Review

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

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A B S T R A C T

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.

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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,”
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”)


3. Results

3.1. Literature search

The study selection process was 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. 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, local density appearance (unilocular/multilocular), tooth displacement/unerupted 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.

2.4. 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, local density appearance (unilocular/multilocular), tooth displacement/unerupted 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 (<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).

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 immunohistochecmistry 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.

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malignant variant of SOT was reported (Ide et al., 1999), but it was also not included in the analysis. Thus, a total of 74 publications (see Supplemental Appendix) were included in the review.

3.2. Description of the studies and analyses

3.2.1. Squamous odontogenic tumor (SOT)

Some studies reporting series of odontogenic studies and including SOTs were found, but their cases were not included here due to lack of enough clinical, radiological and histological information to confirm the diagnosis of SOTs. These include, for example, Simon et al. (2002) with 2 cases, Ladeinde et al. (2005) with 6 cases, Ortega et al. (2007) with 1 case, Avelar et al. (2008) with 1 case, and Nalabolu et al. (2017) with 2 cases.

Table 1 presents demographic and clinical features of all 110 SOTs, 102 central and 8 peripheral lesions. The lesion was more prevalent in men than in women, at a 1.2:1 proportion. Some cases represented multiple lesions in the same individual (Baden et al., 1993; Elmuradi et al., 2016; Hopper et al., 1980; McNeill et al., 1980; Mills et al., 1986; Norris et al., 1984; Pullon et al., 1975), and there was one report describing multicentric disease in three siblings (Leider et al., 1989). The mean age of the patients was 34.8 ± 14.5 and 45.4 ± 23.0 for central and peripheral lesions, respectively. Fig. 2 shows the distribution of the lesions according to age, with a high prevalence in the third and fifth decades of life. The lesions were equally distributed between maxillae and mandibles and between incisors/canine and molar regions, but occurred less frequently in the premolar area (Fig. 3). About 62% of the central lesions showed signs of cortical bone perforation and 90% had a radiological unilocular appearance. Approximately 60% of the central lesions were associated with a tooth displacement/unerupted due to the lesion’s growth. Nearly 5% of the central SOTs presented root resorption of adjacent teeth.

Treatment of the lesions was known in 104 cases (96 central, 8 peripheral lesions), of which 90 consisted of conservative surgery (excisions or enucleations) and 13 cases were treated by marginal or segmental resection. Time of follow-up was given for 66 lesions, with a mean ± SD of 34.7 ± 41.6 months (n = 61) and 44.8 ± 66.1 months (n = 5) for central and peripheral lesions, respectively. There was information about recurrence for 68 lesions (63 central, 5 peripheral), of which 5 recurred (4 central, 6.3%; 1 peripheral, 20%). The interval from initial treatment to the first recurrence ranged from 2 to 20 months for central lesions, with a mean interval of 11.3 ± 7.4 months. The only recurrence in peripheral lesions occurred after 156 months. The race of the patient was reported in 95 cases. Forty-five cases (all central lesions) were diagnosed in blacks, 20 (1 peripheral) in whites, 13 (3 peripheral) in Asians, 14 (3 peripheral) in Hispanics, 1 each in a Turk, a Hispanic, and other, all central lesions.

Table 2 shows the recurrence rate for central SOTs according to some variables. Recurrences occurred only when the lesions were treated by enucleation, but this was the most commonly treatment performed. The age of the patient, location of the lesion (maxilla/mandible), expansion of the osseous region adjacent to the tumor, perforation of cortical bone, and locularity appearance in radiological exams seem to not influence the recurrence rate.

![Fig. 1. Study screening process.](image_url)
Table 1
Demographic and clinical features of squamous odontogenic tumor (SOT) and squamous odontogenic tumor-like proliferations in odontogenic cysts (SOT-LPOC) described in the literature.

<table>
<thead>
<tr>
<th>Variables</th>
<th>Central SOT</th>
<th>SOT-LPOC</th>
<th>p value (^a)</th>
<th>Peripheral SOT</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sample size (n)</td>
<td>102</td>
<td>60</td>
<td>8</td>
<td></td>
</tr>
<tr>
<td>Age (year), mean ± SD (min–max)</td>
<td>34.8 ± 14.5 (8–67)</td>
<td>44.4 ± 7.8 (17–80)</td>
<td>&lt;0.001 (^b)</td>
<td>45.4 ± 23.0 (11–74)</td>
</tr>
<tr>
<td>Men</td>
<td>35.6 ± 14.8 (8–65)</td>
<td>46.6 ± 12.6 (19–60)</td>
<td>0.025 (^b)</td>
<td>52.0 ± 31.1 (30–74)</td>
</tr>
<tr>
<td>Women</td>
<td>33.1 ± 14.0 (10–67)</td>
<td>39.6 ± 15.9 (17–65)</td>
<td>0.265 (^b)</td>
<td>43.2 ± 22.9 (11–61)</td>
</tr>
<tr>
<td>p value (^d)</td>
<td>0.227(^b)</td>
<td>0.269(^b)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Gender, n (%)</td>
<td>57 (56.4)</td>
<td>9 (50)</td>
<td>0.613 (^b)</td>
<td>2 (25)</td>
</tr>
<tr>
<td>Jaw, n (%)</td>
<td>50 (50)</td>
<td>41 (69.5)</td>
<td>0.016 (^b)</td>
<td>4 (50)</td>
</tr>
<tr>
<td>Bone expansion, n (%)</td>
<td>18 (19.1)</td>
<td>3 (17.6)</td>
<td>0.594 (^b)</td>
<td>1 (14.3)</td>
</tr>
<tr>
<td>Symptomatic, n (%)</td>
<td>76 (80.9)</td>
<td>14 (82.4)</td>
<td></td>
<td>6 (85.7)</td>
</tr>
<tr>
<td>Tooth mobility, n (%)</td>
<td>26 (34.2)</td>
<td>1 (7.7)</td>
<td>0.047 (^b)</td>
<td>0 (0)</td>
</tr>
<tr>
<td>Cortical bone perforation, n (%)</td>
<td>53 (62.4)</td>
<td>0 (0)</td>
<td>&lt;0.001 (^b)</td>
<td>-</td>
</tr>
<tr>
<td>Bone erosion, n (%) (^g)</td>
<td>-</td>
<td>-</td>
<td></td>
<td>2 (50)</td>
</tr>
<tr>
<td>Locularity, n (%)</td>
<td>84 (90.3)</td>
<td>57 (100)</td>
<td>0.012 (^b)</td>
<td>5 (100)</td>
</tr>
<tr>
<td>Unilocular</td>
<td>9 (9.7)</td>
<td>0 (0)</td>
<td></td>
<td>0 (0)</td>
</tr>
<tr>
<td>Multilocular</td>
<td>9</td>
<td>3</td>
<td>3</td>
<td></td>
</tr>
<tr>
<td>Tooth displacement/unerupted, n (%)</td>
<td>54 (62.1)</td>
<td>6 (40)</td>
<td>0.109 (^b)</td>
<td>1 (14.3)</td>
</tr>
<tr>
<td>Tooth root resorption, n (%)</td>
<td>33 (37.9)</td>
<td>9 (60)</td>
<td>63.7</td>
<td></td>
</tr>
<tr>
<td>Treatment, n (%)</td>
<td>15</td>
<td>45</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>None</td>
<td>1 (1)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td></td>
</tr>
<tr>
<td>Excision/curettage</td>
<td>2 (2.1)</td>
<td>0 (0)</td>
<td>5 (62.5)</td>
<td></td>
</tr>
<tr>
<td>Enucleation</td>
<td>80 (83.3)</td>
<td>58 (100)</td>
<td>3 (37.5)</td>
<td></td>
</tr>
<tr>
<td>Marginal resection</td>
<td>9 (9.4)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td></td>
</tr>
<tr>
<td>Segmental resection(^h)</td>
<td>4 (4.2)</td>
<td>0 (0)</td>
<td>0 (0)</td>
<td></td>
</tr>
<tr>
<td>Recurrence, n (%)</td>
<td>14</td>
<td>50</td>
<td>1</td>
<td></td>
</tr>
<tr>
<td>Yes</td>
<td>4 (6.3)</td>
<td>0 (0)</td>
<td>0.464 (^b)</td>
<td>1 (20)</td>
</tr>
<tr>
<td>No</td>
<td>59 (93.7)</td>
<td>13 (100)</td>
<td>4 (80)</td>
<td></td>
</tr>
<tr>
<td>Unknown</td>
<td>39</td>
<td>47</td>
<td>3</td>
<td></td>
</tr>
<tr>
<td>Follow-up time (months), mean ± SD (min–max)</td>
<td>34.7 ± 41.6 (2–216; n = 61)</td>
<td>31.8 ± 24.9 (4–96; n = 13)</td>
<td>0.582 (^b)</td>
<td>44.8 ± 66.1 (5–162; n = 5)</td>
</tr>
<tr>
<td>Lesion size (cm), mean ± SD (min–max)</td>
<td>2.2 ± 1.4 (0.5–7.0; n = 85)</td>
<td>1.8 ± 1.4 (0.5–5.0; n = 10)</td>
<td>0.273 (^b)</td>
<td>1.9 ± 1.5 (0.5–5.0; n = 7)</td>
</tr>
</tbody>
</table>

SD — standard deviation.

\(^a\) Comparison of the parameters between central SOT and SOT-LPOC lesions.
\(^b\) The mean age (min–max) of the 42 cases of Parmar et al. (2011) was also included here, but not separately for “men” and “women”, as the information of sex distribution was missing in this study.
\(^c\) Mann–Whitney test.
\(^d\) Comparison of the mean values between men and women.
\(^e\) Pearson's chi-squared test.
\(^f\) Fisher's exact test.
\(^g\) Applied to peripheral lesions only.
\(^h\) Resection with continuity defect.
3.2.2. Squamous odontogenic tumor-like proliferations in odontogenic cysts (SOT-LPOC)

Although some authors have used the term odontogenic keratocyst with SOT-like proliferation in the capsule (Beovide and Kornecki, 1994; Cotten et al., 1982; Hodgkinson et al., 1978), we have not included in the study because these features are within the histopathological spectrum expected for this lesion. Although the odontogenic keratocysts may form epithelial buds, together with the formation of daughter cysts and solid epithelial islands, which may mimic SOT-like proliferation, both conditions are distinct clinicopathologic conditions.

Table 1 presents demographic and clinical features of all 60 SOT-LPOCs. It is important to stress here that the data from Parmar et al. (2011) were incorporated into the analysis only when possible, because the study has not provided information about the variables separately by 42 lesions described. The lesion was equally prevalent in men and women. The mean age of the patients was 44.4 ± 7.8 years. A graphic with age distribution of the patients was not performed for the SOT-LPOCs, due to missing separated information of the 42 lesions described by Parmar et al. (2011). The ages of the remaining 18 patients from the other publications were 17, 19, 21, 31, 31, 42, 45, 45, 47, 49, 53, 54, 55, 57, 60, and 65. The lesions were more prevalent in the maxilla in comparison to the mandible, and at the anterior region in comparison to the posterior region. Of the 42 cases reported by Parmar et al (Parmar et al., 2011), 33 were located in the incisor-canine region, 4 in the premolar, and 4 in the molar region, but no distinction was made concerning the distribution of these precise locations between the maxilla and mandible. Of the other 18 cases of SOT-LPOCs described in the literature, 5 were located in the incisor-canine region, 5 in the premolar area, 6 in the molar region, one in the maxilla, and one in the “right body-angle” area.

All SOT-LPOC lesions showed no signs of cortical bone perforation or root resorption of adjacent teeth, and had a radiological unilocular appearance. Most SOT-LPOCs were associated with a radicular cyst (n = 51), 6 lesions were associated to a dentigerous cyst, one lesion each was associated to a lateral periodontal cyst and a residual cyst, and for one lesion the information was not available and it was not possible to determine what kind of odontogenic cyst the lesion was associated with.

Treatment of the lesions was known in 58 cases, all by enucleation. Time of follow-up was given for 13 lesions, with a mean ± SD of 31.8 ± 24.9 months. There were no recurrences, but this information was available for only 13 lesions. The race of the patient was reported in 57 cases. A total of 43 cases (75.4%) were diagnosed in whites, 9 in blacks, 3 in Asians, 1 in an Indian, and 1 in a Turkish patient.

3.2.3. Central SOT vs. SOT-LPOC

Five factors were statistically significantly different between central SOTs and SOT-LPOCs: mean age of the patients (older patients in SOT-LPOCs), lesion location (SOT-LPOCs more frequently observed in maxillae), mobility of tooth/teeth associated with the lesion (more frequently observed with central SOTs), cortical bone perforation (only observed in central SOTs), and locularity appearance in radiological examinations (all SOT-LPOCs presented a unilocular appearance).

4. Discussion

The present review of the literature revealed that the SOTs and SOT-LPOCs are rare lesions. Peripheral SOTs are even rarer than the central counterparts. Peripheral SOTs and SOT-LPOC occurred in older patients than did central SOTs. The lesions were not commonly associated with patients presenting clinical symptoms,
and there was a fairly equal prevalence distribution between sexes. Radiologically, multilocular lesions were rarely seen. When central SOTs were compared to SOT-LPOCs, they statistically significantly differed in lesion location, prevalence of cortical bone perforation and multilocularity, and in the patients’ mean age.

Even though it was not the aim of the present study to compare the different lesions histologically, it is important to emphasize here that the SOT-LPOC possesses certain histopathological features that overlap with SOT, and it might be difficult to distinguish the lesions microscopically. Thus, it is decisive that clinicopathologic correlation be part of the clinician’s treatment protocol (Parmar et al., 2011). A residual cyst with SOT-LPOC has been misinterpreted as a squamous cell carcinoma arising in a residual epithelium of radicular cysts and attribute it to a response to inflammation. Olivera et al. (1995) also ruled out the participation of inflammation as a proliferative stimulus. These same authors later suggested that SOT-LPOCs could be an early expression of neoplastic change but not a carcinoma or any other tumor (Oliveira et al., 2006; Swinson et al., 2005).

The nature of SOT-LPOCs is not well known. Philipsen and Reichart (1996) believe that the SOT-LPOCs are a result of a reactive, inflammatory hyperplasia of the epithelial cyst lining. Odell and Morgan (1998) favor a budding type of hyperplasia of the lining epithelium of radicular cysts and attribute it to a response to subsiding inflammation because it usually occurs in areas without inflammation. Olivera et al. (1995) also ruled out the participation of inflammation as a proliferative stimulus. These same authors later suggested that SOT-LPOCs could be an early expression of neoplastic change but not a carcinoma or any other tumor (Oliveira et al., 2006). The study of Parmar et al. (2011) corroborated in part some of these hypotheses, as two-thirds of the 42 cases described in their study demonstrated budding of the epithelial cyst lining, which was histologically identical to the squamous epithelial islands in the cyst wall. This implies indirect origin from the rests of Malassez because they are thought to be the source of the cyst lining in radicular cysts (Parmar et al., 2011). However, SOT-LPOCs are not limited to inflammatory cysts and they were also reported in developmental cysts, i.e., dentigerous cysts and glandular odontogenic cysts.

SOT-LPOCs show no evidence of neoplastic transformation to a solid SOT, and their clinical behavior is no more aggressive than the cysts in which they occur (Simon and Jensen, 1985; Wright, 1979). Because of this, it has been suggested that it represents a reactive or regressive state of the residual odontogenic epithelium (Stoelinga et al., 1975; Toller, 1967) or a hamartoid lesion (Unal et al., 1987). Some results of the present review might indicate that SOTs could have a more aggressive behavior than SOT-LPOCs. These include the higher prevalence among SOTs in comparison to SOT-LPOCs of cortical bone expansion and perforation, tooth root resorption, mobility of teeth involved in the lesion, and displacement or blockage of eruption of teeth due to growth of the lesion. Considering all the clinical discrepancies found in our study, it is not plausible to believe in the natural progression of SOT-LPOC to SOT.

Enucleation was the treatment modality most often described in the literature, resulting in recurrence in 5 cases, i.e., 4 central lesions and 1 peripheral lesion. This represents 6.3% and 20% of the central and peripheral SOTs cases, respectively, with a reported follow-up and for which the information about recurrence was available. These figures cannot be ignored. Therefore, the surgical-pathological team must be attentive to the clinicopathologic characteristics of each individual lesion. Some cases of large lesions presenting or not clinical aggressiveness (radiological multilocularity, great bone expansion and cortical bone perforation) were treated by marginal or segmental resection, none of them with recurrences. Although we do not suggest that an aggressive approach is justified for SOT, surgeons must consider all of these data in the surgical treatment planning of the patient.

The results of the present study have to be interpreted with caution because of several study limitations. First, all included studies were retrospective reports, which inherently results in flaws, as demonstrated by gaps in information and incomplete records. Second, many of the cases have a short follow-up, which could have led to an underestimation of the actual recurrence rate, because a longer follow-up period can lead to an increase in the recurrence rate. However, it is difficult to define what it would be considered a short follow-up period to evaluate the recurrence of SOT and SOT-LPOC. Third, the great majority of the cases described were published as isolated case reports or small case series, the great majority of them analyzed by different pathologists.

5. Conclusions

SOT shows a more aggressive biologic behavior than SOT-LPOC, which supports the hypothesis that the two lesions are distinct clinicopathological conditions.
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Conflicts of interest

There are no conflicts of interest to declare.

Appendix A. Supplementary data

Supplementary data related to this article can be found at https://doi.org/10.1016/j.jcma.2017.12.023.