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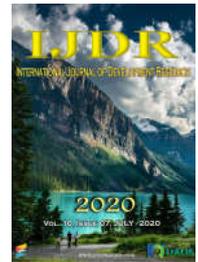
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SURGICAL MANAGEMENT OF UNICYSTIC AMELOBLASTOMA: CONSERVATIVE OR RADICAL APPROACH: A SYSTEMATIC REVIEW

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ABSTRACT

Unicystic ameloblastoma is a benign epithelial odontogenic neoplasm that represents about 5 to 15% of all intraosseous ameloblastomas, its biological behavior is related to its histopathological subtypes, with the mural variant being the most aggressive. The objective of this systematic review was to evaluate in the scientific literature the effectiveness of the conservative and radical surgical approach in the treatment of unicystic ameloblastomas. A systematic literature review was performed using the Science Direct, Embase, Cochrane Collaboration Library, and PubMed/MEDLINE databases. Studies related to conservative and radical surgical therapies for the treatment of unicystic ameloblastomas were selected. The search strategy provided a total of 791 studies. After screening by reading the titles and abstracts, 25 studies were considered potentially eligible and read in full by the evaluators. At the end of the analysis, five articles met all the inclusion criteria and were included in this systematic review. Studies have shown that a conservative surgical approach for the treatment of unicystic ameloblastoma is recommended only for the luminal and intra-luminal variants, and is not recommended for the mural subtype. However, in order to carry out a radical treatment, one must take into account the age and health conditions of the patient. Post-surgical clinical and radiographic monitoring is of fundamental importance for early detection of recurrence.

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INTRODUCTION

Several sequential interactions between the epithelium and the mesenchyme that originate from the neural crest are related to tooth development. The size, shape and structure of the teeth, as well as their position in the arch, are determined by genes that act on dental development and maturation (Nanci, 2013). The occurrence of pathological processes in the remnants of these primordial tissues can lead to the development of tumors that differ in their microscopic appearance and biological behavior (Mahajan, 2013; Kulkarni, 2017).

Ameloblastoma is a benign epithelial odontogenic neoplasm formed by the odontogenic epithelium without the participation of the ectomesenchyme, its pathogenesis is related to the presence of the epithelial rests of Malassez, the rests of Serres and the reduced epithelium of the enamel organ. The lesion has a multilocular radiographic appearance, aggressive behavior and a high rate of recurrence (Costa, 2016 and Li, 2017). In the fourth edition of WHO (2017), ameloblastoma underwent changes in its terminology, being classified according to new studies based on current genetic concepts. The classification has been simplified for solid

ameloblastoma, unicystic ameloblastoma and peripheral ameloblastoma, with each variant presenting a different biological behavior in relation to treatment and prognosis. (El-Naggar, 2017 and Wright, 2017). Unicystic ameloblastoma has less clinical aggressiveness when compared to solid ameloblastoma, representing between 5 to 15% of all intraosseous ameloblastomas (Nagi, 2016). The lesion commonly occurs between the first and fourth decade of life and has no predilection for ethnicity (Li, 2017). It presents clinically as a painless, slow-growing lesion, with greater involvement in the posterior region of the mandible and is often associated with unerupted lower third molars. Radiographically, the lesion displays a delimited unilocular radiolucent image similar to a cyst (Muzio, 2017). Microscopically, the lesion exhibits a cystic cavity, with three types of variants being observed, luminal, intraluminal and mural. The luminal and intra-luminal subtype has an ameloblastomatousepithelium and cell infiltration into the fibrous capsule is not observed. However, the mural pattern exhibits an ameloblastomatous epithelium that infiltrates the capsule in the form of cords or cell nests, with a high recurrence rate (Wright, 2017 and El Naggar, 2017). The treatment of unicystic ameloblastoma varies from a conservative to a radical approach, depending on the size of the lesion and the histopathological subtype. Conservative surgical enucleation is recommended for the luminal and intra-luminal variants (Meshram, 2017). However, in the mural pattern, performing only a surgical enucleation will not allow a complete removal of cellular remnants in the adjacent bone, requiring curettage of the site, use of sclerosing solutions or marsupialization to reduce the size of the lesion (Anchlia, 2016). On the other hand, some studies do not recommend the use of conservative treatment in the mural variant, and radical treatment through marginal resection or aggressive resection of the tumor with a safety margin and adequate bone reconstruction of the segment is indicated. The recurrence rate after conservative treatment varies from 36% in tumors, being significantly higher than the recurrence rate for radical treatment, which varies from 8.3% (Sasaki, 2014). It is observed that currently there are several surgical protocols for the treatment of unicystic ameloblastoma. However, it is not yet completely clear which is the most suitable and which presents the most promising results. Therefore, the objective of this systematic review was to evaluate in the scientific literature the effectiveness of the conservative and radical surgical approach in the treatment of unicystic ameloblastomas.

MATERIALS AND METHODS

This study followed the guidelines of the Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA) Statement (Moher, 2009).

Information of sources and search strategies: A literature search was performed in June of 2020 in the following electronic databases: Science Direct, Embase, Cochrane Collaboration Library, and PubMed/MEDLINE. The following review question was developed according to population, intervention, comparison, and outcome (PICO): "What is the most appropriate surgical management of unicystic ameloblastoma?" and a keyword search was performed. The search was carried out without time and language restrictions. Hand searches were also conducted by cross-checking the reference lists of the included articles. Duplicates were

removed upon identification. The search strategy was based on combinations of the following keywords: ("Ameloblastoma" [tw] OR "Unicystic ameloblastoma"[tw]) AND ("Treatment" [tw] OR "Conservative treatment" [tw] OR "Radical Treatment" [tw]) AND ("Prognostic"[tw]).

Eligibility criteria and Study selection: The reviewer independently screened and assessed potential articles. Studies that did not fulfil the inclusion criteria were excluded. In the first stage, the titles and abstracts of all retrieved reports were screened for potentially eligible studies. The full text articles of the previously identified studies were then examined in detail according to predefined eligibility criteria for inclusion in the qualitative review. Disagreements were solved by discussion between the authors. The inclusion criteria were:

1. Studies that evaluated the treatment modalities for unicystic ameloblastoma and its role associated with the prognosis.
2. Diagnosis of unicystic ameloblastoma obtained after histological examination.
3. At least presented one treatment approach (radical or conservative) of unicystic ameloblastoma and matching recurrence rate.

Exclusion criteria:

1. Animal studies.
2. in vitro studies.
3. Opinion articles.
4. Letters to the editor.
5. Review articles.
6. Interviews, updates.
7. Abstracts.
8. Unpublished studies.

The review authors independently screened the articles for data extraction. Any disagreements were resolved by discussion.

RESULTS

Study selection and characteristics: The search strategy developed in this systematic review identified a total of 791 studies located in the evaluated databases. After screening through reading the titles and abstracts and excluding duplicate articles, 25 studies were considered potentially eligible and read in full by the evaluators. At the end of the analysis, five articles published between 2003 and 2019 met all the inclusion criteria and were selected for the present systematic review. The flowchart applied for the article selection and selection process is illustrated in figure 1. In total, 334 patients diagnosed with unicystic ameloblastoma were evaluated in the five selected studies, with the sample ranging from 5 patients (Junquera, 2003) to 116 patients (Zheng, 2019). Most patients were diagnosed with unicystic ameloblastoma in the fourth decade of life. The average follow-up time ranged from one to nine years. All included studies evaluated different surgical treatment protocols for unicystic ameloblastoma. The main methodological aspects and observed results can be seen in Table 1.

Main results: In the study by Junquera (2003) conservative surgical treatment was performed in 5 cases of unicystic ameloblastoma. Such patients were treated by enucleation and curettage and, in this sample, 40% of cases had at least one

Table 1. Summary of the descriptive characteristics and results of the included studies (n=5)

Author (year)	Numberofpatients	Groups	Age(mean)	Surgerytype	Recurrence(%)	Follow-up	Summary of the results
Junquera (2003)	5	Group I. Conservative treatment (n= 5)	31.4	Enucleation and curettage	40%	1 to 9 years	In the sample treated conservatively, forty percent of unicystic lesions had at least 1 recurrence.
Hong (2007)	91	Group I. Conservative treatment (n= 77) Group II. Resection with bone margin (n= 11) Segmental resection or maxillectomy (n= 3)	34.5	Enucleation, marsupialization followed by enucleation, or enucleation with bone curettage – without safety margin,	13.1%	2 to 14 years	The unicystic type show a relatively low potential for recurrence.
Fregnani (2010)	7	Group I. Conservative treatment (n= 7)	33.2	Curettage associated with cryotherapy.	28.5%	9.7 years (mean)	The two unicystic cases that recurred were of the mural subtype.
Gunawardhan (2010)	115	Group I. Conservative treatment (n= 64) Group II. Resection Surgery (n= 42)	5–80 years	Enucleation (Group I) or Resection Surgery (Group II)	Group I = 27% Group II = 0%		There was no recurrence in the group that underwent surgical resection. In 27% of the cases treated with enucleation, there was recurrence.
Zheng (2019)	116	Group I. Conservative treatment (n= 116)	33.4 (mean)	Marsupialisation	12%	9 years (mean)	Marsupialisation is effective for these patients, with a recurrence rate similar to that of radical treatment. The outcomes can be predicted using characteristics of the lesion such as resorption of the root, perforation of the cortical bone, and histopathological subtypes.

recurrence during the follow-up period. The recurrence rate in the study by Junquera (2003) was the highest among all the studies analyzed, however, other studies such as Hong (2007), Fregnani (2010), Gunawardhan (2010) and Zheng (2019) also showed recurrence rates varying between 12 and 28.5%. Gunawardhana (2010) provided data on mandibular and maxillary ameloblastomas in the Sri Lankan population. Unicystic ameloblastomas were treated conservatively (enucleation) or aggressively (resection surgery). In this study, no cases of recurrence were observed among patients treated through surgical resection, presenting itself as an effective treatment. However, the authors emphasize that important factors such as lesion size, histopathological subtype, age of the patient must be taken into account when choosing treatment, and long-term follow-up of all cases should always be performed. In the study by Hong (2007), the importance of analyzing the histopathological subtype was also emphasized in order to choose the most appropriate treatment for unicystic ameloblastoma, with the mural subtype associated with a higher probability of recurrence compared to other histopathological patterns.

DISCUSSION

Odontogenic tumors are a heterogeneous group of lesions with different biological behaviors and different clinical manifestations. Unicystic ameloblastomas do not have a gender predilection, affecting mostly young adult patients (Costa, 2016 and El Naggar et, 2017). In the present study, it was observed that the affected patients were in the fourth decade of life, and were diagnosed through routine exams, corroborating the findings in the literature.

The differential radiographic diagnosis includes a variety of tumors and cysts of an odontogenic nature, such as myxoma, odontogenic keratocyst, also including tumors and non-odontogenic cysts, such as the central giant cell lesion (El Naggar, 2017). The performance of procedures such as aspiration puncture and incisional biopsy are essential to confirm the diagnosis and establish a correct treatment (Muzio, 2017).

According to Samuel (2014) conservative treatment should be used as a primary approach in patients with unicystic ameloblastoma and radical treatments should only be performed only in cases of recurrence. Rakesh (2010) demonstrates in his study that vigorous bone curettage and chemical cauterization with Carnoy's solution should always be performed after enucleation. Conservative treatment is suggested mainly for young patients between the first and second decade of life, in view of the impacts in the development of the jaw, masticatory function, facial growth and psychosocial aspects that a radical treatment would cause. In the study by Junquera (2003) and Fregnani (2010), all patients were treated by enucleation, curettage and cryotherapy. However, most cases had local recurrence. Therefore, radiographic monitoring of patients should be performed for a minimum period of 10 years, since recurrence after conservative treatment is approximately 7 to 25% (Kahairi, 2008; Pogrel, 2009). Marsupialization is a treatment option that aims to decompress and reduce the injury (Sasaki, 2014). To perform this procedure, a surgical window must be made that presents a communication with the oral cavity, and that is sutured next to the adjacent mucosa. The surgical window must be kept open so that the internal content of the lesion gradually empties out.

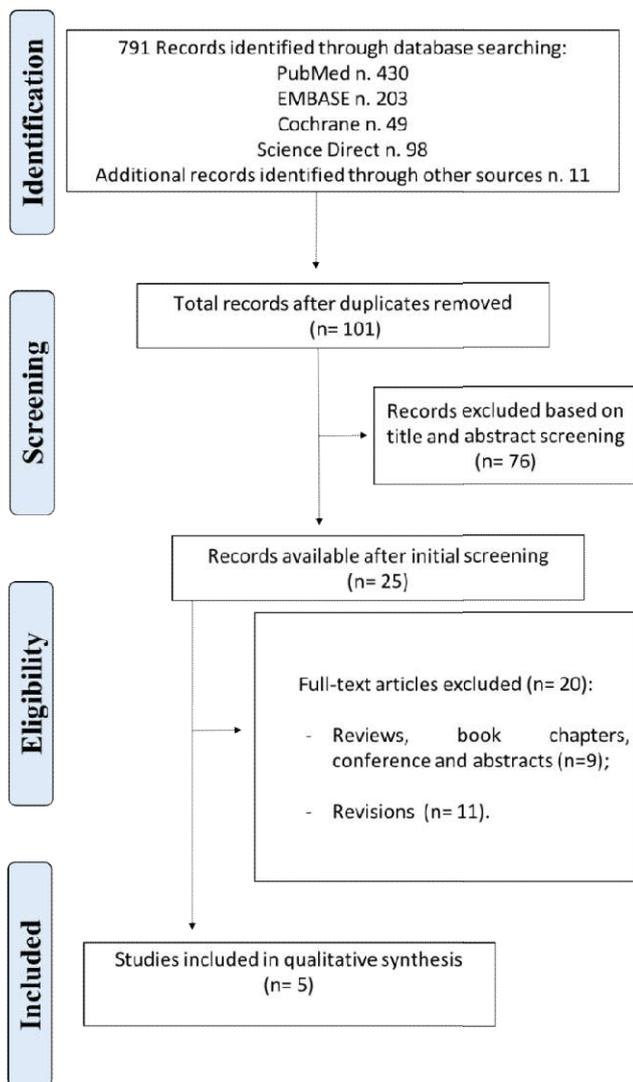


Figure 1. PRISMA flow diagram of screened studies

In the conservative treatment of extensive injuries, this approach allows decompression prior to curettage to occur, thereby avoiding radical treatment (Isolan, 2018). In the study by Zheng (2019), marsupialization presented itself as an effective treatment in reducing unicystic ameloblastoma. However, some complications such as cortical perforation have been associated with this therapy. Despite the good results obtained, the isolated technique should not be considered as a standard procedure because it can cause the lesion to recur (Isolan, 2018). Surgical resection with a safety margin should be performed taking into account factors such as the patient's health status, age, location and tumor extension (Anchlia, 2016). In the study by Gunawardhana (2010), all patients treated by surgical resection did not present recurrence. However, the aesthetic, phonetic and functional problems caused by a possible facial deformity must be taken into account when choosing this therapy. Reconstructive and rehabilitative procedures should be proposed for patients after radical surgery (Nagi, 2016). Rakesh (2010) reported the importance of evaluating the histopathological subtype of unicystic ameloblastoma to choose the appropriate treatment for the patient. The luminal and intraluminal variants have less recurrence, and conservative treatment may be indicated in these cases, especially in young patients. However, conservative treatment is not indicated for the mural variant, which presents a recurrence rate corresponding to 35.7% (Samuel, 2014).

These findings corroborate the study by Hong (2007), in which the importance of performing histopathological exam was highlighted in order to choose the appropriate treatment.

Conclusion

Knowing the biological behavior of unicystic ameloblastoma is essential to establish an adequate therapeutic approach. Microscopic examination is of fundamental importance to elucidate the histopathological subtype of the lesion, and to establish a more accurate prognosis for patients. Studies have shown that a conservative surgical approach for the treatment of unicystic ameloblastoma is recommended only for the luminal and intra-luminal variants, and is not recommended for the mural subtype. It is important to note that the mural variant, due to the infiltration of cells in the capsule of the lesion, is considered the most aggressive variant of unicystic ameloblastomas, and should be treated radically when compared with the other variants, in view of its high recurrence rate. However, in order to carry out a radical treatment, one must take into account the age and health conditions of the patient. Post-surgical clinical and radiographic monitoring is of fundamental importance for early detection of recurrence.

Conflict of Interest: The authors declare no conflicts of interests.

Ethics approval: Not applicable.

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