HHS Public Access

Author manuscript

J Oral Rehabil. Author manuscript; available in PMC 2016 December 01.

Published in final edited form as:

J Oral Rehabil. 2015 December ; 42(12): 942–955. doi:10.1111/joor.12329.

Perspectives on Next Steps in Classification of Orofacial Pain – Part 2: Role of psychosocial factors

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Abstract

This paper was initiated by a symposium, in which the present authors contributed, organised by the International RDC/TMD Consortium Network in March 2013. The purpose of the paper is to review the status of biobehavioural research – both quantitative and qualitative – related to orofacial pain with respect to the etiology, pathophysiology, diagnosis and management of orofacial pain conditions, and how this information can optimally be used for developing a structured orofacial pain classification system for research. In particular, we address: representation of psychosocial entities in classification systems, use of qualitative research to identify and understand the full scope of psychosocial entities and their interaction, and the usage of classification system for guiding treatment. We then provide recommendations for addressing these problems, including how ontological principles can inform this process.

Keywords

Biopsychosocial; classification; orofacial pain; ontology; TMD

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The authors declare that they have no conflicts of interest

1 Introduction

With the notable exception of temporomandibular disorders (TMD), psychosocial factors have received scant attention from most research devoted to orofacial pain (OFP) conditions (1). The persistent absence of psychosocial factors in OFP research seemingly conveys the implicit message that they are unimportant (in comparison to nociceptive processes). However, throughout this paper we will highlight and explain the impact of psychosocial factors and the influence they exert on how disease courses in OFP evolve. Related to this is our relative ignorance of what constitutes the phenotype of a pain condition. The Orofacial Pain: Prospective Evaluation and Risk Assessment study (OPPERA) has provided an immense amount of data describing the phenotype associated with TMD (for summaries, see (2, 3)), but other OFP conditions are less well characterized.

The characterization of OFP conditions must necessarily include attention to psychosocial factors and, despite the level of data provided by OPPERA regarding TMD, the question remains: what level of (psychosocial) phenotypic data should be collected for the characterization of other OFP conditions? Future research identifying the phenotype of other OFP conditions could progress along the same, exhaustive, lines as OPPERA or it could progress perhaps more pragmatically and efficiently by qualitatively examining the patients' experiences caused by the disorder underlying their pain condition, thereby allowing us to develop a more complete set of hypotheses regarding the composition of such phenotypes, and the nature of the disorders leading to these phenotypes. This approach may help identify specific constructs (or variables) of interest, tie these constructs to anatomical and physiological entities, reduce redundant data collection, and explain 'anomalies' within accepted classifications.

The authors of this paper were invited by the International RDC/TMD Consortium Network for a symposium, held at the 2013 IADR General Session in Seattle, in order to review the state of the art regarding the formal development of diagnostic criteria for OFP conditions. The ultimate aim of this examination was to critically appraise the arguments for and against the development of specific Research Diagnostic Criteria for Orofacial Pain (RDC/OFP). The domains and perspectives represented by the authors are: OFP conditions, qualitative research, behavioural medicine, medical classification and related statistical methods, and realism-based ontology . Questions for which answers were sought in this paper included:

1) Are psychosocial constructs identified for TMD useful for orofacial pain?

2) Can qualitative research methods provide significant insights into OFP which might affect how we identify entities as well as axes or dimensions for diagnostic criteria?

3) Are traditional (conservative) approaches to medical classification sufficient for new diagnostic criteria or do they need to be complemented by recent developments in the application of ontological realism?

The other parts of this series of papers cover ontological realism in depth (4) and biomarkers' role in an emerging classification system (5). This paper focuses on questions one and two above and relates its findings in relation to questions one and two to question

three, but an in-depth exploration and explanation of ontology's role can be found in part one of this series.

2 The biopsychosocial model of pain

The biopsychosocial model proposed by Engel (6, 7) has been applied to most types of chronic pain with parallel improvements in its understanding and management (8-11). Alongside the application of this model there have been important advances in our understanding of the biological mechanisms behind chronic pain (12-16). It has been demonstrated that chronic pain involves multiple systems in addition to the nociceptive system. It is this fact that makes classification of OFP conditions so challenging.

Pain conditions undoubtedly vary in the extent to which psychosocial characteristics drive the presenting symptom pattern. Drossman (17) suggests a two-dimensional plot on which various health conditions, including pain conditions, can be mapped (Figure 1). The axes (dimensions) for this plot are disease (biological factors) and illness (psychosocial factors). Conditions can, therefore, range from those with a large role for illness factors, for example chronic abdominal pain, to those with a large role for disease factors, for example an asymptomatic ulcer. Clearly, some conditions can then, therefore, lie in between disease and illness in terms of the factors driving the patient's presentation, and it is, of course, this area bridging both illness and disease that accounts for the immense difficulties in the consultation room. Figure 1 demonstrates one perspective of where OFP conditions such as myofascial TMDs or neuropathic orofacial pain conditions may be positioned in this two-dimensional space. The suggested locations of myofascial TMDs and neuropathic orofacial pain conditions in the plot are not intended to represent a firm consensus in the field. They are also not intended to precisely depict the relative impact versus cause of psychosocial factors, but rather hypothesized relative placements about the role of illness versus disease factors. Indeed, within a certain group of disorders it is likely that specific disorders might ultimately occupy different locations in two-dimensional space. For example, a myofascial TMD *with* painful disc interference would likely occupy a location different from that currently depicted for “myofascial TMD”.

Despite the preliminary mappings in Drossman's two-dimensional biopsychosocial space, it is important to remember that the plotted points represent the likely role of biological and psychosocial characteristics for the average person with a given condition. Psychosocial characteristics themselves may drive care-seeking for an array of conditions (18) to varying degrees. It is therefore important to understand this, the individual patient, and the factors that may affect his or her prognosis for a given condition, rather than only the characteristics of the hypothetical ‘average’ patient. The assessment and understanding of the individual patient with orofacial pain therefore requires the inclusion of psychosocial information. The ‘average’ patient profile only provides an indication of whether a patient is statistically likely to be at risk of psychosocial burden.

2.1 Should an orofacial pain classification be based on the biopsychosocial model of pain?

It is the inclusion of the biopsychosocial model that is perhaps what most distinguishes the RDC/TMD from other classification systems of pain in general. The RDC/TMD explicitly includes separate assessment and classification for psychosocial functioning on a psychosocial “Axis II,” orthogonal to signs and symptoms that lead to primary physical disorder classification in exactly the same manner as Drossman proposes. Should, therefore, an expanded orofacial pain classification system formally include a similar psychosocial assessment intended for all patients with such complaints? The short answer is: Yes. The longer answer involves an understanding of the evolving view of TMDs and the biopsychosocial model.

The use of a dual axis system was included in the RDC/TMD because of awareness of the biopsychosocial nature of virtually every chronic pain condition, including but not limited to TMDs. Axis II instruments were selected to screen patients for psychological status (depression and nonspecific physical symptoms) and to classify patients into a “chronic pain grade” based on characteristic pain and activity interference levels from the Graded Chronic Pain Scale (GCPS) (19, 20). These Axis II measures were intended to serve as screening instruments for the constructs of depression, somatic symptoms, and disability, given their relevance as risk factors for poor clinical outcomes as based on the data available at that time. By identifying patients at risk of poor outcome due to impaired psychosocial functioning, the intent of such screening was that these individuals could be referred for psychological assessment and interventions. Data since the publication of the RDC/TMD have only reinforced the significance of psychosocial factors in treatment response (21-23) at least with respect to improved psychosocial function. Tailoring treatment to patients with compromised psychosocial adaptation has been increasingly shown to be effective (22). Ongoing development of the RD/TMD has resulted in the recent publication of the DC/TMD (24), which maintains the dual axis structure and focus of the parent criteria, but attempts to improve validity and reliability of Axis I by building on the RDC/TMD validation project data (25-30). The DC/TMD has also revised the Axis II to allow a shortened version for everyday clinical practice and full details can be found at the RDC/TMD Consortium's website (http://www.rdctmdinternational.org/TMDAssessmentDiagnosis/DCTMD.aspx).

Documentation of psychosocial dysfunction in orofacial pain conditions other than TMDs has been much less extensive. Does that mean that psychosocial factors are not emerging from the data as prognostic factors for other orofacial pain conditions, or does that mean that psychosocial factors have yet to be systematically evaluated? The biopsychosocial model of pain remains the primary justification for the assessment of psychosocial factors in all orofacial pain conditions and pain conditions in general. What then has been the frequency with which Axis II psychosocial assessments have appeared in the research literature? Limiting our review to English-language research articles retrievable through PubMed including abstract terms of temporomandibular joint disorder or temporomandibular pain and dysfunction syndromes or TMD, and abstract words of patient(s) and RDC, we recently found the following: in 2012 alone, 43 clinical research articles met search criteria by

reporting results of RDC/TMD Axis I to identify and characterize patients, but only 20 of these reported some aspects of RDC/TMD Axis II assessment (31). It might be argued that some authors assessed RDC/TMD Axis II but did not report results in a particular manuscript. If this is the case we would suggest that this leaves the reader unable to assess the effect of psychosocial heterogeneity within the sample on the outcome of an intervention. This may be one of the reasons why outcomes for similar interventions differ markedly, even when Axis I physical diagnoses are identical between studies.

Nearly a decade ago, an extensive review (32) documented the comorbidity between depression and pain, noting their shared biological pathways. Moreover, the review documented that, for a variety of pain conditions, comorbid depression was associated with long-term negative outcomes including functional disability, chronicity, greater use of health care resources, and poorer adherence to prescribed treatment modalities. Consequently, the focus on psychosocial factors in pain patients has been considered primarily from a ‘yellow flag’, or risk, perspective in which psychosocial factors affect prognosis (e.g., (33)). As psychosocial pain research continues to be successful in identifying constructs that affect pain experience as well as developing better measures of those constructs, pain treatment should benefit from the inclusion of therapies that strive to target those constructs. Psychosocial pain treatment overall is, however, at a standstill, given the number of identified relevant constructs versus treatments with demonstrated efficacy. We believe that this represents an opportunity to more carefully evaluate how these constructs should be considered within the context of classification, in order for the disorders to be better conceptualized with respect to treatment models.

Less empirical emphasis has been paid to psychosocial factors as treatment effect modifiers or moderators, or as treatment mediators. Psychosocial factors as treatment effect moderators would be indicated if a particular treatment had a differential effect in the presence of specific psychosocial risk factors. A few notable constructs – fear–avoidance, depression, anxiety, and pain-catastrophizing – exhibit strong models that explain behavioral or CNS mechanisms linking the identified psychosocial construct with pain intensity or persistence. The empirical support for the success, in terms of pain response, of an intervention specific to the respective identified aforementioned constructs is not strong, however, because psychosocial treatments are generally provided as a “package” with multiple potential modes of action. Aside from a few exceptions (34-36), psychosocial factors as treatment effect moderators in pain interventions have been under-examined. Moreover, many studies are often underpowered to detect moderator effects. To the extent that psychosocial factors are or can be identified as moderators, they may help clinicians to answer the question of “what psychosocial intervention works best for whom?” (37). To our knowledge, only one randomized controlled clinical trial (38) has shown that more intensive psychosocial interventions can benefit psychologically high risk patients with musculoskeletal pain, while simpler interventions may benefit lower risk patients. Despite the persuasive hypothesis that matching treatments to patient's biopsychosocial characteristics should improve outcome (39) and efficient care, a recent meta-analytic review (35) concludes that evidence of moderator effects and benefit of targeting treatments in chronic pain patients is currently not strong. On the other hand and from a different perspective, studies specifically examining TMD patients (22) suggest that simple self-care

strategies may be sufficient for psychosocially functional patients, but that impaired patients benefit from more comprehensive treatment including cognitive behavior therapy.

3 Qualitative research is a useful tool to identify psychosocial entities to be represented in future orofacial pain classifications

3.1 Characteristics of qualitative research

Qualitative research (QR) seeks not to enumerate, but to interpret, and build understanding of naturally occurring phenomena from the perspective of the participants (40, 41). To give an example, one might examine the “objective” pain levels of patients undergoing neurosurgery for trigeminal neuralgia (quantitative), but one might also be interested in the factors that drive their desire to undergo such a procedure and their experiences pre- and post-operatively (qualitative). QR can be used in a standalone study, or in combination with quantitative research (42), for example to define the new items required for a new health status measure (an example in orofacial pain is provided by Durham et al (43)), or to help explain unusual or complex results from a quantitative survey.

Data are usually collected in QR by the use of any of four main methods: interviews, focus groups, observation, or documentary analysis (41, 44-46). In-depth summaries of the differences between qualitative methods and their differences to quantitative research methods are available (40, 41).

Subject samples in qualitative research tend to be small, non-probabilistic, and purposive aimed at identifying a depth and breadth of opinion (47). If the study is of a responsive design the sample will also evolve in order to identify any groups of individuals who might give disconfirming evidence of any theory that has been generated.

The natural occurring data “unit” of qualitative research is text. This text can be generated from observations, interviews or focus groups, or it can be directly analyzed from patient submissions (47). The text collected is subject to a coding process (48, 49) whereby the researcher systematically analyses the text at several different levels for recurring themes, or experiences, expressed by participants. From this the data can start to be organized into recurring units and (nascent) theoretical constructs and explanations can be developed in line with the applicable typologies, theoretical paradigms, and philosophical assumptions (50). Wolf et al give a worked example of one approach to coding relevant to chronic orofacial pain (51, 52).

The validity of QR has often been a source of concern for quantitative trained clinical researchers, but there are several simple strategies to help ensure the validity of the data and the theory generated in a qualitative study. These strategies include: independent assessment of transcripts (dual coding by independent researchers); data-rich papers or appendices so that readers are able to read in full relevant portions of the data collected; and triangulation of data/theory by other methods (53-56).

3.2 Assessment of psychosocial factors in orofacial pain using qualitative research

There has been a slowly increasing awareness of the benefits that qualitative research can bring to the examination and understanding of the psychosocial dimension of chronic pain (57, 58).

A brief literature search using PubMed and Web of Science was undertaken in February 2013 to identify relevant papers in the field from 1950 onwards. Table 1 demonstrates the number of hits and the search terms employed. The *a priori* exclusion criteria for a paper were: 1) the paper was not in the English language; 2) the paper did not primarily focus on a named orofacial pain condition; or 3) the paper focused on professional perceptions rather than patient perceptions.

Abstracts were read for all 165 identified papers, of which 147 were original research papers. Of these 147, only 15 had an orofacial pain condition as the primary focus of the study (43, 51, 52, 59-70). Of these 15 papers, 2 involved some element of professional perceptions (61, 66), and the remaining 13 papers provide us with some data (43, 51, 52, 59, 60, 62-65, 67-70).

When the 13 papers were read carefully it was apparent that some authors had reported linked data in two papers (43, 51, 52, 62, 64, 65), with only 10 original qualitative reports available regarding orofacial pain conditions: toothache, temporomandibular disorders (TMDs), persistent dentoalveolar pain, and nonspecific chronic orofacial pain. Table 2 summarizes the studies' characteristics and Table 3 outlines their quality assessment according to Popay et al and the qualitative research appraisal tool (55, 71). Interestingly, contrary to a recent report about the general state of qualitative research in dentistry (72) the quality assessment was reasonably high for papers specific to an orofacial pain condition. This finding should, however, be interpreted with caution as the assessment was performed by only one researcher.

All 13 papers were read, and common and recurring themes or experiences were extracted from them. This was done across all papers irrespective of pain condition and also by condition for TMDs and "toothache". It should be noted at this point that this process was for descriptive purposes only and cannot be considered to be a qualitative metasynthesis for two critical reasons: the methodologies vary between studies, and the data are too sparse and divergent to allow this at present. We wish here to only illustrate the value of the process with respect to the goals of this paper, which is to provide a rich description of orofacial pain in order to better serve purposes of developing classification.

Temporomandibular disorders clearly exemplify the difficulties of radiating pain, which tends to pervade into everyday activities (43, 62, 65, 67). The psychosocial impacts reported across those studies examining TMDs included: embarrassment when eating outside of the home reportedly because of clicking and locking and changes to dietary intake/consistency (43, 67); generalized reduced functional ability (43, 62, 67); relationship effects reportedly mediated both by mood changes and by decreased willingness to be intimate (43, 65, 67); negative mood changes seemingly because of the persistency of the problem and lack of diagnosis (43, 65, 67, 70); reduction in ability to perform in normal employment/school (43,

65, 67); and a degree of helplessness implicitly caused by the unremitting nature of the complaint (43, 65, 67). Another key defining feature reported by some of the studies in TMD was the uncertainty individuals faced when looking for a diagnosis and the lack of legitimacy they often felt in their care-seeking (43, 62, 65). This uncertainty then had consequences for how individuals felt they were supported by close friends and relatives. These feelings around uncertainty and legitimacy were mirrored in the papers examining "non-specific chronic orofacial pain" as were the extensive day-today limitations in activity (51, 52).

"Toothache" seemed to be reported as an acute, and easily conceptualized, problem in the studies examining it (59, 60, 68, 69). "Toothache" could present suddenly as an intense pain, or build gradually to a peak of intensity, but in either case a level of "unbearable" pain was often the trigger to seek care (59, 60, 68, 69). Psychosocial impacts of "toothache" were extremely varied, but those that recurred included: general activity limitation including decreased work productivity (59, 60, 68, 69), sleep disruption (59, 60, 69), and changes in dietary intake which were either abstinence from eating, or changes in consistency of intake (59, 60).

Examining across all orofacial pain conditions it would appear, based on the qualitative research, that there are some common biopsychosocial impacts to all orofacial pain conditions:

- Perceived sleep disturbance
- Activity limitation – social and work
- Changes in dietary choice and consistency
- Distress
- Decreased self-efficacy

Qualitative research in chronic orofacial pain has also led to further developments in understanding chronic orofacial pain including the possibility of a liminal state between health and illness in chronic orofacial pain (62) and its consequences. Qualitative research techniques have also allowed the development of putative screening instruments for the more rare conditions such as Persistent DentoAlveolar Pain disorder (PDAP) (63) grounded in the patients' experiences.

Unsurprisingly this review appears to demonstrate a large number of psychosocial impacts in the chronic conditions as compared to the acute and "curable" toothache conditions. There are also clear implications for the sociology of chronic orofacial pain to be examined in more depth given the expressed problems in obtaining legitimacy and thereby receiving social support for the individual's complaint. More research is required to assess the effect of psychosocial factors as treatment moderators or mediators for orofacial pain (73) (p.423). Qualitative research, if appropriately performed, clearly has potential to not only elucidate factors that play a role in outcome, but also explain how they may do so.

4 Phenotype determination using the Ontology of General Medical Science

4.1 Disorders, diseases, and disease courses

The Ontology of General Medical Science (OGMS) is based on a terminological framework that encompasses diseases, their causes and manifestations, and diagnostic acts and other entities pertaining to the ways diseases are recognized and interpreted in the clinic. The framework was designed to avoid the common problem of entities (for example disease) and evidence for the existence of entities becoming inextricably joined (conflation of entity and evidence for entity) and mutually exclusive so that one wrongly would assume that the disease does not exist unless a particular sign or symptom (evidence) exists (74). Clearly, however, the disease can exist irrespective of whether the sign or symptom is present, and the opposite is also true: the sign or symptom may be present but the disease that the clinician has in mind may not be in existence in the individual with the sign or symptom.

The basic axiom of the OGMS is that every disease rests always on some (perhaps as yet unknown) physical basis. When, for example, there is in a specific patient an elevated level of TNF in the synovial fluid of the TMJ, then this is because some physical structure or substance in the organism is disordered, for instance physical damage of some sort in the TMJ. It is this physically damaged, ‘abnormal’, structure or substance that is known in realist ontology jargon as the ‘*disorder*’. This use of the term ‘disorder’ is thus narrower than the loose manner in which the term is used in medical jargon where typically no systematic distinction is made between ‘disorder’ and ‘disease’.

OGMS states that when such a disorder exists in the organism (human body in this case) then there is a second entity present known as *disposition*. Dispositions are just like, for example, functions, tendencies and propensities and are therefore special types of realizable entities. The use of the term ‘realizable’ in relation to the entity refers to the fact that there must be certain circumstances for a disposition to be realized. What, for instance, we would call “pain on palpation of the Temporomandibular joint (TMJ)”, is the realization of such a disposition, namely of the disposition to report pain when palpating the TMJ: the patient will only report pain on palpation when brought under suitable conditions such as sufficient presence of TNF (75). A patient without such levels of TNF would *still* have the disposition, but not the realization thereof. Similarly, if there is in some body part or organism a disorder, then there is in that organism also the disposition for the organism to act or undergo processes in a certain abnormal way. It is this disposition that in realist ontology jargon is called the *disease*. For OGMS, disease and disorder are thus two distinct entities, but tied together like the two sides of a coin: one cannot exist without the other. In case of TMD, the disorder might be, for instance, a displaced disk or arthrosis in the TMJ, and the corresponding disease then the disposition for pathological processes leading to clicking, pain, limited mobility, etc. From the point of view of OGMS, a term like ‘TMD’ is thus ambiguous as OGMS recognizes Temporomandibular Disorder and Temporomandibular Disease as distinct entities.

It is only when the disease leads to pathological processes (e.g. inflammation) that then a third entity comes into existence: the *disease course* which is formed by all processes of involved body parts, including the entire organism, which realize the disposition. The

disease course includes manifestations that can be recognized as symptoms and signs of the disorder (e.g. pain, crepitus, decreased mobility) or through measurement assays (e.g. laboratory tests, imaging procedures).

4.2 Disease courses and illnesses

The OGMS view remains valid for the patient with mental and psychological issues as exemplified in the Ontology of Mental Disease (76). As an example, certain forms of depression are characterized by morphologic configurations in specific brain regions that differ from the configurations exhibited by healthy individuals such as, for instance, abnormal configurations in or of the serotonin receptors. These configurations thus constitute the disorder in OGMS sense. With this disorder at the level of serotonin receptors then comes the particular disposition for the individual to act in a certain manner commonly recognized as depressive mood and vegetative symptoms. It is this disposition that constitutes the disease that we call ‘depression’. This disposition may then become realized in pathological processes of various sorts for example, disturbed sleeping or altered behavior characterized by a decrease in certain activities. Other pathological processes are those which are part of the disease course which together constitute the more complex process described as ‘distress’ or ‘mental suffering’.

4.3 Towards a phenotype for orofacial pain conditions

OGMS offers three classes which are useful to be included – and further to be subtyped – in an ontology-based classification system for orofacial pain conditions:

- **Phenotype –** A (combination of) bodily feature(s) of an organism determined by the interaction of its genetic make-up and environment.
- **Clinical Phenotype –** *A clinically abnormal phenotype.*
- **Disease Phenotype –** A clinical phenotype that is characteristic of a single disease

Entities that qualify as bodily features are: (1) physical components such as bodily components (e.g., nerve cells, nociceptors, neurotransmitters) and external components (e.g., pathogens, toxins, microbiome); (2) bodily qualities such as cytokine concentrations; (3) bodily processes in which physical components participate, irrespective of them being normal (e.g., neurotransmission and concordant pain sensation), pathological (e.g., phantom pain), or induced through interventions (e.g. hyperesthesia).

Examples of bodily processes that qualify as clinical phenotypes are the aforementioned disturbed sleeping and distress in the meaning of mental suffering. Sleeping and brain processes such as thinking and decision-making are phenotypes determined by the interaction of our genetic make-up and environment. In contrast disturbed sleeping and distress are clinically abnormal in the sense that they are: (a) not part of the life plan for an organism of the relevant type (unlike pregnancy or menopause), and (b) causally linked to an elevated risk of pain, of other feelings of illness, or of death or dysfunction, such that the elevated risk exceeds a certain threshold level (74). Disturbed sleeping and distress therefore qualify further as *clinical phenotypes*.

A phenotype, either disease or clinical phenotype, can exist without being observed. With the advance of technology, the ability to detect more underlying components will expand. The clinical phenotype – for a specific patient – incorporates the abnormal phenotypes realized at each stage of the disease course. A disease phenotype may be a single type of abnormality characteristic of a given disease; or it may be a combination of several manifestations of a disease and clinically normal physical components, ordered in a temporal sequence characteristic of one or more typical disease courses for the given disease.

OGMS has been used as a foundation for a series of domain-specific ontologies three of which will be described in order to provide initial evidence that using such classifications for unproven domains such as orofacial pain may contribute to better understanding of orofacial pain and, in particular, the psychosocial domain. The ***Neurological Disease Ontology*** (ND) is an extension of OGMS that provides a set of classes to represent neurological diseases along with their associated signs and symptoms, assessments, diagnoses, and interventions encountered in the course of clinical practice and research (77). Initial work on ND was focused on the areas of dementia and Alzheimer's disease, multiple sclerosis, and stroke and cerebrovascular disease.

The ***Ontology of Adverse Events*** (OAE) was developed to standardize and integrate data relating to adverse events arising subsequent to medical interventions, as well as to support computer-assisted reasoning. OAE has over 3,000 terms classified in terms of OGMS, the term ‘adverse event’ thereby denoting a pathological bodily process in a patient that occurs after a medical intervention. OAE covers adverse events based on anatomic regions and clinical outcomes, including symptoms, signs, and abnormal processes. It has been used in the analysis of several different sorts of vaccine and drug adverse event data, for example, to analyze vaccine adverse events associated with the administrations of different types of influenza vaccines and to represent and classify the vaccine adverse events cited in package inserts of FDA-licensed human vaccines in the USA (78).

The ***Infectious Disease Ontology*** (IDO) consists of a core ontology (IDO Core) covering terms and relations generally relevant to the infectious disease domain, and a set of disease- or pathogen-specific ontologies developed as extensions from the core. The core IDO imports terms such as “disease”, “disorder”, “disease course”, and “treatment” from OGMS, and provides infectious disease-specific terms such as “pathogen”, “vector”, “herd immunity”, “fomite”, “virulence”, “focal infection”, “carrier”, “seroprevalence”, “epidemic”, and “antibiogram” (79).

The examples provided demonstrate that the OGMS is well accepted in a variety of biomedical domains. Within the domain of pain, OGMS was first used to give an ontologically adequate framework of pain and of other pain-related phenomena (80), building on the definition of pain provided by the International Association for the Study of Pain (IASP). This framework was then used to develop an ontology-based taxonomy for disorders that manifest themselves through the symptom of chronic orofacial pain and are commonly seen in clinical practice and difficult to manage (81). The diagnostic criteria

proposed using this methodology were then used to conduct a systematic review to identify reliable somatosensory evaluation methods for atypical odontalgia patients (82).

OGMS thus offers an ideal framework to categorize the various phenotypes associated with differing pain conditions in function of bodily features and their relationship to known disease types. Whether the use of such ontologies can improve prognosis or better tailor treatment – which would be an ultimate goal in terms of utility – cannot be determined until better classification has been developed and then tested.

5 Recommendations

There is no doubt that future orofacial pain classifications need to include representations for various types of psychosocial entities. Such entities have been demonstrated to play various important roles in the prognosis of pain conditions in general and temporomandibular disorders in particular; moreover, psychosocial entities are core components of emerging concepts of chronicity (83). However, such entities are not at present systematically included in case reports and research studies of other types of orofacial pain. It is here that qualitative research can be used in a systematic manner to help identify and explore any new psychosocial factors and or phenotypes that have yet to be identified and explored in orofacial pain conditions. These may then go on to be represented in future orofacial pain classifications using the framework offered by the Ontology of General Medical Science is a guide for achieving consistency and coherence.

There are some limited qualitative data available from which it is possible to begin to build an understanding of some of the impacts of orofacial pain conditions – to understand the lived meaningfulness of the psychosocial entities and why they matter if our classification system is to truly capture the depth and breadth of the pain experience. Despite a slight increase in the publication of qualitative studies in the last decade, such studies still tend to be sporadic and somewhat uncoordinated in their approach to exploring the biopsychosocial complexities of orofacial pain. Perhaps given recent endorsement from major funding bodies (84) this will change, but a more coordinated and targeted examination of the conditions comprising orofacial pain is urgently needed. This will ensure that advances in patient management remain grounded in the patients' expectations and address the problems that they are experiencing (85). It will also aid the incorporation of the full scope of the respective psychosocial entities identified to be adequately and comprehensibly represented in a classification system

Simultaneous with incorporation of psychosocial entities into a developing classification system is the application of some of those entities in the clinic setting. If psychosocial assessment is not considered a key characteristic in which TMD patients in research studies are described, how likely is it that such characteristics will be assessed in studies on other orofacial pain patients? How much less likely is it that psychosocial characteristics will be routinely assessed in clinical practice of TMDs or other orofacial pain conditions? This paper intends to rectify the lack of attention to psychosocial factors: routine psychosocial assessment of orofacial patients is an inherent part of a biopsychosocial model of care. Given the potential utility of psychosocial factors for reconciling disparate research findings

and the potential of psychosocial factors to guide proper treatment for individual patients and predict prognosis, we suggest that a good argument is needed to *not* screen for psychosocial factors in orofacial pain patients. For the orofacial pain clinician, the primary role of assessment for psychosocial factors is likely, therefore, to involve their role in predicting patient prognosis and, potentially, the need to refer the patient for specialty psychological or psychiatric care to treat comorbid psychosocial problems. An ontological realism-based taxonomy places the elements that need to be addressed in such an assessment in perspective, and an ontological realism-based taxonomy reduces thereby the possibility for incomplete documentation or misinterpretation thereof afterwards.

In summary therefore the recommendations of this review are:

1. Use qualitative research to systematically identify and explore any new psychosocial factors and or phenotypes in orofacial pain.
2. Take a coordinated and targeted approach to future qualitative research in orofacial pain in order to examine the biopsychosocial impact of orofacial pain conditions.
3. Adopt the framework offered by the Ontology of General Medical Science to build future orofacial pain classifications, using data gathered from point 1 above and other sources as appropriate, that intrinsically include psychosocial factors as one necessary part of how pain would be classified.
4. Apply the knowledge gained through research into psychosocial phenotypes into everyday clinical practice both through routine screening for psychosocial comorbidities and appropriate (liaison for) management of these comorbidities.

Acknowledgements

The work described is funded in part by grant 1R01-DE021917-01A1 (W. Ceusters and R. Ohrbach) from the National Institute of Dental and Craniofacial Research.

J Durham is funded by an NIHR Clinician Scientist award NIHR-CS-011-003.

The authors would like to acknowledge and thank Dr Catherine Exley for her helpful critique of an earlier version of parts of this paper as well as Prof. Yair Sharav for his stimulating and insightful discussions.

The views expressed in this publication are those of the authors and not necessarily those of the NHS, the National Institute for Health Research, the Department of Health in the United Kingdom, or the National Institute of Dental and Craniofacial Research in the USA.

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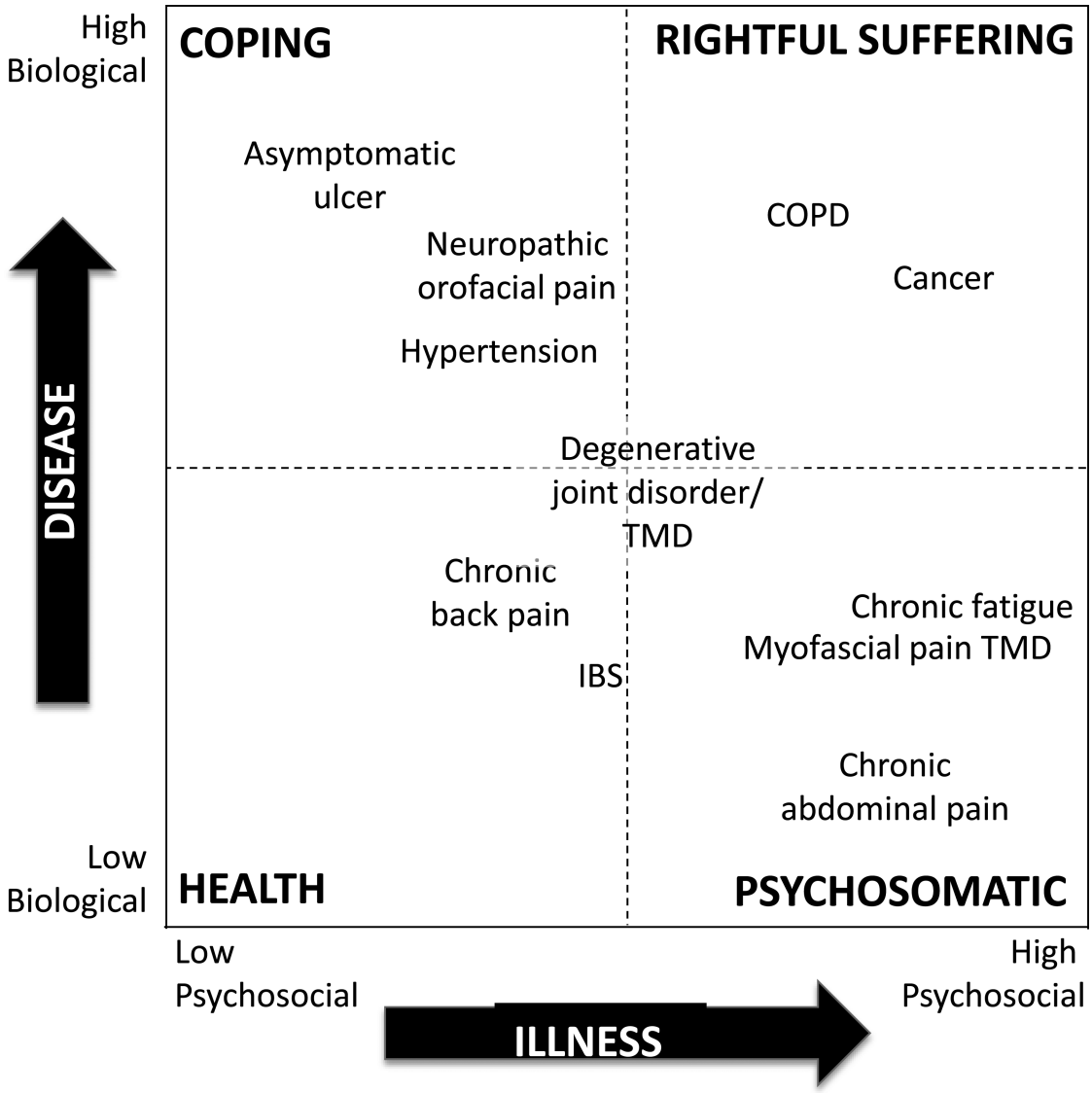

Figure 1. Illness versus disease. Adapted from Drossman (1998)

Traditional bi-axial depiction of Disease vs Illness using estimated typical placement of representative conditions. Adapted from Drossman, 1998.

Table 1

Search strategy and numbers of papers identified

Qualitative					
AND	Dental	Dentistry	Oral	Orofacial	Facial
	1967	1236	3153	26	373
AND pain	11	12	126	1	15

Table 2

Characteristics of thirteen papers included in the review

Citation	Summary of area studied	Location	Sample	Data collection and analysis
(59)	Patients' attending dental emergency clinics perceptions of "toothache"	Dental emergency clinics in two hospitals on weekends, Wales	44 pts (18F;26M). Mean age: 35.3	Interviews. Framework
(60)	Low income persons with toothache	Community, USA	66 participants in 8 focus groups (44F;22M) Age range: <25>65	Focus groups. Analysis described resembles thematic analysis. Used Nvivo.
(62)	Uncertainty in TMDs	Secondary care, England	19 patients (14F;5M). Age range: 18-60	Interviews. Constant comparative method and framework
(43)	Care pathway in TMDs	Secondary care, England	29 patients (23F;6M). Age range: 18-65	Interviews. Constant comparative method and framework
(63)	Perceptions of PDAP	Secondary care, USA	20 patients (15F;5M). Age range: 41-72	Interviews. Constant comparative method and framework
(64)	Experiences of TMDs as a chronic illness	Secondary care, USA	32 individuals from "TMJ" support groups [TMD sufferers] (27F;5M). Age range: 23-69	Interviews. Content analysis
(65)				
(67)	Adolescents' experiences of living with TMD pain	Secondary care, Sweden	21 patients (19F; 2M) Age range: 15-19	Interviews. Content analysis
(68)	Dental emergency patients with toothache	Secondary care, England	35 patients (21F; 14M). Age range: 18-40+	Interviews. Thematic analysis using framework to organise data.
(69)	Coping with tooth pain	Rural dental clinic, USA	50 patients (32F; 18M); Age range: 19-77	Interviews. Constant comparative method using Hyper-research to organise data
(70)	Female's experiences of shamanic healing for TMDs	Primary care, USA	All Female Age range: Unavailable in paper and requested form authors but no response	Interviews. Analysis described resembles thematic analysis. Used ATLAS.ti
(51)	Chronic orofacial pain patients' experiences of consultations	Secondary care orofacial pain clinic, Sweden	14 patients (11F; 3M); Age range: 21-77.	Interviews. Phenomenological approach to analysis identifying meaning units and subsequent to this cores of significance from transcripts
(52)	Chronic orofacial pain patients' experiences of their pain			

J Oral Rehabil. Author manuscript; available in PMC 2016 December 01.

Table 3

Quality assessment of papers included in metasynthesis

Citation	Quality criterion from Popay et al 1998 unless otherwise stated							
	Clear statement of aim (CASP*)	**Qualitative method appropriate (CASP*)**	**Responsive design**	**Theoretical or purposive sampling**	**Adequate description through data of subject/ phenomenon studied**	**Transparency of data collection and analysis**	**Reflexivity of researcher and participant considered (CASP*)**	**Evidence of higher levels of analysis**
(59)	Yes	Yes	No	Convenience	Yes	Yes	Yes	Yes
(60)	Yes	Yes	Yes	Yes	Yes	Yes but possibility valuable data lost or misinterpreted through the summarising rather than recording of focus groups	Yes adjusted moderators according to ethnic mix of the group and conducted in Spanish if necessary. No information given on forward-backward translations	Yes
(62)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
(43)								
(63)								
(64)	Yes	Yes	Unclear	Yes	Yes	Yes	Unclear	Yes
(65)								
(68)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
(67)	Yes	Yes	No	Convenience	Yes	Yes	Yes	Yes
(69)	Yes	Yes	Yes	Yes	Yes	Yes	Unclear	No
(70)	Yes	Yes	Yes	No (part of RCT)	Yes	Yes	Unclear	No
(51)	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
(52)								

Quality criteria are from Popay et al 1998 unless otherwise stated.

*CASP - Critical Appraisal Skills Programme