Journal of Cranio-Maxillo-Facial Surgery 46 (2018) 504–510

Contents lists available at ScienceDirect

Journal of Cranio-Maxillo-Facial Surgery

journal homepage: www.jcmfs.com

Review

Squamous odontogenic tumor and squamous odontogenic tumor-like proliferations in odontogenic cysts: An updated analysis of 170 cases reported in the literature

Bruno Ramos Chrcanovic [a, *], Ricardo Santiago Gomez [b]

[a] Department of Prosthodontics, (Head: Dr. Liselott Lindh, DDS, PhD), Faculty of Odontology, Malmö University, Malmö, Sweden
[b] Department of Oral Surgery and Pathology, (Head: Dr. Henrique Pretti), School of Dentistry, Universidade Federal de Minas Gerais, Belo Horizonte, Brazil

ARTICLE INFO

Article history:
Paper received 20 January 2017
Accepted 18 December 2017
Available online 26 December 2017

Keywords:
Squamous odontogenic tumor
Squamous odontogenic tumor-like proliferations in odontogenic cysts
Odontogenic tumors
Clinical features
Recurrence rate

ABSTRACT

Purpose: To integrate the available data published on squamous odontogenic tumors (SOT) and squamous odontogenic tumor-like proliferations in odontogenic cysts (SOT-LPOC) into a comprehensive analysis of their clinical/radiologic features.
Materials and methods: An electronic search was undertaken in January 2017. Eligibility criteria included publications having enough clinical/radiological/histological information to confirm a definite diagnosis.
Results: A total of 74 publications reporting 110 SOTs (102 central, 8 peripheral) and 60 SOT-LPOC were included. Compared to SOT-LPOC, SOT showed lower mean age, no preference regarding maxilla or mandible localization, significant association with cortical bone perforation, multilocular radiographic appearance, and mobility of the tooth/teeth associated with the lesion. While 5 recurrent SOT were reported after enucleation, no recurrent SOT-LPOC was found.
Conclusions: SOT shows a more aggressive biologic behavior than SOT-LPOC, which supports the hypothesis that the two lesions are distinct clinicopathological conditions.

1. Introduction

According to the World Health Organization (WHO, 2017), the squamous odontogenic tumor (SOT) is a locally infiltrative neoplasm consisting of islands of well-differentiated squamous epithelium in a fibrous stroma. The squamous odontogenic tumor-like proliferations in odontogenic cysts (SOT-LPOC) is an uncommon histologic finding consisting of multiple islands of squamous odontogenic epithelium present in the wall of odontogenic cyst, with aspects similar to those of the SOT, that appears as a solid lesion (Unal et al., 1987; Wright, 1979).

SOT and SOT-LPOC are considered to be rare lesions, and because of that, there are limited details in the literature regarding their clinical and radiologic features. The epidemiological study of such lesions is of great importance because provides information that can improve the diagnostic accuracy and will allow pathologists and surgeons to make informed decisions and to refine treatment plans to optimize clinical outcomes (Chrcanovic and Gomez, 2016, 2017a; b). The aim of the present study was to integrate the available data published in the literature on SOT and SOT-LPOC into an updated, comprehensive, comparative analysis of their clinical and radiologic features, and to report the frequency of recurrence of these lesions.

2. Materials and methods

This study followed the PRISMA Statement guidelines (Moher et al., 2009), an evidence-based minimum set of items for reporting in systematic reviews. PRISMA focuses on ways in which authors can ensure a transparent and complete reporting of this type of research. A review protocol does not exist.

2.1. Search strategies

An electronic search without time restrictions was undertaken in January 2017 in the following databases: PubMed/Medline, Web of Science, and Science Direct. The search for the terms in the database Science Direct was limited to "Title, Abstract, Keyword,"

* Corresponding author. Department of Prosthodontics, Faculty of Odontology, Malmö University, Carl Gustafs väg 34, SE-214 21, Malmö, Sweden. Fax: +46 40 6658503.
E-mail addresses: bruno.chrcanovic@mah.se, brunochrcanovic@hotmail.com (B.R. Chrcanovic), rsgomez@ufmg.br (R.S. Gomez).

https://doi.org/10.1016/j.jcms.2017.12.023
1010-5182/

due to a large initial amount of entries. The following terms were used in the search strategies:

("squamous odontogenic tumor") OR ("squamous odontogenic tumour") OR ("squamous odontogenic tumor-like proliferations in odontogenic cysts") OR ("squamous odontogenic tumor arising in odontogenic cysts") OR ("squamous odontogenic tumor-like proliferations")

Google Scholar was also checked. A manual search of related journals was performed, including *Acta Odontologica Scandinavica, Acta Oto-Laryngologica, Annals of Otology Rhinology and Laryngology, British Journal of Oral and Maxillofacial Surgery, Cancer, Head & Neck, Head and Neck Pathology, International Journal of Oral and Maxillofacial Surgery, Japanese Journal of Oral and Maxillofacial Surgery, Journal of Dental Research, Journal of Craniofacial Surgery, Journal of Cranio-Maxillofacial Surgery, Journal of Japanese Society of Oral Oncology, Journal of the Japanese Stomatological Society, Journal of Laryngology and Otology, Journal of Maxillofacial and Oral Surgery, Journal of Nihon University School of Dentistry, Journal of Oral and Maxillofacial Surgery, Journal of Oral Pathology and Medicine, Journal of the Stomatological Society, Laryngoscope, Oral Diseases, Oral Oncology, Oral Surgery Oral Medicine Oral Pathology Oral Radiology, Otolaryngology–Head and Neck Surgery,* and *Quintessence International*. The reference list of identified studies and the relevant reviews on the subject were also checked for possible additional studies. Publications with lesions identified by other authors as being SOT or SOT-LPOC, even not having the term "squamous odontogenic tumor" or "squamous odontogenic tumor-like proliferations in odontogenic cysts" in the title of the article, were also re-evaluated by an author (R.S.G.) of the present study.

2.2. Inclusion and exclusion criteria

Eligibility criteria included publications reporting cases of SOTs and/or SOT-LPOCs. The studies needed to contain enough clinical, radiological and histological information to confirm the diagnosis. The definitions and criteria of the World Health Organization Classification of Tumors—Head and Neck Tumors book (WHO, 2017), last updated in 2017, were used to diagnose the lesions as SOT or SOT-LPOC. The inclusion criteria for SOT diagnosis included the following histopathological features: presence of islands of differentiated squamous epithelium tightly packed together and showing a flattened peripheral layer, occasional presence of microcystic degenerations with individual cell keratinization and calcification, rare figures of mitosis.

Randomized and controlled clinical trials, cohort studies, case—control studies, cross-sectional studies, case series, and case reports were included. Exclusion criteria were immunohistochemical studies, histomorphometric studies, radiological studies, genetic expression studies, histopathological studies, cytological studies, cell proliferation/apoptosis studies, *in vitro* studies, and review papers, unless any of these publication categories had reported any cases with enough clinical, radiological and histological information. All cases associated with well-defined clinicopathological conditions, such as odontogenic keratocyst, were excluded.

2.3. Study selection

The titles and abstracts of all reports identified through the electronic searches were read independently by the authors. For studies appearing to meet the inclusion criteria, or for which there were insufficient data in the title and abstract to make a clear decision, the full report was obtained. Disagreements were solved by discussion between the authors. The clinical and radiological aspects, as well as the histological description of the lesions reported by the publications were thoroughly assessed by one of the authors (R.S.G.), an expert in oral pathology, in order to confirm the diagnosis of SOT and SOT-LPOC.

2.4. Data extraction

The review authors independently extracted data using specially designed data extraction forms. Any disagreements were resolved by discussion. For each of the identified studies included, the following data were then extracted on a standard form, when available: year of publication, number of patients, patient's sex, age and race, follow-up period, duration of the lesion previously to treatment, lesion location (maxilla/mandible), anterior/posterior location (three categories: [a] anterior: lesions in the incisors/canine region; [b] premolar region; [c] posterior: lesions in the molars/retromolar region), recurrence, recurrence period, lesion size, presence of erosion of the subjacent cortical bone (for peripheral lesions), perforation of cortical bone, locularity radiological appearance (unilocular/multilocular), tooth displacement/unerupted and/or tooth root resorption due to lesion's growth, expansion of osseous region adjacent to the tumor, presence of clinical symptoms, and treatment performed (curettage/excision, enucleation, partial resection, resection with continuity). The lesion size was determined according to the largest diameter reported in the publications. Contact with authors for possible missing data was performed.

2.5. Data analyses

The mean, standard deviation (SD), and percentages were presented as descriptive statistics. Kolmogorov—Smirnov test was performed to evaluate the normal distribution of the variables, and Levene test evaluated homoscedasticity. The performed tests for two independent groups were the Student t-test or Mann—Whitney test, depending on the normality. The Pearson chi-squared or Fisher exact tests were used for categorical variables, depending on the expected count of events in a 2x2 contingency table. The probability of recurrence was calculated for four variables, in odds ratios (95% confidence intervals). The variables were the age of the patients ($\leq$30 years, >30 years), expansion of the osseous region adjacent to the tumor, perforation of cortical bone, and lesion location (maxilla/mandible). The degree of statistical significance was considered $p < 0.05$. All data were statistically analyzed using the Statistical Package for the Social Sciences (SPSS) version 23 software (SPSS Inc., Chicago, IL, USA).

3. Results

3.1. Literature search

The study selection process is summarized in Fig. 1. The search strategy in the databases resulted in 333 papers. The search in Google Scholar resulted in 14 eligible papers not found in the three main databases. A total of 86 articles were cited in more than one database (i.e., were duplicates). The reviewers independently screened the abstracts for articles related to the focus question. Of the resulting 261 studies, 175 were excluded for not being related to the topic. Additional hand-searching of journals and of the reference lists of selected studies yielded 9 additional papers. The full-text reports of the remaining 95 articles led to the exclusion of 21 because they did not meet the inclusion criteria (see Supplemental Appendix). The excluded studies did not have enough clinical, radiological and histological information to confirm the diagnosis of SOT and SOT-LPOC, or were cases of so-called odontogenic keratocysts (Beovide and Kornecki, 1994; Cotten et al., 1982; Hodgkinson et al., 1978) and glandular odontogenic cysts (Patron et al., 1991) with SOT-like proliferation in the capsule. One