

Phantom tooth pain: Cause-effect relationship with tooth manipulation, injury or extraction

By

WorkSafeBC Evidence-Based Practice Group

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June 2008

About this report

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Published: June 2008

About the Evidence-Based Practice Group

The Evidence-Based Practice Group was established to address the many medical and policy issues that WorkSafeBC officers deal with on a regular basis. Members apply established techniques of critical appraisal and evidence-based review of topics solicited from both WorkSafeBC staff and other interested parties such as surgeons, medical specialists, and rehabilitation providers.

Suggested Citation

Martin CW. Phantom tooth pain. Richmond, BC: WorksafeBC Evidence-Based Practice Group; June 2008. Available at:

http://worksafebc.com/health_care_providers/Assets/PDF/phantom_tooth_pain.pdf

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Table of contents

About this report	i
Background	1
Methods	2
Results	2
Summary and conclusion	4
References	5
Appendix 1	26

Background

Recently, the Evidence Based Practice Group (EBPG) was presented with a claim in which a worker, injured in a motor vehicle accident, sustained an injury to the right side of his face and eye. The clinical findings at that time included right retinal detachment, blow out fracture of the right lower orbit with an associated fractured zygoma, perforation of the left tympanic membrane and a clinical basal skull fracture. CT scan confirmed the diagnosis of left temporal extradural haemorrhage. Due to ongoing complaint of pain after various courses of treatment, the right maxillary nerve was then ablated. However, the worker still complained of pain in the distribution of supraorbital and supratrochlear nerves with some sensation over the right cheek and upper lip after the ablation. At some stage, it was thought that two teeth (upper right central and lateral incisivus) contributed to the pain. These two teeth received root canals. However, the pain was only 'a little bit better' for a short period. Hence, the teeth were 'repaired' again after recurrence of the pain and were then extracted. It was found that these were normal teeth. In 1996 (nine years post injury), the neurologist consultant wrote that in August 1994 (seven years post injury), the patient developed pain in the left mandible (as opposed to the right side of the actual injury) in relationship to three teeth. Multiple dental procedures had been done and one of the molars was eventually extracted. This had not made any major impact upon the pain. The other two teeth had been treated with various dental procedures including root canals. The patient felt a slight reduction in pain at that time (1996), and felt the maximum pain was about the area where the missing tooth was and the gum tended to be very uncomfortable. It should be noted that the EBPG could not find any dental report/information pertaining to these three, presumably left, teeth. Subsequently, the injured worker developed chronic pain and was diagnosed with persistent right infraorbital/hemifacial pain. The characteristics of the pain seemed to be neuropathic in nature. This claim related to phantom tooth pain was presented to the Board.

For a long time, teeth, and more recently, dental pulp tissues, have been among the most frequently amputated structures in humans. Regardless of this fact, there is little information available with regard to post amputation pain in the oral cavity, unlike that of phantom limb pain. The term phantom tooth pain (PTP) was introduced in 1978 by Marbach.¹⁶³ Prior to 1978, similar symptoms were known as idiopathic periodontalgia and atypical odontalgia.²⁶⁶ Marbach defined PTP as a syndrome of persistent pain or paresthesia in teeth and other oral tissues that might appear following dental or surgical procedures such as pulp extirpation, apicoectomy or tooth extraction, when nerves were injured by physical trauma or even after routine inferior alveolar nerve blocks if the needle pierced the nerve sheath.¹⁶³ It should be noted that other experts⁵¹ strictly define PTP as unexplained persistent pain at the site of an extracted tooth. Due to the lack of objective findings, Klausner¹³⁵ suggested that PTP was a diagnosis of exclusion.

Subsequent to his 1978 paper,¹⁶³ Marbach published several papers on PTP.^{158-162, 164-167} However, it should be noted that the only epidemiological data presented by him was limited to data coming from one study.¹⁶⁵ Further, it should be noted that the available evidence of PTP was based on subjective patient reporting rather than on clinical examination of orofacial pain problems.¹¹¹ At present, there is no objective method for assessing PTP.¹¹¹

Given these facts, the objective of this rapid systematic review is to investigate whether there is a causal relationship between tooth manipulation, injury or extraction and the subsequent development of phantom tooth pain.

Methods

A systematic literature search was conducted on May 20, 2008. The search, done on MEDLINE and MEDLINE Daily Update commercial medical databases available through Ovid Technologies, was done by employing keywords ((phantom pain) AND (tooth OR teeth OR dentistry)), which also included related terms. The search was not limited to any particular publication years but was limited to publications in the English language and studies involving human subjects.

This search yielded 277 articles.¹⁻²⁷⁷ Upon examination of titles and abstracts, 19 articles^{25,26,51,103,111,133,158-165,169,205,246,256,266} were retrieved in full and critically appraised. Of these 19 articles, 12^{51,103,133,158,160,162-164,205,246,256,266} were further excluded since these articles were expert reviews in nature and did not provide any data that could be employed in assessing association (such as to calculate odds ratios, relative risk or attributable risk). The findings of seven articles^{25,26,111,159,161,165,169} are presented below.

Results

Of the seven articles retained, 2 were case reports (n=1)^{26,169} (Level of Evidence 4. Appendix 1), one was a small size (n=10) ‘case control’¹¹¹ (Level of Evidence 3. Appendix 1), two were moderate size case series (n=25 and 30)^{25,161} (Level of Evidence 4. Appendix 1), one was a ‘case-control’ study¹⁵⁹ (Level of Evidence 3. Appendix 1), and one was a survey¹⁶⁵ (Level of Evidence 4. Appendix 1).

Matwychuk¹⁶⁹ and Battrum et al.²⁶ each presented a case to illustrate the clinical presentation of PTP as well as provide expert reviews on the potential etiology, diagnosis and treatment of PTP. However, given the nature of these reports, there is no information that can be used to calculate the size of the association (i.e. the denominator of the odds ratio or relative risk or attributable risk) between purported exposures and PTP.

Jacobs et al.¹¹¹ reported a ‘case-control’ study to determine whether patients with PTP had an altered sensory perception as compared with pain-free patients. This ‘case control’ was based on a survey of patients of the Department of Periodontology at a university in Belgium. Five hundred McGill Pain Questionnaire-based letters were sent and only 176 (35%) of the questionnaires were returned. Of these 176 participants, ten were identified with potential PTP based on their answers to certain questions. The authors selected ten age- and sex-matched, pain-free controls and explored the association between various psychophysical factors, such as anxiety, depression, agoraphobia, obsessive compulsive behaviour, light pressure sensation, and thermal sensation, with PTP. It should be noted that this is a small ‘case control’ study with a very low response rate exploring many potential associations between psychophysical factors and PTP without any clear hypothesis or statistical adjustment on this issue.

Bates and Stewart²⁵ reported 30 patients diagnosed with PTP. Ninety percent of these patients were female, mean age 58.4 years, and had PTP for an average of 4.4 years. It should be noted that no information is given as to how the diagnosis of PTP was established. Also, there is no information provided regarding the population from which these cases were drawn or potential causes of PTP among these patients. A case series of 25 patients diagnosed with PTP was reported by Marbach.¹⁶¹ This case series suffers the same problems as the other case series by Bates and Stewart.²⁵

Marbach et al,¹⁶⁵ in their article entitled “Incidence of phantom tooth pain”, tried to calculate, retrospectively, the incidence of PTP post endodontic procedures. Five endodontists practicing in five separate private practices were invited to participate in this study. Only one endodontist (20%) agreed to participate. A 10-item questionnaire was mailed to 732 former unselected patients of this participating endodontist (the authors did not provide any information about the study period). Five hundred and ten (70%) responded and only 463 questionnaires (63%) were deemed usable. Presumably, this study explored the association between any kind of endodontic therapy and the development of PTP. Given the information provided in this article we could not discern further the types of endodontic treatments involved. It should be noted that while the authors did not collect any data from general dentists, the data from the patients of the endodontist may have covered both endodontic work as well as any general dental procedures performed by the endodontist. The data cannot be employed to assess the association between any specific endodontic treatments or specific dental treatments and the development of PTP. Based on this data, Marbach et al.¹⁶⁵ estimated the incidence of PTP in this population to be between 3%-6%. It should be noted that, in addition to a high potential for selection bias and the limited variables provided, this study did not provide any data that would enable us to compute any size of association between certain exposures and the development of PTP.

In another article, Marbach¹⁵⁹ compared the psychological factors among patients with PTP with those of control and contrast groups. 115 patients diagnosed with PTP, 151 patients diagnosed

with temporomandibular joint pain dysfunction syndrome (TMPDS) and 137 non-pain controls drawn from the acquaintances of the TMPDS group were included in this study. It should be noted that there was no information on how these samples were drawn. There was also no information on the size of the population from which these samples were drawn. Further, this was an exploratory study in which there was no specific hypothesis to be tested and various potential outcomes were presented.

With regard to establishing causation between certain exposures and outcomes, Sir Bradford Hill's "criteria" are the "standard" employed in epidemiological studies.²⁷⁸ These "criteria" include assessing the strength of the association (as expressed by odds ratio or relative risk or attributable risk), consistency (across different study periods and/or populations), specificity, temporality, biological gradient (dose response relationship), plausibility, coherence, experimental evidence and analogy. At present, none of the studies published provide any data that can help us in investigating the association between certain dental procedures, such as tooth extraction, and the development of PTP.

Summary and conclusion

The EBPG conducted a systematic literature review to investigate the association between tooth manipulation, injury or extraction and the subsequent development of phantom tooth pain. At present, there is no published study that can support such a causal association.

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Appendix 1

WorkSafeBC Evidence-Based Practice Group levels of evidence ^{adapted from 1,2,3,4}

1	Evidence from at least 1 properly randomized controlled trial (RCT) or systematic review of RCTs.
2	Evidence from well-designed controlled trials without randomization or systematic reviews of observational studies.
3	Evidence from well-designed cohort or case-control analytic studies, preferably from more than 1 centre or research group.
4	Evidence from comparisons between times or places with or without the intervention. Dramatic results in uncontrolled experiments could also be included here.
5	Opinions of respected authorities, based on clinical experience, descriptive studies or reports of expert committees.

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