



REVIEW ARTICLE



The prevalence of oral stage dysphagia in adults presenting with temporomandibular disorders: a systematic review and meta-analysis

Órla Gilheaney^a, Sibylle Béchet^a, Patrick Kerr^b, Ciaran Kenny^a , Shauna Smith^a, Rita Kouider^a, Rachel Kidd^a and Margaret Walshe^a 

^aDepartment of Clinical Speech and Language Studies, Trinity College Dublin, Dublin, Ireland; ^bSchool of Medicine, Trinity Biomedical Sciences Institute, Trinity College, Dublin, Ireland

ABSTRACT

Objective: Temporomandibular disorders (TMDs) are the most commonly experienced non-dental orofacial pain disorders, with pain and dysfunction potentially resulting in oral stage dysphagia (OD). However, limited research has been conducted on this condition, with potential negative effects on clinical practice. Therefore, the aim of this study was to determine the prevalence of OD in adults presenting with TMDs, diagnosed as per the Research Diagnostic Criteria for Temporomandibular Disorders or the Diagnostic Criteria for Temporomandibular Disorders protocols.

Material and methods: A systematic review of the literature was completed. Nine electronic databases were searched from inception to January 2017, with no date/language restriction applied. Grey literature, conference proceedings, and reference lists were also searched. Studies presenting original data regarding OD prevalence in adults presenting with TMDs were included if they investigated impaired swallowing, mastication, masticatory pain or fatigue, or weight loss. Study eligibility and quality were assessed by two independent reviewers. Methodological quality was assessed using the Down's and Black tool.

Results and conclusions: This search yielded 20 eligible studies. Swallowing itself was impaired in only 9.3% of patients with TMDs. A range of additional OD signs and symptoms were also commonly reported (e.g. masticatory pain (87.4%) and fatigue (62%)). Study limitations included the small number of studies which were eligible for inclusion. As signs and symptoms of OD are frequently reported by patients with TMDs, psychometrically robust prospective research is warranted to determine current and optimal management of this condition.

ARTICLE HISTORY

Received 26 June 2017
Revised 22 November 2017
Accepted 31 December 2017

KEYWORDS

Dysphagia; temporomandibular joint; temporomandibular joint disorder; prevalence

Introduction

Temporomandibular disorders (TMDs) are a cluster of conditions caused by alterations in the structure and/or function of the temporomandibular joint (TMJ), the wider masticatory muscle system, and/or osseous components, which are commonly characterised by heterogeneous signs and symptoms [1–8]. TMDs are the most frequent orofacial pain disorders of non-dental origin, and are also reported to be the second most common musculoskeletal and neuromuscular disorder after lower back pain, with up to a striking 93% of the general population showing at least one TMD sign or symptom on examination, and 10–20% of these individuals seeking treatment [9–17]. Although the clinical presentation of TMDs are frequently heterogeneous [1], commonly experienced signs and symptoms include: pain, dysfunction, and fatigue of the TMJ and muscles of mastication, limitations of mandibular movement and mouth opening, impaired oral transit, and the potential for unintentional weight loss [2,18–21]. These functional difficulties have the potential to combine to impair typical eating, drinking, and swallowing, causing oral

stage dysphagia (OD), which may subsequently impact on quality of life (QOL) [22–24].

Despite the potential for adults who present with TMDs to develop OD which may impact upon both functioning and well-being, there has been limited epidemiological research into this condition. Also, various methodological limitations within the available evidence render it difficult to determine the true prevalence and nature of TMD-related OD [18]. These issues include: (1) diversity in the definition of key terms such as what constitutes a diagnosis of OD; (2) reported historical use of unspecified subjective assessments which may not have adequate sensitivity to detect OD; (3) increasing use of the Research Diagnostic Criteria for Temporomandibular Disorders [25] and recently updated Diagnostic Criteria for Temporomandibular Disorders [22] protocols in clinical and academic fields, which although standardised, address TMD-related OD in a brief manner; (4) lack of reporting of specific TMD diagnoses of participants; (5) and lack of adequate description of and adjustment for confounding factors. As such, a valid and reliable description of the epidemiology of TMD-related OD is required in order

to describe the nature and significance of this condition, and to provide rationale for improvements in its typical identification and management practices. Therefore, the aim of this study was to conduct a systematic review and meta-analysis of the epidemiology of the following signs and symptoms of OD in adults presenting with TMDs: impaired swallowing and mastication, masticatory pain and fatigue, and unintentional weight loss.

Material and methods

This systematic review was conducted in accordance with The Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement [26] and MOOSE Guidelines for Meta-Analyses and Systematic Reviews of Observational Studies [27]. The protocol was prospectively published on the University of York Centre for Reviews and Dissemination Prospero database (Registration number: CRD42016050846) [28]. For the purpose of this study, the definition of OD was as follows: sensory and/or motor difficulties in the movement of a liquid or solid bolus from the oral cavity to the esophagus, inclusive of concomitant emotional, cognitive, and functional difficulties [29]. Originally, the aim of this study was to study the prevalence of broad oropharyngeal dysphagia [28]. However, on further reading and reflection, the focus was narrowed to purely oral stage dysphagia-related difficulties, as these are most relevant to disorders of the structure or function of the TMJ complex.

Eligibility criteria

All published/unpublished records presenting original prevalence figures were eligible for inclusion, with no language, geographic, or date limitations applied. Case reports were excluded due to criticisms regarding their typically low levels of evidence. Prevalence figures were sought regarding humans aged 18 years and over presenting with oral and/or oral preparatory signs/symptoms of OD related to TMDs, as diagnosed using the RDC/TMD [25] or the DC/TMD [22]. The RDCTMD was the most widely used TMD diagnostic protocol since its publication in 1992 until the 2014 publication of the DC/TMD [30]. This system facilitates the characterisation of both the physical and emotional difficulties associated with TMDs and the protocol was based on the biopsychosocial model of pain. The RDC/TMD protocol includes a comprehensive Axis I physical assessment and Axis II examination of psychosocial functioning and pain-related disability. The more recent DC/TMD system [22] is a dual axis assessment tool which provides evidence-based criteria for clinicians to apply when assessing patients and which facilitates trans-disciplinary communication. This system present a more comprehensive and contemporaneous selection of assessment tools than the RDC/TMD for both the brief screening for and in-depth examination of a spectrum of simple and complex TMDs [22]. No restrictions were applied regarding sex, race, disease duration, severity, age-of-onset, or recruitment location. Individuals were not eligible for inclusion if they had a

history of conditions which may result in mandibular or orofacial dysfunction (e.g. head and/or neck cancer, comorbid/congenital conditions of the maxillofacial area, or orthopaedic or neurological trauma to the orofacial region).

Outcomes of interest

Outcomes of interest in this systematic review included:

1. Impaired deglutition and mastication as reported subjectively and/or detected objectively via clinical examination, interviews, questionnaires, and/or imaging techniques;
2. Masticatory pain as reported in interviews, questionnaires, or as rated using subjective scales;
3. Masticatory fatigue as reported via interviews, questionnaires, subjective scales, or detected using clinical or electromyographic (EMG) assessment; and
4. Unintentional weight loss related to OD as reported by the patient or detected via clinical examinations.

Originally, investigators also aimed to research the prevalence of the consumption of texture modified diets by the cohort of interest within this study [28]. However, in order to provide clarity via the separation of epidemiological and management issues, and to afford sufficient attention to these parameters, this estimate will be separately presented in a subsequent report.

Data sources

A sensitive search strategy incorporating filters, key-text and Medical Subject Headings was systematically employed across databases by two independent reviewers (Appendix 1). The databases searched from inception to January 2017 included: EMBASE, PubMed, CINAHL, Web of Science, Elsevier Scopus, Science Direct, AMED, The Cochrane Database of Systematic Reviews, and ProQuest Dissertations and Theses A & I. All search results were subsequently exported to the Zotero bibliographic system (www.zotero.org). Subsequent to duplicate deletion, two authors independently screened titles/abstracts in order to exclude obviously irrelevant papers. A third author was available to independently mediate disputes, if required. The senior author also conducted hand-searches of annual scientific meeting proceedings of the International Association for Dental Research (published in the *Journal of Dental Research*) and the European Society for Swallowing Disorders (published in *Dysphagia*). Hand-searches of the reference lists of studies ultimately included in the systematic review and meta-analysis were also conducted, with one eligible study identified [31]. Subsequent to the completion of the above systematic searches, the senior author also further searched the Google Scholar database in an effort to identify records not indexed in the initially searched databases, resulting in 13 additional eligible studies [32–44]. Eligible records which were included in the systematic review will be discussed in subsequent sections.

Data extraction process and data items

A previously piloted electronic form [45] was used in data extraction by six independent reviewers. Data were extracted regarding study design, setting, and location, participant demographics, outcome measurement and data sources, prevalence and statistical analysis, among other parameters. Reviewers reached 100% consensus regarding data extracted. One author not involved in data extraction was available to mediate disputes if they occurred. Missing/unclear data was addressed via the senior author contacting primary authors of studies published within the previous 10 years. The figure of 10 years was chosen to accommodate the usual 5-year research retention period and to circumvent exclusion of records which were published more than 5 years ago, but primary authors had retained records past this period for retrospective analysis. Records were excluded following no response to two contact attempts.

Assessment of methodological quality

Two independent reviewers assessed the methodological quality of included studies utilising a previously piloted [45] modified Down's and Black Tool [46] which omitted criteria

deemed irrelevant to the aims of this study (e.g. intervention, adverse events, blinding and randomisation) (Appendix 2). Primary studies which recruited a comparison group were marked out of 18 points, while those without comparison groups were rated out of 16 points, as 2 criteria pertained to the recruitment of a control group.

Summary measures and synthesis of results

Included studies were first described descriptively, with subsequent statistical analysis. Fixed and random effects meta-analyses of prevalence estimates were carried out utilising the Microsoft Excel package [47] and the MedCalc system [48]. Prevalence figures were presented using 95% confidence intervals, with forest plots constructed for all estimates.

Results

Study identification

Systematic searches yielded 10,248 results, as shown in the PRISMA figure (Figure 1). Following the exclusion of 4245 duplicates and 5381 records based on their titles, abstract,

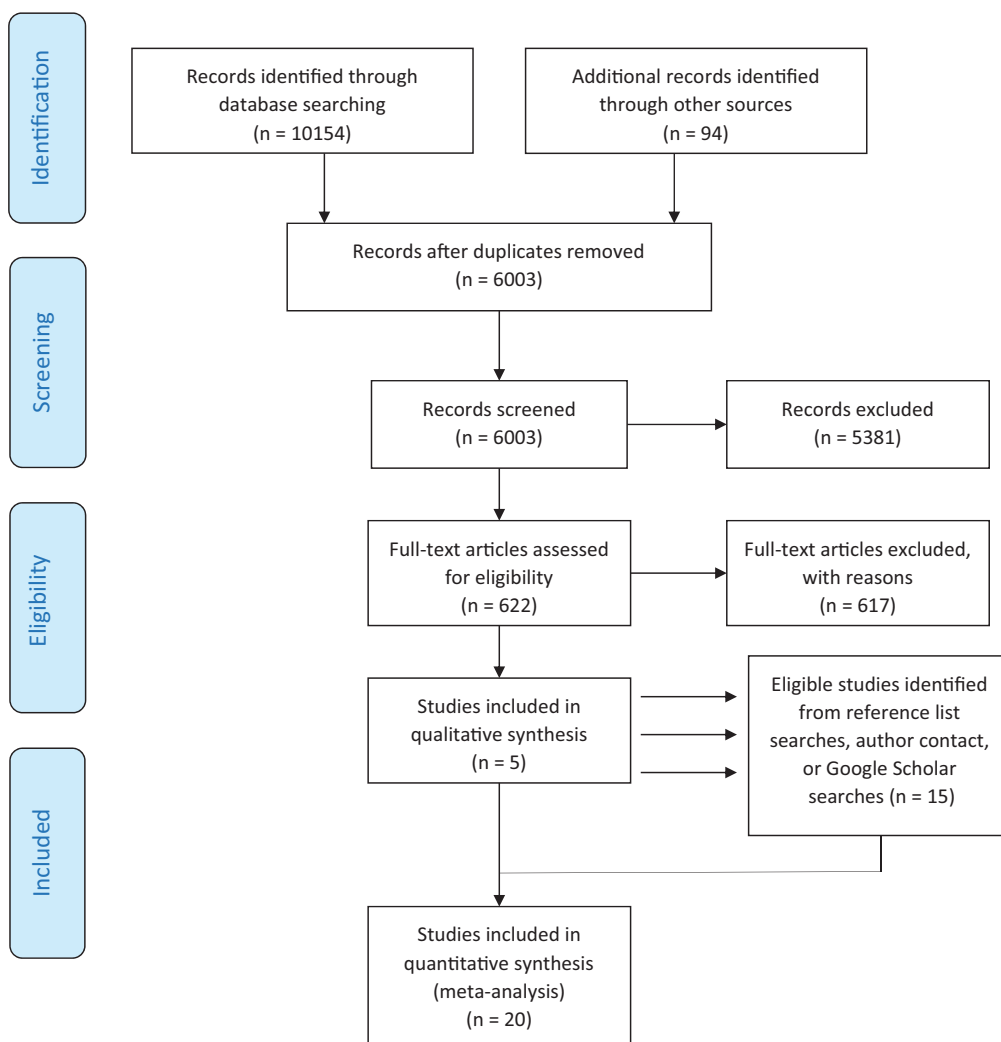


Figure 1. PRISMA diagram.

and/or keywords, two independent reviewers examined 622 full-text records, with a third author available to independently mediate disputes, if required. At this stage, 617 of these full-text records were excluded. Supplementary Google Scholar searches identified 13 further eligible studies [32–44]. An additional eligible article was identified from reference list searches [31] and one further article was identified via author contact [49].

Missing/inconsistent/insufficient data were addressed by the senior author contacting primary authors of studies published within the last 10 years, in order to allow for both the typical 5-year retention period and to also avoid forcible exclusion of studies if they were dated beyond this period yet records were retained for post-hoc analysis. Records were excluded in the case of two unanswered contact attempts. In total, during both foundational and supplementary Google searches, 291 contact attempts were made regarding 155 potentially eligible records. For 117 of these studies, missing data were sought, while 38 communications related to article access. Contact led to the confirmation of 12 eligible studies, the exclusion of 3 studies excluded due to inappropriate participants, 24 excluded due to insufficient data, 98 excluded due to no response, 5 excluded due to inappropriate research objectives, 3 due to use of inappropriate outcome measurement tools, and 10 studies excluded due to inability to access records. Ultimately, 20 studies satisfied the inclusion criteria, had data extracted, and were included in analysis.

Study characteristics

Included study characteristics are described in Table 1.

Half of included records ($n=10$) were case-control studies (50%), while 20% ($n=4$) were descriptive observational studies, 3 (15%) were RCTs, 10% ($n=2$) were cross-sectional studies, and 5% ($n=1$) were prospective cohort studies. Study locations included Europe ($n=7/35\%$), South America ($n=5/25\%$), North America ($n=3/15\%$), Asia ($5\%/n=1$), Australia ($5\%/n=1$), and Israel ($5\%/n=1$). A small cohort did not specify study location ($n=2/10\%$) [36,50]. Study settings varied, with the majority conducted in university clinics ($n=10/50\%$), university teaching hospitals ($n=3/15\%$), and dual study settings ($n=3/15\%$). It was unclear what the settings of two studies were [36,50].

Sources of assessment data varied, with 19 studies (95%) utilising the RDC/TMD [25] and 1 study (5%) [51] employing the DC/TMD system [22]. Objective assessments were employed in six studies (30%) (Table 1). Assessments of mastication were used in eight studies (40%). Visual analogue scales were used in six studies (30%) to gather patient-reported data. Subjective questionnaires were used in 16 studies (80%), with a range of 24 such tools being employed.

Description of participant demographics

Data regarding 1581 patients were included, with the pooled age range of available and eligible participant data being 18–88 years of age. A small cohort of included studies did

not provide mean ages of eligible participants ($n=4/20\%$), while 60% ($n=12$) of eligible studies did not provide details regarding age ranges. A total of 1136 females and 351 males (3.23:1; female: male) were included, while two studies [42,44] did not provide details on gender. The diagnoses with which eligible participants presented are shown in Table 2. The discrepancy between the number of participants recruited ($n=1581$) and the total number of diagnoses presented below ($n=1623$) is due to certain studies applying diagnostic criteria to both TMJs, while others classified patients according to the unilateral TMJ which was most impaired.

The most frequently reported diagnoses were myofascial pain disorder (MFP) ($n=672/41.2\%$), Disc displacements (disc displacement with reduction (DDWR): $n=275/16.9\%$; disc displacement without reduction DDWOR: $n=95/5.8\%$), and arthralgia ($n=190/11.6\%$). A large cohort were classified as presenting with dual diagnoses ($n=229/14\%$), with MFP + DD/osteoarthritis (OA) being the most frequent ($n=91/5.6\%$). Two studies provided unclear diagnoses, classifying participants as presenting with either DD or OA ($n=32$) [39], or MFP combined with DDWR or OA ($n=91$) [42] (Table 2).

Assessment of methodological quality of included studies

Two authors independently reached 100% consensus relating to quality ratings, without disputes. Utilising the Down's and Black tool, studies recruiting a comparison group were awarded an average of 11.3, indicating a mean rating of moderate quality (Table 3).

The items primarily responsible for lower methodological quality ratings were: inadequate/lack of description of whether participants who were prepared to participate were representative of the entire population ($n=12/60\%$); inadequate description of patients who were lost to follow-up ($n=12/60\%$); and inadequate description of the distribution of and adjustment for confounding factors ($n=10/50\%$). Contributing to positive ratings was the judgement that all studies ($n=20$) clearly described primary aims, hypotheses, and outcomes; 95% ($n=19$) utilised accurate outcome measurement tools; 95% ($n=19$) utilised appropriate statistical tests to assess main outcomes; and the characteristics of participants were clearly described in the majority ($n=17/85\%$) of included studies.

Prevalence of investigated outcomes

Based on data extracted from 7 studies ($n=215$ patients) [32,34,41,49,50,52,53], the prevalence of impaired deglutition was estimated to be 9.3% (95% CI: 2.1–20.86%) (Figure 2).

Impaired mastication was reported in 16 studies ($n=1064$ patients) [31–37,40,41,43,49–54], with this prevalence estimated to be 52.67% (95% CI: 37.182–67.91%) (Figure 3).

Masticatory pain was reported in six studies ($n=734$ patients) [31,38,39,42,44,49], with this prevalence estimated to be 87.38% (95% CI: 74–96.39%) (Figure 4).

Table 1. Characteristics of included studies.

Citation/Year/Region/ Setting	Study design	No. of TMD patients studied	Female: male ratio	Mean age (SD) of TMD patients (years)	Mean age (range) of onset (years)	Mean disease duration (range) (months)	Relevant outcome measured	Sources of assessment data	Study quality score: Down's and Black checklist
Baker et al., [41]/2015/ Sweden/University clinic	Randomised control trial (RCT)	34	10.3:1	38.9 (± 15 y)	Unclear	49.1 (unclear)	Impaired swallowing: 5.88%; Impaired mastication: 20.58%	RDC/TMD, X-ray, magnetic resonance imaging (MRI), IMPACT question- naire, visual analogue scale, JFLS- 8, Graded Chronic Pain Scale (GCPS), Symptom Checklist-90- Revised (SCL-90R)	15/18
Wanman [43]/2012/ Sweden/University clinic	Case-control	108	5.23:1	40 (±13)	Unclear	Unclear	Impaired mastication: 62.96%	RDC/TMD, subjective questionnaire, endurance tests, visual analog scale	11/18
Mapelli et al., [34]/2016/ Brazil/University clinic	Case-control	30	9:1	30.5 (Unclear)	Unclear	59.2 (7-240)	Impaired mastication: 76.6%	RDC/TMD, ProTMDmulti question- naire, EMG	13/18
Foteder et al., [35]/2015/ India/University hos- pital clinic	Cross-sectional	195	5.91:1	34.8 (± 17.2)	Unclear	Unclear	Impaired mastication: 54.32%; Masticatory fatigue: 50.61%	RDC/TMD, patient interviews, case history, OHIP-14	12/18
Abrahamsson [37]/2013/ Sweden/University hospital clinic	Case-control	90	1.37:1	22 (± 7.4)	Unclear	Unclear	Impaired mastication: 75.2%	RDC/TMD, visual analog scale, sub- jective questionnaire, self-reported masticatory ability, masticatory performance test, dental exam	16/18
Ferreira et al. [32]/2016/ Italy/Specialised pri- vate practice facial pain clinic and univer- sity hospital clinic	Case-control	12	12:0	37 (±16)	Unclear	39 (unclear)	Impaired swallowing: 0%; Impaired mas- tication: 41.66%; Masticatory fatigue: 66.6%	RDC/TMD, ProTMDmulti, orofacial myofunctional evaluation with scores (OMES), near infrared spectroscopy	15/18
La Touche et al., [42]/ 2015/Spain/Specialised private practice facial pain clinic and public health centre	Case-control	33	Unclear	Unclear	Unclear	Unclear	Masticatory pain: 96.38%; Masticatory fatigue: 98.79%	RDC/TMD, subjective questionnaire, visual analog scale, Pain Catastrophizing Scale (PCS), provo- cation chewing test, pressure pain threshold, maximal mouth open- ing, Neck Disability Index, Headache Impact Test-6	16/18
Dougall et al., [39]/2012/ America/General den- tal practice	Descriptive observational	185	4.28:1	42.68 (unclear)	Unclear	Unclear	Masticatory pain: 89.72%	RDC/TMD, Characteristic Pain Index (CPI), GCPS, chewing performance test, Medical Outcomes Shortform- 36 Status Questionnaire, SCL-90R, Beck Depression Inventory	11/16
Khawaja et al., [50]/ 2015/America/ University clinic	Descriptive observational	94	1.47:1	34.2 (±12.2)	Unclear	Unclear	Impaired mastication: 56.38%	DC/TMD, Oral Behaviours Checklist (OBC), GCPS, CPI, MRI, Patient Health Questionnaire (PHQ), Generalised Anxiety Disorder Scale	11/16
Maffei et al., [51]/2012/ Unclear/Unclear	Descriptive observational	10	1.5:1	Unclear	Unclear	Unclear	Impaired swallowing: 70%; Impaired mastication: 90%	RDC/TMD, VFSS, SLT OD assessment	6/16
De Felicio et al., [52]/ 2012a/Brazil/University clinic	Case-control	30	1:0	30 (±8)	Unclear	Unclear (6-108)	Impaired swallowing: 0%; Impaired mas- tication: 0%	RDC/TMD, EMG, OMES, ProTMDmulti	12/18
Da Silva et al., [49]/2011/ Brazil/University clinic	Case-control	70	0.84:1	53 (unclear)	Unclear	Unclear	Impaired swallowing: 16.6%; Impaired mastication: 11.1%;	RDC/TMD, saliva flow evaluation, xerostomia questionnaire, dental exam	12/18

(continued)

Table 1. Continued

Citation/Year/Region/ Setting	Study design	No. of TMD patients studied	Female: male ratio	Mean age (SD) of TMD patients (years)	Mean age (range) of onset (years)	Mean disease duration (range) (months)	Relevant outcome measured	Sources of assessment data	Study quality score: Down's and Black and checklist
Raphael & Marbach [53]/ 2001/America/ University clinic and general dental practice	RCT	63	1:0	33.7 (±10.9)	Unclear	60 (UNC)	Masticatory pain: 72.2% Impaired swallowing: 7.93%; Impaired mastication: 49.2%;	RDC/TMD, SCL-90, clinical exam, self- reports of pain, functional out- come assessment	16/18
Barros et al., [33]/2008/ Brazil/University clinic	Cross-sectional	132	4.92:1	36.5 (±13.5)	Unclear	Unclear	Impaired mastication: 79.51%; Masticatory fatigue: 33.73%	RDC/TMD, OHIP-14, clinical exam, temporomandibular index	13/16
Radke et al., [36]/2014/ Unclear/Unclear	Descriptive observational	28	1:1	34.5 (±14.0)	Unclear	Unclear	Impaired mastication: 100%	RDC/TMD, magnetic incisor point tracking	7/16
Gonçalves et al., [40]/ 2011/Brazil/University hospital clinic	Case-control	28	28:0	Unclear	Unclear	Unclear	Impaired mastication: 0%	RDC/TMD, anthropomorphic assess- ment, ovulation prediction test, maximum occlusal force, sieve method assessment of masticatory performance	15/18
Brandini et al., [54]/2011/ Australia/Unspecified hospital	Case-control	15	15:0	31.3 (±10.8)	Unclear	Unclear	Impaired mastication: 60%	RDC/TMD, chewing task, numerical rating scale-II, Pain Self-efficacy Questionnaire, Fear-Pain Questionnaire-III, DAS-44, PCS	13/18
Reizmann et al., [31]/ 2007/Germany/ University clinic	Case-control	312	3.16:1	38.6 (±15.6)	Unclear	Unclear	Impaired mastication: 82.07%; Masticatory pain: 68.53%; Masticatory fatigue: 45.91%	RDC/TMD, German version of OHIP, GCPS, Beschwerden-Liste, the Center for Epidemiologic Studies Depression Scale	10/18
Gavish et al., [38]/2002/ Israel/University clinic	Prospective cohort	49	2.76:1	29.2 (±7.8)	Unclear	Unclear	Masticatory pain: 83.67%;	RDC/TMD, clinical exam, experimental chewing task, palpation, visual analogue scale	16/18
Michelotti et al., [44]/ 2002/Italy/University clinic	RCT	63	Unclear	Unclear	Unclear	Unclear	Masticatory pain: 100%	RDC/TMD, anamnestic and clinical scores, measures of pressure pain thresholds, pain during gum chew- ing, and spontaneous pain, visual analogue scales	4/18

Table 2. Diagnoses of participants eligible for inclusion.

Diagnosis	Osteoarthritis	Disc displacement with reduction + myofascial pain	Disc displacement without reduction + arthralgia	Disc displacement with reduction + myalgia	Myofascial pain + osteoarthritis/arthralgia	Disc displacement without reduction + osteoarthritis	Disc displacement with reduction/osteoarthritis	Myofascial pain + osteoarthritis	Arthralgia	Disc displacement without reduction	Arthralgia	Disc displacement with reduction	Myofascial pain
Number of TMD diagnoses reported (n = 1623)	13	30	34	40	34	34	91	59	58	95	190	275	672

Table 3. Down's and Black checklist rating criteria.

Descriptor	Criteria for studies with comparison group	Criteria for studies with no comparison group
Poor quality	0–4	0–3
Fair quality	5–9	4–7
Moderate quality	10–14	8–11
Good quality	15–18	12–16

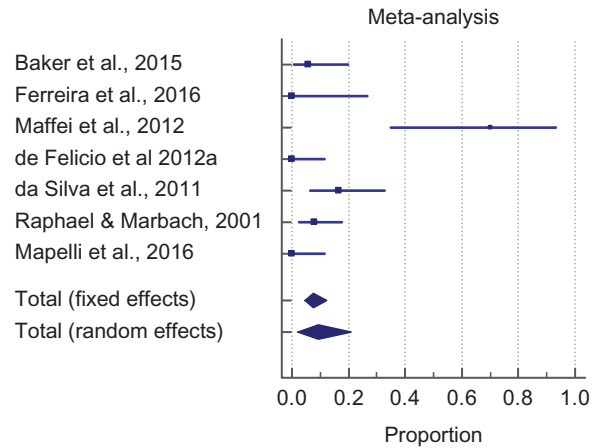


Figure 2. Forest plot of prevalence of impaired swallowing.

Masticatory fatigue was reported in five studies ($n = 577$ patients) [31–33,35,42], with prevalence calculated to be 61.95% (95% CI: 34.16–86.03%) (Figure 5).

No eligible records were identified which estimated the prevalence of weight loss in the cohort of interest, thus preventing analysis.

Discussion

This systematic review and meta-analysis has highlighted that a broad range of signs and symptoms of OD are frequently reported by adult patients with TMDs, although this condition has typically received limited amounts of clinical and research attention. Interestingly, despite this broad range of reported difficulties, the primary outcome under investigation, impaired swallowing, was detected in only 9.3% of included participants. However, potentially contributing to this low prevalence, studies which were eligible for inclusion were assessed to be heterogeneous, which may result in the true prevalence being higher than that estimated within this study. Also, the method by which swallowing was assessed in primary studies may have influenced the overall prevalence estimate, with all eligible studies [32,34,41,49,50,52,53] using subjective assessments, which had varied psychometric properties and were unspecified in certain studies [49,50,53]. The observed reliance on unspecified and varied subjective measures emphasizes the need for both improvements in the reporting of the details and psychometric properties of subjective outcome measurements used, and the frequency of use of objective tools within primary epidemiology studies. Finally, theoretical issues may also impact negatively on the homogeneity, validity, and reliability of research findings in this field. For example: differing theoretical definitions of

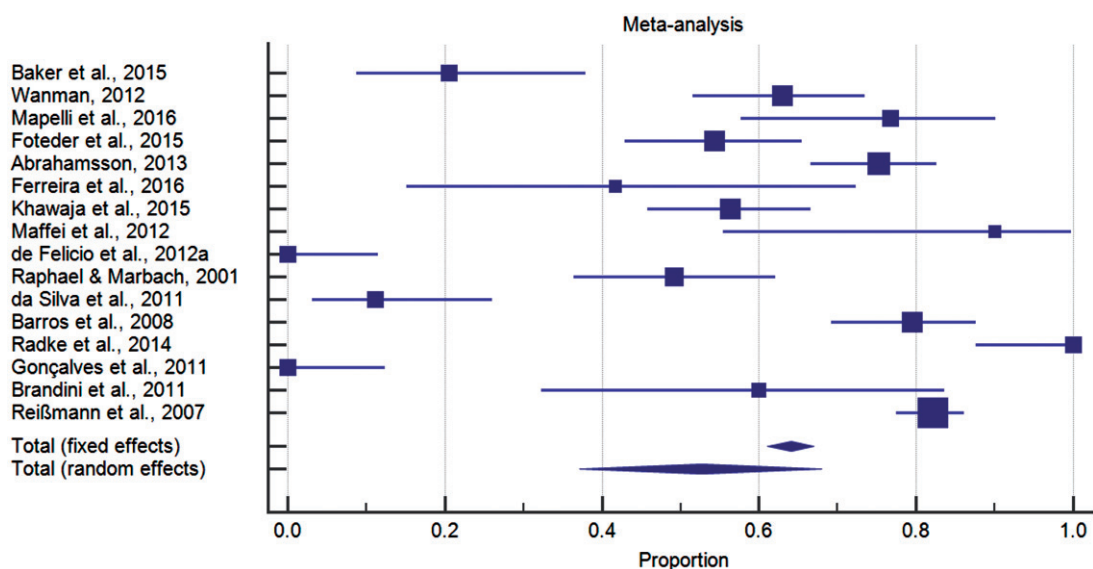


Figure 3. Forest plot of prevalence of impaired mastication.

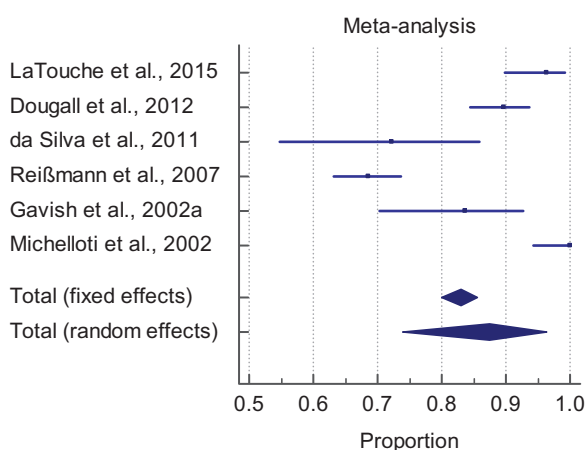


Figure 4. Forest plot of prevalence of masticatory pain.

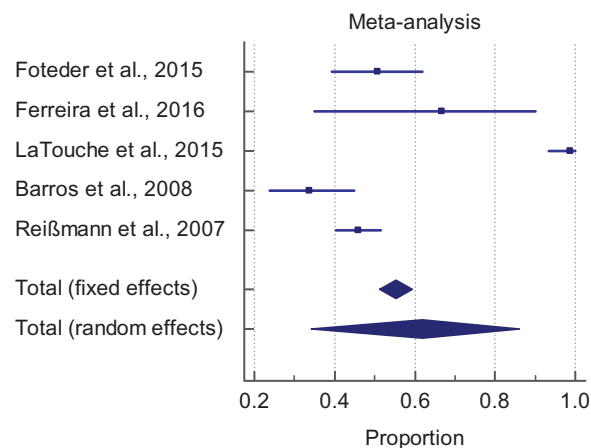


Figure 5. Forest plot of prevalence of masticatory fatigue.

what constitutes a diagnosis of swallowing difficulties in this patient group have been adopted, and many studies which have investigated this parameter have not specified their operational definition of ‘impaired deglutition’ (e.g. impaired transit of the bolus from the oral to the pharyngeal cavity, impaired triggering of the pharyngeal swallow, or impaired protection of the airway, etc.). As such, in critical analysis of studies conducted in this field, researchers are limited in their ability to construct a cohesive picture of findings due to these limitations. As dysphagia caused by a range of aetiologies has documented effects on functioning, levels of activity and participation, and QOL [55], it is essential that this condition receives greater attention within the field of TMD research.

In addition to the investigation of the primary outcome of deglutition, a range of other signs and symptoms were also investigated. For example: impaired mastication was estimated to be experienced by approximately 53% of adult patients presenting with TMDs in this study. However, only 50% of the eligible studies which reported figures on impaired mastication [32,34,37,40,43,50,52] collected data using chewing performance or endurance tasks, with the

remaining studies relying on subjective patient reports [31,33,35,36,41,49,51,53]. As such, there is the potential for the under-identification of perceptually mild masticatory impairments when only subjective data are collected.

However, masticatory impairments are often considered central to the experience of TMDs and therefore, these findings may not be viewed as novel. Yet, it is crucial to note that specific aspects of chewing difficulties have infrequently been discussed in the literature (e.g. masticatory pain or fatigue). In this study, masticatory pain was estimated to be experienced by 87% of individuals, indicating that this may be a commonly experienced difficulty, although the precise pathophysiology of this pain and discomfort across TMD diagnostic classifications is unknown. In addition, masticatory fatigue was present in approximately 62% of individuals with TMDs, as measured using primarily subjective measures [31–33,35,42]. Individuals with chronic pain and related-central sensitization conditions frequently report fatigue as being a primary determinant of well-being, with chronic fatigue being cited as a mediating factor in the relationship between functioning, pain and QOL [56]. However, despite the perceived significance of this symptom, masticatory fatigue

attributed to TMD-related OD has been minimally investigated, and no eligible studies reported prevalence rates using EMG to objectively measure indices of muscle fatigue, endurance and exercise tolerance. As such, although this study provides new evidence regarding the spectrum of possible chewing difficulties beyond that which had been previously discussed, it is evident that further research in this field is required.

Limitations

One limitation of this study was that few records satisfied this systematic review's inclusion criteria. For example: while primary studies reported that patients presented with TMDs, few provided sufficient demographic or diagnostic information (e.g. gender, mean age, DDWR, MFP, etc.) to allow for statistical stratification, with information unavailable on author contact attempts, also. This resulted in the forced exclusion of a large number of studies which may have positively influenced ultimate study findings. Therefore, results are derived from a limited, and potentially unrepresentative, cohort of heterogeneous primary records, with potential for confounding. As such, it is recommended that large-scale psychometrically valid and reliable epidemiological research is conducted in this field to adequately address concerns. A further limitation of this review is the lack of studies which utilised objective measures as sources of assessment data. The heavy reliance on subjective measures alone observed within primary studies in this review may be concerning due to the variable psychometric properties of these tools and their increased potential for the introduction of observer, recall, and detection bias. As such, it is recommended that future studies employ a combination of both subjective patient-report and objective imaging techniques.

Recommendations

Due to the identified high prevalence and varied nature of a range of OD signs and symptoms in adults with TMDs, it is recommended that future robust epidemiological research is conducted in order to both further our understanding of this condition, and to provide a valid and reliable foundation on which subsequent avenues for clinical improvements may be built. In order to appropriately investigate TMD-related OD within future research and clinical contexts, it is also advised that a cohort-specific simple and efficient assessment tool with adequate psychometric properties is developed for use with this patient group. Finally, in order to ensure that the spectrum of potential signs and symptoms of TMD-related OD are identified during definitive evaluations, it is suggested that a broad based assessment protocol is also developed, which encompasses subjective patient reports, functional chewing tasks, and objective assessment measures, as appropriate.

Conclusions

This systematic review and meta-analysis has highlighted that signs and symptoms of OD are consistently reported by

adults presenting with a range of TMD diagnoses. It has also identified a spectrum of methodological limitations within the available literature. This review has, therefore, indicated the need for psychometrically robust epidemiological research which investigates the presence, nature, and impact of TMD-related OD in a valid and reliable manner.



Acknowledgements

The authors would like to acknowledge the contribution of Isolde Harpur to the completion of this manuscript.

Disclosure statement

The authors have no conflicts of interest to declare.

ORCID

Ciaran Kenny  <http://orcid.org/0000-0001-6567-2412>
Margaret Walshe  <http://orcid.org/0000-0003-3924-8073>

References

- [1] Di Fabio RP. Physical therapy for patients with TMD: a descriptive study of treatment, disability, and health status. *J Orofac Pain.* 1997;12:124–135.
- [2] Morales H, Cornelius R. Imaging approach to temporomandibular joint disorders. *Clin Neuroradiol.* 2016;26:5–22.
- [3] Al Zarea BK. Temporomandibular disorders (TMD) in edentulous patients: a review and proposed classification (Dr. Bader's Classification). *J Clin Diagn Res.* 2015;9:6–9.
- [4] Greene CS, Klasser GD, Epstein JB. Revision of the American association of dental research's science information statement about temporomandibular disorders. *J Can Dent Assoc.* 2010;76:115.
- [5] Manfredini D, Chiappe G, Bosco M. Research diagnostic criteria for temporomandibular disorders (RDC/TMD) axis I diagnoses in an Italian patient population. *J Oral Rehabil.* 2006;33:551–558.
- [6] Barghan S, Tetradis S, Mallya SM. Application of cone beam computed tomography for assessment of the temporomandibular joints. *Aust Dent J.* 2012;57:109–118.
- [7] Cooper BC, Kleinberg I. Examination of a large patient population for the presence of symptoms and signs of temporomandibular disorders. *Cranio.* 2007;25:114–126.
- [8] McNeill C, Mohl ND, Rugh JD, et al. Temporomandibular disorders: diagnosis, management, education, and research. *J Am Dent Assoc.* 1990;120:253–263.
- [9] Pedroni CR, De Oliveira AS, Guaratini MI. Prevalence study of signs and symptoms of temporomandibular disorders in university students. *J Oral Rehabil.* 2003;30:283–289.
- [10] Nassif NJ, Al-Salleeh F, Al-Admawi M. The prevalence and treatment needs of symptoms and signs of temporomandibular disorders among young adult males. *J Oral Rehabil.* 2003;30:944–950.
- [11] Taylor M, Suvinen T, Reade P. The effect of grade IV distraction mobilisation on patients with temporomandibular pain-dysfunction disorder. *Physiother Theory Pract.* 1994;10:129–136.
- [12] Bertolucci LE, Grey T. Clinical analysis of mid-laser versus placebo treatment of arthralgic TMJ degenerative joints. *Cranio.* 1995;13:26–29.
- [13] Poveda RR, Bagan JV, Diaz Fernandez JM, et al. Review of temporomandibular joint pathology. Part I: classification, epidemiology and risk factors. *Med Oral Patol Oral Cir Bucal.* 2007;12:292–298.
- [14] Rutkiewicz T, Könönen M, Suominen-Taipale L, et al. Occurrence of clinical signs of temporomandibular disorders in adult Finns. *J Orofac Pain.* 2006;20:208–217.

- [15] Lamot U, Strojan P, Popović KŠ. Magnetic resonance imaging of temporomandibular joint dysfunction-correlation with clinical symptoms, age, and gender. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2013;116:258–263.
- [16] Dworkin SF, Huggins KH, LeResche L, et al. Epidemiology of signs and symptoms in temporomandibular disorders: clinical signs in cases and controls. *J Am Dent Assoc.* 1990;120:273–281.
- [17] Manfredini D, Piccotti F, Ferronato G, et al. Age peaks of different RDC/TMD diagnoses in a patient population. *J Dent.* 2010;8:392–399.
- [18] Nassif N, Talic Y. Classic symptoms in temporomandibular disorder patients: a comparative study. *J Craniomandib Pract.* 2001;19:33–41.
- [19] Tuz H, Onder E, Kısnişi R. Prevalence of otologic complaints in patients with temporomandibular disorder. *Am J Orthod Dentofac Orthop.* 2003;123:620–623.
- [20] Zulgamian BJ, Kahn N, Khattab S. Self-reported symptoms of temporomandibular dysfunction in a female university student population in Saudi Arabia. *J Oral Rehabil.* 1998;25:946–953.
- [21] Liu F, Steinkeler A. Epidemiology, diagnosis, and treatment of temporomandibular disorders. *Dent Clin North Am.* 2013;57:465–479.
- [22] Schiffman E, Ohrbach R, Truelove E, et al. Diagnostic criteria for temporomandibular disorders (DC/TMD) for clinical and research applications: recommendations of the International RDC/TMD Consortium Network and Orofacial Pain Special Interest Group. *J Oral Facial Pain Headache.* 2014;28:6–27.
- [23] Hazazi HSM. A retrospective analysis of in-patient temporomandibular disorders [thesis]. Rutgers The State University of New Jersey, School of Health Related Professions. 2015.
- [24] Bessa-Nogueira RV, Vasconcelos BC, Duarte AP, et al. Targeted assessment of the temporomandibular joint in patients with rheumatoid arthritis. *J Oral Maxillofac Surg.* 2008;66:1804–1811.
- [25] Dworkin SF, LeResche L. Research diagnostic criteria for temporomandibular disorders: review, criteria, examinations and specifications, critique. *J Craniomandib Disord.* 1992;6:301–355.
- [26] Moher D, Liberat A, Tetzlaff J, et al. Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *Ann Intern Med.* 2009;151:264–269.
- [27] Stroup DF, Berlin JA, Morton SC, et al. Meta-analysis of observational studies in epidemiology: a proposal for reporting. Meta-analysis Of Observational Studies in Epidemiology (MOOSE) group. *JAMA.* 2000;283:2008–2012.
- [28] Gilheaney Ó, Zgaga L, Harpur I, et al. The prevalence of oropharyngeal dysphagia in adults presenting with temporomandibular disorders: a systematic review and meta-analysis [Internet]. PROSPERO 2016;CRD42016050846. [cited 2017 Nov 22]. Available from: http://www.crd.york.ac.uk/PROSPERO/display_record.asp?ID=CRD42016050846
- [29] Tanner DC. Case studies in communicative sciences and disorders. Columbus: Pearson Prentice Hall; 2006.
- [30] Truelove E, Pan W, Look JO, et al. Research diagnostic criteria for temporomandibular disorders: validity of axis I diagnoses. *J Orofacial Pain.* 2010;24:35–47.
- [31] Reizsmann DR, John MT, Schierz O, et al. Functional and psychosocial impact related to specific temporomandibular disorder diagnoses. *J Dent.* 2007;31:643–650.
- [32] Ferreira CL, Bellistri G, Montagna S, et al. Patients with myogenic temporomandibular disorders have reduced oxygen extraction in the masseter muscle. *Clin Oral Invest.* 2017;21:1509–1518.
- [33] Barros VM, Seraidarian PI, Côrtes MI, et al. The impact of orofacial pain on the quality of life of patients with temporomandibular disorder. *J Orofac Pain.* 2008;23:28–37.
- [34] Mapelli A, Machado BC, Giglio LD, et al. Reorganization of muscle activity in patients with chronic temporomandibular disorders. *Arch Oral Biol.* 2016;72:164–171.
- [35] Fotedar S, Chauhan A, Bhardwaj V, et al. Oral health-related quality of life in Indian patients with temporomandibular disorders. *J Cranio Max Dis.* 2015;4:42.
- [36] Radke JC, Kull RS, Sethi MS. Chewing movements altered in the presence of temporomandibular joint internal derangements. *Cranio.* 2014;32:187–192.
- [37] Abrahamsson C. Masticatory function and temporomandibular disorders in patients with dentofacial deformities. *Swed Dent J Suppl.* 2012;231:9–85.
- [38] Gavish A, Winocur E, Menashe S, et al. Experimental chewing in myofascial pain patients. *J Orofac Pain.* 2002;16:22–28.
- [39] Dougall AL, Jimenez CA, Haggard RA, et al. Biopsychosocial factors associated with the subcategories of acute temporomandibular joint disorders. *J Orofac Pain.* 2012;26:7–16.
- [40] Gonçalves TM, Sánchez-Ayala A, Ambrosano GM, et al. Female hormonal fluctuation and masticatory function in patients with disc displacement: a case-control study. *Int J Prosthodont.* 2011;24:320–327.
- [41] Baker Z, Eriksson L, Englesson Sahlström L, et al. Questionable effect of lavage for treatment of painful jaw movements at disc displacement without reduction: a 3-year randomised controlled follow-up. *J Oral Rehabil.* 2015;42:742–750.
- [42] La Touche R, Paris-Aleman A, Gil-Martínez A, et al. Masticatory sensory-motor changes after an experimental chewing test influenced by pain catastrophizing and neck-pain-related disability in patients with headache attributed to temporomandibular disorders. *J Headache Pain.* 2015;16:500–514.
- [43] Wänman A. Endurance to physical strain in patients with temporomandibular disorders: a case-control study. *Acta Odontol Scand.* 2012;70:455–462.
- [44] Michelotti A, Steenks MH, Farella M, et al. Short-term effects of physiotherapy versus counselling for the treatment of myofascial pain of the jaw muscles. *J Oral Rehabil.* 2002;29:874.
- [45] Gilheaney Ó, Zgaga L, Harpur I, et al. The prevalence of oropharyngeal dysphagia in adults presenting with temporomandibular disorders associated with rheumatoid arthritis: a systematic review and meta-analysis. *Dysphagia.* 2017;16:1–4.
- [46] Downs SH, Black N. The feasibility of creating a checklist for the assessment of the methodological quality both of randomised and non-randomised studies of health care interventions. *J Epidemiol Community Health.* 1998;52:377–384.
- [47] Neyeloff JL, Fuchs SC, Moreira LB. Meta-analyses and forest plots using a Microsoft excel spreadsheet: step-by-step guide focusing on descriptive data analysis. *BMC Res Notes.* 2012;5:52–57.
- [48] MedCalc Statistical Software version 16.4.3 (MedCalc Software bvba, Ostend, Belgium [Internet]. 2016. Available from: <https://www.medcalc.org>
- [49] da Silva LA, Teixeira MJ, de Siqueira JT, et al. Xerostomia and salivary flow in patients with orofacial pain compared with controls. *Arch Oral Biol.* 2011;56:1142–1147.
- [50] Maffei C, de Mello MM, de Biase NG, et al. Videofluoroscopic evaluation of mastication and swallowing in individuals with TMD. *Braz J Otorhinolaryngol.* 2012;78:24–28.
- [51] Khawaja SN, Nickel JC, Iwasaki LR, et al. Association between waking-state oral parafunctional behaviours and bio-psychosocial characteristics. *J Oral Rehabil.* 2015;42:651–656.
- [52] De Felício CM, Ferreira CL, Medeiros AP, et al. Electromyographic indices, orofacial myofunctional status and temporomandibular disorders severity: a correlation study. *J Electromyogr Kinesiol.* 2012;22:266–272.
- [53] Raphael KG, Marbach JJ. Widespread pain and the effectiveness of oral splints in myofascial face pain. *J Am Dent Assoc.* 2001;132:305–316.
- [54] Brandini DA, Benson J, Nicholas MK, et al. Chewing in temporomandibular disorder patients: an exploratory study of an association with some psychological variables. *J Orofac Pain.* 2011;25:56–67.
- [55] The Royal College of Speech and Language Therapists. RCSLT resource manual for commissioning and planning services for SLCN [Internet]. 2009 [cited 2017 Nov 22]. Available from: https://www.rcslt.org/speech_and_language_therapy/commissioning/dysphagia_manual_072014
- [56] Gold JI, Mahler NE, Yee J, et al. Pain, fatigue, and health-related quality of life in children and adolescents with chronic pain. *Clin J Pain.* 2009;25:407–412.

Appendix 1. Example of database search strategy for PubMed

('Deglutition'[Mesh] OR 'Deglutition Disorders'[Mesh] OR Dysphagia[Title/Abstract] OR Dysphagic[Title/Abstract] OR Deglutition[Title/Abstract] OR Swallow[Title/Abstract] OR Swallows[Title/Abstract] OR Swallowing[Title/Abstract] OR Swallowed[Title/Abstract]) AND ('Mandible'[Mesh] OR 'Temporomandibular Joint'[Mesh] OR 'Temporomandibular Joint Disorders'[Mesh] OR 'Stomatognathic System Abnormalities'[Mesh] OR 'Skull'[Mesh] OR 'Jaw'[Mesh] OR 'Mastication'[Mesh] OR 'Mouth Opening'[Title/Abstract] OR Mandibular[Title/Abstract] OR Mandible[Title/Abstract] OR Temporomandibular[Title/Abstract] OR Stomatognathic[Title/Abstract] OR Masticatory[Title/Abstract] OR Mastication[Title/Abstract] OR Jaw[Title/Abstract] OR Jaws[Title/Abstract] OR Skull[Title/Abstract] OR Skulls[Title/Abstract] OR Cranium[Title/Abstract] OR Calvaria[Title/Abstract] OR Calvarium[Title/Abstract]).

Appendix 2. Down's and Black checklist

	Yes (1 point)	No (0 points)	Unclear (0 points)
Hypothesis/aim/objective explicit			
Main outcomes clearly described in the introduction or methods section			
Characteristics of patients included clearly described			
Distributions of principal confounders in each group of subjects to be compared clearly described ^a			
Main findings clearly described			
Study provides estimates of random variability for main outcomes			
Characteristics of patients lost to follow-up described			
Actual probability values been reported for main outcomes except where probability value is less than 0.001			
Subjects representative of entire population			
Subjects prepared to participate representative of entire population			
Staff, places, and facilities representative			
Any of the results of the study were based on 'data dredging'			
Appropriate statistical tests used to assess main outcomes			
Main outcome measures used accurate (valid and reliable)			
Adequate adjustment for confounding in the analyses			
Patients in different groups or cases and controls recruited from same population			
Subjects in different groups or cases and controls recruited over same time			

^aYes: 2 points; Partially: 1 point; No: 0 points.