Systematic Review and Meta-analysis of Incidence and Prevalence of Orofacial Pain-Global Burden of Diseases

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Abstract

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We conducted a systematic review of the incidence and prevalence of orofacial pain (OFP) from 1980 to 2020, ages 0 to 100, using the Global Burden of Disease protocol, which could then be used to derive global estimates and global OFP burden. Since there is no consensus case definition for OFP, we derived a working definition based on biologic plausibility along with current classifications and study definitions. After reviewing 1962 articles, 38 studies on prevalence and 1 on incidence of OFP met inclusion criteria. The recall periods ranged from pain at the present moment, to one month up to 24 months. We identified a total of 18 case definitions for OFP derived from these 39 studies. We further classified the OFP prevalence and incidence studies according to sub-categories of OFP along with their different recall periods. The mean prevalence of OFP was 32.2%, ranging from 15.1 % to 74.9 %; the incidence rate of OFP was 2.1% per year. We identified 12 subcategories of orofacial pain, and the average number of these subcategories was seven. Adjustment factors for recall period were also calculated, allowing for more accurate estimates of prevalence. OFP is shown to be common within the community.

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I. INTRODUCTION

Pain can be defined as an unpleasant sensory and emotional experience associated with actual or potential tissue damage, or described in terms of such damage. It is a mutually recognizable somatic experience that reflects a person's apprehension of threat to their bodily or existential integrity [1]. The extent of the chronic pain problem often poses a significant economic burden for patients, health services and societies alike and while international comparisons are difficult to make, due to differences in study methods and perspectives employed, it is apparent that pain represents a significant burden on limited healthcare resources across all countries. Pain affects everyone to varying degrees [2],[3]; it seriously impairs the lives of millions of people around the world, thus making it a major public health problem because of its impact on functional, social, physical and psychological outcomes [4]. Epidemiological studies in the area of pain are important because they provide information about its prevalence in different population segments and about factors associated to its etiology and persistence. Such data are useful for health professionals and for the development of programs addressing pain [5].

Orofacial pain (OFP) can be defined as any acute, sub-acute or chronic pain felt in the in the jaw, teeth and/or face, including the forehead, eyes, and temporal region, including referred pain to this region. Currently, there is no standard or universally accepted definition of orofacial pain, and some that exist have no clear-cut anatomical boundaries which can create ambiguity. Hence, before epidemiologic work can commence, there is a need to create a well-defined and anatomically based definition of OFP that is flexible enough to allow the inclusion of most previous prevalence and incidence studies.

Some of the most prevalent and debilitating pain conditions arise from the structures innervated by the trigeminal system (head, face, masticatory musculature, temporomandibular joint and associated structures). OFP can arise from different regions and etiologies including temporomandibular disorders (TMD) that involve the masticatory musculature, the temporomandibular joint (TMJ) or both, trigeminal neuropathic pain and facial pain conditions can arise from injury secondary to dental procedures, infection, neoplasia, or disease or dysfunction of the peripheral and/or central nervous system, neurovascular disorders, such as primary headaches, can present as chronic orofacial pain, such as in the case of facial migraine, where the pain is localized in the second and third division of the trigeminal nerve and odontogenic pain that arises from the teeth. Together, these disorders of the trigeminal system may impact the quality of life of the sufferer, and sometimes, severely [6,7]. The contribution of each of these specific OFP conditions to the entire group is not known, however, and a popular online reference listing the prevalence of individual sub-categories of OFP listed toothache comprising 95% of OFP[7].

OFP lies at the interface of the dental and medical professions, but neither has fully acknowledged that it is within their purview. The result of these disorders crossing professional boundaries has been a lack of coordinated patient care, delays in diagnosis, and many examples of poor-quality care. Given the importance of oral cavity in terms of key functions such as eating and communication, and the psychological significance of face and mouth, with manifestations of Temporomandibular Disorders (TMD) and other conditions being reported with OFP, there are compelling reasons to believe that this group of disorders often can have a major impact on functioning and quality of life of the individual [8].

The WHO global goals for 2020 stressed the need to understand and reduce morbidity from oral and craniofacial diseases and thereby increase the quality of life [9]. The Global Burden of Disease study provides a standardized approach for estimating incidence, prevalence, and Years Lived with Disability (YLDs) and Disability adjusted Life Years (DALYS) by cause, age, sex, year, and geographical location. Measuring non-fatal health loss is one of the most complex endeavors in population health research. Global progress in improving the burden of non-fatal health outcomes has been limited, in part by a predominant focus on mortality rates as a common metric of tracking global health progress. Estimates reported in recent iterations of the Global Burden of Diseases, Injuries, and Risk Factors Study (GBD) have also illustrated differential health outcomes in males and females in certain locations and conditions. Three causes (low back pain, headache disorders, and depressive disorders) have been shown as leading causes of non-fatal health loss for nearly three decades and the increasing burden of non-fatal diseases, injuries, and impairments could pose considerable challenges to health systems and economies not equipped to care for complex and expensive conditions [10]. To date, OFP has not been reported within GBD.

Several studies on OFP have shown unemployment and decreased work effectiveness among affected patients. The proportion of people unable to carry out some activities because of pain ranged from 14% for facial pain to 48% for severe headache [11]. In a postal survey in Canada [42] the authors found that 58% of those experiencing OFP reported one or more behavioral impacts. Odontogenic and TMD pain prevalence have the highest prevalence reported in one prior study, followed by primary headaches, facial pain and oral sores, yet the contribution of each pain condition to OFP prevalence in total is not known. Many factors can influence the prevalence and incidence rates, including age, sex, socio-economic factors and time.

Thus, OFP appears to be a common, sometimes disabling collection of pain disorders that has not been well-characterized nor reported in GBD. In order to estimate the global burden for OFP, a systematic review of OFP prevalence and incidence is needed as an initial step to help derive estimates of DALY's for GBD.

II. AIM AND OBJECTIVES

Aim:

The aim of this study was to conduct a systematic review of prevalence and incidence of Orofacial Pain (OFP) conditions globally from 1980 - 2020 as per the protocol of the Global Burden of Diseases, Injuries, and Risk Factors Study.

Specific study objectives were to:

1. Establish a case definition for orofacial pain
2. Conduct a systematic review of the prevalence of orofacial pain from 1980 to 2020.
3. Conduct a systematic review of the incidence of orofacial pain from 1980 to 2020.
4. Compare and analyze various case definitions of orofacial pain
5. Analyze the influence of recall period on the prevalence rates of orofacial pain
6. Create a GBD Data extraction sheet in Excel for future DisMod (Disease Modeling) regression.

III. METHODS AND APPROACH

III a. General Approach

In order to complete this systematic review and meta-analyses of the prevalence and incidence of OFP, the following aims were completed, step by step. First, current classification systems for orofacial pain were reviewed, and then a working definition of orofacial pain was created, along with anatomical boundaries, so that a case definition could be developed. Second, a systematic review of the prevalence of orofacial pain was conducted, using guidelines from GBD and PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analysis). Third, a systematic review of the incidence of orofacial pain was conducted. Fourth, case definitions for OFP were compared, analyzed and grouped. Fifth, the effect of the recall period on the prevalence rate of orofacial pain was measured. Finally, a data extraction sheet in Excel was created for future DisMod regression for GBD.

III b. Review of Classification of Orofacial Pain

A classification allows the definition of specific entities according to specific characteristics. In medicine and thus for orofacial pain (OFP), the characteristics of such an ideal diagnostic system could be derived from the etiology, pathophysiology, diagnosis, and/or management of a specific disease or disorder. For the clinician, a correct definition and classification of the specific disease entity is important since it assists in planning the management and in discussing the prognosis with the patient. More refined pain classifications require additional knowledge of pain behavior and require a greater diagnostic effort. To classify pain by the location of its source requires an understanding of heterotopic pains and entails the need for diagnostic differentiation between primary pain and its secondary effects.

There are various orofacial pain classification systems described in the literature. All of them have their own strengths and weaknesses. We reviewed the International Association for the Study of Pain, American Academy of Craniofacial Pain, International Headache Society and American Academy of Orofacial Pain classification system(s) in order to decide which system was most appropriate for our study [61].

The classification structure of orofacial pain conditions from the 5th edition of the American Academy of Orofacial Pain (AAOP) guidelines was adopted for this study since it included broad guidelines for assessment and diagnosis to include all clinical phenotypes of OFP related disorders. Figure 2 presents the list of orofacial pain conditions in the AAOP classification system.

III c. Establishing a case definition for Orofacial Pain

Establishing a case definition is an essential and a crucial step in epidemiological studies. A case definition includes criteria for person, place, time, and clinical features. The first step in our study was to construct a strong and a valid case definition, considering the anatomical location and the innervation of the region. In addition, after the prevalence and incidence studies for the systematic review were located, the available scientific work helped to inform whether the case definition was feasible.

III d. Protocol

This systematic review of Prevalence and Incidence of OFP was done according to the Global Burden of Diseases protocol [62].

The literature review and this thesis was conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines as shown in Figure 3 and Table 1.

PICOS question

Our PICOS question components were the following –

Population: global population or Problem: prevalence of OFP is not known

Intervention: none

Comparison: by country, age, sex, recall period and case definition

Outcome: prevalence of OFP

Studies: prevalence and incidence studies on OFP

III e. Study Inclusion and Exclusion Criteria

All prevalence and incidence studies were required to meet specific criteria in order to be included in this review. The inclusion criteria were:

- Multiple categories of orofacial pain were included
- Studies were representative of a population of a country, region or super-region according to the GBD criteria.
- Studies of persons within the age group 0-100 years.
- Studies on both or either males / females.
- Studies done after 1980.

Exclusion criteria were as follows:

- Studies did not provide an overall incidence or prevalence rate.
- Studies reported prevalence/incidence rates exclusively on one sub-category of OFP.
- Studies where orofacial pain prevalence/incidence rates were studied in a cohort with an additional disease or condition and where there was a possibility that the presence of this condition might affect the overall rates of orofacial pain.
- Studies where it was not possible to obtain full text.
- Review studies.

III f. Information sources, data search and selection

An initial search was conducted in December 2018. The databases used to conduct the search included PubMed, Embase, LILACS, SciELO, CINAHL Plus, APA PsycINFO, Scopus and Web of Science. In addition to the above databases, the following resources were also used:

- Gray Literature: Unpublished studies, reports and dissertations, conference abstracts or papers, AHRQ (U.S. Department of Health & Human Services), WHO surveys and Open Grey.

- Other Sources of data: Country Bureau of Statistics and Ministry of Health websites, Disease and Country experts, Major Multinational survey data catalogues and Ministry of Health and Central Statistical Office Websites.

A repeat search was conducted in February 2020 with the same headings and at the same databases with the same mesh terms.

The following mesh terms / search terms were used:

a) PubMed: Mesh Terms: "Facial Pain", " Pain, Referred", "Craniomandibular Disorders", "Stomatognathic Diseases", "Health Services Research", "Epidemiologic Methods"
   Mesh Subheadings: "Epidemiology"

b) Embase: "face pain", "tooth pain", "headache and facial pain", "mouth pain" "temporomandibular joint disorder", "disease burden", "epidemiological data".

c) LILACS, SciELO, CINAHL Plus, APA PsycINFO, Scopus and Web of Science: "Facial Pain", "Pain, Referred", "Craniomandibular Disorders", "Stomatognathic Diseases", "Health Services Research", "Epidemiologic Methods", "Epidemiology"

Additionally, the electronic search was supplemented with hand searches of reference lists of all relevant publications including review articles and textbooks whenever applicable.

III g. Data collection process

Data collection or extraction is a process that involves retrieval of data from various sources. Raw data were extracted for 6 fields (Table 1) and data regarding OFP questionnaires for 3 fields (Table 2). GBD data was extracted into 48 fields as shown below.

III h. Data items

1. field_citation_value
2. page_num
3. source_type
4. location_name
5. location_id
6. ihme_loc_id
7. smaller_site_unit
8. site_memo
9. sex
10. sex_issue
11. year_start
12. year_end
13. year_issue
14. age_start
15. age_end
16. age_issue
17. age_demographer
18. measure
19. mean
20. sample_size

21. unit_type
22. unit_value_as_published
23. measure_issue
24. measure_adjustment
25. representative_name
26. urbanicity_type
27. recall_type
28. recall_type_value
29. case_name
30. case_definition
31. note_SR
32. extractor
33. bundle_name
34. cv_recallbias
35. cv_TMJ Pain
36. cv_TMJ pain %
37. cv_Toothache
38. cv_Toothache %
39. cv_Oral sores
40. cv_Burning mouth
41. cv_Facial pain
42. cv_Muscle pain
43. cv_Eye pain

44. cv_Headache

45. cv_Acute trauma

46. cv_Gum pain

47. cv_Tumour pain

48. cv_Referred pain to orofacial region

Table 3 shows if each study includes or excludes a specific subtype of orofacial pain.

III i. Additional analyses – Recall Period

There were many different recall periods in these different case definitions. As a result of this, we cannot easily compare the different prevalence rates within a single case definition. In order to study the influence of the effect of recall period across case definitions, we performed a meta-analysis. The meta-analysis was performed using the STATA-16 software.

The meta-analysis was done in two steps:

1. *Univariate meta-analyses of recall period for each case definition*
2. *Univariate meta-analyses of definition effect, after adjusting for recall period effect*

STEP 1

The first step was to perform univariate meta-analyses of recall period for each case definition. After matching for age and sex, ratios were obtained within each case definition of the prevalence rates for different recall periods. Below is a detailed description of this method:

Case Definition 1: There are three recall periods: 0 (i.e., point prevalence), 1 and 6. There are multiple studies for each of them. After matching the prevalence rates, the ratio of 1 month: 0 month and 6 months: 0 month for prevalence rates was obtained. The ratios were log transformed and used in STEP 2.

Case Definition 2: There are three recall periods: 0, 3 and 6. There are multiple studies for each of them. After matching the prevalence rates, the ratio of 6 months: 0 month and 3 months: 0 month for prevalence rates was obtained. The ratios were log transformed and used in STEP 2.

Case Definition 3: There are four recall periods – 1, 3, 6 and 24. There are multiple studies for each of them. After matching the prevalence rates, the ratio of 3 months: 1 month, 6 months: 1 month, 24 months: 1 month for prevalence rates was obtained. The ratios were log transformed and used in STEP 2.

Case Definition 4: There are three recall periods – 0, 1, 6 and 12. There are multiple studies for each of them. After matching the prevalence rates, the ratio of 1 month: 0 month, 6 months: 0 month, 24 months: 0 month for prevalence rates was obtained. The ratios were log transformed and used in STEP 2.

Case Definition 5: There are two recall periods 1 and 6. There are multiple studies for each of them. After matching the prevalence rates, the ratio of 6 months to 1 month for prevalence rates was obtained. The ratios were log transformed and used in STEP 2.

Case Definition 6: There is a single recall period of 12 months. There is a single study for this case definition. Therefore, no ratios were calculated.

STEP 2:

The next step was to pool the ratios across case definitions. This wasn't possible for all the case definitions since recall periods were not similar across case definitions. However, we were able to pool for three: 6 to 0, 1 to 0 and 6 to 1.

The ratios were then log transformed (shown above) and labeled "log10 Ratio". The Standard Error was labeled "SE" and was obtained and from prevalence and study population for each case definition for each recall period. The study sample size was labeled 'study size'. The log transformed ratio was then meta-analyzed.

For the Meta-analysis, STATA 16 software was used. The following settings were applied in the software:

1. We have pre-computed effect sizes as the log-transformed ratios. They would be selected for the "Effect size" drop down menu.

2. We have Standard error for each of the ratios. They would be selected from the drop-down menu of "standard error".

3. In the second step, a fixed-effects inverse variance model was selected. The reason why this model was selected was because although we have controlled for age and sex, there are some variables like location that are not controlled for and hence using a fixed-effects model would be more appropriate. The concept behind selecting this model is that whatever effects the omitted variables have on the subject at one time, they will also have the same effect at a later time; hence their effects will be constant, or "fixed."

4. The Study label is how we would label our studies. We have labelled them according to the ratio and the case definition number combination. The study size would be the sample size. For effect – size, we labeled it as Ratio.
5. Next would be the summary and the Forest Plot. They would essentially follow the same model as above and there was no need to change any settings. The below image displays the settings inherited for Summary and a Forest Plot.

Similarly, with the same settings, pooled prevalence for the ratio 1 to 0 and 6 to 1 were obtained across case definitions. The resultant Forest plots and the results have been discussed in the results section.

IV. RESULTS

IV a. Establishing a case definition

The case definition for OFP was adopted as follows:

1. Any pain felt in the mouth, jaws and face above the neck. This includes pain within the oral cavity, pain of dental origin, temporomandibular disorders and pain in the facial region including the forehead, eyes, and temporal region.

2. Pain referred to anywhere in this region from any part of the body.

The anatomical boundaries were as follows: bounded superiorly by the galea aponeurotica, and laterally and inferiorly by mandible and the temporal fossa, it includes all the structures above the neck in this region (see Figure 1). The extent is further divided into the oral and facial regions. Oral pain includes pain in the oral cavity which includes the lips, gingivae, retromolar trigone, teeth, hard palate, lining mucosa, tongue, and floor of the mouth.

Facial pain includes pain in the upper, middle and lower face regions as described below.

- The upper face region contains the forehead, eyes, and temporal region.
- The middle face region contains the nose, cheeks, and ears.
- The lower face region contains the lips, chin, and jaws. The lateral border of the lower face is made up of the angle of the mandible.

Time: A case definition requires delineating a period of time associated with illness onset for the cases under investigation. Pain experienced for one day or more was required as part of our case definition.

IV b. Study selection

A total of 2,875 articles were obtained after the initial database search. Five studies, including one unpublished thesis, were obtained from other sources. After removing the duplicates, we were left with 1,962 studies. All of the 1,962 articles were screened for eligibility using their abstracts. Abstracts were available for all the articles. Where more information was required to determine if the article met inclusion and exclusion criteria, the article was obtained. A second reviewer was consulted if there was any question about eligibility. After these steps, 172 full text articles were obtained to further assess eligibility [Figure 3].

133 full text articles were excluded based on the inclusion and exclusion criteria. Each article was reviewed by two reviewers, PR and MD. The interrater reliability score between reviewers was 0.86. MD had more experience in the field of OFP and made the final decision to include or exclude the article in case of any conflicts.

The following were the categories for exclusion of studies, and the number of studies in each category:

- Hospital/clinic based with biased results: 46
- Single subcategory of Orofacial Pain: 41
- No summary incidence or prevalence rates across all categories of orofacial pain: 4
- Results cannot be generalized to entire population: 22
- OFP associated with another disease: 13
- Article older than 1980: 6

- Measured non-painful conditions via Helkimo index: 1

A total of 39 articles, 1 for Incidence and 38 for Prevalence rates were included in the qualitative review, and 35 were included in the quantitative analysis. The 38 articles obtained for prevalence were from 24 different countries. All of the articles were in the English language except for one article which was in German and no English version was available online. This article [32] was translated into English using Google Translate and UW library resources.

IV c. Study characteristics

The total study size for 38 prevalence studies was 106,161 individuals and for the incidence study 35,464 individuals from 24 countries and 1 country respectively. Overall, the studies estimated a prevalence ranging from 15.1 % to 74.8 % and some of them also reported on disease burden, with the variation in the estimates attributed mainly to different case definitions including variation in recall periods in the study. Table 1 shows a summary of the data obtained during the initial data extraction process. Additional data was extracted for the method and the questionnaire used to evaluate OFP in the study shown in Table 2.

Data in Table 1 shows data extracted from 6 fields – The first one is location. We mapped the data according to the location (Figure 4). There were studies from different countries, including cities and some rural areas. United States has the largest number of studies (5), followed by Brazil (3) and Canada (3). Other countries have either two, one or no studies. The study type and data source were also collected which showed how the study was conducted. Some were conducted as part of

national health surveys in South Korea, Germany, Finland and the United States. Some were conducted in university students, some at certain public schools and one study was also conducted at a nursing home. The sample size ranged from 80 to 42,370 individuals. The prevalence rate extracted from the articles is also shown in the table. Finally, the recall period was extracted. It ranged from point prevalence to 24 months.

Data show that there is variation in the prevalence rates across different regions in the world possibly due to methodological factors like different recall periods, socio-economic factors, different age groups (0-100). Studies also had different methods of examination including postal surveys and telephone interviews which included different questionnaires with some of them followed by a clinical examination.

Data were extracted as shown in Table 2 into 4 fields which were recall period, method of evaluation, study question set summary and assessment. The study question set summary and assessment fields show how the questions were framed or the type of questionnaires used in each article. Although each study asked about presence of pain and used the term "pain", some also used terms like "tenderness", "ache" and "soreness''. The questions asked were framed in different ways and there was no standardized questionnaire used in multiple studies, however there was a general uniformity in what was asked to the study subjects, which was if they have or have had orofacial pain– yes or no. We also found that 13 out of the total 39 studies (33.33%) had questionnaire followed by a clinical examination.

Data was extracted into the Global Burden of Disease Excel spreadsheet from the 39 articles. They were extracted into 48 different fields mentioned in the methods.

There was a single study for Incidence that was conducted in Denmark. It was a large population-based study [49] conducted in a total of 35,464 individuals who were between 18 and 70 years of age. The incidence rate reported was 2.1 % or 2.1 per 100 person-years.

IV d. Data classification and categorizing case definitions

A total of 39 studies (38 Prevalence and 1 Incidence) were obtained. Initially, raw data from each article was extracted for 7 different categories which include geographical location, age, sex, prevalence rates, recall period, OFP sub-categories and methods. Final data were extracted from the 39 articles for 48 different categories which included 12 sub-categories of OFP.

The studies were then categorized into 6 different groups based on their inclusion or exclusion of 12 different sub-categories of OFP. The sub-categories were determined based on a standardized classification, The American Academy of Orofacial Pain Classification, and were further refined based on what was found across all 39 articles. The sub-categories included the following:

1. TMJ Pain
2. Toothache / Odontalgia
3. Mucosal Pain / Oral sores
4. BMS / Tongue Pain

5. Facial Pain (Including but not limited to Trigeminal Neuralgia, Trigeminal Neuropathic Pain, Atypical Facial Pain)
6. Orofacial Muscle Pain
7. Eye Pain
8. Headaches
9. Gum Pain
10. Acute Trauma
11. Tumor/Lesion in the Orofacial region and associated pain
12. Referred Pain

Table 3 shows that none of the 38 prevalence studies included all 12 subcategories of orofacial pain. The range was from 5 up to 10 categories. It shows different sub-types of OFP as extracted from each article. Facial pain was assessed in all the studies. We found that all the studies (100%) have asked about any kind of facial pain symptoms. This was followed by Burning mouth syndrome was assessed second highest (97.4%), followed by TMJ pain and Muscle pain (92.3% of studies each), Oral sores (82%), Toothache and Gum pain (79.4%), Eye pain (53.3%), Primary headaches (46.15%) and Acute trauma (7.6%); least commonly assessed were Pathology Pain (5.1%) and referred pain (5.1%). One of the reasons for this pattern is that all studies considered Facial pain and BMS as an Orofacial pain sub-type and hence these entities were included in their questionnaires. Similarly, TMD pain is an important component of OFP and was measured by almost all the articles. Toothache however was not considered as an OFP sub-type by certain studies and was not measured as a part of every study.

Table 4 shows the case definitions included and different recall periods within each case definition. This table shows how we categorized different case definitions based on different recall period present in all the studies. The case definition 4, sub-type L which included TMJ pain + muscle pain + BMS pain + facial pain + toothache + oral sores + gum pain at point prevalence, had 5 studies, which were the highest across all case definitions. Next were case definition 3 sub-type G (TMJ pain + muscle pain + BMS pain + facial pain + eye pain + headache+ toothache + oral sores + gum pain at 6 months recall period) and case definition 1 sub-type C (TMJ pain + muscle pain + BMS pain + facial pain + eye pain + headache at Point prevalence) in which each had 4 studies.

IV e. Additional analysis – Recall Period

In Table 5, we see the ratios that we obtained from STEP 1. We were only able to obtain ratios from case definitions 1, 2 and 4 since these have Point Prevalence and 6 months recall periods.

Following Meta-analyses, the pooled ratios were obtained for the following recall periods:

1. 6 Month to 0 Month
2. 6 Month to 1 Month
3. 1 Month to 0 Month

6 Month to 0 Month:

The settings were applied as mentioned in the methods, and we found the following results:

As shown in Figure 5, the heterogeneity is 44.18, which is low (< 50% considered as low). There seems to be consistency in the ratios across case definitions indicating recall period effect is similar across case definitions. The forest plot shows that the overall effect size is -0.05, the antilog of which is 0.891251. The standard error is 0.0127. Hence overall, the ratio for 6 to 0 is 0.891 (0.878-0.903). This would mean that studies using a recall period of 6 months had a pooled 10.9 (9.7 – 12.2) % lesser prevalence of reported OFP to that of the point prevalence for the same case definition. The prevalence at 6 months recall period is lower than at point prevalence across all case definitions. This can be possibly due to underreporting (recall bias) poor or incomplete memory recall.

6 Month to 1 Month

Similarly, for 6 months vs. 1 month, the observations in each case definition were matched and then a ratio was obtained from Case Definitions 1 and 5 (Table 6). We see in table that there was some degree of consistency in the ratios of 6 to 1 suggesting there in an influence of recall period and that effect is almost consistent across at least these two case definitions.

Both the ratios imply that the pain prevalence obtained at 6 months is less than that obtained 1 month. This implies that for both the case definitions 1 and 5, fewer people reported pain over a period of 6 months than what is reported over a period of 1 month.

As shown in Figure 6, the heterogeneity is 67.52%, which is high (> 50% considered as high) because of the difference in sample sizes. The forest plot shows that the overall effect size is -0.13,

the antilog of which is 0.7413102. Hence overall, we get a ratio for 6 to 1 which is 0.7413. The standard error is here is calculated to be 0.0178. We obtained a ratio for 6 to 1 as 0.741 (0.723-0.759). This would mean that studies using a recall period of 6 months had a pooled 25.9 (24.1-27.7) % lesser prevalence of reported OFP to that of the 1-month recall period for the same case definition.

The results agree with the suggestion that there is a recall bias, i.e., that when someone is asked if they had experienced pain in the past 6 months, fewer people reported pain as compared to if they were asked about having experienced pain in the past one month because of their inability to recall.

1 Month to 0 Month

Similarly, for 1-month recall vs. point prevalence, the observations in each case definitions were matched and then a ratio was obtained from Case Definitions 1 and 4 (Table 7). We see in Table 7 that the ratio of the prevalence here for 1 to 0 is more than 1. It would mean that for the same case definition, a greater number of people report pain at 1 month than on same day (Point prevalence). This is true for both the case definitions 1 and 4, which indicates pain reporting is uniformly a greater number of people report at 1-month recall than on the same day.

As shown in Figure 7 heterogeneity is 0, which is low (< 50% considered as low) which indicates a great degree of similarity between study data. The forest plot shows that the overall effect size is 0.06, the antilog of which is 1.1220185. Hence overall, we get a ratio for 1 to 0 which is 1.1220. The standard error here is calculated to be 0.0229. By inversion, we get the ratio of 0 to 1 recall

period as 0.892 (0.874 – 0.909). This would mean that point prevalence studies had a pooled 10.8 (9.1-12.6) % lesser prevalence of reported OFP to that of 1 month recall period.

Table 8 is a summary table of the ratios for prevalence rates, with different case definitions, after matching for age and sex. This table shows a summary of the ratios (both pooled and unpooled) for prevalence rates, after matching for age and sex for all the available recall periods in the studies. We could pool 3 ratios because of sufficient studies across case definitions. For interpretation, if the pooled ratio of prevalence rates across case definition for 1 to 0 which was 1.1220, and hence for 0 to 1 it was 0.892 (0.874 – 0.909), it would mean that studies conducted at point prevalence had a pooled 10.8 (9.1-12.6) % lesser prevalence of reported OFP to that of 1 month recall period. Similarly, a pooled ratio of prevalence rates across case definition for 6 to 1 as 0.741 (0.723-0.759). This would mean that studies using a recall period of 6 months had a pooled 25.9 (24.1-27.7) % lesser prevalence of reported OFP to that of the 1-month recall period for the same case definition. The pooled ratio of prevalence rates across case definition for 6 to 0 is 0.891 (0.878-0.903). This would mean that studies using a recall period of 6 months had a pooled 10.9 (9.7 – 12.2) % lesser prevalence of reported OFP to that of the point prevalence for the same case definition. These were the three ratios we could pool, and we could not pool the other ratios since there were insufficient data. These pooled ratios can be used to adjust the prevalence rates for different recall periods, so, in effect, to standardize them to one recall period.

The graph in Figure 8 shows the average prevalence of OFP increases from the point prevalence to 1-month recall period, and then decreases with 6, 12 and 24 month recall period. Possible explanations for this counter-intuitive effect will be discussed below.

When surveys and questionnaire are used to determine the location of a pain or the modalities of its occurrence, there seems to be a need for an additional step to confirm and refine the diagnoses, such as a physical exam to confirm the type of pain, for example, if it is really toothache or is it myofascial pain being referred to tooth. There is a possibility of overreporting of Odontalgia and underreporting of TMD pain since any pain in the jaw will be almost always perceived as a toothache. However, this would not change the overall OFP prevalence. Out of the total 39 studies, only 9 studies studied the proportion of TMD as well as Toothache. These studies are shown in Table 9, along with the prevalence ratio of the two conditions. Table 9 shows that the prevalence rates and the ratio of the two conditions vary due to factors like age, socio-economic factors, methods of study and recall period.

V. DISCUSSION AND CONCLUSION

Va. Summary of evidence

To our knowledge, this is the first study that has summarized all relevant prevalence studies of OFP while identifying the sub-categories along with differing methodological characteristics that can influence the frequency of this condition. Different methods and approaches have been used to conduct the studies. Some are conducted at a national level, some at subnational levels and some are at city levels. Some studies have also been conducted at tertiary clinics and dental hospitals, but these seem to be the main treatment centers in their respective areas so there are minimal chances of any over-reported OFP cases, and the size of the underlying population is known, hence the prevalence can be calculated.

We had a total of 18 case definitions with 6 recall periods. Since OFP is comprised of many sub-categories, each case definition was outlined according to the specific sub-categories of OFP used to measure the estimates. Further, these were divided according to the recall periods used in the study. Unfortunately, there was no study that included all the 12 subcategories of OFP, and every study had one or more sub-categories missing. The recall periods ranged from one month to 24 months, with some studies also being conducted at point prevalence. This is the first study to classify the OFP prevalence and incidence studies according to the sub-categories of OFP with different recall periods.

Variability in the prevalence of orofacial pain has also been shown across socio-economic status in the studies. A greater prevalence of dentoalveolar pain in patients visiting community public health clinics than those visiting private clinics may suggest an association of dentoalveolar pain and patient socioeconomic status because community health clinics are key resources for underserved populations. Other data, including the presence of treatment records for restorative treatment but not dental maintenance and the loss of permanent teeth, may imply past or present poor oral health [41]. Overall prevalence was also higher for low income countries and the studies highlight the importance of educating patients in the dangers associated with self-medication and late presentation for treatment. Many studies [15,12,17,19,20,24,27] have reported that the prevalence of OFP decreases in the older age groups, however it is unclear why. In the all 39 studies, female have higher overall prevalence or incidence rates than males, which has been attributed to biological, hormonal, stress factors and psychosocial characteristics. Gender-specific mechanisms seem to play a role not only when facial pain occurs, but also in the transition from acute to chronic forms.

When we meta-analyzed the ratios of prevalence for different recall periods across case definitions, we found a consistency in the ratios across case definitions indicating recall period effect is similar across case definitions. The studies that used recall period of 6 month had a pooled 10.9 % lesser prevalence of reported OFP to that of the point prevalence for the same case definitions which can be attributed to recall bias. Recall bias is a systematic error that occurs when participants do not remember previous events or experiences accurately or omit details; the accuracy and volume of memories may be influenced by subsequent events and experiences. Similarly, studies that used a recall period of 6 months had a pooled 25.9 % lesser prevalence of reported OFP to that of the 1-month recall period for the same case definitions. When we compared studies conducted at 1 month to that at point prevalence, we found that studies conducted at point prevalence had a pooled 10.8 % lesser prevalence of reported OFP to that of 1 month recall period for the same case definitions.

As previously noted, in the graph (Figure 8) we see that the average prevalence increases from point prevalence to 1-month recall period, and then decreases with 6, 12 and 24 month recall period. All our studies have a standard question "Do you have pain?" or "Did you have pain in the past X months ?" (Table 2). One reasonable explanation for this type of graph pattern would be that there may be 2 factors that control the prevalence rate. One would be a recall bias (which would be dependent on the recall period) and other would be accumulation (because over time a greater number of people will report pain) on prevalence of the pain. What happens with OFP pain is that from current prevalence to 1 month, the "accumulation" of pain has more effect than "recall bias" simply because people are less likely to forget pain in one month. As the recall period increases, the accumulation starts having lesser effect on prevalence than recall bias. The reasons

for this would be that the "accumulation" effect of pain starts to be plateauing, while the recall bias starts to increase with time. So, as we proceed with 6, 12 and 24 months, the "recall bias" gets stronger, while the "accumulation" effect, which is still there, does not increase or have an effect at the same rate. Figure 9 shows, hypothetically, the effect of these two factors on the prevalence of OFP over time that can yield the prevalence pattern shown in figure 8. Adjustment of these factors would likely yield more accurate values for the prevalence of OFP.

V b. Strengths and limitations:

Our strengths include that this was the first study to conduct a systematic review from 1980 to 2020 of prevalence and incidence of Orofacial Pain (OFP) on a global level that identified, compared and analyzed the various case definitions and recall periods used in the studies. We were able to analyze the influence of recall period on prevalence rates and obtain a pattern for the change which has not been done till now to our knowledge. There were some limitations in the study. The studies summarized used different methodologies when trying to measure the prevalence of OFP. Some used telephone interviews, some used postal surveys, some used personal interviews followed by actual physical examination, which can result in different estimates of the frequency of orofacial pain. Another limitation is we only had studies from 24 countries, reflecting the need to conduct more population-based studies on incidence and prevalence of oral facial pain.

V c. Conclusions

Orofacial pain is common in the community, with a mean prevalence of 32.2 %, and is composed of multiple often overlapping pain conditions, such as odontogenic pain, TMD, and facial pain [13]. No OFP studies included all subcategories of pain, although several include the majority of

pain conditions, likely yielding underestimates of the actual prevalence of OFP. We found a reduction in the prevalence of OFP as the time interval for recall increased. A lack of a consensus of the definition of OFP and what pain conditions should be measured and included is a limiting factor for the current systematic review. The next crucial steps would be to use the data and results derived from this study to derive global estimates and also project estimates for countries that don't have any data available using the GBD protocol and methodology. It is evident from these studies that there is general lack of data about the impacts and the burden of orofacial pain conditions. It is vital to conduct more population-based studies to derive incidence and prevalence which would be crucial in calculating the burden and overall impact which is suspected to be significant. There is also a great need for epidemiological studies of orofacial pain to better analyze etiology, including demography and lifestyle, local and mechanical factors and medical history, with the aim of achieving better diagnosis and therapy.

VI. FIGURES AND TABLES

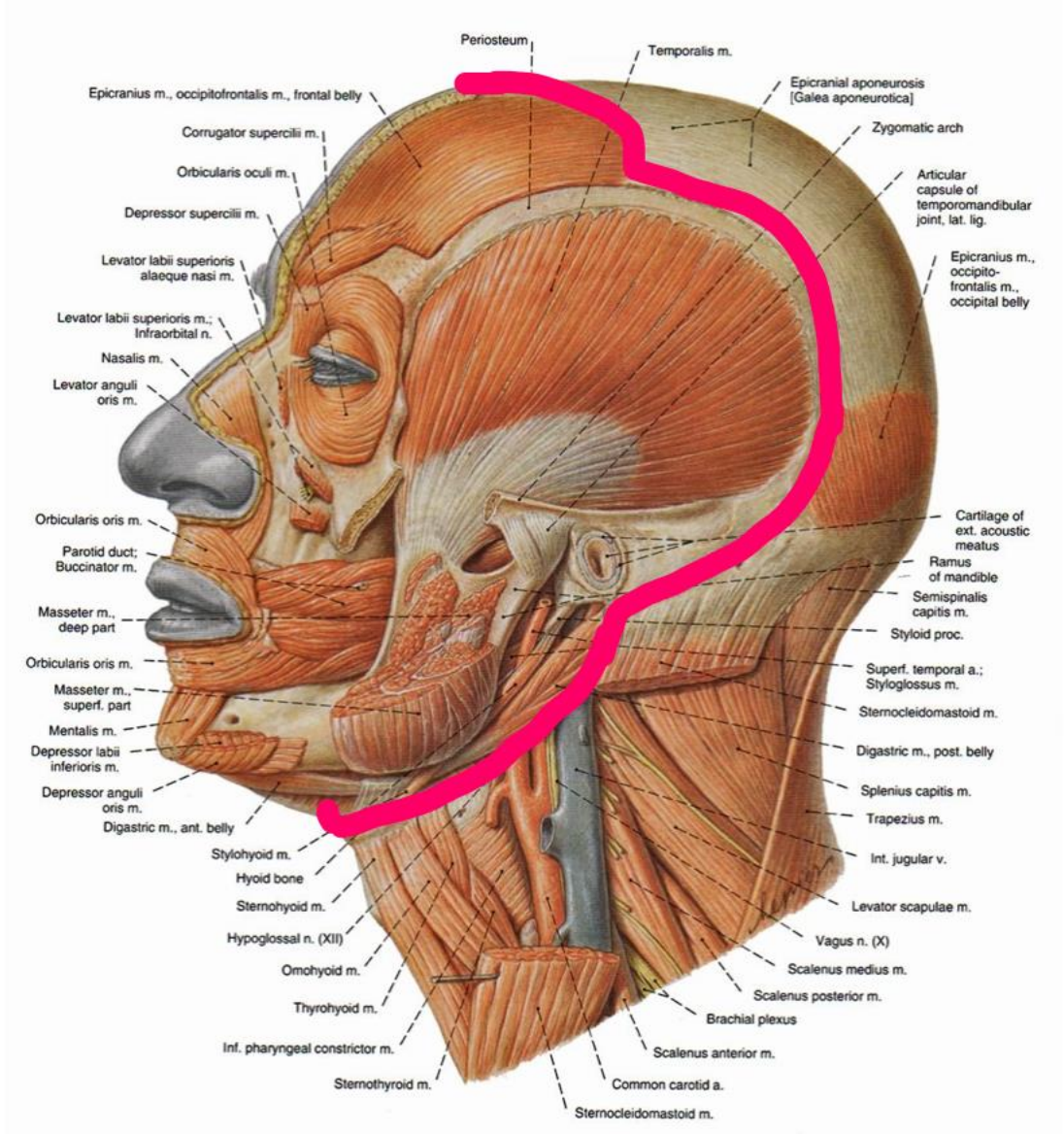

Figure 1: Anatomical Extent of OFP (demarcated by pink line).

The anatomical boundaries were as follows: bounded superiorly by the galea aponeurotica, and laterally and inferiorly by mandible and the temporal fossa, it includes all the structures above the neck in this region.

Vascular and nonvascular intracranial cause of orofacial pain
Headache associated with vascular intracranial disorders (IHS/ICHD-3 code 6.1 to 6.6)
Headache associated with nonvascular intracranial disorders (IHS/ICHD-3 code 7.1 to 7.8)
Primary headache disorders
Migraine (IHS/ICHD-3 code 1.1 to 1.6)
Tension-type headache (IHS/ICHD-3 code 2.1 to 2.4)
Cluster headache and other trigeminal autonomic cephalalgias (IHS/ICHD-3 code 3.1 to 3.5)
Neuropathic pain
Episodic neuropathic pain (IHS/ICHD-3 code 13.1.1, 13.2, 13.3, 13.9)
Continuous neuropathic pain (IHS/ICHD-3 code 13.1.2, 13.10, 13.11, 13.12.2)
Dysesthesia
Intraoral pain disorders
Odontogenic pain
Non odontogenic pain
Oral mucosal pain
Temporomandibular disorders (see Table 5)
Temporomandibular joint disorders
Masticatory muscle disorders
Extracranial causes of orofacial pain and headaches
Pain stemming from tissues or organs in the head and neck (IHS/ICHD-3 code 11.1, 11.3 to 11.5)
Pain stemming from systemic disease (IHS/ICHD-3 code 13.12.1)
Cervicogenic mechanisms of orofacial pain and headaches
Common cervical spine disorders (IHS/ICHD-3 code 11.2, 11.8, 13.2, 13.4)
IHS International Headache Society, *ICHD-3* International Classification of Headache Disorders

Figure 2: The American Academy of Orofacial Pain guidelines.

Classification structure of orofacial pain conditions from the 5th edition of the American Academy of Orofacial Pain guidelines (De Leeuw and Klasser 2013)

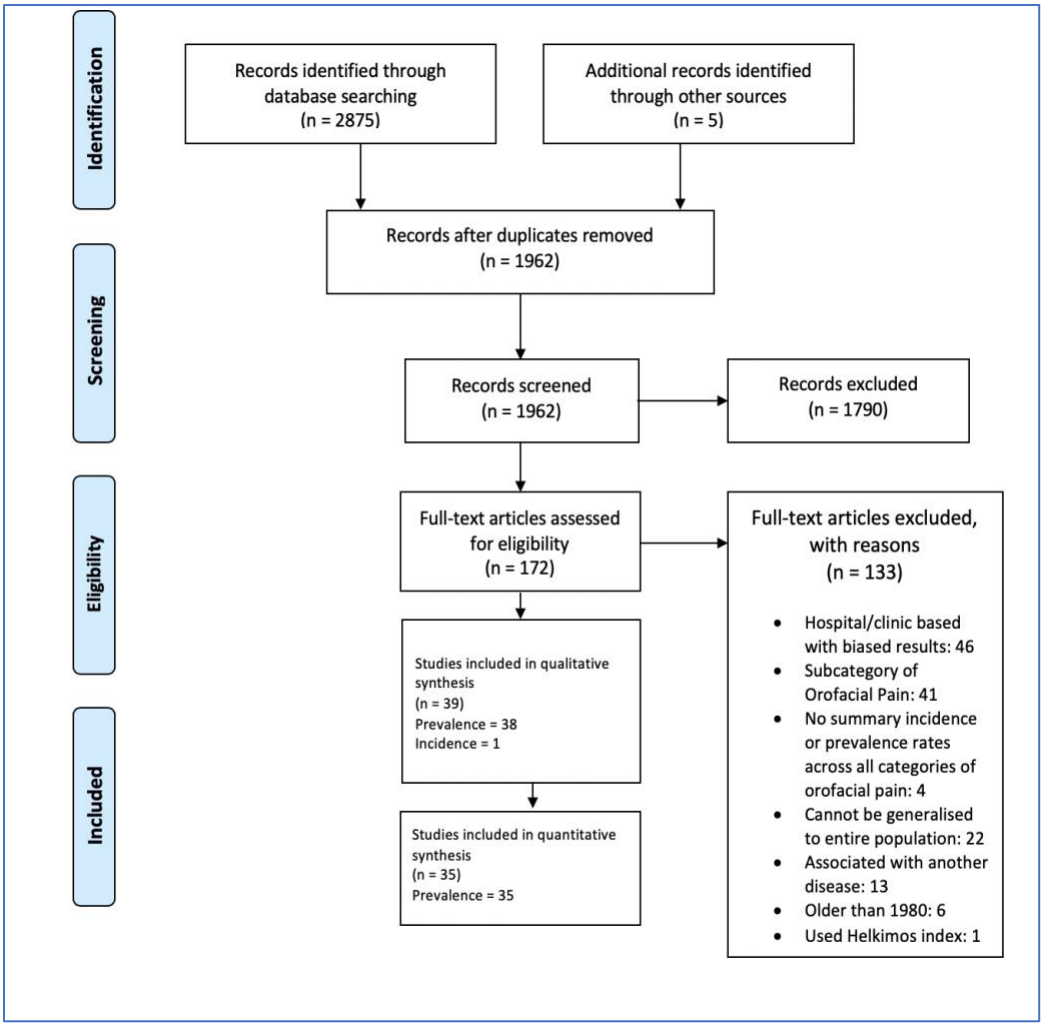

Figure 3: PRISMA flow chart for literature search.

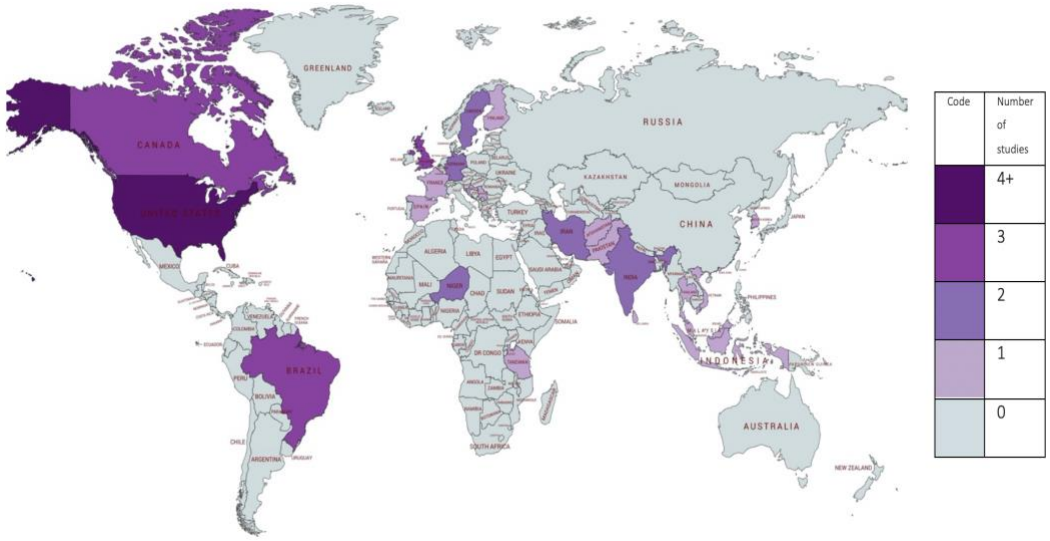

Figure 4: World map shows the number of articles with data on prevalence of OFP by country where the study was conducted.

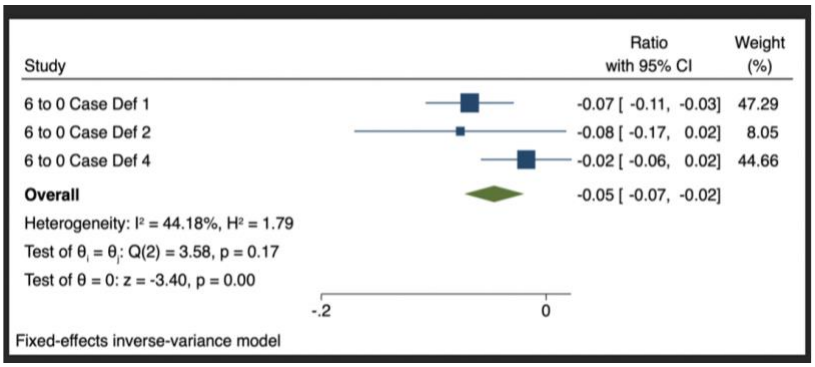

Figure 5: Presents a forest plot for the analyses comparing the prevalence found with a 6-month recall period to the point prevalence for the same case definition

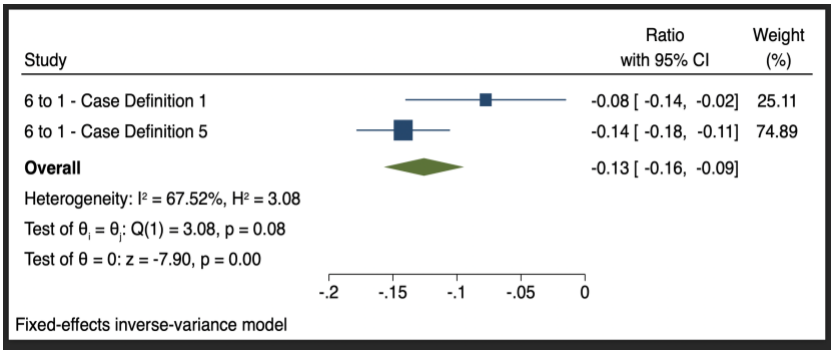

Figure 6: Presents a forest plot for the analyses comparing the prevalence found with a 6-month recall period to the 1-month recall period for the same case definition

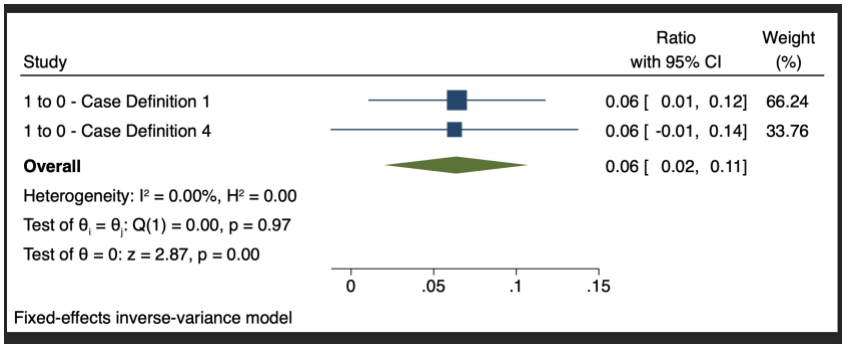

Figure 7: Presents a forest plot for the analyses comparing the prevalence found with a 1-month recall period to the point prevalence for the same case definition

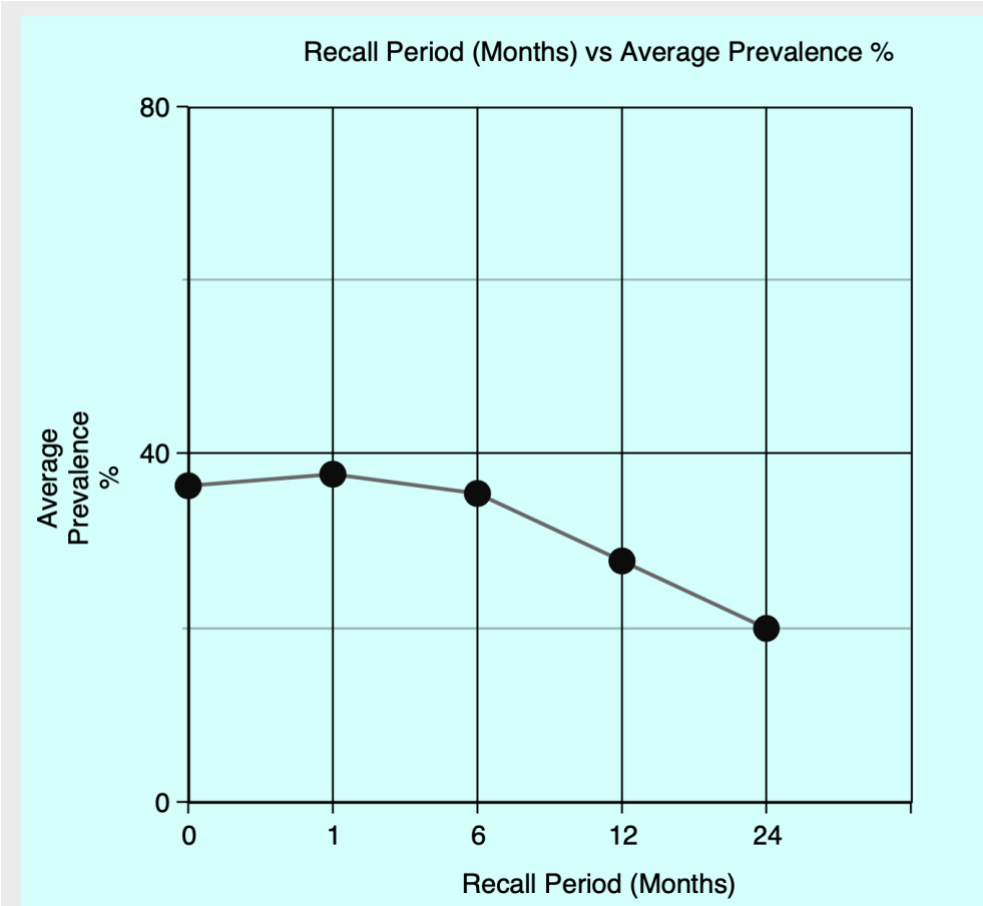

Figure 8: Recall Period vs average prevalence of orofacial pain across all case definitions.

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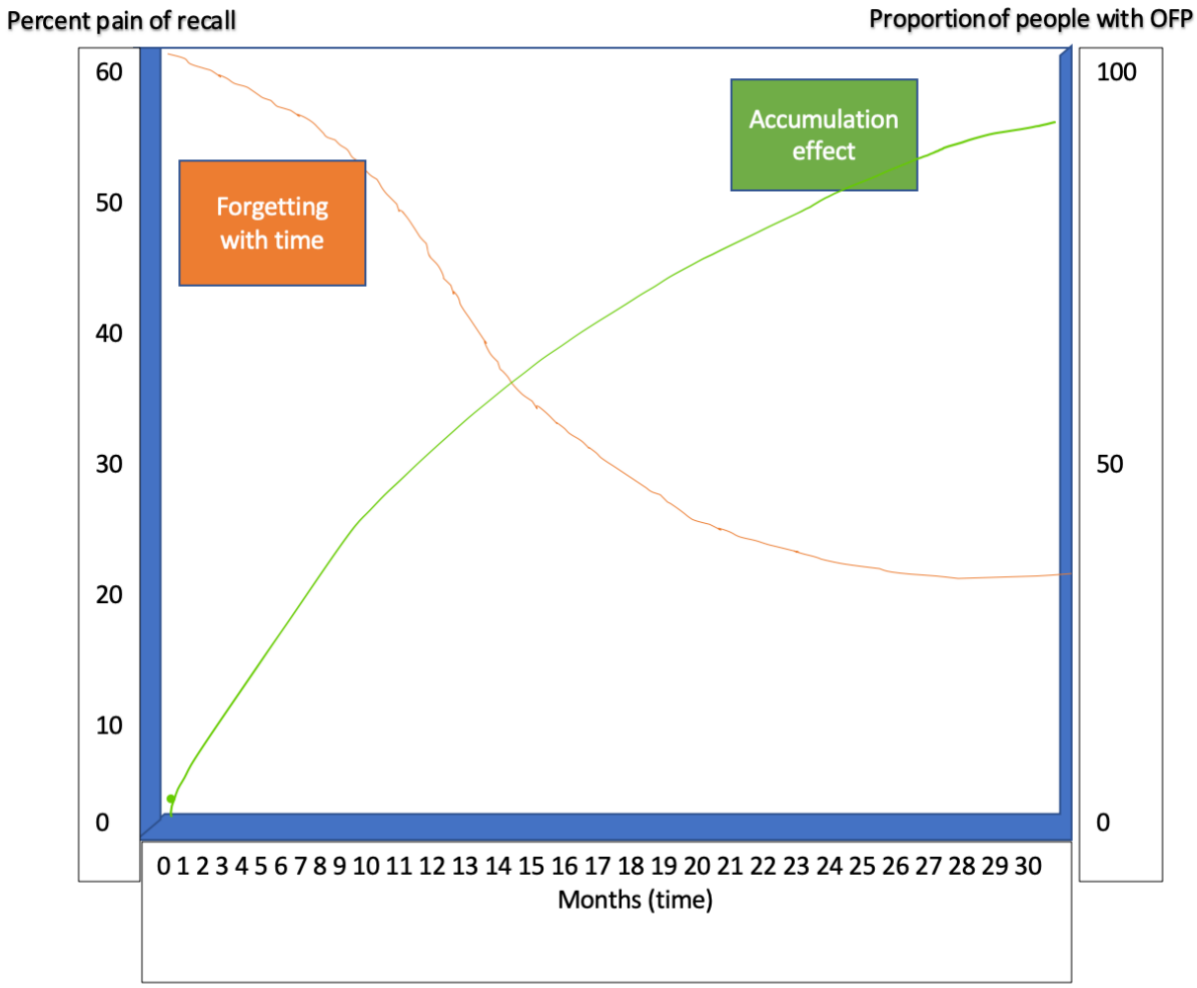

Figure 9: Hypothetical relationship between proportion with orofacial pain with increasing time span (accumulation effect) and the "forgetting" that occurs with distance since the event (decrease in recall of pain).

Table 1: Studies included: Key data

Reference	Location	Type of study & data source	Sex	Study Sample size	Prevalence	Recall Period
11	Cheshire East, UK	General medical practice in South East Cheshire, United Kingdom	Both	2504	26.18 %	1 month
12	São Paulo, Brazil	Volunteers from 68 districts of the city Piracicaba, São Paulo.	Both	400	54.75 %	Point Prevalence
13	West Java, Indonesia	Participants from regencies and three cities in the West Java province were recruited	Both	1551	49.9 %	6 months
14	Seoul, South Korea	Cohort of the Korean National Interview Survey of Oral Health Status (KNISOH), South Korea	Both	1032	42.0 %	6 months
15	United States of America	The overall sample is selected to represent the entire civilian, non-institutionalized, non-military population living in the United States.	Both	42370	28.06%	6 months
16	Madhya Pradesh, India	Children selected from various public and private schools located in Indore city, India	Both	800	17.9%	Point Prevalence

Table 1: Studies included: Key data

17	Finland	Geographically defined area of the two northernmost provinces of Finland.	Both	5696	15%	12 months
18	Florida, USA	Stratified random sample of community dwelling older (65+) north Floridians.	Both	1636	17.4%	12 months
19	Florida, USA	Data were taken from the Florida Dental Care Study (FDCS), a prospective longitudinal study of oral health and dental care.	Both	724	39.3 %	6 months
20	Nigeria	The study center is a tertiary hospital located in Ibadan, the largest city in West Africa	Both	426	74.88%	6 months
21	Serbia	Students from the Medical Faculty and second-year students from the Faculty of Technical Sciences of the University of Kosovska Mitrovica	Both	319	32%	3 months
22	Sri Lanka	Children attending schools in the Education Division of Badulla, province, Sri Lanka	Both	576	25%	2 months
23	Nigeria	Dental Center of University of Port Harcourt Teaching Hospital, Port Harcourt, Nigeria. A major referral center in the southern region of Nigeria.	Both	2426	18.5%	12 months

Table 1: Studies included: Key data

24	São Paulo, Brazil	Population-based cross-sectional study of adult population of the district Ermelino Matarazzo East Zone of Sao Paulo	Both	890	55.5%	6 months
25	Germany	People were surveyed who were representative for the German population according to age, sex and education	Both	2050	19.5%	24 months
26	Karnataka, India	Study area: Study was carried out in urban population of Hubli - Dharwad which are located at a distance of around 420 km north of Bangalore.	Both	1600	26.94%	1 month
27	Hong Kong Special Administrative Region of China	A sample of Cantonese-speaking Chinese people aged 18 years and over living in Hong Kong was interviewed	Both	1222	41.6%	1 month
28	Spain	The study was carried out in Catalonia, a region in the north-east of Spain	Both	5000	22.65 %	6 months
29	West Java	Patients at 35 Indonesian Community Health Centers, in Bandung, Indonesia	Both	700	55.9%	6 months
30	Eastern Sub-Saharan Africa	The study area covered six urban and eight rural study clusters, which had been selected using the WHO Pathfinder methodology in Tanzania.	Both	1759	59 %	12 months

Table 1: Studies included: Key data

31	Cheshire East	General medical practice in South East Cheshire, North West England	Both	1680	19 %	1 month
32	Germany	A Federal Health Survey - German National Health Interview and Examination Survey: Response	Both	7124	16%	6 months
33	Sweden	Inhabitants of West Bothnia were studied for prevalence of symptoms and clinical signs of mandibular dysfunction	Both	1992	18.75%	Point Prevalence
34	São Paulo, Brazil	This cross-sectional, analytical study conducted at the Preventive Dentistry Clinic of the School of Dentistry of Araraquara (UNESP)	Both	80	77.5%	6 months
35	Washington, USA	Patients were taken from general dentists in their clinics was conducted from September 2006 to July 2009 in the Northwest PRECE- DENT research network, a dental practice–based research network.	Both	1639	16.1%	12 months
36	Germany	This survey assessed procedures performed by general dentists in 42 German university hospitals treating patients with chronic orofacial pain (COP).	Both	1593	16%	3 months

Table 1: Studies included: Key data

37	Iran	In elderly subjects referred to the Shiraz Oral Medicine and Prosthodontics Department (Iran)	Both	5,221	11.6 %	Point Prevalence
38	Malaysia	Study in sixteen-year-old students from three rural schools in Kelantan to establish the prevalence of orofacial pain and discomfort. The sampling frame consists of Form Four Malay students (average age 16 years) enrolled in government assisted schools in the Districts of Kota Sham, Bachok, Pasir Putih, Pasir Mas, Tanah Merah and Tumpat in Kelantan.	Both	135	27 %	1 month
39	United Kingdom	Cross-sectional observational data was collected from four nursing homes in London, UK. These are residential settings that are staffed by qualified nurses and can provide nursing care	Both	1110	16.4 %	Point Prevalence
40	Canada	Five hundred randomly selected households on the roster of a group family practice unit (H.S.O.) were sampled. The practices chosen were representative of the general population of the City of Burlington	Both	500	22.9 %	1 month

Table 1: Studies included: Key data

41	Spain	Spanish 35- to 44-year-old adults and more elderly people in the Spanish National Oral Health Survey.	Both	540	34.0%	Point Prevalence
42	Canada	The mail survey was undertaken in the City of Toronto, Canada	Both	1014	53%	1 month
43	England	Participants of a large prospective cohort study of school children in South Wales, United Kingdom (11–13) participated	Both	337	23 %	Point Prevalence
44	Michigan	203 children from a pre-school and kindergarten. 20 miles west of Detroit, Michigan, U.S.A	Both	203	25.1%	Point Prevalence
45	Croatia	The study consisted of patients who came to the dental clinic for regular check-ups, therapy, or because of symptoms arising as a result of pathological conditions of the orofacial region in Croatia	Both	2,735	16.49%	Point Prevalence
46	Iran	The present study was conducted in the Oral and Maxillofacial Medicine, Department of Shiraz School of Dentistry, Shiraz, Iran, for the past 10 years from October 2005 to October 2015	Both	427	18.1%	6 months

Table 1: Studies included: Key data

47	Pakistan	This cross-sectional study was carried out on patients who visited the private dental clinics in Peshawar (in Province Khyber Pakhtunkhwa, Paki- stan) for the dental treatment	Both	4735	63.36%	Point Prevalence
48	Thailand	Randomly selected people living in 10 different districts in Bangkok	Both	1501	30%	6 months

Reference	Location	Type of study and data source	Sex	Sample size	Incidence
49	Denmark	30 dental practices in Ribe County, Denmark.	Both	35464	2.1 %

Table 2: Methods of Evaluation of OFP and specific questionnaires in the study :

Reference No.	Recall Period / Time	Method of Evaluation / measuring OFP	Summary of procedure and question set in the study	Assessment
11	1 month	**Postal questionnaire**	The main question concerning OFP consisted of nine items about various types of pain experienced over the past month. The OFP was defined as present if the respondent answered **positively** to any of the following *Which of the following symptoms have you had during the past month?* *Pain in the jaw joint/s* *Pain in area just in front of the ear/s* *Pain in or around the eyes* *Pain when opening the mouth wide* *Shooting pains in the face or cheeks* *Pain in the jaw joint when chewing food* *Pain in and around the temples* *Tenderness of muscles at the side of the face* *A prolonged burning sensation in the tongue or other parts of the mouth*	Pain experienced in the past 1 month. *Tenderness of muscles at the side of the face*
12	Point Prevalence	**Questionnaire**	Orofacial Pain Questionnaire developed by the authors, with no intention of being a diagnostic tool, the questionnaire is made up of questions aiming at sample demographic characterization (gender, age, education level, economic condition, district of residence), six questions to evaluate the presence of OFP *Do you have pain anywhere in the face region ?Do you have pain in any teeth ? Do you have pain in and around the temples ? Do burning sensation in the tongue ? Do you have painful sores in the mouth or around the lips ?*	Do you have any pain right now ?
13	6 months	**Questionnaire**	Interviewed by using an Indonesian version this questionnaire *Have you had the following symptoms during the past 6 months?* • *Toothache* • *Pain in the jaw joint/s* • *Pain in area just in front of the ear/s* • *Pain in or around the eyes* • *Pain when opening the mouth wide* • *Shooting pains in the face or cheeks* • *Pain in the jaw joint when chewing food*	Very detailed Questionnaire Pain experienced in the past 6 months.

			• *Pain in and around the temples* • *A prolonged burning sensation in the tongue or other parts of the mouth* *** for detailed Questionnaire see article* *Additionally, they were also asked if they experienced two or more symptoms.*	
14	6 months	**Telephone Interview**	A Telephone interview was conducted, and the following questions were asked as a part of the survey. *Jaw joint pain: In the past 6 months, did you have pain in the jaw joint or in front of the ear?* *Face pain: In the past 6 months, did you have pain in the face or cheeks? Tooth pain: In the past 6 months, did you have a toothache ?Oral sores: In the past 6 months, did you have painful sores in the mouth or around the lips?* *Burning mouth: In the past 6 months, did you have a burning sensation on the tongue or other parts of the mouth?*	Pain experienced in the past 6 months.
15	6 months	**Questionnaire**	A questionnaire was used to assess the OFP Prevalence. People reported that they had experienced at least one of the five types of orofacial pain during the past 6 months. *All individuals were asked "During the past six months, did you have more than once. . pain in the jaw joint or in front of the ear; a dull, aching pain across your face or cheek (excluding sinus pain);a prolonged, unexplained burning sensation in your tongue or any other part of your mouth; painful sores or irritations around the lips or on the tongue, cheeks or gums;*	Pain experienced in the past 6 months
16	Point Prevalence	**Questionnaire +Examination**	A standardized structured pro forma was developed to collect the required information. *The first part of the pro forma was used to collect information on sociodemographic characteristics like age, gender, socioeconomic status, type of school and previous dental visit, etc. The socioeconomic status was assessed using the Prasad scale. The second part of the questionnaire dealt with the presence/absence of orofacial pain*	Do you have any pain right now ?

			Based on that they were examined and classifies into Toothache, TMD pain, Oral sores, Facial pain, and Burning mouth sensation.	
17	12 months	**Questionnaire**	A Questionnaire was developed for the purpose of this study. The major part of the questionnaire concerned the following topics: *1. Have you had pain or ache in the face during the last 12 months? No/yes* *2. Have you had symptoms in the area of jaw joint during the last 12 months? No/yes* *3. Did you have painful sores in your mouth during the last 12 months? No/yes* *4. Did you have burning sensation in your tongue or any other part of your mouth during the last 12 months? No/yes* *5. Did you experience headaches during the last 12 months? No/yes*	Pain experienced in the past 12 months. "ache"
18	12 months	**Telephone interviews**	A Telephone interview was conducted, and the following questions were asked as a part of the survey. *Jaw joint pain. During the past 12 months did you have pain in the jaw joint or in front of the ear more than once?* *Face pain and headaches. During the past 12 months did you have a dull aching pain across your face or cheek more than once?* *Oral sores. During the past 12 months did you have painful sores or irritations around the lips or on the tongue, cheeks, or gums more than once?* *Burning mouth. During the past 12 months did you have a prolonged, unexplained burning sensation in your tongue or any part of your mouth more than once?* *Toothache. During the past 12 months did you have a toothache.*	Pain experienced in the past 12 months.
19	6 months	**Questionnaire**	A Questionnaire was developed for the purpose of this study. The major part of the questionnaire concerned the following topics: *Jaw joint pain: In the past 6 months did you have pain in the jaw joint or in front of the ear?* *Face pain: In the past 6 months did you have a dull aching pain across your face or cheek?*	Pain experienced in the past 6 months.

			Toothache: In the past 6 months did you have toothache pain? *Temperature sensitivity: In the past 6 months, have you had teeth that are sensitive to hot or cold fluids?* *Pain when chewing: In the past 6 months, have you had tooth pain while chewing?* *Oral sores: In the past 6 months, did you have painful sores or irritations around the lips or in your mouth?* *Burning mouth: During the past 6 months, have you had a burning sensation in your tongue or any other part of your mouth?*	
20	6 months	**Questionnaire**	Pain characteristics such as etiology, family history, location, duration, trigger aggravating or relieving factors, clinical diagnosis and treatment modalities in the last 6 months : *Have you had the following symptoms during the past 6 months?* *Pain in the jaw joint/joints or pain in the area just in front of the ear?* *Toothache ?* *Pain in the gums ?* *Shooting pain in the face ?* *Burning sensation in the tongue ?*	Pain experienced in the past 6 months.
21	3 months	**Questionnaire**	The questionnaire included items on the types of pain respondents had experienced during the previous 3 months. Orofacial pain was considered to be present if respondents answered positively to any of these questions: *In the previous 3 months did you have:* *a. toothache?* *b. pain in the area just in front of the ear?* *c. pain when opening your mouth or chewing food?* *d. pain in or around the eyes?* *e. pain in the temples?* *f. shooting pain in the face or cheeks?* *g. a prolonged burning sensation of the tongue or other parts of the mouth?*	Pain experienced in the past 3 months.
22	2 months	**Questionnaire**	The data were collected by means of a pre-tested questionnaire. These elicited information on past experience of oral pain and the degree of oral pain assessed using a three-point scale: mild, moderate and severe pain; trigger factors and possible reasons for the pain. *They were asked if they had the following symptoms during the past 2 months :*	Pain experienced in the past 2 months.

			Toothache ? Oral sores ? Pain in the jaw joints ? Pain in the face or cheeks ? Burning sensation in your tongue ?	
23	12 months	**Questionnaire + Examination**	Questionnaire was created to ask for pain in teeth, gum pain, oral sores, acute trauma or any tumor pain in the jaw joints or any facial pain *The patients were later examined to ascertain the specific causes of pain; these were also documented in the special forms. The history taking and examination were carried out by the same examiner for all the patients.*	Pain experienced in the past 12 months.
24	6 months	**Questionnaire**	Patients were asked if they experienced any pain complaint in the last 6 months 1.*Headache 2. Pain at the tongue 3.Toothache 4. Facial pain 5. Oral sores*	Pain experienced in the past 6 months.
25	Point Prevalence	**Questionnaire**	Questionnaire is a screening instrument for identification of persons with somatoform disorders. It is strictly based on the criteria of the classification systems DSM-IV [1] and ICD-10 [15] which specified symptom lists for the diagnosis of somatoform disorders. A certain number of symptoms is required along with various inclusion and exclusion criteria. *Among these items were 10 items measuring the prevalence of somatoform pain symptoms: head or facial pain*	Do you have any pain right now ?
26	1 month	**Questionnaire**	Questionnaire was created to ask for pain – *Did you have any of the following symptoms do you had during past one month?* *Pain in jaw joints, Pain in the area just in front of ears, Pain in / around the eyes, Pain when opening the mouth wide open, Shooting pain in the face/ jaw joints, Pain in jaw joints while chewing food, Pain in /around temples/side of forehead, Pain in /around temples/side of forehead when touched, Frequent headaches because of pain in orofacial region, Toothache, Painful gums, Painful sore spots/ulcers in your mouth, Prolonged burning sensation in tongue other*	Pain experienced in the past 1 month.

			parts of your mouth, Sensitivity in your teeth due to hot/ cold foods.	
27	1 month	**Questionnaire**	The data were collected by means of a pre-tested questionnaire. In the past one month, have you had any of these types of pain? *The pain symptoms included toothache, pain in the teeth with hot or cold liquids, pain in the jaw joints, pain in the jaw while chewing, pain in the jaw joint / s while opening the mouth wide, pain in the face in front of the ear, a prolonged burning sensation in the tongue or other parts of the mouth, and sharp shooting pains across the face, head and / or cheeks*	Pain experienced in the past 1 month.
28	6 months	**Questionnaire**	A Questionnaire was developed for the purpose of this study *Patients were asked if they experienced any pain complaint in the last 6 months* *-in the head, face or teeth region, regardless of its intensity and duration. The presence or absence of pain and its body location during the previous 6 months were determined. When more than one location was reported by the interviewee, he or she was asked which had been the most troublesome. All subsequent data obtained related only to this pain location for practical purposes*.	Pain experienced in the past 6 months.
29	6 months	**Questionnaire**	A Questionnaire was developed for the purpose of this study *Have you had the following symptoms during the past 6 months?* *Toothache* *Pain in the jaw joint/s* *Pain in area just in front of the ear/s* *Pain in or around the eyes* *Shooting pains in the face or cheeks* *Pain in the jaw joint when chewing food* *Pain in and around the temples* *A prolonged burning sensation in the tongue or other parts of the mouth*	Pain experienced in the past 6 months.

30	12 months	**Questionnaire**	The questionnaire was constructed from a list of oral health indicators summarized in the report of the *Consensus Workshop for Selecting Essential Oral Health Indicators in Europe* held at the University of Granada, Spain. https://ec.europa.eu/health/ph_projects/2002/monitoring/fp_monitoring_2002_a2_frep_03_en.pdf	Pain experienced in the past 12 months.
31	1 month	**Questionnaire + Examination**	A Questionnaire was developed for the purpose of this study *Patients were asked if they have had any pain in their face, mouth or jaws that has lasted for one day or longer' during the past month*	Pain experienced in the past 1 month
32	6 months	**Questionnaire**	With the help of a questionnaire - *Pain in the face and tongue, in the chewing muscles, head and in the temporomandibular joint or in the ear area in the was assessed in the last 6 months*	Pain experienced in the past 6 months.
33	Point Prevalence	**Questionnaire + Examination**	A Questionnaire was developed for the purpose of this study *The questions in the interview concerned the occurrence of headaches and migraine, the duration and location of these symptoms and other pain symptoms from the oral and neck area, and awareness of various oral parafunctions such as grinding or clenching of teeth and biting habits.* *The questions were constructed to be answered yes or no, and the participants were allowed to discuss, after interviews, the participants were examined clinically.*	Do you have any pain right now ?
34	6 months	**Questionnaire**	A Questionnaire was developed for the purpose of this study *They had: "spontaneous toothache; toothache caused by hot/cold liquids or sweets; prolonged burning tongue sensation; pain in the jaw or during chewing; pain when opening the mouth, in the face or around/behind the eyes". The possible*	Pain experienced in the past 6 months.

			answers "yes" or "no" determined the prevalence of orofacial pain. Those who answered yes were also asked about the intensity of pain on a scale of 1 (mild pain) to 4 (very severe).	
35	12 months	**Questionnaire**	A questionnaire was used to assess the OFP Prevalence. *For collecting data on diagnosis and treatment of oral conditions, and specifically the presence of pain in the orofacial tissues during the past 12 months.* *In the past 12 months :* *Did you have pain in the jaw joint or in front of the ear ?* *Did you have pain across your face or cheek ?* *Did you have toothache ?* *Did you have painful sores ?* *Did you have pain in your tongue ?*	Pain experienced in the past 12 months.
36	3 months	**Questionnaire**	A Questionnaire was developed for the purpose of this study *In the past 3 months did you have :* *Pain in the jaw joint/s ;Pain when opening the mouth wide ; Shooting pains in the face or cheeks Pain in the jaw joint when chewing food ;Pain in and around the temples ; A prolonged burning sensation in the tongue or other parts of the mouth*	Pain experienced in the past 3 months.
37	Point Prevalence	**Questionnaire + Examination**	A questionnaire was used to assess the OFP Prevalence. *Do you have* *a)headaches b) pain in the face c) pain in the jaw joints/front of the ears d) pain at the tongue ?* *The questions were constructed to be answered yes or no and he the participants were examined.*	Do you have any pain right now ?
38	1 month	**Questionnaire + Examination**	Subjects were first asked to complete a self-administered questionnaire concerning	Pain experienced in the past 1 month.

			episodes of pain experienced in the past 1 month. 1. *Toothache ?* 2. *Soft tissue ?* 3. *TMJ ?* 4. *Through Hot / Cold Fluids ?* *This was followed by an interview to check on the accuracy of the responses about their orofacial pain episodes. Subsequently the subjects were examined clinically to diagnose the cause of pain / discomfort and to record their dental caries status, periodontal status and presence of traumatized teeth. However, no further investigations such as radiographs or pulp vitality testing were done to confirm the preliminary diagnosis.*	
39	Point Prevalence	**Questionnaire + Examination**	Participants were questioned about if they experienced any *Toothache, Oral sores, Pain in the jaw joints, Pain in the face or cheeks.* *A brief oral examination was conducted*	Do you have any pain right now ?
40	1 month	**Questionnaire**	All participants in the study were questioned *Have you or any family member experienced any noteworthy pain in the head and face within the past 1 month ?*	Pain experienced in the past 1 month.
41	12 month	**Questionnaire + Examination**	All participants were questioned about if they experienced any sort of pain because of problems with their mouth, teeth, or dentures in the previous 12 months. *The replies of the participants concerning pain or eating problems were recorded on a Likert-type scale (0 = never, 1 = hardly ever, 2 = sometimes, 3 = fairly often, and 4 = very often). The prevalence of impact was estimated using the sometimes threshold to visualize the pro- portion of subjects suffering from pain or eating problems with a certain frequency. A consented, standardized clinical oral examination for caries, periodontal disease, temporomandibular joint (TMJ) function, and prosthodontic status was performed by calibrated examiners.*	Pain experienced in the past 12 month.
42	1 month.	**Questionnaire + Examination**	Participants were questioned about if they experienced any *Pain in the teeth with hot or cold fluids or sweet things, Toothache, Pain in the jaw*	Pain experienced in the past 1 month.

			joints, Pain in the face just in front of the ear, Pain the jaw while chewing, Pain in the jaw joint when opening the mouth wide, Sharp shooting pains across the face or cheeks. A prolonged burning sensation in the tongue or other parts of the mouth *These patients completed the questionnaire prior to a clinical examination.*	
43	Point Prevalence	**Questionnaire**	Orofacial pain (OFP) was defined as present if the participant reported having at least one of the following: *pain from the joint of the jaw; pain or discomfort when opening the mouth wide; pain in the muscles of the jaw, bleeding gums, tongue pain, stiffness in the jaw and headaches.*	Do you have any pain right now ?
44	Point Prevalence	**Questionnaire + Examination**	Interviewed concerning acute pain, jaw movement impairment, TM joint sounds, parafunctions, and pain history. *Do you have any pain in the jaws during chewing such things as gum?* *Do you have any problems in opening your mouth?* *Do you have any facial pain ?* *Are you aware of or has anyone told you that yon grind your teeth?* *Pain history. Headache, earache, neck pain and jaw- joint pain frequencies were recorded.* *Care was taken when questioning to determine if pain was derived from pulpitis, sensitive tooth areas, or periodontal tissues. The TM Joints were palpated laterally and posteriorly, during mandibular rest and during jaw movements. Soreness and tenderness were recorded on a three- graded scale: 0 = no pain; 1 - slight soreness on left and/or right side; 2=distinct pain and/or palpebral reflex on left and/or right side.*	Do you have any pain right now ? "Soreness and tenderness"
45	Point Prevalence	**Questionnaire + Examination**	A Questionnaire was developed for the purpose of this study *Do you have pain anywhere in the face region or tongue ? Do you have pain in any teeth ? Do you have pain in the jaw joint ? Do you have pain in the area in front of your ear ? Do*	Do you have any pain right now ?

			you have pain in or around the eyes ? Do you have pain in and around the temples ?	
46	6 months	**Questionnaire**	OFP was measured with the help of a questionnaire . *Did you experience pain or soreness in the past 6 months ? Y/N. OFP was classified using criteria of IASP that modified to four categories; Temporo-Mandibular Disorders (TMD), neuralgia, psychological facial pain, BMS and others (headache, odontogenic, sinusitis, etc.)*	Pain experienced in the past 6 months. *"Soreness"*
47	Point Prevalence	**Questionnaire + Examination**	*A specially designed questionnaire was prepared by the authors.* *The patients were examined by authors in their clinics by using the same questionnaires and clinical methods. The questionnaire included age, sex and various pain characteristics. They were assessed for Toothache, Shooting pains in the face or cheeks, Pain in the jaw joint/s, Pain in the jaw joint when chewing food, Pain in and around the temples, burning sensation in the tongue or other parts of the mouth.*	Do you have any pain right now ?
48	6 months	**Questionnaire**	The questionnaire was designed to collect information on current and past experiences about orofacial pain in the past 6 months. The questionnaire was composed of three parts. Sociodemographic data including age, gender and educational level were obtained from the questionnaire. The alternatives for education level were 'lower than bachelor's degree' and 'higher than bachelor's degree'. *Orofacial pain was queried as follows:* *(1) In the past 6 months, did you have aching pain across your face or cheek?* *(2) In the past 6 months, did you have painful sores or irritations around the lips or on the tongue, cheeks or gums?*	Pain experienced in the past 6 months.
49	Incidence Study	**Questionnaire + Examination**	The following variables were recorded for each patient: *I) age, 2) sex, 3) regularity of dental care (i.e. at least one annual consultation during the last 5 yr., yes/no), 4) pain region, 5) pain level a) moderate: causing discomfort, but*	Incidence

			endurable, b) severe: causing considerable discomfort, difficult to endure, c) very severe: unendurable, 6) duration of pain, 7) cause of the pain, 8) influence on daily functions, and 9) consumption of analgesics within the last 24 h, yes/no.	

Table 3: Distribution of Orofacial Pain subtypes for each included study.

Reference	TMJ	Toothache	Oral Sores	BMS	Facial Pain	Muscle Pain	Eye Pain	Primary Headaches	Acute Trauma	Gum Pain	Pathology Pain	Referred Pain	Total	Percentage of sub-type measured.
	1	**1**	**1**	**1**	**1**	**1**	**1**	**1**	**1**	**1**	**1**	**1**	**12**	**100%**
11	1	0	0	1	1	1	1	1	0	0	0	0	6	50%
12	0	1	1	1	1	0	0	0	0	1	0	0	5	42%
13	1	1	1	1	1	1	1	1	0	1	0	0	9	75%
14	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
15	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
16	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
17	1	1	1	1	1	1	1	0	0	1	0	0	8	67%
18	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
19	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
20	1	1	1	1	1	1	0	0	0	1	0	0	5	42%
21	1	1	1	1	1	1	1	1	0	1	0	0	9	75%
22	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
23	1	1	1	1	1	1	0	0	1	1	1	1	10	83%
24	0	1	1	1	1	0	0	0	0	1	0	0	5	42%
25	1	1	1	1	1	1	1	1	0	1	0	0	9	75%
26	1	1	1	1	1	1	1	1	0	1	0	0	9	75%
27	1	1	1	1	1	1	1	1	0	1	0	0	9	75%
28	1	1	1	1	1	1	1	0	0	1	0	0	8	67%
29	1	1	1	1	1	1	1	1	0	1	0	0	9	75%
30	0	1	1	1	1	0	0	0	0	1	0	0	5	42%
31	1	1	1	1	1	1	1	1	0	1	0	0	9	75%
32	1	0	0	1	1	1	1	1	0	0	0	0	6	50%
33	1	0	0	1	1	1	1	1	0	0	0	0	6	50%
34	1	1	1	1	1	1	1	1	0	1	0	0	9	75%
35	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
36	1	0	0	1	1	1	1	1	0	0	0	0	6	50%
37	1	0	0	1	1	1	1	1	0	0	0	0	6	50%
38	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
39	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
40	1	0	0	1	1	1	1	1	0	0	0	0	6	50%
41	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
42	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
43	1	0	0	1	1	1	1	1	0	0	0	0	6	50%
44	1	0	0	1	1	1	1	1	0	0	0	0	6	50%
45	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
46	1	1	1	1	1	1	1	1	0	1	0	0	9	75%
47	1	1	1	1	1	1	0	0	0	1	0	0	7	58%
48	1	1	1	1	1	1	1	0	0	1	0	0	8	67%
49	0	1	1	0	1	0	0	0	1	0	0	0	5	33.3%
Percentage of studies that measured each sub-type	92.3 %	79.4%	82.0%	97.4%	100%	92.3%	53.3%	46.15%	7.6%	79.4%	5.1 %	5.1%		

Table 4 : Case definitions with sub-types and different recall periods

CASE DEFINTION NUMBER	CASE DEFINITION SUB-TYPE	CASE DEFINITION	NUMBER OF STUDIES
1	A	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + EYE PAIN + HEADACHE **1 MONTH RECALL PERIOD**	2
	B	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + EYE PAIN + HEADACHE **6 MONTHS RECALL PERIOD**	1
	C	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + EYE PAIN + HEADACHE **POINT**	4
2	D	TOOTHACHE + ORAL SORES + BMS PAIN + FACIAL PAIN + GUM PAIN **POINT**	2
	E	TOOTHACHE + ORAL SORES + BMS PAIN + FACIAL PAIN + GUM PAIN **3 MONTHS RECALL PERIOD**	1
	F	TOOTHACHE + ORAL SORES + BMS PAIN + FACIAL PAIN + GUM PAIN **6 MONTHS RECALL PERIOD**	3
3	G	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + EYE PAIN + HEADACHE+ TOOTHACHE + ORAL SORES + GUM PAIN **6 MONTHS RECALL PERIOD**	4
	H	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + EYE PAIN + HEADACHE+ TOOTHACHE + ORAL SORES + GUM PAIN **3 MONTHS RECALL PERIOD**	1
	I	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + EYE PAIN + HEADACHE+ TOOTHACHE + ORAL SORES + GUM PAIN **24 MONTHS RECALL PERIOD**	1
	J	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + EYE PAIN + HEADACHE+ TOOTHACHE + ORAL SORES + GUM PAIN **1 MONTH RECALL PERIOD**	3
4	K	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + TOOTHACHE + ORAL SORES + GUM PAIN **6 MONTHS RECALL PERIOD**	3
	L	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + TOOTHACHE + ORAL SORES + GUM PAIN **POINT**	5
	M	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + TOOTHACHE + ORAL SORES + GUM PAIN **12 MONTHS RECALL PERIOD**	2
	N	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + TOOTHACHE + ORAL SORES + GUM PAIN **1 MONTH RECALL PERIOD**	2
5	O	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + TOOTHACHE + ORAL SORES + GUM PAIN + EYE PAIN **12 MONTHS RECALL PERIOD**	1
	P	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + TOOTHACHE + ORAL SORES + GUM PAIN + EYE PAIN **6 MONTHS RECALL PERIOD**	1
	Q	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + TOOTHACHE + ORAL SORES + GUM PAIN + EYE PAIN **1 MONTH RECALL PERIOD**	1
6	R	TMJ PAIN + MUSCLE PAIN + BMS PAIN + FACIAL PAIN + TOOTHACHE + ORAL SORES + GUM PAIN + ACUTE TRUAMA + REFFERED PAIN + TUMOUR PAIN **12 MONTHS RECALL PERIOD**	1

Table 5: Ratios for case definitions comparing 6-month to point prevalence

Case Definition	Ratio	Log10 Ratio	Standard Error	Study size
1	0.855	-0.068033	0.0197	7531
2	0.838	-0.076375	0.0478	642
4	0.96	-0.017728	0.0203	22350

Table 6: Ratios for case definitions comparing 6-month to 1-month prevalence

Case Definition	Ratio	Log10 Ratio	Standard Error	Study size
1	0.8365	-0.0775	0.0317	1533
5	0.7212	-0.1419	0.0184	2723

Table 7: Ratios for case definitions comparing 1-month to point prevalence

Case Definition	**Ratio**	**Log10 Ratio**	**Standard Error**	**Study size**
1	1.159	0.064083	0.0271	873
4	1.155	0.062581	0.0380	762

Table 8: Summary table of the ratios for prevalence rates, with different recall periods (age and sex matched)

Recall Period in months	Ratio
1 to 0 (Pooled)	1.12 (1.0-1.14)
6 to 0 (Pooled)	0.89 (0.87 - 0.90)
6 to 1 (Pooled)	0.74 (0.72-0.75)
12 to 0	0.51 (0.49-0.53)
3 to 1	0.97 (0.93 – 1.0)
24 to 1	0.76 (0.80 – 0.73)

Table 9 : Comparison of Odontogenic and TMD across different recall periods

Reference	Odontogenic	TMD	Ratio	Recall Period
13	15 %	10 %	1.5	6 months
15	12.2 %	5.3 %	2.3	6 months
16	10.1 %	4.3 %	2.3	Point
18	12.2 %	7.7 %	1.5	12 months
19	12.0 %	8.3 %	1.4	6 months
26	15.44 %	11.3 %	1.3	1 month
31	32 %	18.4 %	1.7	1 month
38	19.3 %	8.1 %	2.3	1 month
42	14.1 %	9.1 %	1.5	1 month
Average			1.76	

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