



Systematic review of oral ulceration with bone sequestration



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ARTICLE INFO

Article history:

Paper received 27 August 2015

Accepted 25 November 2015

Available online 22 December 2015

Keywords:

Oral ulceration
Bone sequestration
Bone exposure
OUBS
Idiopathic
Osteonecrosis

ABSTRACT

Background: This article represents the first systematic review entirely dedicated toward a disease called oral ulceration with bone sequestration (OUBS). We performed this review in order to further define and outline this disease. A secondary interest was to recognize the prevalence and importance of OUBS in relation to other oral disorders accompanied by ulceration and bone exposure.

Material and methods: The systematic review was registered with PROSPERO (registration number CRD42015024294) and performed in cooperation with Harvard's Countway Library. Searches were built using MeSH terms and proximity operators previously mentioned in OUBS descriptions. Database searches were performed through EMBASE, Medline, and PubMed, followed by a handsearch of bibliographies for relevant articles. Articles were assessed against eligibility and inclusion criteria centering on bone exposure without known etiologic cause. We sought to gather information on patient age, sex, anatomical location, clinical presentation, and comorbidities. PRISMA guidelines were followed.

Results: The searches identified 766 records total. Despite considerable inspection, we found only 8 articles qualifying for our review. In the 8 articles, there were a total of 24 patients fulfilling the criteria of OUBS. Although some abstracts mentioned idiopathic nature, most authors presented clinical cases with probable causes to ulceration and sequestration. The mean age of these patients was 43.21 ± 11.94 years. The male to female ratio was 3:1. The predominant area of occurrence was the mandible ($n = 23$, 95.8%).

Conclusion: The representation of OUBS in the literature remains scarce. More data must be generated and gathered on the concept of OUBS so as to determine the true incidence and importance of this disease. Despite rare occurrences of conditions characterizing OUBS, the recent discussion of this topic in the scientific community calls for more knowledge to be brought forth, with great benefit to patients suffering from ulcerative diseases and osteonecrosis.

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1. Introduction

Oral ulceration with bone sequestration (OUBS) has been used variably by authors and practitioners to characterize oral ulceration and bone sequestration without an etiologic cause (Khan et al., 2015). The most common locations reported for ulceration and subsequent bone exposure occur at the mylohyoid ridge, mandibular tori, palatal tori, and mandibular exostosis (Farah and Savage, 2003). No precise definition for OUBS has been provided or

accepted, and there has been recent debate in the literature pertaining to the disorder's conceptualization (Khan et al., 2015; Otto et al., 2015a). It is a rare condition which contains cases previously recognized by the term 'lingual mandibular sequestration and ulceration' (Peters et al., 1993). Almazrooa and Woo (2009) define OUBS as, "spontaneous sequestration of the lingual mandibular bone, usually in the area of the mylohyoid ridge, in patients with no significant underlying systemic condition." Previously, to the best of our knowledge, only one review has been attempted on OUBS (Khan et al., 2015).

The objective of this current systematic review was to distinguish between various ulcerative disorders of the oral cavity and that of OUBS. Although oral ulceration is common, it is normally self-limiting and the progression to bone exposure and

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sequestration is extremely rare (Farah and Savage, 2003). Medical practitioners still must be aware of the local, systemic, and anatomical predispositions which lead to increasingly destructive ulcerative disorders. Our secondary interest concerned assessing the prevalence and importance of OUBS in relation to other oral maladies associated with bone exposure. The determination of a general prevalence value will be significant since OUBS has recently been stressed as a potential differential diagnosis pertaining to ONJ (Khan et al., 2015). We sought to collect articles and case reports mentioning idiopathic bone exposure or sequestration in order to begin constructing a framework for characterizing this concept.

2. Materials and methods

This systematic review was performed according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher et al., 2009). Searches were conducted on 13th July 2015 after being registered with PROSPERO (registration #CRD42015024294). No date limits were applied for the beginning of the search in order to obtain all relevant articles. Research experts at Harvard's Countway Library were consulted for expertise in article identification. Databases were screened based on the MeSH and Emtree terms developed in conjunction with Harvard's Countway Library. The search was ended on 13th July 2015.

We searched Embase, MEDLINE, and PubMed for articles that mentioned key words for oral ulceration with exposed, necrotic,

idiopathic, unexplained, spontaneous, or sequestered bone. Controlled vocabulary terms (both MeSH and Emtree) were used when available and appropriate. Searches were linked by OR operators and intersections of concepts linked consecutively with AND statements. Proximity operators (NEAR/n) allowed for detection of words possibly combined in unpredictable ways. Appendix 1 contains search algorithms used. Each search contains field tags which tell interfaces which indexes to search. A final handsearch through bibliographies of relevant articles was conducted in order to accumulate as much information on OUBS as possible.

Inclusion criteria were met if: 1) the abstract made any discussion on oral ulceration and bone sequestration without providing an etiologic cause; 2) an 'idiopathic' or an 'unknown' etiology was stated in the abstract regardless of pathologic mechanisms discussed; 3) full article in English language was available. Records were excluded if one of the following criteria pertained: 1) language other than English; 2) pertaining diagnostic methods unrelated to OUBS; 3) pertaining to animals and unrelated to OUBS; 4) pertaining to therapy unrelated to OUBS; 5) pertaining to topics irrelevant to OUBS; 6) etiologic cause of oral ulceration or bone sequestration was given in abstract (i.e. antiresorptive drugs, osteomyelitis, osteoradionecrosis, trauma, neoplasia, etc). This systematic review followed the PRISMA guidelines (6).

3. Results

The searches returned a total of 766 records. After duplicates were removed, 726 records remained (Fig. 1). All 726 articles were

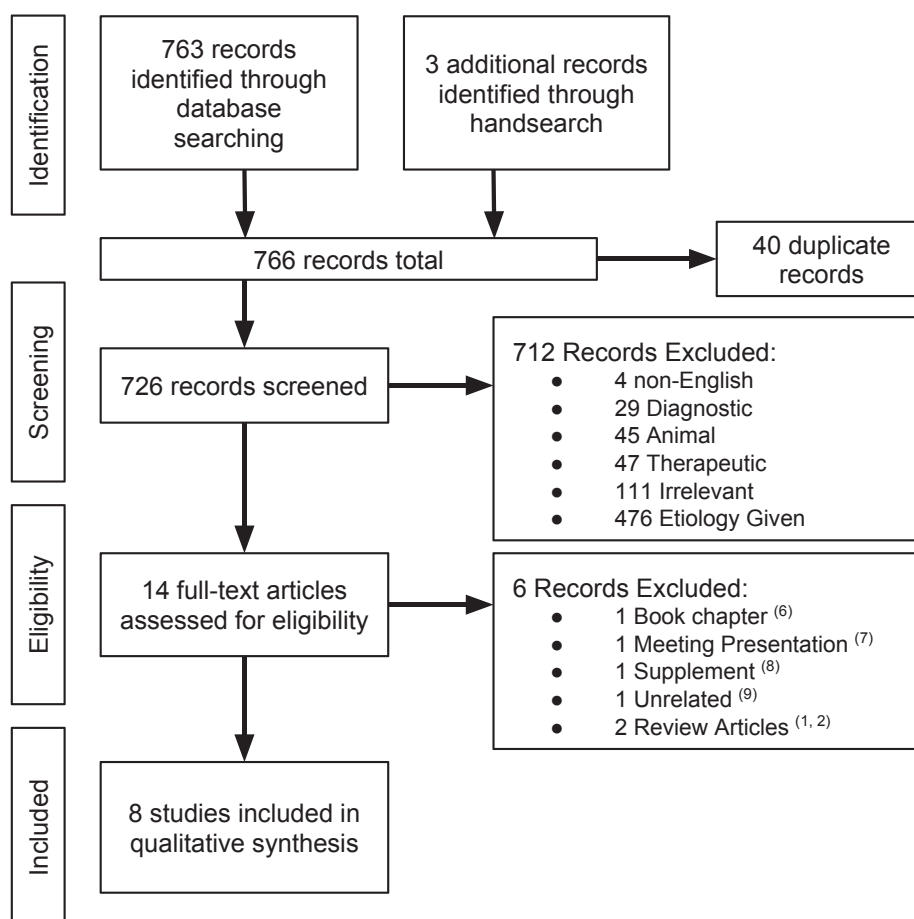


Fig. 1. Flow chart for record selection.

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