

Oral Rehabil. Author manuscript; available in PMC 2012 June 26.

Published in final edited form as:

J Oral Rehabil. 2012 March; 39(3): 161–169. doi:10.1111/j.1365-2842.2011.02247.x.

Classifying orofacial pains: a new proposal of taxonomy based on ontology

D. R. NIXDORF*, M. T. DRANGSHOLT†, D. A. ETTLIN‡, C. GAUL§, R. DE LEEUW¶, P. SVENSSON**, J. M. ZAKRZEWSKA††, A. DE LAAT‡‡, and W. CEUSTERS§§

*Division of TMD & Orofacial Pain and Department of Neurology, University of Minnesota, Minneapolis, MN †Departments of Oral Medicine and Dental Public Health Sciences, University of Washington, Seattle, WA, USA ‡Clinic for Masticatory Disorders, Removable Prosthodontics, Geriatrics, and Special Care Dentistry, University of Zurich, Zurich, Switzerland §Department of Neurology, University Duisburg-Essen, Essen, Germany ¶Orofacial Pain Center, University of Kentucky, Lexington, KY, USA **Department of Clinical Oral Physiology and Department of Oral Maxillofacial Surgery, Aarhus University, Aarhus, Denmark ††Orofacial Pain Unit, Eastman Dental Hospital, University College London, London, UK ‡†Department of Oral and Maxillofacial Surgery, Catholic University of Leuven, Leuven, Belgium §§Ontology Research Group, New York State Center of Excellence in Bioinformatics and Life Sciences, University at Buffalo, Buffalo, NY, USA

SUMMARY

Propose a new taxonomy model based on ontological principles for disorders that manifest themselves through the symptom of persistent orofacial pain and are commonly seen in clinical practice and difficult to manage. Consensus meeting of eight experts from various geographic areas representing different perspectives (orofacial pain, headache, oral medicine and ontology) as an initial step towards improving the taxonomy. Ontological principles were introduced, reviewed and applied during the consensus building process. Diagnostic criteria for persistent dento-alveolar pain disorder (PDAP) were formulated as an example to be used to model the taxonomical structure of all orofacial pain conditions. These criteria have the advantage of being (i) anatomically defined, (ii) in accordance with other classification systems for the provision of clinical care, (iii) descriptive and succinct, (iv) easy to adapt for applications in varying settings, (v) scalable and (vi) transferable for the description of pain disorders in other orofacial regions of interest. Limitations are that the criteria introduce new terminology, do not have widespread acceptance and have yet to be tested. These results were presented to the greater conference membership and were unanimously accepted. Consensus for the diagnostic criteria of PDAP was established within this working group. This is an initial first step towards developing a coherent taxonomy for orofacial pain disorders, which is needed to improve clinical research and care.

Keywords

Non-odontogenic tooth pain; chronic tooth pain; atypical odontalgia; phantom tooth pain
consensus; taxonomy; ontology; diagnosis; diagnostic criteria

Introduction

Orofacial pain, ¹ is a common symptom and its prevalence is estimated to be around 13–26% in the community (1–3). In some of these people, the pain is persistent and does not have a readily identifiable local aetiology, which results in poor response to existing treatments (4). One major reason for this unfortunate situation is the general state of confusion surrounding the diagnosis and classification of persistent orofacial pain disorders, specifically chronic tooth-related pain disorders, which in turn has inhibited progress in research and clinical care (5, 6). As the development of diagnostic criteria for various orofacial pain disorders has proceeded largely without 'structure' (7), an international consensus collaboration to initiate the development of a taxonomy for orofacial pain disorders based on ontology was organised by the International RDC-TMD Consortium initially focusing on clarifying terminology and their relationships, so that standardised multisite data collection can occur and future research, such as cluster analysis methods, can be applied to refine these criteria.

Current problems with diagnostic criteria for painful orofacial disorders

Various sorts of pain can be experienced in the head and neck region, and some of them are symptoms of pain disorders that have existing diagnostic taxonomies such as temporomandibular disorders (TMDs), primary or secondary headache disorders, as well as neuropathic pain disorders. These diagnoses are based on separate diagnostic criteria that are at different stages of development and for which it is still unknown whether they truly correspond to distinct disorder types and if so, to what degree their manifestations have overlapping characteristics that make it difficult to distinguish these disorders clinically. The current situation is further complicated by the use of multiple terms to describe what are often considered similar entities, such as with atypical odontalgia, phantom tooth pain, deafferentation pain and trigeminal neuropathy (8). Confusion also has occurred in the different interpretations of the meanings of terms because the same terms are often used for different purposes (i.e. pain = sensation as a symptom AND as a disorder AND as a diagnosis), and sometimes the same terms are used for different entities (i.e. trigeminal neuralgia for a specific entity AND pain of neurogenic origin occurring within the trigeminal nerve distribution). This is compounded by the general lack of a clear operationalisation of these diagnostic criteria, resulting in research findings that are difficult to interpret. These problems highlight the need to perform a thorough terminological 'clean up' of the domain of orofacial pain disorders. This enhanced terminology would improve communication and research to assist in reaching the ultimate goal of improving patient care.

The development of the RDC /TMD, which by most has been deemed to be successful, has started to address many of the diagnostic shortcomings mentioned earlier (9). Despite the benefits this system offers, it has not succeeded in gaining widespread clinical application. Four speculative reasons why this may be occurring are (in no specific order) excessive detail needed in the diagnostic process to arrive at a diagnosis within a clinical environment, lack of consensus within and between different clinician groups, a lack of a coherent overarching taxonomy based on ontological principles and the difficulty to prognosticate treatment outcomes. Therefore, to build from the experiences gained from almost two decades of working with the RDC /TMD, the long-term goals of this consensus process are to:

 $^{^{1}}$ We use from here on the term 'pain' to denote pain as a symptom experienced by a patient and 'pain disorder' to denote a disorder for which pain experiences are the most prominent feature.

1. Develop, using ontologically sound principles, an accurate representation of orofacial pain disorders comprising a taxonomy as its back bone and a set of corresponding terms and definitions and

2. allow rapid implementation of the resulting taxonomy into clinical practice by making it easy to use.

Materials and methods

Application of ontological principles

The word 'ontology' has two meanings. In philosophy, it denotes the study of the nature of being, such as whether an entity exists or not, similarities and differences between such entities and how they relate to each other. In computer science, the word denotes an artefact that functions as a representation of a domain such as human anatomy, nosology and so forth (10). Most development methods for ontologies in the computer science sense have their origin in concept-based terminology, because their advocates believe that the way in which concepts are organised mimics the way reality is organised. These methods use computational techniques that, when applied correctly, guarantee the logical consistency of such representation. Unfortunately, logical methods alone cannot detect representational errors (11) that are resultant from flaws in the concept-based approach itself (12). An example of such error is stating that fictitious pain, such as feigning the pain of a headache to avoid work, is a special sort of pain - which is an error because there actually is in such case no pain symptom present at all. Many existing biomedical terminologies, ontologies and classification systems contain such flaws because they rest on incomplete, inconsistent or confused accounts of basic terms pertaining to, for instance, diseases, diagnoses and clinical phenotypes. The National Cancer Institute Thesaurus (NCIT), for example, identifies 'Chronic Phase of Disease' as a subtype of 'Finding', which it defines as 'Objective evidence of disease perceptible to the examining physician (sign) and subjective evidence of disease perceived by the patient (symptom) (http://ncit.nci.nih.gov/ncitbrowser/ConceptReport.jsp?dictionary=NCI

%20Thesaurus&code=C3367). This definition implies that a disease does not exist except as one or other form of evidence. Thus, the NCIT authors do not distinguish between disease and evidence of disease. The reason why this conflation is problematic is revealed when we need to represent in an ontology the link between observable clinical phenomena and hypothesised unobservable biological causes: a belief about some cause is only truly *about* something when the cause exists; although that does not invalidate the existence of the belief itself, it does require extreme caution in the development of ontologies that are faithful to reality.

The ontological methodology presented here combines the computer science and philosophical approaches to ontology and is based on the idea that representations, including diagnostic criteria, must be compatible with future advances in empirical science (13) and must therefore distinguish between:

- 1. Level L1: the level of reality (e.g. infected pulpal tissues that trouble the patient and are therefore referred to as 'on the side of the patient'),
- 2. Level L2: the level of cognitive representations and experiences of this reality, as embodied in observations, interpretations, beliefs, and desires on the part of patients, clinicians, and others (e.g. the pain resulting from infected pulpal tissue, which is experienced only by the individual, L1 the clinicians cannot directly experience their patient's pain and also the belief of the clinician that the patient indeed suffers pain, L2) and

3. Level L3: the level of publicly accessible concretisations of cognitive representations (L2) in information artefacts of various sorts, such as in ontologies, terminologies, health records and diagnostic classification systems, as well as in verbal or written statements by the patient, for instance statements that the patients experiences pain.

Making these distinctions allows us to differentiate between disorders and diseases on the one hand that exist in first-order reality (L1), and diagnoses on the other, which are formulated in a clinician's mind (L2) or in an entry in a health record (L3). These distinctions are further used in our analysis of the clinical characteristics for orofacial pain as reported in the published literature (Table 1).

Further, our ontological methodology distinguishes *particulars* from *universals* and *classes*. Particulars are entities like a specific person, such as John Doe, or the specific facial pain from which John (and he alone) has been suffering since last year. Universals, in contrast, are repeatable entities that have particulars as their instances, such as human being and facial pain, while classes are collections of particulars that share one or more characteristics that are not essential for the particulars that instantiate a given universal, such as human beings with facial pain: having facial pain is not essential for something to be a human being. Thus, John Doe is an instance of human being, and his facial pain is an instance of the universal pain. Diagnoses are about disorder and disease instances; published diagnostic criteria are about disorder and disease universals or classes.

Like other areas of health care, the field of orofacial pain struggles with such ontological issues, because content experts deriving various diagnostic criteria are usually clinicians and researchers familiar to the subject matter but ignorant of the process of thinking in terms of particulars and universals, as well as of levels of reality. Consequently, conflations exist in published diagnostic classification systems because fundamental underlying principles were not followed, resulting in improper representation, miscomprehension and confusion in communication.

To avoid repletion of past errors, we applied ontological principles, during a recent international consensus meeting, to assist in developing a classification system amenable to future advances in the field.

Results

Consensus development process

A type of orofacial pain encountered in clinical practice with considerable taxonomical confusion is persistent pain present in the dento-alveolar areas of the mouth, often referred to as 'atypical odontalgia' (6). This condition served as an example for the application of ontological principles to develop diagnostic criteria. Descriptive aspects reported about this sort of pain and the various contexts in which instances thereof appear were delineated and discussed in terms of the L1 /L2 /L3 distinction, thereby disambiguating descriptions in the literature based on whether they denote phenomena that can be observed in individual patients or patient populations (Table 1). For example, a description such as 'the pain is mainly unilateral' can mean (i) that when a specific patient has pain, there is usually only pain on one side, (ii) that when a population of pain patients is observed, most of these patients exhibit pain on one side or (iii) both situations occur.

The expert in ontological realism further explained the basics of the Ontology of General Medical Science (OGMS) (14), which are summarised in Fig. 1. From this perspective, multiple signs and symptoms including their nature and position under OGMS' framework were discussed and key descriptive clinical features were identified (Table 2), while other

candidate features were discarded (Table 3). Consensus on the observations that predominate in patients became the diagnostic criteria that exist in the observable domain (Fig. 1).

The most challenging tasks were to assess (i) which terms used in the domain correspond with real entities, (ii) what real entities need to exist for certain signs and symptoms to manifest themselves, (iii) to what degree do distinct pain disorders lead to similar types of signs and symptoms, and (iv) to what extent can individual patients be suffering from distinct pain disorders at the same time, yet exhibiting manifestations that can be explained by the presence of only one particular pain disorder. Clinical experts need an adequate terminology to describe, with sufficient discriminative power, the various observable phenomena, thereby making sure to name each distinct entity differently to avoid confusion (i.e. word 'pain' being simultaneously used as an observation, an entity, and a term to label multiple entities; see Fig. 1). This approach does not require the aetiology of the phenomena to be known but nevertheless guarantees that when the underlying pathophysiology becomes clearer in the future, the terminology used to describe the entities will still remain valid.

The discussion therefore centred on the ideal terminology. Because the pain in case of what is currently believed to be denoted by the term 'atypical odontalgia' is long lasting and not acute, the term 'chronic', defined as 3 months or more (IASP definition), was initially agreed upon to be adequate. The ontologist then pointed out that under this definition of 'chronic', the criterion of having lasted longer than 3 months is a criterion about when the term 'chronic' may be used to describe a particular pain to have consistent descriptions cross-patients. At the level of the pain entity in reality itself, there is indeed no magical line, which when crossed suddenly transforms the existing non-chronic pain into a chronic pain a par with a caterpillar transforming in a butterfly. If one is allowed to name a particular pain instance chronic in line with the definition for chronic used, then that particular pain was already chronic from its very start, although it could not be known yet. The pain is in most patients continuous, present more than 90 days of 180 (15); the anatomical location is usually in and around teeth, or in the alveolar bone where teeth once were located (dentoalveolar) – following the precedent of regionally defining pain disorders (16, 17). The term 'persistent' was considered in the place of *chronic* and *continuous* for two reasons: (i) chronic is thought to also capture the negative emotional experience, which for the disorder may or may not be present and (ii) continuous suggests that the symptom of pain is uninterrupted, which is untrue in some instances. The term 'persistent' is not ideal because it presumes that the outcome of treatment is a failure to relieve the symptom of pain, which is contrary to long-term goal of these collaborative efforts. Looking at this from another perspective, the term 'persistent' relates to the clinical observation that local surgical treatments to the dento-alveolar tissues is ineffective at relieving the pain symptom of this disorder, which is useful perception to promulgate at the present time given the failure of contemporary approaches to treatment. Furthermore, the term 'persistent' is the term recently proposed to describe a similar pain-related phenomenon presenting within the muscles of mastication (18) and is therefore a consistent terminology that can be used to expand this taxonomy. Finally, because pain in ontological terms is a symptom, this entity would better be described as a pain disorder, a specific disease entity.² Thus, all together, this results in the term 'persistent dento-alveolar pain' disorder, which may be shortened for everyday use to PDAP.

²The Ontology of General Medical Science (OGMS, Fig. 1) distinguishes between disorders and diseases. A disorder forms the pathor-morphological basis that creates the disease. The term 'pain disorder' as we use it here is in OGMS terms a disease rather than a disorder, and the term 'pain disease' would therefore be more appropriate. However, we decided to keep the term 'pain disorder' as this term – in contrast to the term 'pain' itself – is consistently used in the domain, namely in the sense of a disease as viewed in OGMS.

Changing the operationalised criteria used to define this entity, PDAP, does not affect whether this entity exists or not. Individuals with this pain disorder may express the signs and symptoms differently, but this does not affect the reality of whether they have the pain disorder or not. Therefore, conceptually, the criteria to define this pain disorder can and do vary based on the setting in which it is being used. This allows researchers investigating the underlying mechanisms of this pain disorder, those most interested in studying individuals who are known to have this pain disorder (true positive cases), to maximise the specificity of the diagnostic criteria. This will come at the expense of sensitivity, meaning that some individuals with the actual pain disorder may be excluded because the diagnostic criteria used were too stringent (false negative cases). Conversely, epidemiological researchers will need to strike a balance between diagnostic sensitivity and specificity, but will want to use diagnostic criteria that may not necessarily involve extensive examination procedures or difficult to access testing, such as imaging techniques. These situations highlight the fact that for research and clinical purposes, the criteria used to define the pain disorder may differ, but this does not change the underlying reality or the terminology used to refer to it. For this key reason, following ontological principles lead to the development of the diagnostic criteria for PDAP defined by the four components included within the name and developed recommendations for operationalisable diagnostic criteria (Fig. 2).

During the development of the inclusion and exclusion criteria, one potentially vexing question remained: how to best exclude pain disorders that are caused by a local aetiology, such as inflammatory disorders that may linger 3 months or more? Currently, the only methods are to perform a clinical examination along with appropriate diagnostic imaging to exclude known dental and other orofacial pain disorders that may have the overlapping symptom of continuous or near continuous pain.

Subgroups and a taxonomic structure were also mapped out. As these pain disorders can either be precipitated by known traumatic events or arise in the absence thereof (19), a primary and secondary form was suggested, in line with the existing pain taxonomy (20). This subdivision delineates potentially causal factors, so that future research can explore his relationship more fully. Subjects with sensory changes detectable by neurological examination, or more precise quantitative sensory testing, can be separated from those who do not demonstrate these changes; which is consistent with a more specific classification of pain disorders having neuropathic underpinnings (21).

Discussion

Advantages and limitations with new taxonomy and nomenclature

There are several advantages to this ontologically based taxonomic approach. The pain phenomenon is concisely described, so that all health practitioners could potentially make the diagnosis about the presence of this sort of pain with minimal training. No specialised equipment is necessary to derive the diagnosis beyond clinical examination and appropriate imaging that is currently employed in practice. In addition, the diagnosis is operational, mutually exclusive from other types of pain disorders and could be used in epidemiological studies. Furthermore, subtypes exist, such as secondary PDAP in association with sensory abnormalities, which would meet accepted criteria for trigeminal neuropathic pain disorder (21). Such accepted subtypes would allow orofacial pain specialists, neurologists and others, to better define and communicate disease categories by mechanism, aiding in diagnosis and treatment. Future research will help to estimate the prevalence of each of these subtypes, and cluster analysis techniques can be used to explore relationships with similar entities. Another advantage is that this taxonomic approach could fit within existing diagnostic systems such as the International Criteria of Headache Disorders (ICHD-II) of the International Headache Society (20). Finally, initiatives to develop or improve diagnostic criteria for pain disorders

in other anatomical locations in the orofacial region could benefit from the same approach, such as persistent zygomatic pain disorder and persistent oral mucosal pain disorder, thus producing a parallel classification system without overlap and less confusion.

There are several limitations to this taxonomical approach. It still is based on expert opinion, but given the lack of research data present on orofacial pain taxonomy, other than TMDs and trigeminal neuralgia, there is no better option to pursue at this juncture. Multisite data collection efforts with the aim to provide evidence-based improvements on this taxonomy should be a focus of future research to move beyond opinion. The introduction of new terminology can be problematic for recognition purposes. This approach was deliberately performed as to not make *a priori* assumptions on underlying mechanisms for this pain disorder, or groups of pain disorders, and to avoid confusion regarding major conceptual change of the definition if a previous term was retained. In addition, it was thought to be favourable in helping gain widespread acceptance, especially as it is complimentary with the Ontology for Dental Research being developed (22).

It should be noted that the ontology underlying the diagnostic criteria for orofacial pain, once fully developed as exemplified by the criteria for PDAP, can be considered the first axis of a multiaxis classification system, such as the Research Diagnostic Criteria for TMD (23). Within such a multiaxis classification, people who are classified as having the pain disorder in question (axis I) are then further classified on the variable amount of psychosocial distress present (axis II). Suggestions on how to approach the delineation of this second axis is beyond the scope of this current project, but discussion on the topic is encouraged among the interested groups to help shape and form the taxonomy.

Anticipated future activities

The derivation of the diagnostic criteria for PDAP has produced a taxonomical model for one chronic orofacial pain disorder that can be applied to other anatomical areas. The purpose of this article was to disseminate this information regarding the process, underlying rationale and outcome, of the recent international consensus meeting. The ontological, expert-based approach was described to stimulate discussion at local, national and international levels and garner feedback, such as response letters to the editor, as an essential part of the larger consensus development process. It is anticipated that a Delphi process will be performed to incorporate the information and further refine the diagnosis to derive a widespread consensus among healthcare experts (24–27) for a taxonomy of orofacial pain disorders. Also anticipated is the development of a systematic multicentre data collection method to produce larger robust data sets to further perform research and refine this taxonomy.

Conclusions

Consensus for the diagnostic criteria, with favourable properties, for PDAP was established as an example on how ontological principles can be used to improve related taxonomy. This is an initial first step towards developing a harmonised taxonomy for orofacial pain disorders, which is needed to improve clinical research and patient care.

Acknowledgments

Supported by International RDC /TMD Consortium Network of IADR, Orofacial Pain Special Interest Group of IASP, Department of Health's NIHR Biomedical Research Centre UCL /UCLHT (J. M. Zakrzewska), NIH K12-RR023247 (D. R. Nixdorf), NIH R21-DE018768 (M. T. Drangsholt) IASP Bruun Travel grant (M. T. Drangsholt and P Svensson), National Center for Biomedical Ontology and NIH R01-DE021917 (W. Ceusters) and Medotech.

References

1. Locker D, Grushka M. Prevalence of oral and facial pain and discomfort: preliminary results of a mail survey. Community Dent Oral Epidemiol. 1987; 15:169–172. [PubMed: 3474103]

- 2. Lipton J, Ship JA, Larach-Robinson D. Estimated prevalence and distribution of reported orofacial pain in the United States. J Am Dent Assoc. 1993; 124:115–121. [PubMed: 8409001]
- 3. Macfarlane TV, Blinkhorn AS, Davies RM, Kincey J, Worthington HV. Orofacial pain in the community: prevalence and associated impact. Community Dent Oral Epidemiol. 2002; 30:52–60. [PubMed: 11918576]
- 4. Lewis MAO, Sankar V, De Laat A, Benoliel R. Management of neuropathic orofacial pain. Oral Surg Oral Med Oral Path Oral Rad and Endod. 2007; 103(suppl 1):S32:e1–e24.
- Woda A, Pionchon P. A unified concept of idiopathic orofacial pain: clinical features. J Orofac Pain. 1999; 13:172–184. [PubMed: 10823031]
- Lavigne GJ, Sessle BJ. The Vancouver symposium: a unique focus on nerve damage and neuropathic trigeminal pain. J Orofac Pain. 2004; 18:277.
- 7. Woda A. A rationale for the classification of orofacial pain. 2007; 15:209–222.
- 8. Melis M, Lobo SL, Ceneviz C, Zawawi K, Al-Badawi E, Maloney G, et al. Atypical odontalgia: a review of the literature. Headache. 2003; 43:1060–1074. [PubMed: 14629241]
- 9. Schiffman EL, Truelove EL, Ohrbach R, Anderson GC, John MT, List T, et al. The research diagnostic criteria for temporo-mandibular disorders. I: overview and methodology for assessment of validity. J Orofac Pain. 2010; 24:7–24. [PubMed: 20213028]
- 10. Smith B, Ceusters W. Ontology as the core discipline of biomedical informatics: legacies of the past and recommendations for the future direction of research. 2007:104–122. ????? ????
- 11. Ceusters W, Smith B, Kumar A, Dhaen C. Mistakes in medical ontologies: where do they come from and how can they be detected? In: Pisanelli DM. Ontologies in medicine. Proceedings of the workshop on medical ontologies. 2004:145–163.
- 12. Smith B, Ceusters W, Temmerman R. Wüsteria. Stud Health Technol Inform. 2005; 116:647–655. [PubMed: 16160331]
- 13. Smith B, Ceusters W. Ontological realism: a methodology for coordinated evolution of scientific ontologies. Appl Ontol. 2010; 5:139–188. [PubMed: 21637730]
- 14. Scheuermann R, Ceusters W, Smith B. Toward an ontological treatment of disease and diagnosis. Summit on Translat Bioinforma. 2009; 1:116–120. [PubMed: 21347182]
- 15. Von Korff M, Dunn KM. Chronic pain revisited. Pain. 2008; 138:267–276. [PubMed: 18226858]
- Headache Classification Subcommittee of the International Headache Society. Classification and diagnostic criteria for headache disorders, cranial neuralgias and facial pain. Cephalalgia. 1988; 8:1–96.
- 17. Merskey, H.; Bogduk, N. Classification of chronic pain: descriptions of chronic pain syndromes and definitions of pain terms. Seattle, WA: IASP Press; 1994.
- 18. Benoliel R, Svensson P, Heir GH, Sirois D, Zakrzewska J, Oke-Nwosu J, et al. Persistent orofacial muscle pain. Oral Dis. 2011; 17:23–41. [PubMed: 21382137]
- Woda A, Pionchon P. A unified concept of idiopathic orofacial pain: pathophysiologic features. J Orofac Pain. 2000; 14:196–212. [PubMed: 11203755]
- 20. Headache Classification Subcommittee of the International Headache Society. The international classification of headache disorders, 2nd edition (ICHD-II). Cephalalgia. 2004; 24:9–160. [PubMed: 14979299]
- Treede RD, Jensen TS, Campbell JN, Cruccu G, Dostrovsky JO, Griffin JW, et al. Neuropathic pain: redefinition and a grading system for clinical and research purposes. Neurology. 2008; 70:1630–1635. [PubMed: 18003941]
- 22. Smith B, Goldberg LJ, Ruttenberg A, Glick M. Ontology and the future of dental research informatics. J Am Dent Assoc. 2010; 141:1173–1175. [PubMed: 20884915]
- 23. Dworkin SF, LeResche L. Research diagnostic criteria for temporomandibular disorders: review criteria, examinations and specifications, critique. J Craniomandib Disord. 1992; 6:301–355. [PubMed: 1298767]

 Cramer C, Estein JB, Sheps SB, Schecter MT, Brusser JR. Modified Delphi survey for decision analysis for prophylaxis of post-radiation osteonecrosis. Oral Oncol. 2002; 38:574–583. [PubMed: 12167435]

- 25. Mease PJ, Clauw DJ, Arnold LM, Goldenberg DL, Witter J, Williams DA, et al. Fibromyalgia syndrome. J Rheumatol. 2005; 32:2270–2277. [PubMed: 16265715]
- Ko DT, Wijeysundera HC, Zhu X, Richards J, Tu JV. National Expert Panel. Canadian quality indicators for percutaneous coronary interventions. Can J Cardiol. 2008; 24:899–903. [PubMed: 19052669]
- 27. Sherman PM, Hassall E, Fagundes-Neto U, Gold BD, Kato S, Koletzko S, et al. A global, evidence-based consensus on the definition of gastroesophageal reflux disease in the pediatric population. Am J Gastroenterol. 2009; 104:1278–1295. [PubMed: 19352345]
- 28. Jacobs R, Wu CH, Goossens K, De Laat A, Van Loven K, Antonis Y, et al. A case-control study on the psychophysical and psychological characteristics of the phantom tooth phenomenon. Clin Oral Investig. 2002; 6:58–64.
- 29. Allerbring M, Haegerstam G. Chronic idiopathic orofacial pain. A long-term follow-up study. Acta Odontol Scand. 2004; 62:66–69. [PubMed: 15198384]
- 30. List T, Leijon G, Helkimo M, Oster A, Svensson P. Effect of local anesthesia on atypical odontalgia a randomized controlled trial. Pain. 2006; 122:306–314. [PubMed: 16564621]
- 31. List T, Leijon G, Helkimo M, Oster A, Dworkin SF, Svensson P. Clinical findings and psychosocial factors in patients with atypical odontalgia: a case-control study. J Orofac Pain. 2007; 21:89–98. [PubMed: 17547120]
- 32. List T, Leijon G, Svensson P. Somatosensory abnormalities in atypical odontalgia: a case-control study. Pain. 2008; 139:333–341. [PubMed: 18571324]
- 33. Williams N. The factory model of disease. Monist. 2007; 90:555-584.

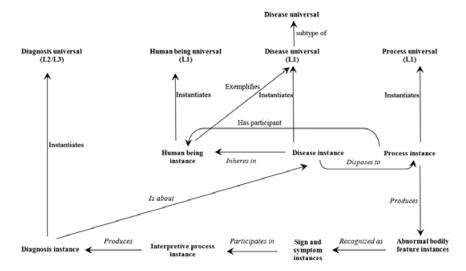


Fig. 1.

Conceptual framework for Ontology of General Medical Science (OGMS). The OGMS pursues a view of disease as resting in every case on some (perhaps as yet unknown) physical basis (Williams, 2007) (33). When, for example, there is a persistent pain in some body part of a person, this is because (1) some physical structure or substance in the person is disordered (e.g. there is a gingival lesion or damage to a nerve) as a result of which (2) there exists a disposition for the person to undergo processes that can be qualified as being pathological. In many cases, patients thus harbour disorders before the associated dispositions are realised in changes some of which may become observable. Once observable, these changes are usually first recognised by patients (symptoms) and subsequently observed by clinicians (signs). All changes brought about by a disorder constitute the disease phenotype that can exist without being observed. Indeed, as technology advances, our ability to detect the underlying components of a disease phenotype will expand. What is observed, including erroneous beliefs about manifestations, becomes interpreted and leads to a diagnoses.

PDAP

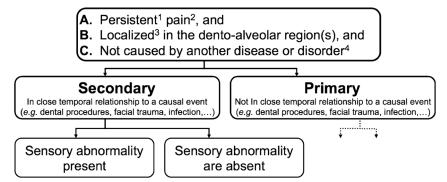


Fig. 2. Diagnostic Criteria for PDAP

Criteria:

¹persistent meaning pain present at least 8 h per day 15 days or more per month for 3 months during

²pain is defined as per IASP criteria (includes dysesthesia)

³localised meaning the maximum pain defined within an anatomical area

 $^{^4}$ extent of evaluation non-specified (dental, neurological examination +/-) imaging, such as intra-oral, CT and / or MRI).

 Table 1

 Review of the clinical characteristics reported in published literature

General clinical concept		tions in the context of what is currently referred to as Odontalgia'	Ontologic referent	al category / intended
Sensation	a.	Pain and / or dysesthesia	a.	Symptom (L2) / individual patient
Pain intensity	a.	Variable, mild to severe and may change over time	a.	Symptom (L2) / individual patient
Anatomical location	a.	In and around teeth, or where teeth were previously located, not moving or changing over course of months	a.	Body part (L1) / individual patient
	b.	Maxillary teeth tend to be affected more often than mandibular, and posterior teeth more often that anterior teeth	b.	Information entity (L3) / patient population
Laterality	or re	d b) Mainly unilateral; may have more than one focus and / ferral to ipsilateral opposite arch or same arch contralateral	a.	Body part (L1) / individual patient
	side		b.	Information entity (L3) / patient population
Temporal characteristics	a.	Continuous or near continuous pain, not defined by paroxysms without provocation.	a.	Temporal region (L1) / individual patient
	b.	In clinic populations, pain often present for months to years, often reported refractory to various treatments	b.	Information entity (L3) / patient population
Character of pain	a.	Variable; dull, aching, throbbing and most other descriptors	a.	Symptom (L2) / individual patient
Age of onset	a.	Variable; clinical presentation commonly in 4th and 5th decades of life	a.	Information entity (L3) / patient population
Gender ratio – F / M	a.	Female preponderance; clinical populations range from 3 to 8 females to 1 male.	a.	Information entity (L3) / patient population
Precipitating events	a.	Commonly reported to occur in relation to (1) various dental treatments such as root canal treatment and tooth extractions, and (2) trauma and upper respiratory infections, while (3) also without any observed precipitating event	a.	Information entity (L3) / patient population
Clinical presentation	a.	Absence of an identified local pathology	a.	Sign (L1) / individual patient and conclusion (L2) clinician
Somatosensory findings	a.	Evidence for neuropathy / loss of function in some cases, but not present in all cases, while evidence of hyperalgesia / allodynia / gain in function in others – suggesting heterogeneity	a.	Information entity (L3) / patient population
Anaesthetic blockade	a.	Equivocal, with variable response suggesting heterogeneity	a.	Information entity (L3) / patient population
Sleep	a.	Thought not be disturbed, but little evidence present	a.	Hypothesis (L2 / L3) / patient population
Comorbid disorders	a.	Regional pain disorders = headache disorders and TMD suggested	a.	Information entity (L3) / patient population

General clinical concept	Manifestations in the context of what is currently referred to as 'Atypical Odontalgia'	Ontological category / intended referent	
	Psychosocial = depression, anxiety, somatisation suggested		
	Functional syndromes = widespread pain, irritable bowel syndrome suggested		
	General medical = unknown		

Where: L1 = first-order reality.

L2 = second-order reality, such as cognitive representations, experiences, beliefs, interpretations and desires in clinicians, patients and others which are about L1-entities.

L3 = third-order reality, which are accessible concretisations of L2, such as classification systems, terminologies and health records, as well as statements by clinicians or patients.

An explicit distinction is made between literature reports about phenomena that have variable characteristics within a single patient and those that describe variations in patient populations.

Data for this table were obtained from the following references: Woda & Pionchon, 1999 (5); Jacobs et al., 2002 (28); Melis et al., 2003 (8); Allebring & Haegerstram, 2004 (29); List et al., 2006 (30); List et al., 2007 (31); List et al., 2008 (32).

Table 2

Features separating individuals with PDAP from those without it

Key features: patient report of \dots

episode(s) of dysesthesia, otherwise referred to as 'pain' which:

is anatomically located in the dento-alveolar region innervated by the trigeminal nerve exhibits a continuous or near continuous temporal course

is present more often than not

is not primarily characterised by acute paroxysms

cannot be explained by the presence of another disease or disorder

 Table 3

 Clinical features discussed and the reasons why they were not included

Other features discussed	Reasons for not including
Precipitated by events, such as dental procedures and trauma, that are deemed to be causative for this disorder	Documented cases of initiation of this disorder without a specific cause being identified
Presumed to be of neuropathic origin, consistent with the criteria published by Treede <i>et al.</i> , 2008 in Neurology (21)	This has not been conclusively demonstrated, especially in the light of occurrences of the disorder for which no specific identifiable cause
Quality of pain	Pain quality is highly variable between individuals and similar qualities used to describe the symptom of pain from different disorders
Pain intensity	Somatosensation of pain is highly variable between individual (vs. a migraine headache disorder is deemed to be present when aura and autonomic features occur in an individual without experiencing pain)
Findings on clinical examination and imaging that are not specific to the disorder	Something can only be observed when it exists. Whether something existing is observable depends on the level of rigour applied to determine its presence, as well as state and availability of technology (sensitivity & specificity) (e.g. just because the entity <i>love</i> cannot be imaged does not mean it is non-existent). On the other hand, both our senses and technology can lead us to believe something exists, while it is not the case (e.g. hallucinations and imaging artefacts). Finally, it is not because something truly exists, that it has any relationship with the pain in the context of which observations are made