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Journal of Sleep Medicine & Disorders

Review Article

Management of Medically Unexplained Chronic Orofacial Pain: Justification for Behavioural Interventions

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Submitted: 07 January 2016
Accepted: 22 January 2016
Published: 25 January 2016
ISSN: 2379-0822

Keywords
- Management of chronic oro-facial pain
- Unexplained chronic oro-facial pain
- Behavioural interventions

Abstract

Chronic Orofacial Pain (COFP) is currently treated according to a biomedical model, particularly in primary care, where management often involves invasive and irreversible treatments that are not evidence based and do not improve outcomes for patients. Studies have shown that COFP shares a number of risk factors and maintaining features with other medically unexplained conditions/symptoms (MUS), which are associated with psychological, social and behavioural dysfunctions. Evidence from studies of COFP and other MUS suggests that CBT based therapies may be effective in targeting both maintaining and exacerbating features and subsequently improve outcomes for patients. This piece positions COFP as the MUS of dentistry, affording a unique perspective in terms of exploring possible non-invasive management techniques, which should be a priority for future testing.

INTRODUCTION

Chronic Orofacial Pain (COFP) can be attributed to a number of disorders of the face, jaw and mouth, and is an encompassing term that includes clinical conditions such as temporomandibular joint disorder (TMD), burning mouth syndrome, atypical odontalgia and atypical facial pain (or persistent idiopathic facial pain). Although the site of the pain or discomfort may differ, when they become chronic and intractable, these clinical presentations display a common cluster of several symptoms, and a unified concept has been proposed based on their shared characteristics [1]. Oral pain includes pain experienced in the mouth, gums and teeth, and facial pain includes pain originating below the orbitomeatal line, above the neck and anterior to the ears [2]. The condition is considered chronic or long–term when symptoms have been experienced for a period of at least three months. As with many other medically unexplained symptoms, patients with chronic orofacial pain are often subjected to extensive diagnostic investigations from which an organic cause is rarely found. A diagnosis of COFP is therefore reached when other dental diseases have been excluded. Historically, COFP is a well recognised condition particularly in secondary care [3-5] and is commonly diagnosed in hospital outpatient clinics [6]. It is also common in the general population with a prevalence of 7% [7].

CURRENT MANAGEMENT

Agostoni, Frigerio, & Santoro [8] describe current treatment of persistent idiopathic facial pain as *"difficult and unsatisfactory"*. Most chronic orofacial pain disorders are ultimately referred to, and managed by, dentists and most patients undergo unnecessary dental procedures while a diagnosis is sought [9].Management of COFP tends to focus on correction of mechanical factors such as bruxism (teeth grinding) and malocclusion (irregular bite), using physical therapies such as occlusal splints, occlusion adjustment and surgery [10,11]. Evidence suggests that these procedures show limited efficacy for chronic sufferers [10,12,13] and research into the use of commonly used/applied medical interventions to treat oro-facial pain has found that symptoms often persist following treatment using these procedures [10,14,15]. Studies on the use of occlusal splint therapy, a commonly used method to treat TMD, have found weak evidence to suggest that it is beneficial [12,16,17] and some forms of surgery, such as tooth extractions and the replacement of the temporomandibular joint with a prosthesis may result in iatrogenic harm to patients [18].

There is therefore a lack of evidence to support the use of current dental treatments, which may in themselves be invasive and have the potential to cause irreversible damage.

COFP as a medically unexplained symptom.

Pain in the face or mouth in its acute form is most commonly related to periodontal disease and dental caries and, less frequently, damage or trauma to the teeth and mouth and can often be successfully treated by routine dental procedures. However when the condition presents as chronic, diagnosis, treatment and management become more difficult and inconsistent. Long term symptoms (experienced for more than more than 3 months) often cannot be attributed to a pathological or medical origin by clinicians, or the original pathology has long since been resolved while symptoms remain [19] . COFP can be diagnosed according to distinct characteristics, many in common with other idiopathic (of unknown origin) disorders, differentiating it from other dental conditions for which a clear underlying pathology can be found. Symptoms are often reported in idiosyncratic ways with patients often using pain descriptors such as burning, nagging, aching, and tingling [20]. Stress is likely to be reported as an exacerbating factor, the pain site is often described as poorly localised, and symptoms tend to remain for a persistent or chronic duration. In addition, psychosocial disability, multiple consultations and co-morbidities, such as teeth grinding or clenching and reporting of other unexplained syndromes are reported frequently by COFP patients [21-23].

Other chronic conditions, which are diagnosed by an assessment of symptoms, rather than detection of pathology include those characterised by pain (e.g. fibromyalgia and lower back pain), tiredness (e.g. chronic fatigue syndrome) and gastric discomfort (Irritable bowel syndrome). Medically unexplained symptoms (MUS) are common, with more than a quarter of primary care patients in England having unexplained chronic pain, causing similar levels of disease and disability as conditions with an identifiable physical pathology [24]. Most medical specialities have a diagnostic category for MUS, with common symptoms, epidemiology and responses to treatment. For example, gastroenterology has irritable bowel syndrome and rheumatology has fibromyalgia) [6]. There is evidence to suggest that COFP may be not only a dental MUS but part of a wider spectrum of unexplained disorders [7]. Similar social, psychological and behavioural traits have been found in individuals suffering from other MUS conditions and COFP [25-27]. High levels of health anxiety, reassurance seeking behaviour, reporting of recent adverse life events and unhelpful behaviours in response to illness have been found in patients suffering from COFP, chronic widespread pain, irritable bowel syndrome and chronic fatigue [21,28].

Psychological models of MUS conditions

The role of psychological factors in the development and maintenance of MUS has been hypothesised for decades [29] and more recently has been formally recognised; predisposing factors, including prior learning and consequent behaviours relating to illness, have been proposed as basic components in the development of all chronic medically unexplained pain conditions [30]. Emotions, beliefs, attitudes, expectations, the meaning of pain to the individual and social and environmental factors can influence a person's complex perceptual experience of pain and relate to how successfully an individual can adjust to life with illness [31]. Social and psychological factors have also been specifically associated with COFP. A systematic review of 59 epidemiological studies looking at the antecedents of COFP identified that those affected by psychological factors such as stress, depression and anxiety are more likely to have symptoms associated with COFP [32] and more recently, a prospective epidemiological study showed that that subjects reporting anxiety, depression and health anxiety at baseline had a two to four fold increased risk of developing COFP at a two year follow up [28]. This preliminary work offers evidence of associations between psychological factors and COFP, however it does not provide insight into mechanisms and causal processes. An examination of other literature and theoretical perspectives in the area of MUS syndromes and the use of relevant psychological treatments could be helpful in gaining an understanding of how an effective intervention for COFP might function. However, it is important to note that MUS is a somewhat controversial term that is sometimes regarded as carrying pejorative connotations and as dichotomizing conditions as either physical or psychological. It can be argued that the term oversimplifies complex problems; however we have retained this term because the clinical conditions within this definition are well understood as they share common characteristics. That said, we do acknowledge its limitations and also note that a condition does not need to be considered medically unexplained for the need for CBT and vice versa i.e. the efficacy of CBT is not dependent on medical explanation.

Evidence supporting Cognitive Behavioural Therapy (CBT) as a treatment for COFP

A number of systematic, critical reviews and clinical trials have found that interventions based on Cognitive Behaviour Therapy (CBT) have produced some evidence of effectiveness for chronic pain patients for a number of outcomes (Dworkin et al.,1994;Hoffman et al.,2007;Morley et al.,1999;Turner et al.,2007;Williams et al.,2012) [33-37]. Evidence from other studies further supports the efficacy of CBT in the management of a variety of chronic pain disorders, including TMD [33, 35, 37, 38-43]. Additionally, a review of 31 clinical trials of treatment for somatisation and MUS syndromes found that physical symptoms improved more following a CBT based intervention than for control groups in 71% of studies [40]. Evidence suggests therefore that CBT may be an effective psychological intervention in improving outcomes for COFP.

There is specific evidence to suggest that a CBT intervention might be an effective way of managing COFP [14,15,47]. A recent Cochrane systematic review [44] found some evidence to support the use of CBT interventions for COFP. The studies included in this review were small and showed medium to high risk of bias, therefore results should be treated with caution. It is difficult to ascertain from many studies of psychological interventions, which elements are effective and the mechanisms which could bring about changes in patients tend not to be addressed. Morley et al. and Williams et al [35,37] note that the studies included in their systematic review of CBT for chronic pain in adults vary

considerably in the quality and quantity of the interventions reported, with a lack of explicit accounts of the procedures involved. CBT is a complex treatment comprising discrete components and the current literature tells us little about the specific mechanisms that bring about change in this condition particularly when it is administered in combination with other approaches like anti-depressant medication [45].

Despite the weak evidence to support the use of CBT for COFP, Aggarwal et al. [44] recommended that this type of non-invasive intervention should be considered in preference to other invasive and irreversible treatments currently used, which have limited or no efficacy. In concurrence with this conclusion, the theoretical significance of CBT models of treatment and its possible application to COFP will be considered as psychological therapies like CBT have the potential to target modifiable psychological factors that lead to the onset and persistence of COFP.

Possible mechanisms of a CBT based intervention for COFP

CBT has been effective to varying degrees in managing mental health, MUS and chronic pain conditions. The main factors that influence the experience of pain and that might be amenable to psychological therapies like CBT include memory, cognitive appraisal/hypervigilance and catastrophising. Figure 1 outlines how some of these factors can be specifically related to COFP symptoms. CBT treatment can target cycles of maladaptive cognitions (such as catastrophising) and unhelpful behaviours [46], which are considered to be maintaining features of dysfunction which perpetuate an individuals' experience of distressing symptoms. Deary et al. [47] proposes a CBT model of MUS where predisposing factors (early adverse experiences, personality, genetics and life events) can result in a general inability to tolerate distress and a high sensitisation towards physiological sensations. Consequently, subsequent experiences of physical symptoms and general distress are augmented, resulting in normal or benign bodily sensations being perceived as signs of a more serious malaise. Furthermore, these heightened negative experiences are perpetuated by a combination of harmful physiological arousal (hypocortisolism), social factors (e.g. medical uncertainty), unhelpful cognitive processes (e.g. catastrophising) and negative behaviours (avoidance of symptoms, recuperative illness response). The mechanisms of this perpetual cycle is analogised with a self-constructing and self-maintaining biological system known as autopoiesis [48]. This work has largely been conducted in relation to other MUS and further research needs to be carried out before any mechanisms specifically relating to CBT for COFP can be described. Nevertheless, this theoretical framework has been adapted to provide a basic model relating to COFP, and is illustrated in Figure 1.

In this model, a maintaining cycle, or vicious circle is experienced, where unhelpful thoughts and behaviours relating to COFP interact to perpetuate physical and psychological symptoms. Negative cognitions relating to COFP might be worries that symptoms are signs of a more serious condition and feelings of failure or helplessness as withdrawal from normal routines takes place. Unhelpful behaviours might include avoiding certain social situations, avoiding hard foods, clenching and grinding of teeth and withdrawing from work and routine activities. A CBT based intervention would aim to break this cycle by utilising techniques which target behavioural and cognitive elements. It is proposed that breaking this negative pattern will have a positive effect on the remaining areas of the model, thus symptom severity will be reduced.

Kroenke and Swindle [40] denote CBT related techniques for MUS conditions in relation to changing illness perceptions (also sometimes referred to interchangeably as illness representations, illness cognitions and illness beliefs) and adverse behaviours. Illness perceptions influence the type of related behaviours and coping strategies adopted by patients which may affect or have implications for the severity of symptoms reported.

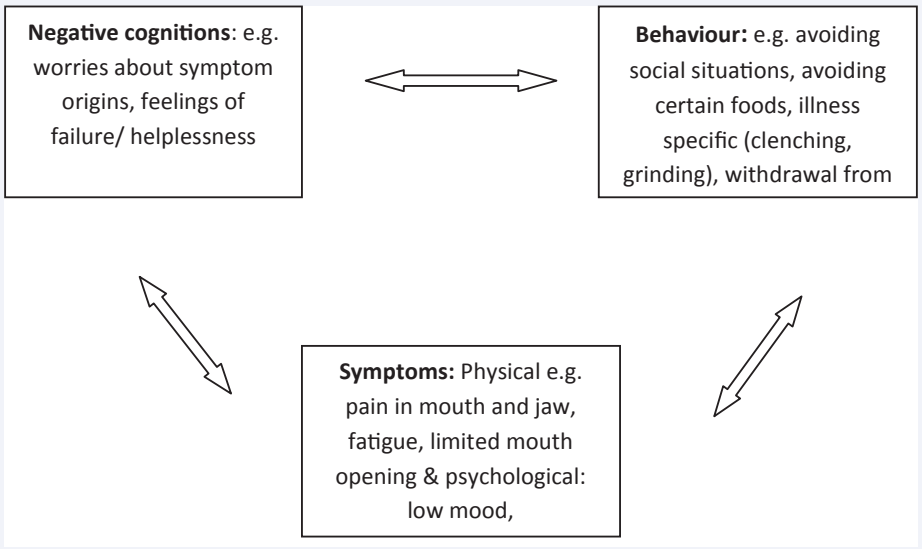

Figure 1 A cognitive behavioural model of COFP. In the model, a maintaining cycle, or vicious circle is experienced, where unhelpful thoughts and behaviours relating to COFP interact to perpetuate physical and psychological symptoms.

Behavioural techniques may relate to behavioural activation for negative symptoms (such as withdrawal from normal activity) and modification techniques for positive symptoms (such as hyperactivity during periods of feeling well). However, whilst psychological therapies like CBT address the cognitive and behavioural aspects, it is important to understand that pain inherently involves the experience of physical symptoms which for COFP have been described in figure 1 above. It is important to integrate these into biopsychosocial models that can explain to patients how physical symptoms, psychological distress and social problems may be linked via physiological mechanisms. For example, in relation to COFP, distress and anxiety can manifest as unconscious grinding or clenching of teeth leading to sustained contraction of muscles of the face. This produces pain which provokes further anxiety, which in turn produces prolonged muscle spasm at trigger points, as well as vasoconstriction, ischaemia and release of pain producing substances. This will then further reduce physical activity, and consequently muscle flexibility, tone, strength and physical endurance leading to the commonly observed physical symptoms of COFP such as limited mouth opening and the feeling that teeth are not fitting properly (figure 1). Therefore biomedical approaches targeted towards reversing teeth clenching and/or grinding need to also take into account the underlying psychosocial stressors that are perpetuating this behaviour so that both can be targeted by a biopsychosocial approach.

Implications for future research and clinical management

Although the evidence and mechanisms discussed above suggest that a CBT based intervention may benefit patients with COFP, much work is still needed in addressing several gaps in the design of such interventions.

COFP presents with varying levels of severity and patients display a spectrum of pain related behaviours. Future interventions should be tailored to the needs of patients by targeting the number and difficulty of such behaviours, perhaps by first determining specific behaviours associated with COFP. These behaviours could then be targeted, by incorporating appropriate components within an intervention. Indeed, none of the interventions in the systematic review [44] encompassed patient views. The Medical Research Council's guidance for "Developing and Evaluating Complex Interventions to Improve Health" [49] acknowledges that designing and testing time consuming interventions that are not feasible and acceptable to patients is a waste of resources.

There is emerging evidence [50] that whilst dentists are able to diagnose COFP, they are unable and unwilling to manage it. Training in psychosocial management of COFP for dentists at undergraduate and postgraduate levels is unreported within the literature and rare in UK dental schools. Therefore, the feasibility of delivering psychosocial interventions such as CBT also needs careful evaluation.

There is a need to move from the current biomedical approach, particularly in primary care where patients initially present, to a biopsychosocial approach that encompasses patient beliefs, emotions and behaviour in the management of COFP. Key challenges to make this a reality include:

- Early identification and management of COFP
- Development of evidence-based interventions that target modifiable perpetuating and exacerbating factors
- Structured evidence-based training programmes for dentists and GPs at undergraduate and postgraduate level

Perhaps we need to learn from the management of other functional pain disorders. Screening patients for targeted treatment for back pain in primary care has been shown to improve treatment efficiency using a stratified care, biopsychosocial model [51]. Using this system, patients identified as low risk using a screening tool are not over treated, whilst those at high risk are not kept from receiving more intensive or specialist pain management. Certainly, levels of physical and psychological functioning vary amongst COFP disorders. For example, some TMD patients display low levels of pain and psychosocial distress, while those with persistent idiopathic facial pain may have more severe and intractable symptoms of the same nature. A screening tool for COFP that stratifies patients according to the intensity of their symptoms has the potential to improve treatment efficiency using CBT approaches.

That said, while certain symptom clusters appear common within long-term conditions (e.g. anxiety and depression), this does not mean that all these disorders should be treated similarly or are medically unexplained. Indeed these additional symptom clusters need to be addressed and CBT could be a modality of choice. The decision to use CBT approaches clearly has to be diagnosis led. The diagnosis of chronic oro-facial pain remains firmly based on history and examination as there are currently no objective tests to confirm the diagnosis. Time is needed to take a comprehensive history including identification of potential psychosocial factors and investigation of the impact of pain on the patient's quality of life. A careful oral examination including basic cranial nerve examination is important to determine whether the pain is dental or non-dental in origin. Of the dental causes cracked tooth can be very difficult to diagnose. Trigeminal neuralgia can also easily be mistaken both by patient and dentist as toothache or chronic oro-facial pain.

It is therefore crucial to establish an early diagnosis to avoid unnecessary dental treatment, excessive use of drugs e.g. over the counter drugs, and inappropriate surgery. Unnecessary treatment can expose practitioners to litigation particularly where such treatment results in iatrogenic harm. Once a diagnosis has been established, the decision of onward referral for CBT can be made based on the psychosocial evaluation of the patient. The role of the dental practitioner is to exclude dental disease and onward referral for CBT will be through liaison with GPs and /or in association with the Improving Access to Psychological Treatment (IAPT) team specialist centres. The IAPT services have recently extended their services to provide psychological therapies for people with long-term conditions and medically unexplained symptoms. The British pain society have acknowledged that this will inevitably include chronic pain conditions and have welcomed this inclusion albeit with caution. Their specific cautionary notes apply to our earlier discussion that a condition need not be medically unexplained to receive a psychological therapy and

vice versa. They strongly suggest that chronic pain management needs a biopsychosocial approach and whilst psychological therapies are important they need to be offered in conjunction with appropriate medical and physical therapies. Indeed at an international level, this approach is also being recognised in the re-classification of ICD-11 and DSM V where diagnoses are being related to their specialist areas with a wider recognition for the role of psychological factors and according to Goldberg [52], this reclassification will encourage general practitioners and hospital specialists to assess anxious and depressive symptoms whenever they are faced with unexplained somatic symptoms or other psychological symptoms. Therefore clinicians would recognise somatic symptoms in a depressed patient and provide an explanation for the physical symptoms. Similarly, clinicians diagnosing unexplained or somatic symptoms would also inquire into psychosocial co-morbidities. It remains to be seen how these approaches will move forward in clinical settings but it certainly provides hope that integrated biopsychosocial management of chronic oro-facial pain could become a reality in the next 5-10 years.

CONCLUSION

COFP can be positioned within a realm of Medically Unexplained Symptoms as the MUS of dentistry. Research has suggested that psychological factors play a part in the generation and maintenance of MUS conditions, including COFP. However COFP is often treated mechanistically particularly in primary care where it is managed according to a biomedical model. Evidence from studies of COFP and other conditions associated with medically unexplained symptoms suggests that CBT may be effective in targeting maintaining features and improving outcomes for patients, and relevant theory concurs with this evidence, offering a model for bringing about change. CBT can be a very useful coping strategy for patients who present with chronic oro-facial pain. Whilst CBT may not be curative it can certainly help patients cope with their pain. It can be offered in addition to any other forms of therapy that patients receive and such an integrated biopsychosocial approach should certainly be considered early in primary care settings where it could have a positive impact for patients with chronic oro-facial pain in the future. However, although there is plausible evidence to support a CBT based intervention to treat COFP, we have identified several limitations of current CBT based treatments. There is therefore a need to design an intervention for COFP that is feasible and acceptable to patients and that can intervene at varying levels of severity and pain related behaviours that present amongst patients with COFP. Medical research council guidance [49] provides an explicit framework from which to design such interventions and early management in primary care using such interventions should be a priority for future testing.

FUNDING

This work was funded by a Clinician Scientist award held by Vishal Aggarwal and issued by the National Institute of Health Research – grant number CS/2008/08/001. The views expressed in this publication are those of the authors and not necessarily those of the NHS, the National Institute for Health Research or the Department of Health. There are no conflicts of interest to declare.

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Cite this article

Goldthorpe J, Lovell K, Peters S, McGowan L, Aggarwal VR (2016) Management of Medically Unexplained Chronic Orofacial Pain: Justification for Behavioural Interventions. J Sleep Med Disord 3(1): 1038.