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Developing a Research Diagnostic Criteria for Burning Mouth Syndrome: Results from an International Delphi Process

Short Running Title: A Research Diagnostic Criteria for BMS

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Abstract (250 words)

Objective: To develop a beta version of a preliminary set of empirically-derived research diagnostic criteria (RDC) for burning mouth syndrome (BMS) through expert consensus, which can then be taken into a test period before publication of a final RDC/BMS.

Design: A 6 round Delphi process with twelve experts in the field of BMS was used. The first round formed a focus group during which the purpose of the RDC and the definition of BMS was agreed upon, as well as the structure and contents. The remaining rounds were carried out virtually via email to achieve a consensus of the beta version of the RDC BMS.

Results: The definition of BMS was agreed to be “an intraoral burning or dysaesthetic sensation, recurring daily for more than 2 hours per day over more than 3 months, without evident causative lesions on clinical examination and investigation”. The RDC was based upon the already developed and validated RDC/TMD and formed three main parts: patient self-report; examination; psychosocial self-report. A fourth additional part was also developed listing aspirational biomarkers which could be used as part of the BMS diagnosis where available, or to inform future research.

Conclusion: This Delphi process has created a beta version of an RDC for use with BMS. This will allow future clinical research within BMS to be carried out to a higher standard, ensuring only patients with true BMS are included. Further validation studies will be required alongside refinement of the RDC as trialling progresses.

Keywords: burning mouth syndrome, research diagnostic criteria, orofacial pain, oral dysaesthesia
glossodynia, stomatodynia

Introduction

Burning mouth syndrome (BMS) is characterised by persistent daily intraoral burning or dysaesthetic sensation, often combined with taste alterations and dry mouth, which cannot be explained by any clinically evident oral or systemic pathology and lasts for more than two hours per day for more than three months¹. This was previously referred to as primary BMS. Symptoms of burning in the oral cavity can be caused by underlying medical and dental causes, previously referred to as secondary BMS. It is therefore important to exclude any intraoral burning symptoms which can be attributed to a causative lesion(s), in order to make the correct diagnosis of BMS.

BMS affects between 0.7 to 18% of the population²⁻⁷ and is most common in post-menopausal women⁸⁻¹⁰. The wide range in BMS prevalence reported may be due to a number of factors, including definition used of BMS, and also the lack of a diagnostic criteria for BMS³⁻⁷. The aetiology of BMS is poorly understood, however is likely to be complex and multifactorial which makes management challenging¹¹.

There are a range of underlying causative lesions which can cause intra oral burning, including: candida infection, oral lichen planus, hyposalivation, contact mucosal reactivity, medications, anaemia, deficiencies in vitamin B12 or folic acid, Sjogren's syndrome, diabetes, and hypothyroidism^{3,11}. It is therefore prudent to exclude all of these causative factors in order to correctly diagnose BMS, this can be done with a thorough patient history and examination, as well as intra oral swabs or smear for candida, salivary gland function and flow studies, and haematological investigations.

In order to be able to correctly and reproducibly diagnose BMS, and therefore carry out comparable research in terms of aetiology and management, research diagnostic criteria (RDC) are required. Utilizing well-established methods within science for creating measures and concepts, an RDC for temporomandibular disorders (TMDs) was developed based on 5 core principles: biopsychosocial model, epidemiologic data, dual-axis for classifying disease and assessing the person, operationalized criteria and examination specifications, and recognition that the initial procedures, though required to initiate collection of useful data, would inevitably undergo revision. The 1992 publication was followed by extensive international research on TMDs using the same methods, which created a critical mass of information sufficient to demonstrate the viability of the RDC/TMD approach; this in turn led to a large-scale project for assessing all aspects of the RDC/TMD¹², which in turn led to a large consensus workshop and eventual publication of the DC/TMD¹³. Without the RDC/TMD and subsequently the DC/TMD, extensive studies on TMD etiology, mechanisms, disease progression, and treatments would either have not been possible or would have contributed very little to our understanding of TMDs. Consequently, it is entirely fair to claim that TMD knowledge and research would not be at its present state had the RDC/TMD not emerged to consolidate a field that was highly chaotic by the late 1980s¹²⁻²². Moreover,

since its development the RDC/TMD has served as a template for similar developments in RDC for persistent back pain²³ and for persistent pain more generally²⁴.

The aim of the present study was to therefore develop a beta version of a preliminary set of empirically-derived RDC for BMS through expert consensus. This beta version can then go on to further testing in appropriate environments, refinement and revision, before publication of a finalised version of the RDC/BMS.

Methods

Use of the Delphi process has been successfully used elsewhere within dental research for development of guidelines based on expert consensus²⁶. The number of participants required for a Delphi process varies dependent on purpose and the literature but was set as a minimum of seven²⁶⁻²⁸. Fourteen international experts were invited to take part in the Delphi process, with two declining involvement as they were no longer in the field or unavailable. Of the other twelve experts seven were able to meet face-to-face to begin the Delphi process and the other five participated remotely. The experts were identified from the literature and membership to the International Network for Orofacial Pain and Related Disorders Methodology (INFORM) of the International Association for Dental Research. With the exception of the first author, all had more than 8 years' experience of managing patients with BMS.

The first meeting (round 1) was held face to face at the International Association for Dental Research's annual meeting (San Francisco 2017). INFORM hosted the meeting, with seven of the study authors in attendance (CC, JD, MK, TR, CNH, YI, RO), with no expenses or incentives being offered. All participants received study-specific documentation prior to the meeting, including copies of relevant literature^{2,29-36} and an outline of the areas for discussion. The areas for discussion initially included: the definition of BMS and the components to be included in the RDC, and any further aspirational research required within the area.

The first meeting was in the form of a focus group, moderated by the senior author (JD), with minutes taken by the first author (CC). The first author was not involved decision making for the RDC/BMS. The meeting was structured around the topics for discussion, with the definition of BMS first being agreed upon, followed by the purpose and structure of the RDC. The contents of the RDC were then discussed, listed and structured into the relevant axes/parts of the RDC using a flipchart. Potential investigations to include were discussed, along with their availability at various centres internationally (for example the availability of QST). Instruments to be included for psychosocial assessment were also considered, along with the length, validity and potential relevance to BMS, as well as the number of instruments to use in

the RDC. The meeting concluded with discussion of biomarkers and aspirational future research within BMS. The minutes and drafted RDC structure were then used to draft a copy of the RDC/BMS by the first author to send to all participants. All participants were asked to add further critique and revisions to the draft RDC/BMS in an iterative process by email. Once all comments were received the RDC/BMS was revised by the first author and resubmitted to the panel for cross checking and any further revisions. Comments and responses were displayed in all copies of the evolving RDC/BMS document so the panel could see responses and revisions made. This process continued until no further revisions were suggested and all authors were in agreement, at this point (round 6) the beta version of the RDC/BMS was considered complete.

Results

Definition of BMS

Given the ongoing nature of the Delphi process and the involvement of several members in the IASP, American Academy of Orofacial Pain and the International Classification of Orofacial Pain (encompassing INFORM, IHS, AAOP and IASP) the definition of BMS for this RDC was revised to be that defined by the International Classification of Orofacial Pain 2020¹: “an intraoral burning or dysaesthetic sensation, recurring daily for more than 2 hours per day over more than 3 months, without evident causative lesions on clinical examination and investigation”, whereby the pain must have a burning quality and be felt superficially in the oral mucosa, and investigations include both clinical and laboratory based findings¹.

It was decided that the aim of this RDC/BMS was to exclude any intraoral burning symptoms which can be attributed to one or more causative lesions. It was agreed that an exception to this is a patient who was considered to have a mucosal pain disorder as classified by the ICOP¹, who following initiation of treatment for the causative lesion(s) continued to report a persistent burning symptom. In this case the patient can then be further classified as having BMS by the RDC²⁵. This RDC may also be used for patients who have had burning symptoms for less than 3 months, which would then be classified as probable BMS¹.

Structure and Content of the RDC

Given that an RDC/TMD had previously been developed for TMD, with further revisions and validation¹²⁻²², the participants agreed to use this as a basis for development of the RDC/BMS, in terms of its structure and development model. It was therefore decided to construct three data collection tools: symptom self-report, examination, and psychosocial self-report. Collectively, the information from these three tools

supports two axes: a physical diagnosis (and disease characterization), and psychosocial status of the person. The psychosocial status was important to include given the wealth of evidence regarding psychosocial factors in BMS pain^{37,38}, which are discussed in detail elsewhere³⁹. A third axis was also developed which included aspirational biomarkers. These were biomarkers that were considered to be potentially useful in diagnosing BMS, but which not all centres may have available, or biomarkers that would be useful to collect information on in BMS patients for future research. The third axis is not considered mandatory as part of the beta version of the RDC/BMS, but will be refined as the RDC/BMS is trialled and revised, with relevant validated biomarkers added as they emerge.

The full beta version of the RDC/BMS protocol is published as an appendix to this paper, as well as online on the INfORM website, but the following is an overview of the content of the data collection tools:

Part 1: Symptom Self-Report.

This instrument was designed in four parts to be completed by the patient prior to their initial patient assessment, and includes questions to ensure the patients' symptoms map to the definition of BMS.

The first part focuses on the primary symptom description and includes questions relating to: patient demographics, symptom description, symptom location, onset of symptoms (to map to the BMS definition of symptoms for longer than 3 months), duration of symptoms (to map to the BMS definition of more than 2 hours per day), diurnal variation, exacerbating and relieving factors, and symptom intensity. Symptom description uses both the short form of the McGill pain questionnaire, version 2⁴¹ plus additional words patients with BMS often use to describe their symptoms as experienced by the authors. It was recognised that patients use a variety of terms to describe their pain and therefore it was not practical to create an exhaustive list of terms, instead a free text option was also included. Symptom location is recorded by use of the pain diagram which has already been validated in the DC/TMD¹³, with an additional diagram of the tongue (based upon a standard oral cancer map) so patients are able to clearly mark the areas they suffer their burning symptoms, and to map to the definition of BMS in symptom location. In addition, the clinician should refer to this diagram with the patient during their verbal history and examination to ensure that their symptoms are experienced within the superficial mucosa as per the definition of BMS. Symptom intensity uses the graded chronic pain scale (GCPS V2), which has also been validated previously⁴².

The second part relates to potential associated features, including changes in taste, xerostomia, and specific food, drink and activities which may either exacerbate or relieve the burning symptoms. The third part enquires about illnesses and medications known to cause burning symptoms, as well as relevant

dental and social history questions. The final part is a daily diary that patient would be asked to take away and complete over the following month. This diary captures information on length of daily pain to also map to the BMS definition of symptoms for more than two hours per day.

Part 2: Examination.

The intent behind this instrument is that the health professional would provide an examination during their initial assessment of the patient which would be standardised with the aim being to exclude the following: salivary disorders, mucosal disease (Vessiculo-bullous, infective, autoimmune (Lupus), chronic mucocutaneous inflammation (Lichen Planus), Idiopathic (erythema migrans), trauma (chemical, thermal, radiation, mechanical), anaemia, metal and other allergies. There are three subsections to the examination, including a full extra oral exam, intra oral exam (including soft and hard tissues, dentures and appliances if relevant) and then further investigations. The extra and intra oral examination is set to replicate a standard patient clinical examination, with further points to consider specific to BMS diagnosis. Further investigations included haematological tests, candida swab or smear, Quantitative sensory testing (QST) and patch testing where clinical history and clinical exam indicate these. The QST protocol used in the RDC has been published elsewhere⁴³, and where centres may not have access to the equipment to carry out QST an alternative QualST protocol is included as an alternative⁴⁴. Imaging studies were not considered to be routinely required in the diagnosis of BMS, however could be considered aspirational where clinicians deem them to be relevant to the diagnosis.

Part 3: Psychosocial.

This instrument for the psychosocial axis was largely based on the DC/TMD Axis II; however, the panel decided it should be pain-specific rather than TMD-specific. Two versions were developed, a long and short, both also being patient self-report. The decision to produce a long and short version related to the RDC/BMS being developed as a beta version, therefore the two versions could be evaluated clinically before being refined further in terms of their content and constructs included in the final version of the RDC/BMS. The short version therefore includes all instruments the authors deemed to be essential based on current knowledge, and the long version contains further instruments which were relevant but the authors believed needed further evaluation before being included in the definitive RDC/BMS. The short version included: PHQ-4 (46) and an ultra-brief catastrophising scale^{46,47}, in addition the already completed GCPS V2 and pain diagram from Part I would also be referred to. The long version included the PHQ-9⁴⁷⁻⁵¹ and GAD-7⁵², replacing the PHQ-4, the PHQ-15^{53,54}, ultra-brief pain catastrophising scale, the OBC⁵⁵, PROMIS short form v1.0 Sleep Disturbance, and again reference to the already completed GCPS V2

and pain diagram in Part I. The instruments used in the RDC/BMS can be added to where required in individual circumstances where the clinician deems further psychosocial assessment is required.

For some instruments the timescales were adapted to allow ease of completion for the patient, so all timescales fell within a two week to three month window, as well as reflect the 3 month diagnostic timeline of BMS. The instruments with altered timescales include: Short-form McGill (1 week to 30 days); GCPS (6 months to 30 days); ultra brief catastrophizing scale (6 months to 3 months); PHQ-15 (4 weeks to 30 days); OBC (1 month to 30 days). The wording of the questions in the GCPS were also revised to reflect the nature of BMS symptoms being intra oral. The self-evaluation tool from the PHQ instruments was also added to the PHQ-15.

Part 4: Aspirational Biomarkers.

Following discussion with all participants it was decided that a section on biomarkers was important to include in the RDC/BMS, however would be an aspirational component to finalise following production of the beta version RDC/BMS. Other psychosocial domains for consideration were also discussed, including: prior abuse or neglect, self-efficacy, somatosensory amplification, and psychosocial stress.

Further aspirational work

The initial meeting was concluded with a discussion on further aspirational work that is required within the remit of BMS. These included the need for qualitative work to define the phenomenon of BMS, studies on peripheral measurements of dopamine, studies examining the potential link between BMS and vulvodynia, proteomic studies, and the methods used to olfactory and taste test in patients with BMS. In addition the need for QST studies in patients diagnosed with BMS with use of the RDC was highlighted in the initial meeting, as well as throughout further rounds of the Delphi process. Particular studies highlighted as being required in relation to QST were the use of qualitative sensory testing compared to QST, the use of local anaesthesia compared to QST to phenotype BMS, and the role of conditioned pain modulation in BMS patients.

Discussion

BMS is known to have a significant impact on quality of life, with 0.7-18% of the population suffering from the condition²⁻⁷, yet correct diagnosis and management of patients' symptoms remains challenging, with little high quality evidence available. A major barrier in research is that differing definitions of what BMS is are often used, resulting in patients who may not truly have BMS being included in clinical research^{39, 56-58} and in non-replicated studies.

The agreed definition of BMS as a result of this Delphi process was “an intraoral burning or dysaesthetic sensation, recurring daily for more than 2 hours per day over more than 3 months, without evident causative lesions on clinical examination and investigation”¹. In addition, it was agreed that exceptions can be made for patients considered to have a mucosal pain disorder as classified by the ICOP¹, who following initiation of treatment for the causative lesion(s) continues to report a persistent burning symptom, and for patients who have had burning symptoms for less than 3 months, which would then be classified as probable BMS¹ through use of the RDC.

This Delphi process has produced an expert based standardised approach to diagnosis of BMS through use of a research diagnostic framework, which will allow both researchers and practitioners to identify cases of BMS. The RDC/BMS is based on the definition of BMS set by the ICOP, and it is important to note that the RDC does not create a new classification for BMS, instead it allows the ICOP definition to be operationalised by creating a structured assessment and clinical examination. This, in turn, will allow the development and refinement of the classification criteria of BMS as well as ensure that only patients who fit the clinical criteria for BMS be included in clinical research, and allow high quality research in multiple centres to be carried out in order to fully understand the aetiology and management. For practitioners, the RDC/BMS supports greater confidence in clinical decision-making and identification of patients with putative BMS who nevertheless fall outside the current boundaries of the disorder and which therefore warrant additional clinical investigation.

The current RDC/BMS protocol is considered a beta *test* version. The decision to create a beta version allows the RDC to undergo an explicit period for evaluation, during which the RDC/BMS can be trialled at multiple centres, allowing revisions and refinements, as well as data collection on BMS, before a finalised RDC/BMS protocol is published. For example, the primary symptom list may be refined to include further terms patients frequently use to describe their pain, and instruments for the psychosocial assessment can be refined and prioritised from the short and long versions to create a definitive psychosocial axis. It is of particular importance that the current long and short versions of the psychosocial part are evaluated to ensure that the instruments selected are correct and essential for the final RDC/BMS. For example, the authors decided to include the OBC in the RDC/BMS in order to capture information on oral behaviours such as oral parafunction, this was considered important given the potential for guarding associated with pain elsewhere in the body, which may therefore contribute to sustained BMS symptoms. The OBC is not routinely used in the diagnosis of BMS, however the authors believed it was important to capture all potentially relevant information in the beta version of the RDC/BMS. Use of a beta version which can then go out for real-world testing also allows centres to feedback on the availability and practicality of

tests included in the RDC/BMS listed as aspirational, such as salivary flow rate and QST. During the Delphi process inclusion of these tests was considered to be essential, however there were concerns amongst the authors as to the availability of equipment required, or the time required to carry out such tests in publicly funded healthcare services, and for these reasons they were listed as being aspirational rather than essential so the RDC/BMS can be adopted as widely as possible. As the RDC/BMS is a beta test version and has not been evaluated in patients, this should be born in mind when using it to diagnose BMS as it should be used with clinical interpretation.

Use of the Delphi process has highlighted areas of research required within BMS. In particular, a list of aspirational biomarkers has been produced, these can be used within diagnosis of BMS, or to guide future research.

The use of QST and QualST remain an important area in diagnosis of chronic orofacial pain, such as BMS, and this was highlighted in both the initial face to face discussion and during later virtual discussion. It was agreed that research needs to continue comparing QST to QualST, particularly in BMS patients, as well as to other comparators such as local anaesthesia in order to phenotype BMS. Mechanistic studies are indeed needed to further the understanding of BMS pathophysiology. A further point generated through the Delphi process was the problem of QST in BMS patients when there is a midline distribution of BMS symptoms with no clear painful and non-painful side. There are also technical methodological issues, for example with pressure algometry on the tongue where there is a lack of firm base for isolation to carry out pressure pain thresholds. Work is currently being carried out in this area however, further adaption of existing protocols will be needed in these patients. The outcome of this can be included in the final version of the RDC/BMS.

Further research areas within BMS were also highlighted to include qualitative work to define the phenomenon of BMS, therefore allowing clinicians and researchers to more fully understand the symptoms and features, which would allow revision of the symptom descriptors used within part 1 the RDC/BMS. Other potential areas for research include the best methods for olfactory and taste testing in patients with BMS, evaluation the association of dopamine levels and BMS, exploring the role of the altered pain modulatory system, as well as investigation of a potential link between BMS and other chronic pain conditions, including vulvodynia in female patients.

Conclusion

An RDC for BMS is now available for use and trial within research studies and potentially also for clinical practice. Use of this RDC should allow clinical research within BMS to be carried out to a higher standard,

ensuring only patients with true BMS are included. Further validation studies will be required alongside refinement of the RDC/BMS as clinical trialling progresses.

Author Contributions

C Currie, R Ohrbach, and J Durham contributed to conception, design, data acquisition and interpretation, drafted and critically revised the manuscript. R De Leeuw, H Forssell, Y Imamura⁵, SK Jääskeläinen, M Kouttris, C Nasri-Heir, H Tan, T Renton, P Svensson contributed to data acquisition and interpretation, and critically revised the manuscript.

All authors gave their final approval and agree to be accountable for all aspects of the work.

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