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Diagnostic Studies of Temporomandibular Disorders: Challenges From an Epidemiologic Perspective

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Adequate data on the incidence, prevalence, natural history, and clinical course of temperomandibular disorders (TMD) and other chronic pain conditions are largely lacking, though the need to derive such basic data is recognized by clinicians, researchers, and public health agencies. This paper discusses challenges to the epidemiologic study of TMD diagnosis. These challenges include:

- Case definition: There is currently poor agreement regarding which combinations of clinical and psychosocial findings differentially define cases of TMD
- Differentiation of normal variation *v* pathophysiologic signs: To what extent do commonly gathered clinical measurements constitute pathophysiologic signs of TMD *v* reflect normal biologic variation
- Reliability of clinical measurement: Factors influencing reliability of clinical signs and reliability of examiners have not been adequately assessed
- Progressive *v* self-limiting disease activity: Do TMD subtypes represent a continuum of pathologic disease activity, or nonmutually exclusive categories describing largely symptomatic pain conditions that are selflimiting or stable.

It is recommended that epidemiologic studies not be constrained by *a priori* definitions of TMD subtypes, but continue to gather data on clinical signs and symptoms that have theoretical and clinical relevance to mandibular dysfunction and psychosocial status. An approach is proposed for development of reliable and valid criteria of TMD subtypes suitable for epidemiologic research.

Epidemiologic research, the investigation of the distribution and determinants of disease, aims to provide a scientific basis for efforts to prevent and control disease, illness, and disability. Clinical epidemiologic studies may examine the efficacy and risks of specific treatments.[1] Adequate data on the incidence, prevalence, risk factors, natural history and clinical course of temperomandibular disorders (TMD) and other chronic pain conditions are largely lacking.[2,3] Increasingly, the need to derive such basic data is recognized as a necessary objective by biomedical and pain researchers as well as by clinicians and public health agencies.[3–5]

The present paper discusses challenges that arise in developing and using TMD diagnostic data within the context of epidemiologic research and provides epidemiologic perspectives on reliability and validity of TMD diagnostic data. TMD is the term recommended by the American Dental Association[6] for what is presumed to be a range of painful conditions affecting orofacial and dental structures. These conditions are thought to be differentiated principally by the extent to which they involve the muscles of mastication [eg, myofascial pain dysfunction (MPD)][7] and/or the temporomandibular joint [eg, internal derangements (ID) or degenerative joint disease (DJD)].[8] Persistent pain and mandibular dysfunction are the most important and common manifestations of TMD. Relief from chronic orofacial pain is the principal reason persons seek treatment.[9] The emergence of chronic pain syndrome or dysfunctional chronic pain, although typically with less disability than back pain, has been demonstrated for TMD.[10,11]

Disease classification is not only a prerequisite for epidemiologic study, but one of its major goals.[12] Application of epidemiologic methods to TMD diagnostic entities currently encounters unique challenges that are less salient when TMD signs and symptoms are considered individually. The most important of these challenges, discussed more fully below, include case definition, differentiation of normal variation *v* pathophysiologic signs, reliability of clinical measurement, and progressive *v* selflimiting disease activity.

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Table 1. Prevalence of TMD Subtype Diagnoses in Clinic Cases, Community Cases, and Controls

	Prevalence (% of subjects)		
	MPD	*ID1*	*DJD*
Clinic cases (N = 247):			
Eversole & Machado	26.3	26.3	18.2
University of Washington	63.6	27.1	20.6
Community cases (N = 120):			
Eversole & Machado	30.0	25.8	10.8
University of Washington	37.5	18.3	5.0
Community controls (N = 209):			
Eversole & Machado	28.2	23.4	9.1
University of Washington	0.0	11.5	0.0

Table 2. Agreement of Two Diagnostic Schemes

A Internal derangement (type 1) diagnoses in clinic cases*

University of Washington	Present	Absent	Total
Present	29	38	67
Absent	36	144	180
Total	65	182	247

kappa = 0.236

B DJD diagnosis in clinic cases*

University of Washington	Present	Absent	Total
Present	8	43	51
Absent	37	159	196
Total	45	202	247

kappa = −0.034

* Eversole and Machado[17]

Other well known uncertainties abound, including continuing controversies over nomenclature; etiologic *v* functional approaches to diagnosis; relationship of diagnosis to the broad array of available treatments; criteria for assessing treatment outcome; and the associations between objective and subjective measurement methods. These complex issues all impinge on epidemiologic studies evaluating the reliability and validity of TMD diagnoses.

CASE DEFINITION

Defining Clinical Subtypes

A fundamental challenge to epidemiologic studies of TMD is the problem of "caseness." It is evident that epidemiologic studies cannot meaningfully depict the incidence, prevalence, risk factors, natural history, or clinical course of TMD subtypes without adequate operational criteria for distinguishing among the purported subtypes of TMD (for example, distinguishing cases of MPD from cases of ID or DJD). Although extensive literature is available regarding criteria for diagnosing specific subtypes of TMD,[13,14] few available diagnostic schemes specify in measurable terms the clinical signs that contribute to one form of TMD over another.

Even when comprehensive schemes are offered, critical differences emerge in the criteria for differential diagnosis. LeResche et al.[15,16] compared two comprehensive diagnostic schemes: 1) the published criteria provided by Eversole and Machado,[17] a mutually exclusive classification system, and 2) the clinical decision criteria for diagnosing TMD used at the Orofacial Pain Clinic of the University of Washington,[15,16] a system that allows for multiple diagnoses. Both schemes were applied to the analysis of clinical data gathered on the same cases as part of an on-going longitudinal epidemiologic study of TMD conducted jointly by the University of Washington and the Center for Health Studies of Group Health Cooperative of Puget Sound. A summary of relevant results is shown in Tables 1, 2A, and 2B. Table 1 reveals that the distribution of TMD subtypes differs markedly depending upon which set of diagnostic criteria is used. For example, the proportion of cases diagnosed as having MPD was more than twice as high using University of Washington criteria (64%) compared with the criteria of Eversole and Machado[17] (26%). By contrast, ID (type I) and DJD occurred with equal frequency among clinic cases, using either scheme. However, as Table 2 indicates, even where prevalence rates were comparable, the diagnostic schemes showed poor agreement for assigning individuals the same diagnosis; for example, the two schemes were mutually contradictory for about one-third of cases, for both ID (Table 2A: 74/247 cases; kappa = 0.236) and DJD (Table 2B: 80/247; kappa = −0.034).

These findings are troublesome because both schemes use criteria that are clinically relevant and are in widespread use. Yet these schemes, like most available TMD diagnostic schemes, have not been empirically tested in terms of reliability or validity. Epidemiologic studies based on one or another of these schemes may well lead to significantly different conclusions about the occurrence, causes, and course of TMD subtypes.

TMD as a Chronic Pain Condition

TMD is commonly regarded as a psychophysiologic disorder[18,19] although psychologic or psychosocial variables are not incorporated in current TMD diagnostic schemes. One implication of the view that TMD is a psychophysiologic disorder is that its expression may include psychological distress, especially anxiety and depression[20] and psychosocial impairment,[21] especially the emergence of chronic pain behaviors and chronic pain syndrome[11] or, synonymously, dysfunctional chronic pain.[22] To this extent, at least, TMD may be best understood as a chronic pain condition, similar in major respects to other important

Table 3. Prevalent Cases of Selected Pain Conditions: Comparison of Temporal Dimensions, Pain Intensity, Interference, Activity Limitation, and Treatment Seeking

	Pain conditions				
	TMD	*Head*	*Back*	*Abdomen*	*Chest*
Years since initial onset					
Mean	8.3	13.9	10.5	10.2	6.2
Median	6	12	8	7	4
Frequency in last 6 mo					
One brief episode	9.8%	9.7%	9.1%	11.8%	17.4%
More than half the days	27.7	16.4	28.8	15.3	13.9
Duration (h/day)					
<4 h	54.5%	34.4%	39.9%	65.1%	75.7%
≥9 h	27.3	29.4	32.2	14.5	13.0
Usual intensity (10-pt scale)					
Mean	4.3	5.9	4.7	5.1	4.3
Percent 'severe'	16.4	39.3	15.3	30.9	21.1
Usual interference (7-pt scale)					
Mean	2.2	3.7	3.0	3.1	2.5
Percent unable to carry on some activities	14.1	48.1	29.5	32.2	22.9
Activity limitation days last 6 mo					
Mean/case	0.5	3.9	3.8	1.2	1.2
Percent of cases with 1 + days	9.1	39.6	23.6	28.6	15.3
Percent seeking treatment					
Prior 6 mo	23.1	20.9	26.8	29.1	34.7
No. of cases	123	263	411	172	118
Prevalence (%)	12%	26%	41%	17%	12%

chronic pain states such as back pain and headache. Data summarized from a study of Von Korff et al.[23] (Table 3) support this view. For several pain parameters (years since initial onset, frequency, duration, intensity, and treatment seeking), TMD does indeed resemble other chronic pain conditions. However, TMD had a somewhat lower impact than some other pain conditions, eg, back pain and headache, on extent of interference and activity limitation days due to pain.

Current concepts of chronic pain derived from Fordyce,[24] Sternbach,[25] Turk and others,[26,27] suggest it may be at least as important to differentiate severe, persistent, and disabling chronic pain dysfunction from recurrent but not disabling pain states, as it is to differentially diagnose, for example, internal derangements, from other forms of TMD. An empirical approach to grading dysfunctional chronic pain has been developed by Von Korff and colleagues[11] using epidemiologic data. The highest grade of dysfunctional chronic pain, reflecting severe and persistent pain associated with seven or more disability days due to pain, was observed in about 16% of TMD clinic cases. These cases were found to be more likely to be psychologically impaired and to show higher rates for both use of health care and use of pain medications.

Taken together, current theories of chronic pain and available data support the need for classification criteria that differentiate the commonplace experience of recurrent or persistent pain from a chronic pain problem characterized by suffering, dysfunction, and a poor prognosis. If the dysfunctional aspects of chronic pain are not appropriately identified (even in the presence of an adequate biomedical/physiologic diagnosis), then treatments evolving from an incomplete assessment of TMD may yield ineffective or even deleterious outcomes. Thus, the complexities of classification of clinical TMD subtypes are compounded by the complexities of chronic pain classification.

Appreciable attention has been devoted to the diagnosis and classification of chronic pain. A multiaxial approach to the diagnosis and classification of chronic pain conditions, including TMD, seems to be emerging as most appropriate for the study of chronic pain states.[28–30] Such an approach recognizes the need for multidimensional assessment, so that measurement is not limited to defining cases only by physical findings, but is extended to include measures of psychological and psychosocial levels of function and impairment.

DIFFERENTIATION OF NORMAL VARIATION VERSUS PATHOPHYSIOLOGIC SIGNS

Epidemiologic studies can contribute invaluable perspectives regarding the relevance of clinical signs for diagnosing TMD. Evidence has been accumulating,[31] for example, that occlusion, once identified as a principal etiologic factor in some types of TMD, may not be relevant to the

diagnostic assessment of TMD. Similarly, Gale and coworkers[32] and we[33] have found that temporomandibular joint sounds detected during jaw function may be episodic, fluctuating (seemingly spontaneously) over time, and hence of unknown clinical significance for many TMD cases. Several workers[31,34] point out that for many TMD-related clinical variables, including joint sounds, occlusal variables, muscle palpation tenderness, and range of motion, little data are available to allow valid distinctions to be made between those clinical measurements that fall within the range of normal biologic variability and those clinical measurements that are valid indicators of pathophysiology.

The challenge of identifying signs with adequate specificity to be used as TMD diagnostic criteria in epidemiologic studies of TMD is paradoxical. To contribute data useful for diagnostic studies, logic investigations must include those clinical signs deemed adequate for formulating a differential diagnosis clinically. Yet, as we have seen, there remains uncertainty over which clinical measurements are valid markers of pathology, and whether they can appropriately be incorporated in epidemiologic studies.

Nevertheless, epidemiologic studies are uniquely suited for evaluating the validity of putative clinical signs, principally by comparing the distribution of clinical findings among cases contrasted with controls and then observing, through longitudinal studies, the prognostic value of clinical findings over time.

RELIABILITY OF CLINICAL MEASUREMENT

Our experience with reliability issues[35] in epidemiologic studies of TMD reinforces the need to attend to examiner reliability with rigor and reveals an interesting source of variability in clinical measurement not necessarily associated with examiner reliability. We found that training of examiners to rigid and carefully specified criteria is a crucial prerequisite for obtaining reliable data in TMD epidemiologic studies. Table 4 compiles published data[35] that demonstrate the effect of training on improving reliability among field examiners gathering data in our on-going epidemiologic study of TMD. Table 4 further reveals that, despite training, reliability remains low and only marginally acceptable for certain clinical measurements, notably detection of joint sounds. Table 4 indicates that those clinical measures that are associated with finely graded measuring instruments (eg, rulers for measuring vertical range of motion) tend to be associated with higher estimates of examiner reliability than measures scored categorically or involving subjective estimates on the part of the examiner (eg, assessing response to muscle palpation, classification of occlusion).

Table 4. Interexaminer Reliability Vertical Range of Motion and Joint Sounds on Opening: Intraclass Correlation. Classification of Occlusion, Jaw-Opening Patterns and Pain on Palpation and Function: Kappa Statistics

	Examiners	
Variables	*Initial*	*Retrained*
Vertical range of motion		
Opening without pain	0.90	NA
Opening unassisted	0.96	NA
Opening assisted	0.98	NA
Jaw-opening pattern	0.42	0.70
Occlusion	0.40	0.78
Palpation		
Extraoral muscles	0.47	0.65
Intraoral muscles	0.27	0.61
Temporomandibular joint	0.47	0.52
Function		
Lateral excursion	0.62	0.70
Protrusive excursion	0.73	NA
Joint sounds		
Palpation	0.39	0.68
Stethoscope	0.25	0.26

An additional source of variability in clinical measurement was uncovered through the repeated assessment of joint sounds. We observed that, for many individuals, joint sounds varied considerably from trial and that such seemingly spontaneous variability is relatively independent of issues related to examiner reliability. The particularly challenging aspect of this finding relates to distinguishing short- and long-term stability *v* change in biologic phenomena from reliability *v* unreliability of measuring procedures.

In any event, a critical influence on the adequacy of clinical findings concerns the reliability of clinical measurement. Clinical measurements in medicine, in general, are known to be associated with poor reliability, that is, reveal poor consistency among examiners.[36] Within the context of research, reliability of clinical measurement can be improved through training and calibration of examiners.[37] The World Health Organization requires epidemiologic studies to include estimates of reliability of measurement.[38] Because the validity of any measure can only be equal to or less than its reliability,[37] epidemiologic data gathered through clinical examiners with unkown reliability is, of unknown validity and hence of questionable use in population-based studies of diagnosis.

PROGRESSIVE VERSUS SELFLIMITING DISEASE ACTIVITY

Considerations of case definition, validity, and reliability of clinical findings aside, cross-sectional population-based studies of TMD can illuminate the problem of TMD diag-

nosis only in limited fashion. If TMD includes a set of chronic and episodic conditions, then longitudinal studies are essential to confirm the usefulness of diagnostic paradigms for an understanding of the etiology and natural history of these conditions.

Some chronic diseases lend themselves to etiologically based paradigms. Other chronic diseases (eg, hypertension) are classified based on manifestations rather than on etiologic principles. In either case the ability of a disease classification to explicate or predict disease progression is an important feature of the validity of the classification scheme.

Although there is abundant evidence that chronicity is a characteristic of TMD, there is little evidence that chronicity of TMD is typically associated with progressive pathophysiologic deterioration of the hard or soft tissues and structures that comprise the stomatognathic apparatus. There is even less evidence that chronicity of TMD leads to progressive impairment of important biologic functions associated with those tissues and structures, such as chronic impairment of respiration, deglutition, mastication, communication, and verbal or nonverbal facial expression of emotion.[34]

Rugh and Solberg[39] speculated that TMD may represent a set of largely selflimiting conditions whose prominent characteristic is that they do not become associated with significant physical deterioration or impairment. There is agreement[40,41] for example, that major clinical signs and symptoms of TMD do not increase in prevalence with age. Opposingly, we have presented evidence that the prevalence of TMD-related pain diminishes appreciably after the age of 65.

Analogous findings with regard to gender are equally provocative. We have previously reported no significant gender differences for the most important clinical signs of TMD, with the exception of vertical range of motion (males show a consistently larger range of vertical jaw motion, which we attribute to gender differences in physical stature). Although the preponderance of females in TMD clinic populations is well known, there is presently no evidence that the pathobiology of TMD pursues a different form of disease activity in one sex versus another, although such a theoretical possibility cannot be dismissed.

Thus, the application of longitudinal epidemiologic methods can elucidate whether the course of chronic TMD is associated with pathobiologic disease progression that results in significant impairment or, conversely, whether TMD is typically a selflimiting episodic illness whose chronicity is not typically associated with biologic deterioration and loss of physical capacities.

These considerations point to the importance of identifying the most significant end points in studies of the clinical course of TMD. For example, classification of neoplasm has been evaluated in terms of prediction of tumor progression and consequent mortality: classification of hypertension has been evaluated in terms of prediction of myocardial infarcts, stroke, and associated mortality. These are measurable biologic end points that a largely biomedical model of disease could reliably predict. If the most significant "end points" of TMD are persistent pain and disability, these represent biobehavioral endpoints for which a largely biomedical model of disease is unlikely to be adequate.

CONCLUSIONS AND RECOMMENDATIONS

Disease can be defined, descriptively, as combinations of signs and symptoms, or by the anatomic structures involved. Disease can also be classified by etiology and pathogenic mechanisms, or, alternatively, by organ derangements and their effects on function.[42] New concepts of "illness" and "illness behavior"[27,43] redefine disease as involving integrated biologic, psychologic, and social processes. The shift to a bio-psycho-social[44] model of disease affirms the view that studying disease without studying its impact on the individual on social roles and on social institutions (eg, the health care delivery system) results in an incomplete understanding of disease and associated dysfunction and disability.

The implications of a bio-psycho-social model for epidemiologic studies of TMD are to broaden the scope of research to include not only assessment of disease activity (biologic state of the disease), but also pain and suffering (perception and appraisal of pain and emotional status), impairment (loss or capacity at the organ level), and dysfunction (disturbance or lack in ability to perform at the person level, eg, disability).

With regard to the diagnosis of TMD, as with any chronic pain condition, there is already at least tacit recognition by many clinicians that understanding the patient's problem means more than understanding the patient's pathophysiology. It is reasonable to suggest two foci for investigation when TMD patients are examined. One relates to the assessment of disease activity in the pursuit of a clinical diagnosis. The second focus is on the functional status of the patient, that is, how to change the patient's pain behavior and psychosocial status to ameliorate dysfunction independent of, or in conjunction with, the biologically focused TMD diagnosis.

Recommendation for Approaches to Studying the Diagnosis of TMD

Epidemiologic investigations into the diagnosis of TMD should be responsive to issues of case definition, adequacy of clinical assessment, reliability of clinical measure-

ment and improved delineation of end points, and understanding TMD as a progressive or selflimiting pain and dysfunction condition. In addition, population-based investigations of diagnostic schemes for TMD should be multiaxial in design, reflecting present-day understanding of the biological, psychological, and social impact of chronic pain conditions on impairment, dysfunction, and disability.

Recommendations from Currently Available TMD Data

Because of the lack of reproducible diagnostic criteria suitable for epidemiologic research, it is not possible to derive reliable prevalence estimates for diagnostic subtypes of TMD. In effect, we still know very little about the descriptive epidemiology of MPD, ID, and DJD, presumably the most common forms of TMD. The approach we have taken, as discussed, has been to systematically gather data on clinic cases and community cases and controls for the broadest array of putative clinical signs and symptoms, but without regard to a priori definitions for TMD subtypes. Such data are amenable to subsequent analyses from diverse theoretical and clinical perspectives.[15,16] Diagnostic algorithms based on alternative sets of operationally defined clinical and behavioral criteria can yield prevalence estimates and permit investigation of natural history and clinical course of TMD diagnostic categories so defined.

Recommendation 1. For the short-term, in light of limited present information, especially regarding reliable and valid means for end-point assessments, we recommend continuing the described epidemiologic approach to diagnostic studies of TMD. Examination and interview items for studies are available from our own investigations. Other investigators[45–48] have provided useful compilations of clinical examination and/or questionnaire items, all showing good agreement, which are suitable for use in large scale studies.

Recommendations Based on Experiences in Related Fields

It is important to note that comparable problems of evolving useful diagnostic and classification schemes have been intensively examined in two related clinical fields—rheumatic disease[49] and spinal disorders.[50] In both cases, concerted multidisciplinary efforts have resulted in diagnostic approaches based on functional assessment. This approach has direct implications for the design of epidemiologic studies as well as for treatment and research. In both instances public health agencies and the private sector joined in supporting a sustained effort to organize available scientific and clinical information; also, in both cases, weight was given to evidence that met criteria for scientific admissability.

The endeavour in the field of rheumatic disease[49] was more research oriented. It yielded methodologic criteria for assessing biomedical and psychological functioning, and for choosing appropriate therapeutic interventions based on comprehensive reviews of the literature and the distilled views of an interdisciplinary group of clinicians and researchers.

An alternative potentially useful model for studying the problem of diagnosis may be found in the approach taken by the Quebec Task Force on Spinal Disorders.[50] This multidisciplinary task force included specialists from medicine, surgery, behavioral medicine, occupational and physical therapy, economics, biostatistics, and epidemiology. Their report acknowledged that pain is the primary, and often the only, symptom in the vast majority of spinal disorder cases; that it is difficult to identify the precise origin of pain; that pain remains nonspecific and typically impossible to corroborate with clinical observations or through histologic studies; and, that there is often no modification of tissue observable through current methods. They found the terminology for spinal disorders to be poorly specified and the literature replete with diagnostic terms.

The task force identified lack of uniformity in diagnostic terminology as a major barrier and a key challenge to their study of the problem of diagnosis. The description they provide concerning the state of the art of diagnosis of spinal disorders bears remarkable similarities to the problems encountered in the study of TMD diagnosis.

The approach taken by the Quebec Task Force has many elements in common with the methods used for the coordinated study of research into the diagnosis and management of rheumatoid disease, but with major differences. The task force approach explored issues in greater depth and developed detailed scientific criteria that were applied in an exhaustive review of the literature. This review was used to derive decision-making matrices to allow scientifically based assessment and functional classification of spinal disorders, and a rational basis for managing those disorders, including the specification of endpoint criteria for respective stages of their classification system. Finally, the task force specified areas of future research.

Recommendation 2. Based on the above efforts, a long-term recommendation for TMD research is to develope scientific criteria for:

a. a comprehensive diagnostic scheme for TMD that includes specification of measures for assessing disease activity, impairment of physical function, and psychosocial function

b. specification of a rational clinical decision-making system for identifying treatments consistent with diagnostic assessment, including the specification of the most significant end-points for TMD treatment outcome
c. development of short, intermediate, and long-term research objectives, including, at the earliest stages, carefully designed epidemiologic studies, to further knowledge about the pathobiology of TMD and its personal and social sequelae

Recommendation 3. An additional longer-term recommendation, either as an alternative or subsequent to the second recommendation, is to identify a multidisciplinary panel of experts to evaluate the feasibility of applying to TMD the study approach taken by the Quebec Task Force for the scientific assessment of spinal disorders.

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