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Impacted teeth “kissing molars": systematic review

Dentes impactados "kissing molars": revisão sistemática

Impactan dientes "kissing molars": revisión sistemática

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Abstract

Background: The purpose of this systematic review was to gather more information on impacted teeth called "kissing molars", and their treatment, allowing the oral surgeon to guide the clinical resolution. Methods: An electronic survey was conducted in five databases and manual search, in accordance with the Guidelines for Preferred Reporting Items for Systematic Reviews (PRISMA). The inclusion criteria were studies in which at least one patient was diagnosed with "kissing molars"; published in English, since 2000. Results: Fifteen studies were included for the analysis, totaling 37 patients evaluated, being 21% (8) bilateral cases, 54% (20) unilateral cases and 24% (9) cases were not specified. In 40% (15) patients presented some symptoms that led to the diagnosis. The treatment performed in most cases was dental extraction 89% (33 patients) with post-operative complications in 8% of the cases. Conclusion: The findings of this systematic review suggest a complement in the classification and standardization of this clinical condition, to help in the planning and treatment.

Keywords: Third molar; Tooth; Kissing molars; Impacted molars; Systematic review.

Resumo

Fundamento: O objetivo desta revisão sistemática foi reunir mais informações sobre os dentes incusos denominados "kissing molars" e seu tratamento, permitindo ao cirurgião oral orientar a resolução clínica. Métodos: Foi realizada uma pesquisa eletrônica em cinco bancos de dados e busca manual, de acordo com as Diretrizes para Itens de Relatórios Preferenciais para Revisões Sistemáticas (PRISMA). Os critérios de inclusão foram estudos em que pelo menos um paciente foi diagnosticado com "kissing molars"; publicados em inglês, desde 2000. Resultados: Quinze estudos foram incluídos para análise, totalizando 37 pacientes avaliados, sendo 21% (8) casos bilaterais, 54% (20) casos unilaterais e 24% (9) casos não especificados. Em 40% (15) os pacientes apresentavam alguns sintomas que levaram ao diagnóstico. O tratamento realizado na maioria dos casos foi extração dentária 89% (33 pacientes) com complicações pós-operatórias em 8% dos casos. Conclusão: Os achados desta revisão sistemática sugerem um complemento na classificação e padronização dessa condição clínica, para auxiliar no planejamento e tratamento.

Palavras-chave: Terceiro molar; Dente; Kissing molars; Molares impactados; Revisão sistemática.

Resumen

Antecedentes: el objetivo de esta revisión sistemática fue recopilar más información acerca de los dientes incubados llamados "molares besantes" y su tratamiento, lo que le permitió al cirujano oral guiar la resolución clínica. Métodos: Se realizó una búsqueda electrónica en cinco bases de datos y una búsqueda manual, de acuerdo con las Directrices para los Ítems Preferidos de Reporte para Revisiones Sistemáticas (PRISMA). Los criterios de inclusión fueron estudios en los que al menos un paciente fue diagnosticado con "molares besantes"; publicado en inglés, desde 2000. Resultados: Se incluyeron 15 estudios para análisis, totalizando 37 pacientes evaluados, siendo 21% (8) casos bilaterales, 54% (20) casos unilaterales y 24% (9) casos no especificados. En el 40% (15) los pacientes presentaron algunos síntomas que llevaron al diagnóstico. El tratamiento realizado en la mayoría de los casos fue la extracción del diente en el 89% (33 pacientes) con complicaciones postoperatorias en el 8% de los casos. Conclusión: Los hallazgos de esta revisión sistemática sugieren un complemento en la clasificación y estandarización de esta condición clínica, para ayudar en la planificación y el tratamiento.

Palabras clave: Tercer molar; Diente; Kissing molars; Molares impactados; Revisión sistemática.

1. Introduction

Us described as an extremely rare alteration in the literature, the term "kissing molars" is used to describe alterations in the positioning of impacted permanent molars, which present with the occlusal surface in contact with each other in a single follicular space, with the roots pointing in opposite directions (Gulses, Varol, Sencimen, & Dumlu, 2012; Van Hoof, 1973) (Figure 1).

Figure 1 - X-ray exemplifying the “kissing molar”.

Source: Authors.

This alteration was initially reported by Van Hoof in 1973, and due to its rarity, its etiology is still unknown, without predilection for gender or age, are more frequent unilateral as the involvement between the second and third molars, but according to the cases described in the literature, may present bilaterally, involving other molars (first, second, third and/or fourth molars) and be associated with cysts (Boffano & Gallesio, 2009; Menditti et al., 2015; Van Hoof, 1973).

Due to this rarity, few studies are found in the literature, and there is a lack of standardization in the information about this change and especially in the treatment. The objective of this review is to gather more information about this condition and its treatment, allowing the clinician to guide the clinical resolution of these patients.

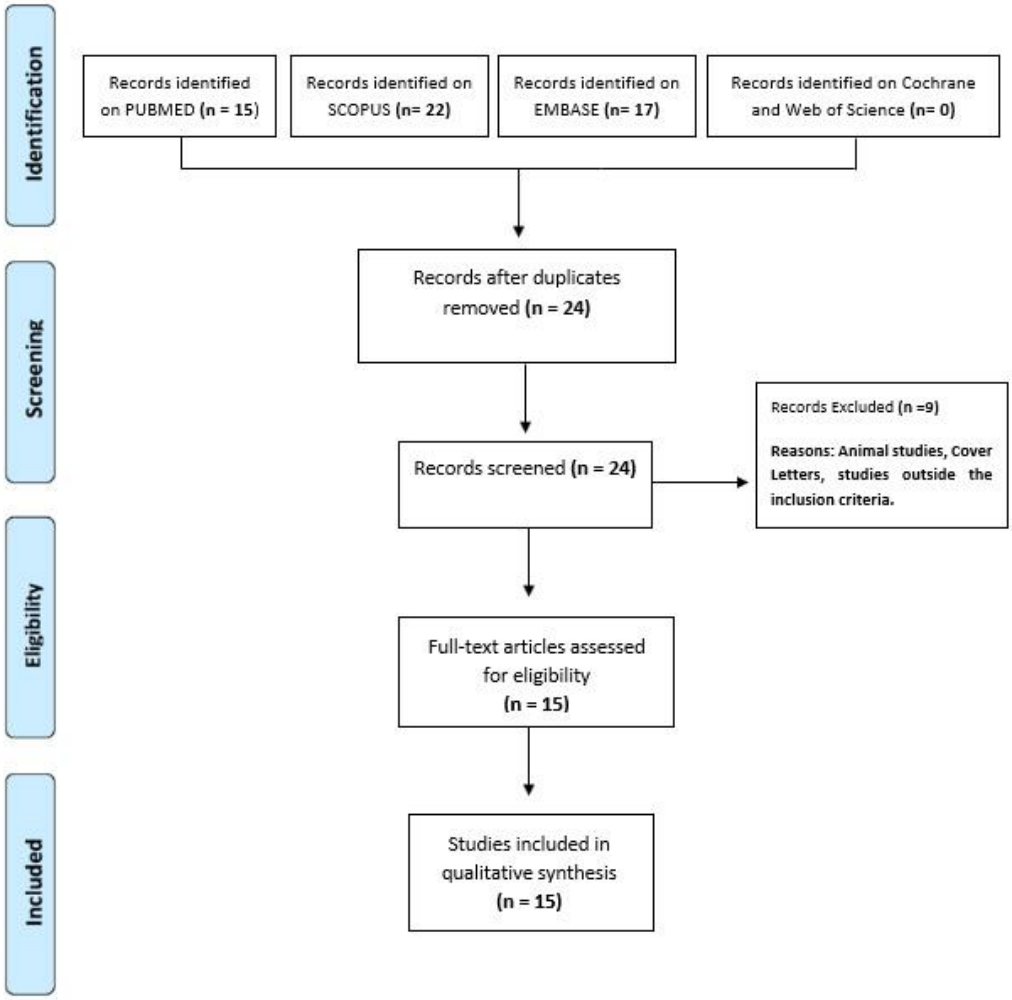
2. Metodology

This systematic review was conducted in accordance with the Guidelines on Preferred Reporting Items for Systematic Reviews (PRISMA). Following the PRISMA standards, using the method adapted from the PICO structured question, in this case the PEO format (Population,

Exposure and Outcome) is applied (Honório & Santiago, 2018; Kahn, 2003). For the work, the question was established: "what are the clinical implications related to patients diagnosed with "kissing molars" ?", in which (P) represented patients diagnosed with "kissing molars", who performed treatment (E) and evaluated in relation to epidemiological data, treatment methods and complications (O). Inclusion criteria were studies in which at least one patient was diagnosed with "kissing molar"; published in English; with year restriction from 2000 onwards. The exclusion criteria were animal studies, publications entitled "Letters to the Editor" and articles in languages other than English.

To Research Strategy two reviewers (W.P.P.S. and C.L.) were conducted independent research on PubMed / MEDLINE, Embase, Scopus, Web of Science and Cochrane Library databases for articles published until March 2020. The keywords selected were: "Kissing Molars" AND "Impacted Molars" for the search strategies in the five databases, in addition to conducting the search through manual search. The selection process of the articles was carried out in two stages: (1) reading of the title and abstract; and (2) full text analysis. Two reviewers (W.P.P.S. and C.L.) conducted independent and blind analysis based on eligibility criteria. Divergences of opinion on the inclusion or exclusion of a study were resolved by consensus, with the involvement of a third reviewer when necessary. Data were extracted from selected articles after full text analysis, and all articles were evaluated for bias risk (Figure 2).

Figure 2 - Flow chart of the literature search and screening process.



Source: Authors.

For data extraction one reviewer (W.P.P.S.) performed the data extraction and another (C.L.) verified the information through the tabulation of demographic characteristics and data on the study populations, interventions, and results of interest.

3. Results and Discution

Study selection

The initial search of the articles according to the PEO methodology was performed using the keyword combinations described above and a total of 54 studies were obtained, 15 articles were found in the Medline/PubMed database, 22 articles in the Scopus database, 17 articles in

the Embase database, in the Web of Science and Cochrane databases no studies were found, the manual search was performed and seven articles were found. After the removal of duplicate articles, the study abstracts were read and articles that did not fit the inclusion and exclusion criteria were eliminated, remaining a total of 15 articles selected for inclusion in the study.

Data items

In the 15 selected studies (Table 1), 37 patients (100%) were reported, 16 men (43%) and 21 women (57%), with ages ranging from 10 to 56 years, with a mean of 29.33 years. 22 patients were reported as healthy, 1 patient with a history of mental retardation, 1 patient diagnosed with Down syndrome, 1 patient with perinatal cerebral Anoxia, 1 patient with hypercholesterolemia and mucopolysaccharidosis, 1 patient with asthma and mucopolysaccharidosis, and in 8 patients no studies were reported.

Table 1 - Summary of the studies included.

Author	Year	Gender/ Age	Location / Teeth	Radiography	Medical Conditions	Symptomatology	Treatment	Postoperative Complications	Histology
Boffano *et al*	2009	M/42	3rd and 4th right	Panoramic	NR	Pain	Tooth extraction LA/ calcium sulfate grafting	No Complications	No Lesions
Gulses *et al*	2012	F/26	2nd and 3rd	No Reported (NR)	NR	No Symptoms	Tooth extraction	No Complications	NR
		F/32	2nd and 3rd	NR	NR	Associated cyst	Tooth extraction	No Complications	Dentigerous Cyst
		M/44	3rd and 4th	NR	NR	No Symptoms	Tooth extraction	No Complications	NR
		M/23	2nd and 3rd	NR	NR	No Symptoms	Tooth extraction	Inferior alveolar nerve paresthesia (4 months)	Follicular Cyst
		M/16	1st and 2nd	NR	NR	Associated cyst	Tooth extraction	Inferior alveolar nerve paresthesia (6 months))	Dentigerous Cyst
		F/37	2nd and 3rd	NR	NR	No Symptoms	Tooth extraction	No Complications	NR
		F/22	2nd and 3rd	NR	NR	No Symptoms	Tooth extraction	No Complications	Follicular Cyst
		M/27	3rd and 4th	NR	NR	Associated cyst	Tooth extraction	Inferior alveolar nerve paresthesia (3 months)	Dentigerous Cyst
		F/20	2nd and 3rd	NR	NR	No Symptoms	Tooth extraction	No Complications	NR
Sá Fortes *et al*	2012	M/33	2nd and 3rd Bilateral	TCCB	No Comorbidity	No Symptoms	local anesthetic tooth extraction	No Complications	Dentigerous Cyst
Giraldi Neto *et al*	2012	F/30	2nd and 3rd Left	Panoramic	No Comorbidity	Pain and inflammation	local anesthetic tooth extraction	No Complications	No Lesions
		F/22	2nd and 3rd Left	Panoramic and TCCB	No Comorbidity	No Symptoms	local anesthetic tooth extraction	No Complications	No Lesions
Guven *et al*	2013	M/32	2nd and 3rd right	Panoramic	Down Syndrome	Pain	local anesthetic tooth extraction	No Complications	No Lesions
		F/13	1st and 2nd right	Panoramic	Hipercolesterolemia Mucopolysacoid	No Symptoms	local anesthetic tooth extraction (2nd molar) and Orthodontics (1st molar)	No Complications	NR
		M/50	1st and 2nd left	Panoramic	No Comorbidity	No Symptoms	Refused treatment	-	-
		F/23	2nd and 3rd right	Panoramic and TCCB	Asthma Mucopolysacoid	No Symptoms	local anesthetic tooth extraction	No Complications	NR

Jannu *et al*	2014	F/18	2nd and 3rd Bilateral	Panoramic	No Comorbidity	No Symptoms	tooth extraction general anaesthesia	No Complications	NR
Nedjat-Shokouhi *et al*	2014	M/18	2nd and 3rd Bilateral	Panoramic	No Comorbidity	Associated cyst	tooth extraction general anaesthesia and Conservative treatment (left side)	No Complications	Dentigerous Cyst
Kiran *et al*	2014	F/18	2nd and 3rd Bilateral	Panoramic	No Comorbidity	Bilateral Edema	tooth extraction general anaesthesia / Gel Foam	No Complications	Follicular Cyst
Gonzalez-Perez *et al*	2015	F/29	2nd and 3rd right	Panoramic and TCCB	Anoxia Cerebral Perinatal	Extraoral Edema	tooth extraction general anaesthesia / calcium phosphosilicate graft	No Complications	NR
		M/35	2nd and 3rd right	Panoramic and TCCB	No Comorbidity	ATM Pain and associated Cyst	local anesthetic tooth extraction	No Complications	Dentigerous Cyst
		F/29	2nd and 3rd right	Panoramic	No Comorbidity	Pain ATM Muscles	Conservative treatment	-	-
Anish *et al*	2015	M/35	2nd and 3rd Bilateral	Panoramic occlusal x-ray	No Comorbidity	No Symptoms	NR	NR	NR
Menditti *et al*	2015	F/35	1st and 2nd Bilateral	Panoramic	No Comorbidity	No Symptoms	local anesthetic tooth extraction	No Complications	Dentigerous Cyst (right side)
		M/56	3rd and 4th Bilateral	Panoramic and TCCB	No Comorbidity	No Symptoms	local anesthetic tooth extraction	No Complications	Inflammatory Tissue
		F/30	3rd and 4th Left	Panoramic and TCCB	No Comorbidity	No Symptoms	local anesthetic tooth extraction	No Complications	Follicular Cyst
Arjona-Amo *et al*	2016	M/35	2nd and 3rd right	Panoramic and TCCB	No Comorbidity	Pain	local anesthetic tooth extraction	No Complications	No Lesions
		F/24	3rd and 4th Left	Panoramic and TCCB	No Comorbidity	Local discomfort	local anesthetic tooth extraction	No Complications	Follicular Cyst
		F/26	2nd and 3rd right	Panoramic	No Comorbidity	Pain and inflammation	local anesthetic tooth extraction / bovine bone grafting	No Complications	No Lesions
		F/31	2nd and 3rd right	Panoramic	Mental Retardation	Pain and inflammation	local anesthetic tooth extraction	No Complications	No Lesions
Zerener *et al*	2016	F/38	2nd and 3rd Bilateral	Panoramic	No Comorbidity	Right side edema	local anesthetic tooth extraction	No Complications	No Lesions
Barros *et al*	2017	F/10	1st and 2nd right	Panoramic	No Comorbidity	Surgical treatment report 3 years ago	Orthodontics	-	Cyst removal (not specific)

Yanik *et al*	2017	M/40	2nd and 3rd Left	Panoramic	No Comorbidity	Pain and Edema	local anesthetic tooth extraction	No Complications	Dentigerous Cyst
		M/32	2nd and 3rd Left	Panoramic	No Comorbidity	Pain and Edema	local anesthetic tooth extraction	No Complications	No Lesions
		M/35	3rd and 4th right	Panoramic and TCCB	No Comorbidity	Pain	local anesthetic tooth extraction	No Complications	No Lesions
		F/29	2nd and 3rd right	Panoramic	No Comorbidity	No Symptoms	local anesthetic tooth extraction	No Complications	No Lesions

*NR – Not Reported *M – Male *F – Female Source: Authors.

The evaluation of location in the studies included 21% (8) bilateral cases, 54%(20) unilateral cases, and 24%(9) cases were not specified. Among the percentage of unilateral cases, there was a predilection of 65% (13) on the right mandible and 35% (7) on the left. The most involved teeth were the 2nd and 3rd lower molars, found in 25 cases (6 bilateral). Then the 3rd and 4th molars, in 7 cases (1 bilateral), and finally the involvement of the 1st and 2nd molars, being reported in 5 cases (1 bilateral). In 40% (15) patients presented some symptoms that resulted in the diagnosis, being reported: pain (9 cases), inflammation (3 cases), edema (5 cases), alterations in the temporomandibular joint region (2 cases). In 45% (17) of the patients there were no symptoms. Another 5% (5) cases were diagnosed with association of cysts but did not specify the presence of other symptoms.

Diagnosis process

The methods used for diagnosis in 72% (27) cases were through panoramic radiography, being complemented in eight cases with cone-beam computed tomography (CBCT) and one case with occlusal radiography. Only 2% (1) cases were diagnosed with CBCT. Another 24% (9) cases did not specify the type of radiographic examination used.

Treatment performed

The treatment performed in most cases was exodontia 89% (33 patients), being under local anesthesia (20) or general anesthesia (4), nine cases were not specific regarding the type of anesthesia. Orthodontics was performed in one case, and in another case a combination of exodontics and orthodontics was performed. The conservative treatment was performed in one case, there was the report of a patient who presented bilaterally, but who performed exodontia on only one side and conservative treatment on the other side. The use of bone grafting after

exodontia was performed in three patients and Foam Gel in one patient. Only one case was not reported, and one patient refused the treatment.

Post-operative complications

Post-operative complications were present in 8% (3) cases, both of temporary paresthesia, being resolved in 3, 4 and 6 months respectively.The presence of pathologies associated with teeth was reported in 40% (15) cases, being diagnosed as changes in the dental follicle or inflammatory tissue in seven cases, and Dentigerous Cyst in eight cases.

Discussion

Although there are several articles reporting the incidence of permanent molar impaction, there are few studies on "kissing molars" due to their rarity. Like the first cases described in the literature, the diagnosis of ¨kissing molars¨ is usually obtained through "findings" in routine imaging exams, and panoramic radiography is usually requested to investigate a dental absence (Mardini & Gohel, 2017). In our study we can correlate this information, since the diagnosis of this alteration was made in 45% of the patients through the imaging examination because the patients did not present symptoms. Only 15 patients (40%) presented signs or symptoms, causing the professional search for investigation and resolution.

Panoramic radiography is reported as one of the main complementary methods for diagnosis in dentistry, due to the possibility of general visualization of anatomical structures in the middle and lower third of the face (Mardini & Gohel, 2017). In 72% of the cases, panoramic radiography was used as the main diagnostic method, and in cases that presented the need for more details for the operative planning, CBCT was requested (8 cases). CBCT presents itself as a high quality exam due to the detailed visualization of anatomical structures, however some services or patients may not have access to this exam due to the higher cost, complementary techniques such as radiography in occlusal incidence is a viable and low-cost method that helps in surgical planning (Anish et al., 2015; Eskandarlou, Poorolajal, Talaeipour, Talebi, & Talaeipour, 2014; Giraldi Neto et al., 2012; Halperin-Sternfeld, Machtei, & Horwitz, 2014; Sá Fortes, Júnior, Modolo, & Mackowiecky, 2014; Takeshita, Chicarelli, & Iwaki, 2015).

Its etiology, prevalence and incidence are not yet defined, the article by Nakamura et al (1992) reports a possible correlation with systemic changes in patients, such as mucopolysaccharidosis and other disorders that cause changes in tooth eruption (Nakamura et

al., 1992). However, after the publication of this study in 1992, only 2 of the 37 patients reported had been diagnosed with mucopolysaccharidosis (Güven, Akbulut, KURŞUN, & ÖZTAŞ, 2013), another three patients were diagnosed with some type of mental retardation (Arjona-Amo et al., 2016; Gonzalez-Perez, Infante-Cossio, Sanchez-Sanchez, Valdivieso-del-Pueblo, & Robles-Garcia, 2015; Güven et al., 2013) and 22 patients were reported as healthy.

The slight predilection for women (57%) than for men (43%) should not be considered as a factor, due to the low number of cases, as well as the great variation in the age group (10 - 56 years, average of 29.33 years) reported in the initial diagnosis.

The main characteristics of "kissing molars" are the sharing of teeth in a single follicle and having contact with their occlusal surfaces, Arneja et al. believes that the presence of a tooth cyst in any of the teeth involved, could not share the same follicle and thus there would be no contact between impacted teeth, considering that this is an uncommon case of impaction (Arjona-Amo et al., 2016). The presence of lesions associated with teeth was reported in 40% (15) cases, being diagnosed as changes in the dental follicle or inflammatory tissue in seven cases, and Dentigerous Cyst in eight cases.

Classification

The proposal to classify "kissing molars" appeared in 2012 by Gulses et al. in a radiological study, based on the teeth involved. Being Class I: first and second molar impaction; Class II: second and third molar impaction; Class III: third and fourth molar impaction (Gulses et al., 2012). Other authors have contributed to the improvement of this classification, such as the importance of the proximity of the occlusal surfaces of the teeth involved, thus defining a "true shape" (True Kissing Molars) of this change, considering the variation with cystic or dental follicle dilatation for each class (I, II and III), and not being associated with diseases/syndromes. The form of "Pseudos-Kissing Molars" considering cases of syndromes or that do not present a close contact to the occlusal surfaces (Gulses et al., 2012; Menditti et al., 2015).

The evaluation of location in the studies included 21% (8) bilateral cases, 54%(20) unilateral cases, and 24%(9) cases were not specified. Among the percentage of unilateral cases, there was a predilection of 65% (13) on the right mandible and 35% (7) on the left. The most involved teeth were the 2nd and 3rd mandibular molars, found in 25 cases (6 bilateral). Then the 3rd and 4th molars, in seven cases (1 bilateral), and finally the involvement of the 1st and 2nd molars, being reported in five cases (1 bilateral).

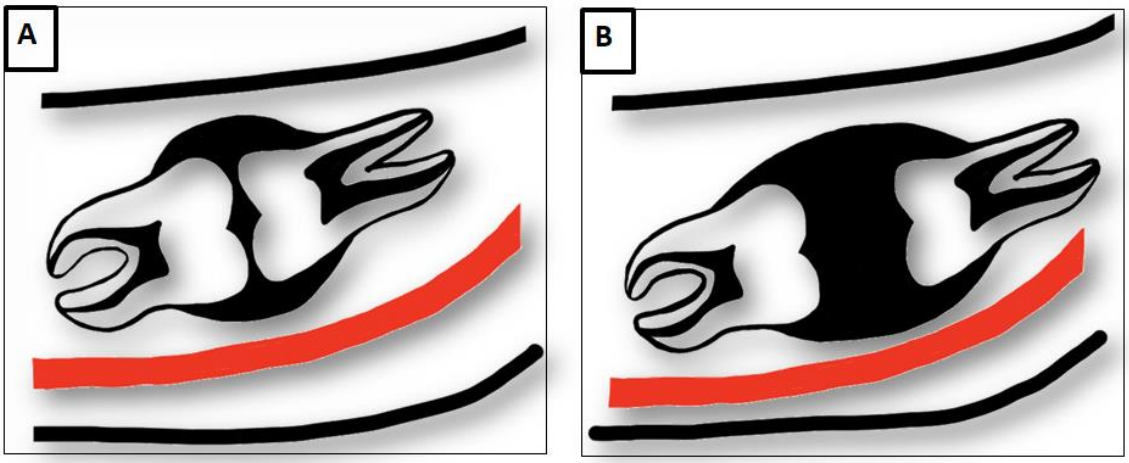
The authors suggest a complement in the classification to standardize the diagnosis for future studies, considering the previous classifications and adding the uni and bilateral presence. Consequently assisting in surgical planning and treatment. (Table 2) (Figure 3).

Table 2 – New classification (considering the previous classifications and adding the uni and bilateral presence).

RELATED TEETH	DENTAL FULLICLE	LOCATION
CLASS I - 1st and 2nd molar	A - no dilation - occlusal contact. B - with dilatation.	1 - Unilateral 2 - Bilateral
CLASS II – 2nd and 3rd molar	A - no dilation - occlusal contact. B - with dilatation.	1 - Unilateral 2 - Bilateral
CLASS III - 3rd and 4th molar	A - no dilation - occlusal contact. B - with dilatation.	1 - Unilateral 2 - Bilateral

Source: Authors.

Figure 3A – Dental follicle with no dilation (occlusal contact); **3B** – Dental follicle with dilatation.



Source: Authors.

The histopathological examination must be performed, the association of teeth with a hyperplastic dental follicle are reported, as well as the presence of an odontogenic cyst. Dental follicle hyperplasia is a disease that performs a 3-5mm expansion around the unerupted tooth crown and its histological analysis reveals a large amount of fibrous connective tissue and collagen fibers of the dental follicle, islands of odontogenic epithelium, mesenchymal

multinucleated giant cells and calcified mineralized areas, which can be confused with odontogenic fibroma (Menditti et al., 2015). Tooth cysts are the second most common type of dental cysts, representing 49% of all cystic lesions, and are usually associated with impacted or unerupted tooth crowns (Gulses et al., 2012; Kiran, Bharani, Kamath, Manimangalath, & Madhushankar, 2014).

The presence of lesions associated with teeth was reported in 40% (15) cases, being diagnosed as alterations in the dental follicle or inflammatory tissue in seven cases, and Dentigerous Cyst in eight cases, reinforcing the importance of performing histopathological examination.

The treatment of choice in most cases was the surgical, with exodontia (89%) of the teeth involved. Detailed planning to avoid trans and postoperative complications is essential to define the treatment plan The degree of difficulty should be assessed through details such as the location and depth of the teeth involved, the position and shape of the roots, the relationship of the teeth to the ascending branch, mandibular body and lower alveolar nerve canal (Jannu, KSN, GK, Veena, & Kamath, 2014).

Surgical planning should consider the use of fixation plates and screws in case of mandibular fracture, the use of bone grafts and hemostatic materials (Carvalho & do Egito Vasconcelos, 2011). The use of bone grafting after exodontia was performed in three patients and Foam Gel in one patient (Kiran et al., 2014).

Postoperative complications are influenced according to the degree of difficulty due to tooth positioning, with alveolar osteitis, infection, bleeding, paresthesia, severe pain or edema, and more rarely mandibular fracture and iatrogenic displacements to adjacent anatomical regions being commonly reported in the literature (Bouloux, Steed, & Perciaccante, 2007; Brauer, 2009; Neto et al., 2012). Post-operative complications were present in 8% (3) cases, both of temporary paresthesia, being resolved in 3, 4 and 6 months, respectively.

The performance of the exodontia under local anesthesia or the performance under general anesthesia should be evaluated by the professional according to each case, considering the patient's age, the patient's collaboration and the surgical planning (Yanik, Ayranci, Isman, Buyukcikrikci, & Aras, 2017; Zerener, Bayar, Altug, & Kiran, 2016). The surgical procedure was performed under local anesthesia in 20 cases and under general anesthesia in four cases, demonstrating that it is a procedure, which when well planned, can be performed under local anesthesia.

The presence of a single or bilateral patient as mentioned above by the authors will influence the treatment plan, in bilateral cases the professional should evaluate the possibility

of performing the exodontia in a single session or in two, there was only one report of a patient who presented bilaterally that performed the exodontia in only one side and the conservative treatment in the other side (Nedjat-Shokouhi & Webb, 2014).

The conservative treatment together with the periodic control through radiographs is possible in some situations, as well as in everyday cases of impacted third molars, a controversial issue in the literature (Bagheri & Khan, 2007; Miclotte, Agbaje, Spaey, Legrand, & Politis, 2018; Osaki, Nomura, Hirota, & Yoneda, 1995; Steed, 2014), only one case was reported as an initial and definitive treatment option (Gonzalez-Perez et al., 2015).

The association of surgical treatment with orthodontics was reported in one of the four cases presented by Güven et al., an associated impaction of the first and second lower molars (Class I) and opted for exodontia only of the second molar with orthodontics in the first and third molars of the region (Güven et al., 2013). Only in one case was orthodontic treatment found, even though it is a viable form of treatment and when well planned and executed to be successful, some factors must be considered, such as the patient's age and the inclination of the teeth involved (Barros, Janson, Chiqueto, Ferreira, & Rosing, 2018), However, the authors of this review, after carefully evaluating the studies involved, do not consider the report presented by Barros et al, as a manifestation of true "kissing molars", being considered by the classification proposals, as a "pseudo kissing molars" due to the inclination of the teeth involved.

4. Conclusion

Therefore, even being a rare condition and a few manuscripts were found, this condition should be considered by oral surgeons, especially to get early diagnosis and then, reducing the complications. Also, it was suggested a complement in the classification and standardization of this clinical condition, to help in the planning and treatment.

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