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REVIEW ARTICLE

WILEY ORAL DISEASES

Odontogenic myxoma: An updated analysis of 1,692 cases reported in the literature

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The aim of the present study was to integrate the available data published on odontogenic myxoma (OM) into a comprehensive analysis of its clinical/radiological features. Electronic search undertaken in January/2018, looking for publications reporting cases of OM. A total of 377 publications were included. We identified 1,692 lesions, and 695 were used for the analysis of recurrence. There is a predominance of OMs in females and in mandibles. OMs usually present with bone expansion, asymptomatic cortical perforation, and a multilocular appearance. Lesion location (maxilla/mandible), bone expansion, cortical bone perforation, locular radiological appearance, tooth resorption, odontogenic epithelial rests, or angular septa are not associated with recurrence. While curettage (31.3%) showed the highest recurrence rate, marginal resection (1.3%) and segmental resection (3.1%) showed the lowest values. Enucleation + peripheral osteotomy (6.7%) showed better results than enucleation (13.1%) or enucleation + curettage (12.7%). In comparison with unilocular lesions, multilocular ones were significantly more prevalent in mandibles, more often presented expansion and cortical bone perforation, had larger mean size, and were more often treated by segmental resection. Conservative surgical procedures are associated with higher probability of recurrence of OM. Taking into consideration the recurrence rate and morbidity associated with different surgical treatments, tumor enucleation followed by peripheral osteotomy should be considered as the first therapeutic choice.

KEYWORDS
clinical features, fibromyxoma, myxofibroma, odontogenic myxoma, odontogenic tumors, recurrence rate

1 | INTRODUCTION

Odontogenic myxoma (OM) is a benign odontogenic neoplasm characterized by stellate and spindle-shaped cells dispersed in an abundant myxoid extracellular matrix, which may contain odontogenic epithelium. Its histogenesis is most likely related to the odontogenic ectomesenchyme of a developing tooth. The neoplasm often behaves in a locally aggressive and infiltrating fashion. It has been listed as the third most frequent odontogenic tumor after odontoma and ameloblastoma (WHO, 2017). The aim of this study was to integrate the available data published in the literature on OM into an updated comprehensive comparative analysis of their clinical and radiological features, as well as the frequency of recurrence.

2 | MATERIALS AND METHODS

This study followed the PRISMA Statement guidelines (Moher, Liberati, Tetzlaff, Altman, & Grp, 2009).

2.1 | Search strategies

An electronic search without time restrictions was undertaken in December 2017 in the following databases: PubMed/Medline, Web of Science, Science Direct, J-Stage, and Lilacs. The following terms were used in the search strategies:

((((myxoma) OR myxofibroma OR fibromyxoma) OR "odontogenic myxoma")) AND ((((maxilla) OR mandible) OR jaw) OR maxillofacial)

Google Scholar was also checked. A manual search of related journals, 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*, was performed. The reference list of the 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 OM, even not having the term "odontogenic myxoma" in the title of the article, were also re-evaluated by an author of this study.

2.2 | Inclusion and exclusion criteria

Eligibility criteria included publication reporting cases of OM. Cases of OM described in epidemiological studies on odontogenic tumors were listed to analyze age distribution and its prevalence between different genders.

Only the studies containing enough clinical, radiological, and histological information to confirm the diagnosis were included in the analyses of recurrence. For these cases, 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. Hybrid tumors containing parts of OM were not considered for this study, as they may behave differently from non-hybrid OM. Only central lesions were considered. Studies presenting the possibility of duplicated cases, usually originated from clinical series from the same service or University or other publications, were excluded.

The definitions and criteria of the World Health Classification of Tumors—Head and Neck Tumors book (WHO, 2017), last updated in 2017, were used to diagnose a lesion as OM.

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 resolved 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.) of this study, an expert in oral pathology, to confirm the diagnosis of OM.

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, perforation of cortical bone, locularity appearance in radiological examinations (unilocular/multilocular), the presence or absence (in radiological appearance) of fine, angular, or straight trabeculations (here called as "*angular septa*"), association of the lesion with a tooth (the tooth can either be erupted with the entire root(s) encompassed by the lesion or unerupted encompassing the entire tooth), tooth displacement and/or tooth root resorption due to lesion's growth, expansion of osseous region adjacent to the tumor, the presence of clinical symptoms, the presence of odontogenic epithelial rests, and treatment performed (curettage, excision, enucleation, partial/marginal resection, resection with continuity, debulking, performance of additional curettage, and/or peripheral osteotomy). 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 | 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's test evaluated homoscedasticity. The performed tests for two independent groups were Student's *t*-test or Mann–Whitney test, depending on the normality. Pearson's chi-squared or Fisher's exact tests were used for categorical variables, depending on the expected count of events in a 2 × 2 contingency table. The probability of recurrence was calculated for some variables, whenever possible, in odds ratio (95% confidence interval). 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 25 software (SPSS Inc., Chicago, IL, USA).

3 | RESULTS

3.1 | Literature search

The study selection process is summarized in Figure S1 (Appendix S1). The search strategy in the databases resulted in 4,793 papers. Search in Google Scholar resulted in 19 eligible papers not found in the five main databases. A number of 1,547 articles were cited in more than one database (duplicates). The reviewers independently screened the abstracts for those articles related to the aim of the review. Of the resulted 3,265 studies, 2,801 were excluded for not being related to the topic or not presenting clinical cases. Additional hand searching of journals and of the reference lists of selected studies did not yield additional papers. The full-text reports of the remaining 464 articles led to the exclusion of 71 because they did not meet the inclusion criteria. Moreover, 16 studies were excluded because their cases were from the same service or University of the cases from other publications, presenting the possibility of duplicated cases. Thus, a total of 377 publications were included in the review (Appendix S1).

3.2 | Description of the studies and analyses

A total of 377 publications reporting OMs were included in the present review, reporting 1,692 odontogenic myxomas. Table 1 presents demographic and clinical features of all cases, of which 695 lesions were used for more a detailed characterization. The lesion was more prevalent in women than in men, at a 1.36:1 proportion. The mean age of the patients was 28.6 years (range 0–80), being more elevated in women. Figure 1 shows the distribution of the lesions according to age, with the highest prevalence in the third and then second and fourth decades of life. The lesions were more prevalent in the mandible in comparison with the maxilla, and at the posterior region in comparison with the anterior region (Figure 2). For those cases with available information, 86 of 1,261 lesions (6.8%) crossed the midline of the jaws, and the maxillary sinus was significantly affected in 163 of 344 lesions (47.4%) located in the maxilla. Eighteen lesions reached the coronoid process and 20 the condylar process of the mandible. The race of the patient was reported in 425 cases, of which 171 cases (40.2%) were diagnosed in Asians, 90 in Whites, 71 in Blacks, 60 in Indians, 11 in Arabs, 10 in Hispanics, seven in Turks, four in Persians, and one in other. Odontogenic epithelial rests were seen in 13.0% of the cases.

About 75% of the lesions showed signs of cortical bone perforation, 62.9% of the lesions had a radiological multilocular appearance, and 34.7% of the OMs showed the presence of angular septa. Nearly 20% of the lesions presented root resorption of adjacent teeth, and 53.8% of the cases showed tooth displacement and/or uneruption due to lesion's growth. The lesions had a mean size of 4.7 ± 2.8 cm (min–max, 0.5–25.0; *n* = 322). The correlation between lesion size

TABLE 1 Demographic and clinical features of central odontogenic myxomas described in the literature

Variables	
n	1,692
Age (years), mean ± *SD* (min–max)	28.6 ± 14.2 (0–80; *n* = 1,655[a])
Men	26.6 ± 16.3 (0–80; *n* = 299)
Women	28.2 ± 15.0 (1–79; *n* = 383)
p value[b]	0.013
Gender, *n* (%)[a]	
Men	705 (42.4)
Women	957 (57.6)
Unknown	30
Jaw, *n* (%)[a]	
Maxilla	726 (43.6)
Mandible	939 (56.4)
Unknown	27
Bone expansion, *n* (%)	
Yes	502 (93.5)
No	35 (6.5)
Unknown	158
Symptomatic, *n* (%)	
Yes	92 (17.2)
No	444 (82.8)
Unknown	159
Cortical bone perforation, *n* (%)	
Yes	283 (75.1)
No	94 (24.9)
Unknown	318
Locularity, *n* (%)	
Unilocular	187 (37.1)
Multilocular	317 (62.9)
Unknown	191
Angular septa, *n* (%)	
Yes	181 (34.7)
No	341 (65.3)
Unknown	173
Tooth displacement/unerupted, *n* (%)	
Yes	253 (53.8)
No	217 (46.2)
Unknown	225
Tooth root resorption, *n* (%)	
Yes	82 (19.2)
No	344 (80.8)
Unknown	269

(Continues)

TABLE 1 (Continued)

Variables	
Odontogenic epithelial rests, *n* (%)	
Yes	65 (13)
No	435 (87)
Unknown	195
Treatment, *n* (%)	
None	3 (0.5)
Curettage	47 (8.6)
Enucleation	194 (35.3)
Marginal resection	103 (18.8)
Segmental resection[c]	199 (36.2)
Debulking	2 (0.4)
Radiotherapy[d]	1 (0.2)
Unknown	146
Recurrence, *n* (%)	
Yes	44 (9.3)
No	431 (90.7)
Unknown	
Follow-up time (months), mean ± *SD* (min–max)	49.1 ± 66.2 (0–420; *n* = 426)
Lesion size (cm), mean ± *SD* (min–max)	4.7 ± 2.8 (0.5–25.0; *n* = 322)

SD, standard deviation.
[a]It also considers data from epidemiological studies of odontogenic tumors.
[b]Comparison of the mean age between men and women (Mann–Whitney test).
[c]Resection with continuity defect.
[d]Radiotherapy was applied as the main therapy in one case, but also as an adjunctive therapy in five cases (one curettage, two enucleations, and two segmental resections). Chemotherapy was applied as an adjunctive therapy in one case (one enucleation).

and age was very weak and not statistically significant ($y = 5.045 - 0.013x$, $R = 0.076$, $R^2 = 0.006$, $p = 0.174$).

Time of follow-up was informed for 426 lesions, with a mean ± *SD* of 49.1 ± 66.2 months (min–max, 0–420). One hundred and thirty-two lesions (31.0%) were followed up to 12 months and 53.8% for up to 24 months. Treatment of the lesions was known in 549 (of 695, 79%) cases, of which 44.3% of the cases consisted of conservative surgery (35.3% enucleations, 8.6% curettage, and 0.4% debulking), 55% of the cases were treated by resection (36.2% resection with continuity and 18.8% partial/marginal resection), and 0.7% by radiotherapy or no treatment. Additional curettage was used in 98 cases, mostly after enucleation (*n* = 88), but also after marginal resection (*n* = 5), segmental resection (*n* = 3), and debulking (*n* = 2). Additional curettage was followed by peripheral osteotomy in three of these cases, all treated by enucleation. Additional peripheral osteotomy was used alone (without previous additional curettage) after enucleation (*n* = 15), marginal resection (*n* = 1), segmental resection (*n* = 2), and when one case was treated only by curettage (without a previous enucleation).

There were 44 recurrences (9.3%) in 475 lesions. There was no information about recurrence for 220 lesions. The interval from

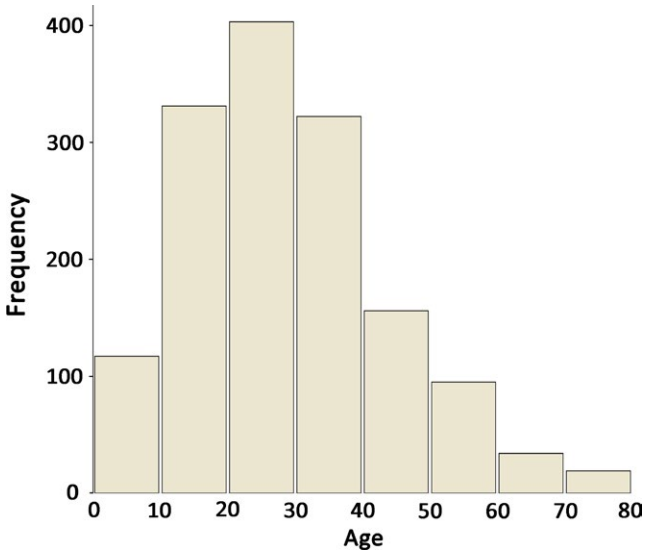

FIGURE 1 Distribution of odontogenic myxomas according to age (for the cases which the patients' age were informed, *n* = 1,478)

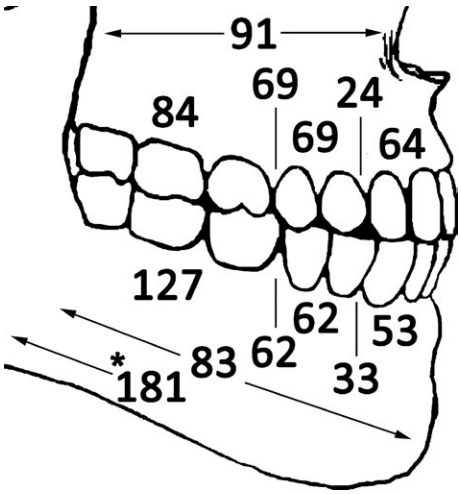

FIGURE 2 Topographical distribution of the known precise locations (*n* = 1,002) of odontogenic myxomas. Cases involving multiple regions (or an entire quadrant) are indicated between arrows. Numbers at the top and bottom of the lines indicate cases involving both adjoining regions: anterior/premolar and premolar/molar. The asterisk (*) indicates the number of lesions from the mandibular body or anterior region that reached the angle and/or ramus. For the rest of the lesions (*n* = 690), the location was the "maxilla" (*n* = 124), "mandible" (*n* = 121), "anterior maxilla" (*n* = 35), "posterior maxilla" (*n* = 88), "anterior mandible" (*n* = 48), "posterior mandible" (*n* = 124), "palate" (*n* = 9), "maxillary sinus" (*n* = 51), "whole mandible" (*n* = 10), "nasolabial groove" (*n* = 12), "mandibular angle" (*n* = 20), "mandibular ramus" (*n* = 9), "mandibular body" (*n* = 4), "mandibular condyle" (alone) (*n* = 4), "mandibular ramus-condyle" (*n* = 2), "mandibular angle-ramus" (*n* = 2), and unknown (*n* = 27). For those cases with available information, 86 of 1,261 lesions (6.8%) crossed the midline of the jaws, and the maxillary sinus was significantly affected in 163 of 344 lesions (47.4%) in the maxilla. Eighteen lesions reached the coronoid process and 20 the condylar process of the mandible

TABLE 2 Treatment recurrence—for the lesions with available information about treatment and recurrence

Treatment	Recurrence/total (% recurrence)
Curettage	10/32 (31.3)
Enucleation	8/61 (13.1)
Enucleation + curettage	10/79 (12.7)
Enucleation + peripheral osteotomy	1/15 (6.7)
Marginal resection	1/80 (1.3)
Segmental resection[a]	5/161 (3.1)

[a]Resection with continuity defect.

initial treatment to the first recurrence (information available for 38 of the 44 recurrences) ranged from 3 to 180 months after treatment, with a mean interval of 24.5 ± 30.5 months. Sixteen recurrences (42.1%) occurred within 12 months after treatment and 73.7% within 24 months. Table 2 shows the recurrence rate according to the treatment performed. Curettage was the treatment with the highest rate of recurrence (10/32, 31.3%), followed by enucleation (8/61, 13.1%). Additional curettage after enucleation (10/79, 12.7%) had a similar recurrence rate in comparison with enucleation alone (13.1%), but the performance of peripheral osteotomy decreased the recurrence rate (1/15, 6.7%). Treatment by resection, regardless whether marginal or segmental, had the lowest recurrence rate, 1.3% (1/80) and 3.1% (5/161), respectively.

Lesions smaller than 2 cm were most often treated by enucleation (in 50% of the cases; and 27.3% by marginal resection, 18.2% by curettage, and 4.5% by segmental resection), as well as those lesions with size between 2 and 5 cm (39.8% treated by enucleation, 26.6% by marginal resection, 17.5% by segmental resection, and 16.1% by curettage). Neoplasms between 5 and 10 cm and those larger than 10 cm were most often treated by segmental resection—52.9% and 61.5% of the cases, respectively.

Table 3 shows the recurrence rate according to some variables. None of the variables analyzed showed a statistically significant

TABLE 3 Recurrence rate for central odontogenic myxomas according to different factors—for the lesions with available information about both recurrence and the factors here included

Factor	Recurrence/total (% recurrence)	*p* value	Odds ratio (95% CI)	*p* value
Jaw				
Maxilla	20/214 (9.3)	0.955[a]	1	
Mandible	24/261 (9.2)		0.982 (0.527, 1.831)	0.955
Bone expansion				
No	3/23 (13)	0.280[b]	1	
Yes	31/339 (7.8)		0.562 (0.158, 1.995)	0.372
Cortical bone perforation				
No	4/81 (4.9)	0.070[a]	1	
Yes	25/207 (12.1)		2.644 (0.890, 7.854)	0.080
Locularity				
Unilocular	8/135 (5.9)	0.404[a]	1	
Multilocular	19/229 (8.3)		1.436 (0.611, 3.377)	0.406
Tooth root resorption				
No	13/227 (5.7)	0.061[b]	1	
Yes	7/53 (13.2)		2.505 (0.947, 6.625)	0.064
Odontogenic epithelial rests				
No	30/317 (9.5)	0.374[b]	1	
Yes	3/46 (6.5)		0.667 (0.195, 2.282)	0.519
Angular septa				
No	13/215 (6.0)	0.415[a]	1	
Yes	10/119 (8.4)		1.426 (0.605, 3.358)	0.417

CI, confidence interval.
[a]Pearson's chi-squared test.
[b]Fisher's exact test.

TABLE 4 Comparison of demographic and clinical features of unilocular and multilocular central odontogenic myxomas

Variables	Unilocular	Multilocular	*p* value
n	187	317	
Age (years), mean ± *SD* (min–max)	28.5 ± 16.7 (1–80; *n* = 187)	29.8 ± 15.0 (2–79; *n* = 316)	0.402[a]
Gender, *n* (%)			
Men	80 (44.2)	130 (41.1)	0.544[b]
Women	101 (55.8)	184 (58.6)	
Unknown	6	3	
Jaw, *n* (%)			
Maxilla	93 (49.7)	105 (33.1)	<0.001[b]
Mandible	94 (50.3)	212 (66.9)	
Unknown	0	0	
Bone expansion, *n* (%)			
Yes	140 (88.6)	257 (95.2)	0.011[b]
No	18 (11.4)	13 (4.8)	
Unknown	29	47	
Symptomatic, *n* (%)			
Yes	24 (15.5)	52 (19.7)	0.280[b]
No	131 (84.5)	212 (80.3)	
Unknown	32	53	
Cortical bone perforation, *n* (%)			
Yes	75 (61.5)	145 (78.4)	0.001[b]
No	47 (38.5)	40 (21.6)	
Unknown	65	132	
Tooth displacement/unerupted, *n* (%)			
Yes	82 (50.6)	137 (53.7)	0.536[b]
No	80 (49.4)	118 (46.3)	
Unknown	25	62	
Tooth root resorption, *n* (%)			
Yes	30 (20.3)	41 (17.8)	0.553[b]
No	118 (79.7)	189 (82.2)	
Unknown	39	87	
Odontogenic epithelial rests, *n* (%)			
Yes	16 (11.3)	33 (13.7)	0.508[b]
No	125 (88.7)	208 (86.3)	
Unknown	46	76	
Treatment, *n* (%)			
Curettage	10/146 (6.9)	20/253 (7.9)	0.721[b]
Enucleation	78/146 (53.4)	70/253 (27.7)	0.001[b]
Marginal resection	26/146 (17.8)	40/253 (15.8)	0.662[b]
Segmental resection [c]	32/146 (21.9)	123/253 (48.6)	<0.001[b]
Recurrence, *n* (%)			
Yes	8 (5.9)	19 (8.3)	0.404[b]
No	127 (94.1)	210 (91.7)	
Unknown	52	88	
Follow-up time (months), mean ± *SD* (min–max)	35.1 ± 43.7 (1–276; *n* = 123)	42.8 ± 50.8 (0–420; *n* = 202)	0.040[a]
Lesion size (cm), mean ± *SD* (min–max)	3.5 ± 1.7 (0.7–8.0; *n* = 101)	5.5 ± 3.0 (1.5–25.0; *n* = 150)	<0.001[a]

[a]Mann–Whitney test.
[b]Pearson chi-squared test.
[c]Resection with continuity defect.

influence on the recurrence rate. Table 4 shows a comparison between lesions of different radiological appearance. Multilocular lesions were statistically significantly more prevalent in mandibles than in maxilla in comparison with lesions with a unilocular radiological appearance. Moreover, multilocular OMs more often presented bone expansion and cortical bone perforation than the unilocular myxomas and presented larger mean size in comparison with the unilocular ones. Multilocular tumors were more often treated by segmental resection and less often by enucleation than unilocular tumors.

4 | DISCUSSION

The aim of the present study was to integrate the available data published in the literature on OM into an updated comprehensive comparative analysis of their clinical and radiological features, as well as the frequency of recurrence. A review of pathological lesions is important because it provides information that can improve diagnostic accuracy, allowing pathologists and surgeons to make informed decisions and refine treatment plans to optimize clinical outcomes (Chrcanovic & Gomez, 2016, 2017a,b).

Many publications were excluded from the present literature review after analysis of the clinical, radiographic, and microscopical features available. This calls the attention to the fact that authors need to better describe their findings. When it comes to OM in particular, the histopathological features play an important role, because dental papillae could be erroneously interpreted as OM when no radiographic or surgical information is available (Li, Sun, & Luo, 2006). Moreover, myxoid change in other neoplasms may mimic OMs (Halfpenny, Verey, & Bardsley, 2000). In that sense, many of the previous reviews could have included cases that may have the diagnosis questioned.

The present review observed a predominance of OMs in females and in mandibles, which is in agreement with previous studies (Martínez-Mata et al., 2008; Noffke, Raubenheimer, Chabikuli, & Bouckaert, 2007). OMs usually present with bone expansion with asymptomatic cortical perforation, and radiographically, the

tumor is commonly multilocular. Although the presence of angular septa raises the suspicion of OM, this was not observed in most cases. Neither the presence of odontogenic epithelial rests nor of angular septa seems to have an impact on the biologic behavior of the OMs. Although there was a higher prevalence of cortical bone perforation in recurrent OM in relation to non-recurrent OM, it was not statistically significant. OMs with a multilocular radiological appearance are more prevalent in mandibles in comparison with unilocular lesions, also showing higher prevalence of bone expansion and of cortical bone perforation, and having larger mean size than the unilocular ones. Still, there was no statistically significant association between these factors and an increase in recurrence rate.

The difference in treatment was the only factor suggested to have a statistically significant influence on the recurrence rate of OMS. Curettage showed the highest risk for recurrence. It seems that an additional curettage after enucleation does not have any impact on the recurrence rates in comparison with enucleation alone, but the performance of additional peripheral osteotomy after enucleation may decrease the chance of recurrence. These relatively high recurrence rates with curettage and enucleation can be attributed to the lack of encapsulation and the subtle local invasion of neoplasms between cancellous bones beyond radiographically visible margins (Pahl, Henn, Binger, Stein, & Remberger, 2000), which the peripheral osteotomy after enucleation seems to be able to remove, but not the additional curettage. Recurrences of OMs could be more related to incomplete removal than to the intrinsic biologic behavior of the neoplasm (Batsakis, 1987). Despite the small number of cases treated with enucleation followed by peripheral osteotomy and the lower recurrence with marginal resection, it is of the authors' opinion that tumor enucleation followed by peripheral osteotomy should be the first choice of treatment due to the lower associated morbidity. However, cases compromising segments of the mandible in its whole caudal–cranial extension with extensive cortical thinning and large areas of cortical perforation should rather be treated by resection with continuity. These cases usually present no bone support left for the performance of an enucleation.

The results of the present study have to be interpreted with caution because of its limitations. First, all included studies were retrospective reports, which inherently result in errors, with incomplete records. Second, many of the published cases had a short follow-up, which could have led to an underestimation of the actual recurrence rate. However, it is hard to define what it would be considered a short follow-up period to evaluate the recurrence of these neoplasms. Considering that over half of the recurrences occurred after 1-year postsurgery and about one-third of the lesions were followed up to 12 months, the actual recurrence rate might be underestimated. Third, many of the cases described were published as isolated case reports or small case series.

5 | CONCLUSIONS

More conservative surgical procedures are associated with higher probability of recurrence of OM. Despite the fact that resection showed the lowest recurrence rates, surgeons should bear in mind that tumor enucleation followed by peripheral osteotomy should be considered as the first choice of treatment due to the associated lower morbidity.

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CONFLICT OF INTEREST

The authors declare that they have no conflict of interest.

AUTHORS' CONTRIBUTION

Bruno R. Chrcanovic involved in conception and design and statistical analysis, acquired the data, interpreted the data, drafted the article, revised the article critically for important intellectual content, and approved the final version to be published; Ricardo S. Gomez involved in conception and design, interpreted the data, revised the article critically for important intellectual content, and approved the final version to be published.

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SUPPORTING INFORMATION

Additional supporting information may be found online in the Supporting Information section at the end of the article.

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