that patients with comorbidities should be seen by specialists with training in pain management [26]. Therapies range from soft diet, splint, and physiotherapy, drugs, psychological and surgical. Many of these have been evaluated in RCTs and there are several systematic reviews [65-67] but the quality of many of the trials is poor. A variety of surgical procedures including arthocentesis and arthroscopy can be used but they should only be used if there are functional signs [68].

Trigeminal neuralgia

Trigeminal neuralgia (TN) defined by The International Association for the Study of Pain (IASP) "as a sudden usually unilateral severe brief stabbing recurrent episodes of pain in the distribution of one or more branches of the trigeminal nerve." [69] and has been shown to have a significant impact on quality of life [70]. TN presents most commonly in the lower two branches of the trigeminal nerve. Very often it presents intraorally with triggers around the teeth [71] and hence many patients will undergo irreversible dental treatment unnecessarily [72].

It is easy to misdiagnosis TN as TMD especially if the TMD is unilateral as it is a far more common condition [73] whereas GPs tended to over diagnosis this condition [9]. Although it is often considered that TN is easy to diagnosis there is an increasing understanding that TN has a varied presentation and that some patients report considerable amount of less intense burning or dull pain after the main sharp attack of pain which can be present for more than 50% of the time. These have variously been labelled as type 2 [74] or TN with concomitant pain [75]. Neurosurgeons have started to report that outcomes are different in these groups [76]. Of the trigeminal autonomic cephalalgias SUNA (short unilateral neuralgiform pain with autonomic symptoms) needs to be considered as a potential differential as it can present in the lower face [77].

Symptomatic causes of TN need to be excluded e.g. tumours, often benign, multiple sclerosis and A-V malformations. Imaging is now considered to be part of the routine workup and some centres will use qualitative sensory testing [78,79].

Management is first medical with carbamazepine or oxcarbazepine with second line drugs being lamotrigine and baclofen [80,81]. If patients develop significant side effects or have poor pain control then surgery needs to be considered [78,79]. There are very few RCTs of surgical treatments [82] and evidence is based mainly on cohort data [78,79]. Microvascular decompression is the only procedure that is non-destructive and gives the longest pain free interval 70% pain free at 10 years [83]. For patients not suitable for this procedure ablative procedures include radiofrequency thermocoagulation glycerol rhizotomy, balloon compression or Gamma knife and these give 50% pain relief for four years but patients risk sensory changes which impact on quality of life [78,79]. Patient support groups are invaluable in providing further information and helping patients make decisions about treatments [84].

Glossopharyngeal neuralgia

This is a very rare condition and has the same features as TN except for location and the two conditions have

also been reported to co-exist. Pain can be felt deep within the ear but more commonly in the back of the tongue and throat. Talking and swallowing are particular trigger factors. Medical management is as for TN and in poorly controlled patients microvascular decompression is the surgery of choice [85].

Anaesthesia Dolorosa/post traumatic trigeminal neuropathy

Anaesthesia dolorosa is a term used by the neurosurgeons to denote pain after surgical damage to the trigeminal nerve most commonly at the level of the Gasserian Ganglion which occurs after ablative procedures for TN [74]. When the cause is due to other trauma e.g. fractures the term post traumatic trigeminal neuropathy is used. Both of these conditions develop within 3-6 months of the traumatic incident. The distribution is varied depending on the extent of the trauma but often in trigeminal neuralgia patients it extends to all three divisions of the trigeminal nerve. Patients report hyperalgesia, allodynia, hypoaesthesia and hypoalgesia very often described as "ants crawling over the face". There are often associated extensive psychological factors e.g. anger, depression, present and management is extremely difficult. In trauma patients the symptoms are often less severe and more localised. Patients report a poor response to drugs used for neuropathic pain and addition of cognitive behaviour therapy may be of value.

Persistent Idiopathic facial pain/Atypical facial pain

This condition has often been used as a bucket term to include all other facial pain that does not fit into other criteria as these patients do not have any sensory or physical signs. A recent small study on this group of patients using voxel-based morphotometry indicates that they do have similar changes to other chronic pain patients in parts of the brain associated with pain [86]. Epidemiological studies and psychiatric assessments suggest that psychological factors play a role in this condition [1,12] and that many of these patients are likely to have other chronic pain elsewhere [7]. This is generally a continuous pain which does not follow a neurological distribution. The pain often gradually becomes more diffuse and involves a larger area of the head and neck. Although often described as nagging and dull it can have sharp exacerbations [87]. All investigations are normal although Forssell et al. have shown that some patients labelled with this condition may on neurophysiological testing have abnormalities similar to trigeminal neuropathic pain [88]. Management is difficult and all types of approaches have been used - pharmacotherapy and behavioural therapy [89,90]. A multi-disciplinary approach using a combination of antidepressants drugs and cognitive behaviour therapy may be the best form of management [91].

Giant cell arteritis

Although principally occurring in the temple region jaw movement can induce more widespread pain and the tongue can be affected to such an extent that it appears cyanosed. The criteria are well established and any patient over 50 years should have an ESR and C reactive protein test. Temporal artery biopsy has been used as the gold standard but it is now suggested that ultrasonography and MRI scanning may be equally diagnostic [92]. Systematic steroids need to be commenced promptly to prevent blindness and other systemic effects [93].

Facial migraine/Neurovascular orofacial pain

There are several papers that have documented migraine like features of the lower face or as Benoliel et al. have called them neurovascular orofacial pain [94,95]. Yoon et al. [96] assessed migraine sufferers in the population and they established that this condition was rare but found that 9% of patients with migraine can have symptoms in the lower face i.e. V2 and 3 distribution but it was very rare to have symptoms only in the lower face. Benoliel et al. in their series of 328 patients diagnosed this condition in 23 patients. The pain was episodic or chronic, high severity, located in the lower half of the facial, (bilateral or unilateral), seven reported nausea and photophobia, eight autonomic features. It was not clear whether the headache was disabling and whether there was associated aura. Response to anti-migraine therapy including triptans was not provided. Oberman in their series of 7 showed all responded to triptans and three to prophylactic measures [94].

Conclusions

There are many different causes of orofacial pain and given the wide range of aetiologies management is also varied so that diagnosis is important in order to use the correct care pathway [40]. It is crucial to take a multidimensional approach to these patients and they are best managed in centres which have multi-disciplinary teams including pain specialists, oral surgeons, liaison psychiatrists, headache neurologists, neurosurgeons, clinical psychologists, physiotherapists and radiologists [4].

Competing interests

The author declare that she has no competing interests.

Acknowledgement

JZ undertook this work at UCL/UCLHT, who received a proportion of funding from the Department of Health's NIHR Biomedical Research Centre funding scheme

Received: 22 February 2013 Accepted: 9 April 2013 Published: 25 April 2013

References

 Taiminen T, Kuusalo L, Lehtinen L, Forssell H, Hagelberg N, Tenovuo O, Luutonen S, Pertovaara A, Jaaskelainen S (2011) Psychiatric (axis 1) and personality (axis11) disorders in patients with burning mouth syndrome or atypical facial pain. Scandinavian J Pain 2:155–160

- Aggarwal VR, Joughin A, Zakrzewska JM, Crawford FJ, Tickle M (2011) Dentists' and specialists' knowledge of chronic orofacial pain: results from a continuing professional development survey. Prim Dent Care 18:41–44
- Durham J, Exley C, Wassell R, Steele JG (2007) 'Management is a black art'-professional ideologies with respect to temporomandibular disorders. Br Dent J 202:E29–3
- Hals EKB, Stubhaug A (2011) Mental and somatic co-morbidities in chronic orofacial pain conditions: Pain patients in need of multiprofessional team approach. Scandinavian J Pain 2:153–154
- Beecroft EV, Durham J, Thomson P (2013) Retrospective examination of the healthcare 'journey' of chronic orofacial pain patients referred to oral and maxillofacial surgery. Br Dent J 214:E12
- Wirz S, Ellerkmann RK, Buecheler M, Putensen C, Nadstawek J, Wartenberg HC (2010) Management of chronic orofacial pain: a survey of general dentists in german university hospitals. Pain Med 11:416–424
- Aggarwal VR, McBeth J, Zakrzewska JM, Lunt M, Macfarlane GJ (2006) The epidemiology of chronic syndromes that are frequently unexplained: do they have common associated factors? Int J Epidemiol 35:468–476
- Mueller D, Obermann M, Yoon MS, Poitz F, Hansen N, Slomke MA, Dommes P, Gizewski E, Diener HC, Katsarava Z (2011) Prevalence of trigeminal neuralgia and persistent idiopathic facial pain: a population-based study. Cephalalgia 31:1542–1548
- Koopman JS, Dieleman JP, Huygen FJ, De Mos M, Martin CG, Sturkenboom MC (2009) Incidence of facial pain in the general population. Pain 147:122–127
- Drangsholt M, LeResche L (1999) Temporomandibular disorder pain. In: Crombie IK, Croft PR, Linton SJ, LeResche L (ed) Epidemiology of Pain. IASP, Seattle, pp 203–233
- Janal MN, Raphael KG, Nayak S, Klausner J (2008) Prevalence of myofascial temporomandibular disorder in US community women. J Oral Rehabil 35:801–809
- Aggarwal VR, Macfarlane GJ, Farragher TM, McBeth J (2010) Risk factors for onset of chronic oro-facial pain–results of the North Cheshire oro-facial pain prospective population study. Pain 149:354–359
- Macfarlane TV, Blinkhorn AS, Davies RM, Kincey J, Worthington HV (2004) Predictors of outcome for orofacial pain in the general population: a fouryear follow-up study. J Dent Res 83:712–717
- Rudich Z, Lerman SF, Gurevich B, Shahar G (2010) Pain specialists' evaluation of patient's prognosis during the first visit predicts subsequent depression and the affective dimension of pain. Pain Med 11:446–452
- Kenny DT (2004) Constructions of chronic pain in doctor-patient relationships: bridging the communication chasm. Patient Educ Couns 52:297–305
- Petrie KJ, Frampton T, Large RG, Moss-Morris R, Johnson M, Meechan G (2005) What do patients expect from their first visit to a pain clinic? Clin J Pain 21:297–301
- Wolf E, Birgerstam P, Nilner M, Petersson K (2006) Patients' experiences of consultations for nonspecific chronic orofacial pain: A phenomenological study. J Orofac Pain 20:226–233
- Wolf E, Birgerstam P, Nilner M, Petersson K (2008) Nonspecific chronic orofacial pain: studying patient experiences and perspectives with a qualitative approach. J Orofac Pain 22:349–358
- 19. Bass C, Jack T (2002) Current approaches in chronic pain. Clin Med 2:505–508
- Galli U, Ettlin DA, Palla S, Ehlert U, Gaab J (2010) Do illness perceptions predict pain-related disability and mood in chronic orofacial pain patients? A 6-month follow-up study. Eur J Pain 14:550–558
- Gerrits MM, Vogelzangs N, van OP, van Marwijk HW, van der Horst H, Penninx BW (2012) Impact of pain on the course of depressive and anxiety disorders. Pain 153:429–436
- Ochsner KN, Ludlow DH, Knierim K, Hanelin J, Ramachandran T, Glover GC, Mackey SC (2006) Neural correlates of individual differences in pain-related fear and anxiety. Pain 120:69–77
- 23. Tang NK, Goodchild CE, Hester J, Salkovskis PM (2010) Mental defeat is linked to interference, distress and disability in chronic pain. Pain 149:547–554
- Arnow BA, Blasey CM, Constantino MJ, Robinson R, Hunkeler E, Lee J, Fireman B, Khaylis A, Feiner L, Hayward C (2011) Catastrophizing, depression and painrelated disability. Gen Hosp Psychiatry 33:150–156
- 25. Sansone RA, Sansone LA (2012) Chronic pain syndromes and borderline personality. Innov Clin Neurosci 9:10–14
- Cairns B, List T, Michelotti A, Ohrbach R, Svensson P (2010) JOR-CORE recommendations on rehabilitation of temporomandibular disorders. J Oral Rehabil 37:481–489

- Clack GB, Allen J, Cooper D, Head JO (2004) Personality differences between doctors and their patients: implications for the teaching of communication skills. Med Educ 38:177–186
- 28. Trost Z, Vangronsveld K, Linton SJ, Quartana PJ, Sullivan MJ (2012) Cognitive dimensions of anger in chronic pain. Pain 153:515–517
- Rahman A, Tasnim S (2007) Twelve tips for better communication with patients during history-taking. Scientific World Journal 7:519–524
- Langewitz W, Denz M, Keller A, Kiss A, Ruttimann S, Wossmer B (2002)
 Spontaneous talking time at start of consultation in outpatient clinic: cohort study. BMJ 325:682–683
- 31. Marvel MK, Epstein RM, Flowers K, Beckman HB (1999) Soliciting the patient's agenda: have we improved? JAMA 281:283–287
- Robinson JD, Heritage J (2006) Physicians' opening questions and patients' satisfaction. Patient Educ Couns 60:279–285
- Street RL, Jr, Haidet P (2011) How well do doctors know their patients? Factors affecting physician understanding of patients' health beliefs.
 J Gen Intern Med 26:21–27
- Verheul W, Sanders A, Bensing J (2010) The effects of physicians' affectoriented communication style and raising expectations on analogue patients' anxiety, affect and expectancies. Patient Educ Couns 80:300–306
- 35. Anonymous (2004) The International Classification of Headache Disorders: 2nd edition. Cephalalgia 24(Suppl 1):9–160
- 36. Nixdorf DR, Drangsholt MT, Ettlin DA, Gaul C, de LR, Svensson P, Zakrzewska JM, DE LA, Ceusters W (2012) Classifying orofacial pains: a new proposal of taxonomy based on ontology. J Oral Rehabil 39:161–169
- Benoliel R, Eliav E, Sharav Y (2010) Classification of chronic orofacial pain: applicability of chronic headache criteria. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 110:729–737
- 38. Smith JH, Cutrer FM (2011) Numbness matters: a clinical review of trigeminal neuropathy. Cephalalgia 31:1131–1144
- Hegarty AM, Zakrzewska JM (2011) Differential diagnosis for orofacial pain, including sinusitis, TMD, trigeminal neuralgia. Dent Update 38:396–408
- Zakrzewska JM (2009) Orofacial Pain. Oxford University Press, Oxford
 Durham J, Exley C, John MT, Nixdorf DR (2013) Persistent dentoalveolar
- pain: the patient's experience. J Orofac Pain 27:6–13
 42. Baad-Hansen L, Leijon G, Svensson P, List T (2008) Comparison of clinical findings and psychosocial factors in patients with atypical odontalgia and temporomandibular disorders. J Orofac Pain 22:7–14
- 43. Baad-Hansen L (2008) Atypical odontalgia pathophysiology and clinical management. J Oral Rehabil 35:1–11
- Baad-Hansen L, List T, Jensen TS, Svensson P (2006) Increased pain sensitivity to intraoral capsaicin in patients with atypical odontalgia. J Orofac Pain 20:107–114
- Nixdorf DR, Moana-Filho EJ, Law AS, McGuire LA, Hodges JS, John MT (2010) Frequency of persistent tooth pain after root canal therapy: a systematic review and meta-analysis. J Endod 36:224–230
- Renton T, Adey-Viscuso D, Meechan JG, Yilmaz Z (2010) Trigeminal nerve injuries in relation to the local anaesthesia in mandibular injections. Br Dent J 209:E15
- 47. Renton T, Yilmaz Z (2011) Profiling of patients presenting with posttraumatic neuropathy of the trigeminal nerve. J Orofac Pain 25:333–344
- Benoliel R, Zadik Y, Eliav E, Sharav Y (2012) Peripheral painful traumatic trigeminal neuropathy: clinical features in 91 cases and proposal of novel diagnostic criteria. J Orofac Pain 26:49–58
- Meyer RA, Bagheri SC (2011) Clinical evaluation of peripheral trigeminal nerve injuries. Atlas Oral Maxillofac Surg Clin North Am 19:15–33
- Woda A, Dao T, Gremeau-Richard C (2009) Steroid dysregulation and stomatodynia (burning mouth syndrome). J Orofac Pain 23:202–210
- Albuquerque RJ, de LR, Carlson CR, Okeson JP, Miller CS, Andersen AH (2006) Cerebral activation during thermal stimulation of patients who have burning mouth disorder: an fMRI study. Pain 122:223–234
- 52. Grushka M (1987) Clinical features of burning mouth syndrome. Oral Surg Oral Med Oral Pathol 63:30–36
- Zakrzewska JM, Forssell H, Glenny AM (2005) Interventions for the treatment of burning mouth syndrome. Cochrane Database Syst Rev 1, CD002779
- Gremeau-Richard C, Woda A, Navez ML, Attal N, Bouhassira D, Gagnieu MC, Laluque JF, Picard P, Pionchon P, Tubert S (2004) Topical clonazepam in stomatodynia: a randomised placebo-controlled study. Pain 108:51–57
- Grushka M, Epstein J, Mott A (1998) An open-label, dose escalation pilot study of the effect of clonazepam in burning mouth syndrome. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 86:557–561

- Sardella A, Lodi G, Demarosi F, Bez C, Cassano S, Carrassi A (2006) Burning mouth syndrome: a retrospective study investigating spontaneous remission and response to treatments. Oral Dis 12:152–155
- Dworkin SF, LeResche L (1992) Research diagnostic criteria for temporomandibular disorders: review, criteria, examinations and specifications, critique. J Craniomandib Disord 6:301–355
- Fillingim RB, Ohrbach R, Greenspan JD, Knott C, Dubner R, Bair E, Baraian C, Slade GD, Maixner W (2011) Potential psychosocial risk factors for chronic TMD: descriptive data and empirically identified domains from the OPPERA case—control study. J Pain 12:T46—T60
- Slade GD, Bair E, By K, Mulkey F, Baraian C, Rothwell R, Reynolds M, Miller V, Gonzalez Y, Gordon S, Ribeiro-Dasilva M, Lim PF, Greenspan JD, Dubner R, Fillingim RB, Diatchenko L, Maixner W, Dampier D, Knott C, Ohrbach R (2011) Study methods, recruitment, sociodemographic findings, and demographic representativeness in the OPPERA study. J Pain 12:T12–T26
- Ohrbach R, Fillingim RB, Mulkey F, Gonzalez Y, Gordon S, Gremillion H, Lim PF, Ribeiro-Dasilva M, Greenspan JD, Knott C, Maixner W, Slade G (2011) Clinical findings and pain symptoms as potential risk factors for chronic TMD: descriptive data and empirically identified domains from the OPPERA case—control study. J Pain 12:T27—T45
- 61. Schiffman E, Ohrbach R, Truelove E, Look J, Andersen G, Goulet JP, List T, Svensson P, Gonzales GR, et al. (2013) 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 Group. Submitted
- Schiffman E, Ohrbach R, List T, Anderson G, Jensen R, John MT, Nixdorf D, Goulet JP, Kang W, Truelove E, Clavel A, Fricton J, Look J (2012) Diagnostic criteria for headache attributed to temporomandibular disorders. Cephalalgia 32:683–692
- Reissmann DR, John MT, Schierz O, Seedorf H, Doering S (2012) Stressrelated adaptive versus maladaptive coping and temporomandibular disorder pain. J Orofac Pain 26:181–190
- Dahlstrom L, Carlsson GE (2010) Temporomandibular disorders and oral health-related quality of life. A systematic review. Acta Odontol Scand 68:80–85
- List T, Axelsson S (2010) Management of TMD: evidence from systematic reviews and meta-analyses. J Oral Rehabil 37:430–451
- Fricton JR, Ouyang W, Nixdorf DR, Schiffman EL, Velly AM, Look JO (2010) Critical appraisal of methods used in randomized controlled trials of treatments for temporomandibular disorders. J Orofac Pain 24:139–151
- Mujakperuo HR, Watson M, Morrison R, Macfarlane TV (2010)
 Pharmacological interventions for pain in patients with temporomandibular disorders. Cochrane Database Syst Rev 10, CD004715
- Guo C, Shi Z, Revington P (2009) Arthrocentesis and lavage for treating temporomandibular joint disorders. Cochrane Database Syst Rev 4, CD004973
- Merskey H, Bogduk N (1994) Classification of chronic pain. Descriptors of chronic pain syndromes and definitions of pain terms. IASP Press, Seattle
- Tolle T, Dukes E, Sadosky A (2006) Patient burden of trigeminal neuralgia: results from a cross-sectional survey of health state impairment and treatment patterns in six european countries. Pain Pract 6:153–160
- Bowsher D (2000) Trigeminal neuralgia: a symptomatic study on 126 successive patients with and without prevoius intervention. Pain Clin 12:93–101
- Law AS, Lilly JP (1995) Trigeminal neuralgia mimicking odontogenic pain. A report of two cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 80-96-100
- Drangsholt M, Truelove E (2001) Trigeminal neuralgia mistaken as temporomandibular disorder. J Evid Base Dent Pract 1:41–50
- Limonadi FM, McCartney S, Burchiel KJ (2006) Design of an artificial neural network for diagnosis of facial pain syndromes. Stereotact Funct Neurosurg 84:212–220
- Obermann M, Yoon MS, Ese D, Maschke M, Kaube H, Diener HC, Katsarava Z (2007) Impaired trigeminal nociceptive processing in patients with trigeminal neuralgia. Neurology 69:835–841
- Miller JP, Magill ST, Acar F, Burchiel KJ (2009) Predictors of long-term success after microvascular decompression for trigeminal neuralgia. J Neurosurg 110:620–626
- 77. Cohen AS, Matharu MS, Goadsby PJ (2006) Short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing

- (SUNCT) or cranial autonomic features (SUNA)–a prospective clinical study of SUNCT and SUNA. Brain 129:2746–2760
- Cruccu G, Gronseth G, Alksne J, Argoff C, Brainin M, Burchiel K, Nurmikko T, Zakrzewska JM (2008) AAN-EFNS guidelines on trigeminal neuralgia management. Eur J Neurol 15:1013–1028
- Gronseth G, Cruccu G, Alksne J, Argoff C, Brainin M, Burchiel K, Nurmikko T, Zakrzewska JM (2008) Practice parameter: the diagnostic evaluation and treatment of trigeminal neuralgia (an evidence-based review): report of the Quality Standards Subcommittee of the American Academy of Neurology and the European Federation of Neurological Societies. Neurology 71:1183–1190
- 80. Wiffen PJ, Derry S, Moore RA, McQuay HJ (2011) Carbamazepine for acute and chronic pain in adults. Cochrane Database Syst Rev 1, CD005451
- Wiffen PJ, Derry S, Moore RA (2011) Lamotrigine for acute and chronic pain. Cochrane Database Syst Rev 2, CD006044
- Zakrzewska JM, Akram H (2011) Neurosurgical interventions for the treatment of classical trigeminal neuralgia. Cochrane Database Syst Rev 9, CD007312
- 83. Zakrzewska JM, Coakham HB (2012) Microvascular decompression for trigeminal neuralgia: update. Curr Opin Neurol 25:296–301
- Zakrzewska JM, Jorns TP, Spatz A (2009) Patient led conferences—who attends, are their expectations met and do they vary in three different countries? Eur J Pain 13:486–491
- Patel A, Kassam A, Horowitz M, Chang YF (2002) Microvascular decompression in the management of glossopharyngeal neuralgia: analysis of 217 cases. Neurosurgery 50:705–710
- Schmidt-Wilcke T, Hierlmeier S, Leinisch E (2010) Altered regional brain morphology in patients with chronic facial pain. Headache 50:1278–1285
- 87. Zebenholzer K, Wober C, Vigl M, Wessely P, Wober-Bingol C (2006) Facial pain and the second edition of the international classification of headache disorders. Headache 46:259–263
- 88. Forssell H, Tenovuo O, Silvoniemi P, Jaaskelainen SK (2007) Differences and similarities between atypical facial pain and trigeminal neuropathic pain. Neurology 69:1451–1459
- Jensen R, Mitsikostas DD, Wober C (2011) Persistent idiopathic facial pain. In: Martelletti P, Steiner TJ (ed) Handbook of headache. Springer, Practical Management, pp 506–511
- 90. List T, Axelsson S, Leijon G (2003) Pharmacologic interventions in the treatment of temporomandibular disorders, atypical facial pain, and burning mouth syndrome. A qualitative systematic review. J Orofac Pain 17:301–310
- 91. Harrison SD, Glover L, Feinmann C, Pearce SA, Harris M (1997) A comparison of antidepressant medication alone and in conjunction with cognitive behavioural therapy for chronic idiopathic facial pain. In: Turner JA, Wiesenfeld-Hallin Z (ed) Jensen TS. IASP Press, Seattle, pp 663–672
- 92. Meisner RJ, Labropoulos N, Gasparis AP, Tassiopoulos AK (2011) How to diagnose giant cell arteritis. Int Angiol 30:58–63
- Kale N, Eggenberger E (2010) Diagnosis and management of giant cell arteritis: a review. Curr Opin Ophthalmol 21:417–422
- Obermann M, Mueller D, Yoon MS, Pageler L, Diener H, Katsarava Z (2007) Migraine with isolated facial pain: a diagnostic challenge. Cephalalgia 27:1278–1282
- Benoliel R, Birman N, Eliav E, Sharav Y (2008) The International Classification of Headache Disorders: accurate diagnosis of orofacial pain? Cephalalgia 28:752–762
- Yoon MS, Mueller D, Hansen N, Poitz F, Slomke M, Dommes P, Diener HC, Katsarava Z, Obermann M (2010) Prevalence of facial pain in migraine: a population-based study. Cephalalgia 30:92–96

doi:10.1186/1129-2377-14-37

Cite this article as: Zakrzewska: Multi-dimensionality of chronic pain of the oral cavity and face. The Journal of Headache and Pain 2013 14:37.