Prevalence of oral mucocele in children and adolescents: systematic review and meta-

analysis

Prevalência de mucocele oral em crianças e adolescentes: revisão sistemática e metanálise

Prevalencia de mucocele oral en niños y adolescentes: revisión sistemática y metanálisis

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Abstract

Background. Mucocele is a benign and asymptomatic pseudo cystic lesion that develops secondarily to the leakage or retention of mucous material from the salivary glands, mainly from the minor salivary glands. Aim. The objective was to assess the available evidence related to the prevalence of oral mucoceles in children and adolescents. Design. Relevant articles were searched in electronic databases of PubMed via MEDLINE, Embase and Scopus. Two review authors independently assessed the risk of bias in the included articles and extracted data. Results. From 638 potentially eligible articles, fifteen relevant articles were included. The overall risk of bias was low for all the studies. All selected studies exhibited appropriate data and were included in the meta-analysis. Regarding publication bias, the statistical analysis did not identify asymmetry in the funnel graph (p=0.867). Conclusion. Considering the global analysis of the included studies, it was found that, the pooled prevalence of mucoceles was 1.77% (CI: 1.19% - 2.65%). It is suggested that future studies consider the classification of the particularities of each clinical presentation of patients seen in dental offices, in order to establish better morphological and anatomical definitions of the lesions concerning mucoceles. Studies that seek to standardize variables - related to age group, time interval, geographic region, genetic formation of the population, examined locations and classification of diseases into subgroups are also important. The certainty of the evidence was considered low.

Keywords: Mucocele; Pediatric dentistry; Children.

Resumo

Introdução: A mucocele é uma lesão pseudocística benigna e assintomática que se desenvolve secundariamente ao extravasamento ou retenção de material mucoso das glândulas salivares, principalmente das glândulas salivares menores. Objetivo: O objetivo foi avaliar as evidências disponíveis relacionadas à prevalência de mucoceles orais em crianças e adolescentes. Desenho: Os artigos relevantes foram pesquisados nas bases de dados eletrônicas do PubMed via MEDLINE, Embase e Scopus. Dois revisores avaliaram independentemente o risco de viés nos artigos incluídos e os dados extraídos. Resultados: De 638 artigos potencialmente elegíveis, quinze artigos relevantes foram incluídos. O risco geral de viés foi baixo para todos os estudos. Todos os estudos selecionados apresentaram dados apropriados e foram incluídos na meta-análise. Em relação ao viés de publicação, a análise estatística não identificou assimetria no gráfico de funil (p=0,867). Conclusão: Considerando a análise global dos estudos incluídos, verificou-se que a prevalência conjunta de mucoceles foi de 1,77% (IC: 1,19% - 2,65%). Sugere-se que estudos futuros considerem a classificação das particularidades de cada apresentação clínica dos pacientes atendidos em consultórios odontológicos, a fim de estabelecer melhores definições morfológicas e anatômicas das lesões referentes às mucoceles. Estudos que busquem padronizar variáveis - relacionadas à faixa etária, intervalo de tempo, região geográfica, formação genética da população, locais examinados e classificação das doenças em subgrupos - também são importantes. A certeza da evidência foi considerada baixa.

Palavras-chave: Mucocele; Odontopediatria; Crianças.

Resumen

Introducción: El mucocele es una lesión pseudoquística benigna y asintomática que se desarrolla secundaria a la extravasación o retención de material mucoso de las glándulas salivales, especialmente de las glándulas salivales menores. Objetivo: El objetivo fue evaluar la evidencia disponible relacionada con la prevalencia de mucoceles orales en niños y adolescentes. Diseño: Se realizaron búsquedas de artículos relevantes en las bases de datos electrónicas de PubMed a través de MEDLINE, Embase y Scopus. Dos revisores evaluaron de forma independiente el riesgo de sesgo en los artículos incluidos y los datos extraídos. Resultados: De 638 artículos potencialmente elegibles, se incluyeron quince artículos relevantes. El riesgo general de sesgo fue bajo para todos los estudios. Todos los estudios seleccionados presentaron datos apropiados y se incluyeron en el metanálisis. En cuanto al sesgo de publicación, el análisis estadístico no identificó asimetría en el funnel plot (p=0,867). Conclusión: Considerando el análisis global de los estudios incluidos, se encontró que la prevalencia combinada de mucoceles fue de 1,77% (IC: 1,19% - 2,65%). Se sugiere que futuros estudios consideren la clasificación de las particularidades de cada presentación clínica de los pacientes atendidos en los consultorios odontológicos, a fin de establecer mejores definiciones morfológicas y anatómicas de las lesiones relacionadas con los mucoceles. También son importantes los estudios que buscan estandarizar variables relacionadas con el grupo de edad, el intervalo de tiempo, la región geográfica, la composición genética de la población, los lugares examinados y la clasificación de enfermedades en subgrupos. La certeza de la evidencia se consideró baja. Palabras clave: Mucocele; Odontopediatría; Niños.

1. Introduction

The presentation of oral disease in pediatric patients is significantly different from that in adult patients. This difference can be explained by the cell renewal during the active growth process in the child and the variable profile of the immune cells. Mucocele is a benign and asymptomatic pseudocystic lesion that develops secondarily to the leakage or retention of mucous material from the salivary glands, mainly from the minor salivary glands. This leakage is usually caused by the rupture of the ducts or the presence of calculus (sialolith). Mucoceles can be congenital or appear immediately after birth, but are rare in children under one year of age (Bhargava, et al., 2014; Minguez-Martinez, et al., 2010). It is usually related to local trauma, although some cases do not report a history of trauma and are more frequent in children and young adults (Neville et al., 1998). The clinical presentation of oral mucoceles may vary depending on the depth of the lesion. Those located just below the mucosa present superficially, with the clinical features of a vesicle or blister; those located in the upper submucosa comprise the classic mucoceles, with the clinical characteristic of a nodule (Rashid et al., 2008).

Regarding the prevalence of mucoceles, there is a variation according to their location, but some authors claim that the lower lip is the most common place it takes place (Choi et al., 2019; Graillon et al., 2019), although they can also be found in the buccal mucosa, tongue and floor of the mouth (Laskaris, 2000; Adachi et al., 2011). Knowledge of its main clinical and

management characteristics is important to assist health professionals in clinical practice (Dhanuthai et al., 2007; Lapthanasupkul et al., 2015).

Currently, there is little literature on the extent and prevalence of oral diseases in children. The evidence found about this type of condition in children is often related to caries, periodontal disease, malocclusion and dental trauma, of an epidemiological nature

It is noteworthy that many studies indicate the prevalence of stomatological lesions in the oral cavity, such as mucoceles, but none of these studies is concerned with grouping these variables and presenting an overall prevalence of mucoceles in children. Therefore, the aim of the present study was to determine the prevalence of oral mucoceles in children and adolescents through a systematic review.

2. Methodology

Registration and Protocol

This systematic review was written following all the steps recommended by the PRISMA guide (Preferred Reporting Items for Systemic Reviews and Meta-Analyses). The protocol for this review is registered in the PROSPERO (International Prospective Register of systematic Review- CRD42021273324).

Eligibility criteria and studies selection

This review sought to answer the following guiding question: What is the prevalence of oral mucoceles in children and adolescents? The question was formulated according to the PEOS strategy, namely: P (population) = children and adolescents; E (exposure) = with oral mucocele; o (outcome) = prevalence; S (type of study) in observational studies.

After identifying the articles, they were exported to EndNote Basic (© 2015 THOMSON REUTERS), the manager's online version (ENDNOTE, 2015) to remove duplicates. Then, the studies were selected in two phases. In phase 1, three reviewers (FMF, GFCG, RPLOM), independently and in triplicate, analyzed titles and abstracts to identify eligible studies.

The selection of studies, however, was made with the agreement of the examiners. Any discrepancies were solved by group discussion and a fourth reviewer was involved where necessary (GT). Thus, titles and abstracts referring to (1) the presence of mucoceles were considered eligible; (2) in children and adolescents. In the second phase of the evaluation, the articles were read in full to verify if they contained the information of interest to carry out the systematic review. As exclusion criteria, the following items were considered: (1) not having data on the prevalence of mucoceles; (2) be a case report; (3) be a literature review study; (4) be a systematic review study; (5) be a personal opinion study; (6) be a differential diagnosis study.

Data and records were extracted and stored using a Microsoft® Excel Spreadsheet (Microsoft, Inc, Redmond, Wash., USA).

Sources and research strategies

A systematic literature research was carried out in three electronic databases: Pubmed/MEDLINE, Embase and Scopus until July 2021. The search was restricted to articles published in English, without restrictions from previous years and including as keywords: mucocele, dental care, pediatric dentistry. The search strategies are shown on table 1 below. In addition, the gray literature (OpenSigle/Opengrey) was examined to identify potentially eligible studies that might not have been identified during searches on conventional platforms. Afterwards, records were selected by discarding duplicate journals and articles that did not

include the keywords defined in the title or abstract. The bibliographic search was carried out for the Pubmed/MEDLINE database and adapted to the others.

Data collection process and collected items

The same three reviewers independently extracted data from the selected studies. For all included studies, the following information was recorded: title, authors, journal, database, type of study, country, age, sex, sample size, location and prevalence of mucocele.

Risk of bias in individual studies

To assess the risk of bias in the studies included in this review, two reviewers (FMF, GT) independently and in duplicate, considering the outcome analyzed, which is the prevalence of oral mucoceles in children and adolescents, adopted the JBI Critical Appraisal Checklist instrument for Studies Reporting Prevalence Data, prepared by the Joanna Briggs Institute (Mun et al., 2015). Reviewers scored articles that met the methodological criteria evaluated for the selection domains and outcome.

JBI Critical Appraisal Checklist for Studies Reporting Prevalence Data

The JBI - Critical Appraisal Checklist for Studies Reporting Prevalence Data - is a research tool that compiles critical questions of internal and external relevance that should be considered when analyzing prevalence data. The checklist can be used in different study modalities, surpassing only cross-sectional cuts and covering all studies that report prevalence data (Munn et al., 2014)."

The instrument described here is composed of nine items that deal with sample selection, characteristics of individuals, accounting for outcomes, data analysis and confounding factors. For each of these items, it is possible to mark "Yes", if the indicated criterion is adequately described in the study; or "No", in case the study does not meet the indicated criteria; there is also the possibility of the "Uncertain" option, when there is no information, or it is only partially described; or even "Not Applicable" (Munn et al., 2015). Therefore, this feature was applied to all studies listed in the review, regardless of their design. The evaluation criteria for the checklist relate the following aspects: guarantee of a representative sample; ensuring optimal recruitment; guarantee of adequate sampling; guarantee of coherent description and report of study, subject and configuration; ensuring that the scope of data from the identified sample is adequate; assurance that the condition was measured reliably and objectively; ensuring proper statistical analysis is performed; ensuring that confounding factors, subgroups and differences were identified and accounted for.

Summary of results

Initially, the I2 test was conducted to assess heterogeneity between studies. Meta-analyses were conducted using the random model and considering the prevalence of mucoceles as an outcome. Publication bias was checked by the Begg test or funnel plot when ten or more studies were available in the meta-analysis. In addition to this, sensitivity analysis was performed. All analyzes were performed using meta and metafor packages in RStudio, version 1.3.95. (PBC, Boston, USA). The proportion and the 95% confidence interval were calculated. The certainty of evidence was rated using the Ranking of Recommendations, Assessment, Development and Evaluation (GRADE) approach performed at https://www.gradepro.org/.

3. Results and Discussion

A systematic literature search identified 3385 possibly relevant studies, referring to 882 publications from PubMed/MEDLINE, 1076 from Scopus and 1427 from Embase. Among these, 1535 studies were duplicates and 1212 (65.51%) were not included (Kappa = 0.79 and 0.86). The main reason for not including the studies was because they were not about mucocele (84.98%) and were not in children or adolescents (15.01%). From this, 638 articles (34.48%) were analyzed in full, and 623 (97.64%) were excluded, as they did not report data on the prevalence of mucocele (39.32%), they were not a clinical study (14.44%), were not in English (4.01%), were inaccessible studies (12.35%) or had incomplete results (29.85%). Finally, 15 articles (2.35%) met the eligibility criteria and were included in this systematic review, as shown in Figure 1. All selected studies exhibited appropriate data and were included in the meta-analysis.



Figure 1. Study selection flowchart.

Source: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. PLoS Med 6(7): e1000097. 10.1371/journal.pmed1000097. Available on www.prisma-statement.org.

Characteristics of the included studies

The studies were published in 11 different journals, the oldest study was published in 2006, while the most recent in 2020. The designs of the included studies varied in retrospective, descriptive and cohort studies, with patients from various geographic contexts (South America, North America, Asia and Europe, with greater emphasis on Brazil, which makes up 40% of the studies). Regarding age group, only 01 study evaluated newborn children, the others evaluated a broader age group, including children and adolescents. In total, results were found from 276,712 biopsies performed, in which the prevalence of mucoceles in children in individual studies ranged from 0.4 to 11.5%. As for the region affected by mucocele, the lower lip was the most common place in the studies showed, being identified in 47% of the studies. Only 02 studies showed the characteristics of mucoceles, and their sizes ranged between 0.9 and 1.97 cm. The summary of the characteristics of the fifteen studies included is presented in Table 1.

Table 1. Details of the search in PubMed, Scopus and Embase databases.

Pubmed	((((((mucocele[MeSH Terms]) OR (mucoceles[MeSH Terms])) OR (mucoceles)) OR (mucocele)) AND
	((("dentistry"[MeSH Terms] OR "evidence based dentistry"[MeSH Terms] OR "pediatric dentistry"[MeSH Terms]
	OR "dental care for children" [MeSH Terms] OR ("paediatric dentistry" [All Fields] OR "pediatric dentistry" [MeSH
	Terms] OR ("pediatric"[All Fields] AND "dentistry"[All Fields]) OR "pediatric dentistry"[All Fields]) OR ("dental
	care"[MeSH Terms] OR ("dental"[All Fields] AND "care"[All Fields]) OR "dental care"[All Fields]) OR ("dental
	clinics"[MeSH Terms] OR ("dental"[All Fields] AND "clinics"[All Fields]) OR "dental clinics"[All Fields] OR
	("dental"[All Fields] AND "clinic"[All Fields]) OR "dental clinic"[All Fields]) OR (("mouth"[MeSH Terms] OR
	"mouth"[All Fields] OR "oral"[All Fields]) AND "care"[All Fields]) OR ("manage"[All Fields] OR "managed"[All
	Fields] OR "management s"[All Fields] OR "managements"[All Fields] OR "manager"[All Fields] OR "manager
	s"[All Fields] OR "managers"[All Fields] OR "manages"[All Fields] OR "managing"[All Fields] OR
	"management"[All Fields]) OR ("diagnosis"[MeSH Terms] OR "diagnosis"[All Fields] OR "diagnostic"[All
	Fields] OR "diagnostical"[All Fields] OR "diagnostically"[All Fields] OR "diagnostics"[All Fields]) OR
	("diagnosable"[All Fields] OR "diagnosi"[All Fields] OR "diagnosis"[MeSH Terms] OR "diagnosis"[All Fields]
	OR "diagnose" [All Fields] OR "diagnosed" [All Fields] OR "diagnoses" [All Fields] OR "diagnosing" [All Fields]
	OR "diagnosis"[MeSH Subheading]) OR ("epidemiology"[MeSH Subheading] OR "epidemiology"[All Fields] OR
	"prevalence"[All Fields] OR "prevalence"[MeSH Terms] OR "prevalence"[All Fields] OR "prevalences"[All
	Fields] OR "prevalence s"[All Fields] OR "prevalent"[All Fields] OR "prevalently"[All Fields] OR "prevalents"[All
	Fields]) OR ("risk factors"[MeSH Terms] OR ("risk"[All Fields] AND "factors"[All Fields]) OR "risk factors"[All
	Fields] OR ("risk"[All Fields] AND "factor"[All Fields]) OR "risk factor"[All Fields]) OR ("therapeutics"[MeSH
	Terms] OR "therapeutics"[All Fields] OR "treatments"[All Fields] OR "therapy"[MeSH Subheading] OR
	"therapy"[All Fields] OR "treatment"[All Fields] OR "treatment s"[All Fields]) OR ("epidemiology"[MeSH
	Subheading] OR "epidemiology"[All Fields] OR "incidence"[All Fields] OR "incidence"[MeSH Terms] OR
	"incidences"[All Fields] OR "incident"[All Fields] OR "incidents"[All Fields]) OR ("aetiologie"[All Fields] OR
	"aetiologies"[All Fields] OR "aetiology"[All Fields] OR "etiologies"[All Fields] OR "etiology"[MeSH
	Subheading] OR "etiology"[All Fields] OR "causality"[MeSH Terms] OR "causality"[All Fields]))))) AND
	((((((oral) OR (buccal)) OR (lip)) OR (mouth)) OR (labium)) OR (mucosa))
Scopus and Embase	(mucocele OR mucoceles) AND ("dentistry" OR "evidence-based dentistry" OR "pediatric dentistry" OR "dental
	care for children" OR "pediatric dentistry" OR "pediatric dentistry" OR "dental care" OR "management" OR
	"diagnosis" OR "prevalence" OR "risk factors" OR "therapeutics" OR "therapy" OR "treatment" OR "aetiology"
	OR "etiology") AND (oral OR buccal OR lip OR mouth OR labium OR mucosa)

Source: Own authorship.

Publication bias

Regarding publication bias, the statistical analysis did not identify asymmetry in the funnel graph (p=0.867), as shown in Figure 2.







Risk of bias of individual studies

It was found that 100% of the articles had a low risk of bias (Table 2).

Data summary

All 15 studies included in this review were grouped to perform the quantitative synthesis. The data were presented by the forest plot, which summarizes the results of the meta-analysis. As the value of the inconsistency test (I2) showed high heterogeneity between the outcomes, the random effect was adopted.

Considering the global analysis of the included studies, it was found that, as shown in Figure 3, the pooled prevalence of mucoceles was 1.77% (CI: 1.19% - 2.65%).

Sensitivity analysis was performed, however, no study seemed to strongly influence the results (figure 4). Low evidence certainty was judged according to the GRADE, indicating a limited confidence in the effect estimate and a certain degree of uncertainty in the findings.

			Table 2. (characteristics of	of the studies.			
Title	Source	Country	n	Gender	Age (Years)	Prevalence (%)	Place of mucocele	Characteristics of the lesion
An analysis of oral and maxillofacial pathology found in children over a 30-year period	Jones; Franklin (2006)	GB	53.666	M: 343 F: 388	0 to 16	1,4	-	-
A retrospective study of paediatric oral lesions from Thailand	Dhanutai et al. (2007)	TH	8.314	-	0 to 16	2,0	-	-
Retrospective review of pediatric oral lesions from a dental school biopsy service	Lima et al. (2008)	BR	9.465	-	0 to 14	1,1	-	-
Retrospective review of pediatric oral lesions from a dental school biopsy service	Shah et al. (2009)	US	75.215	-	0 to 16	1,5	Lower lip	-
Characteristics of the oral cavity of the newborns of Blumenau – SC	Schmitt et al. (2012)	BR	270	-	0 to 1	0,4	Lower lip	-
Paediatric oral pathology in a Chilean population: a 15-year review	Zuñiga et al. (2013)	CL	2.620	-	0 to 16	11,5	-	-
Retrospective study of biopsied oral and maxillofacial lesions in pediatric patients from Southern Taiwan	Lei et al. (2014)	TW	36.264	-	0 to 15	0,6	-	-
A multicenter retrospective cohort study on pediatric oral lesions	Martins-Filho et al. (2015)	BR	4.690	-	0 to 18	2,9	Lower lip	-
Retrospective analysis of 1286 oral and maxillofacial biopsied	Abdullah et al. (2016)	IQ	10.720	M: 26 F: 38	0 to 15	0,6	-	-
				0				

Table 2. Characteristics of the studies.

lesions of Iraqi children over a 30 years period								
Distribution of oral and maxillofacial lesions in pediatric patients from a Brazilian southeastern population	Ataíde et al. (2016)	BR	34.318	-	0 to 16	2,7	Lower lip	-
Epidemiological survey of mucus extravasation phenomenon at an oral pathology referral center during a 43 year period	Bezerra et al. (2016)	BR	11.589	-	0 to 20	3,7	Lower lip	Nodular lesion, smooth surface, same color, 0.9 cm mucosa
Oral and maxillofacial lesions in children and adolescents	Santos et al. (2018)	BR	4.492	-	0 to 19	3,5	Lower lip	Size: 1,97cm
Paediatric oral pathology in Thailand: a 15-year retrospective review from a medical teaching hospital	Taweevisit et al. (2018)	TH	3.887	M: 31 F: 56	0 to 16	2,2	Lip, tongue and mucosa	-
Analysis of oral pathology in an Australian paediatric population: A retrospective study	Bardwhaj; Prabhu (2019)	AU	15.691	M: 83 F: 90	0 to 18	1,1	Lip and mouth floor	-
A retrospective 11-year study on lip lesions attended at an oral diagnostic service	Barros et al. (2020)	BR	5.511	-		1,4	Lower lip	-

Remarks: GB = Great Britain, TH = Thailand, BR = Brazil, US = United States, CL = Chile, TW = Taiwan, IQ = Iraq, AU = Australia. Source: own authorship



Figure 3. Meta-analysis of the pooled prevalence of mucoceles in children and adolescents.

Source: own authorship.

Figure 4. Sensitivity analysis.

Study	Events per 100 observations	Events	95%-CI	12
Omitting Barros CC et al., 2020	+	1.71	[1.66; 1.76]	99.00
Omitting Taweevisit M et al., 2018	+	1.70	[1.65; 1.75]	99.00
Omitting Ataíde AP et al., 2016	+	1.56	[1.51; 1.61]	99.00
Omitting Bezerra TM et al., 2016		1.62	[1.57; 1.67]	99.00
Omitting Martins-Filho PR et al., 2015	+	1.68	[1.64; 1.73]	99.00
Omitting Zuñiga MD et al., 2013	+		[1.56; 1.66]	
Omitting Shah SK et al., 2009	+		[1.72; 1.84]	
Omitting Lima Gda S et al., 2008	+	1.73	[1.68; 1.78]	99.00
Omitting Dhanuthai K et al., 2007		1.70	[1.65; 1.75]	99.00
Omitting Jones AV et al., 2006	+	1.78	[1.72; 1.83]	99.00
Omitting Abdullah BH et al., 2016	+		[1.70; 1.80]	
Omitting Bhardwaj, V & Prabhu, N., 2019	+		[1.69; 1.79]	
Omitting Xavier Santos RL et al., 2018	+		[1.63; 1.72]	
Omitting Lei F et al., 2014	i -	1.87	[1.82; 1.93]	99.00
Omitting Schmitt BHE et al., 2012	+	1.71	[1.66; 1.75]	99.00
Fixed effect model	· · · · · · · · · · · ·	1.70	[1.66; 1.75]	
	-1.5 -1 -0.5 0 0.5 1 1.5			

Source: own authorship.

This is the first systematic review and meta-analysis to present the global prevalence of mucoceles in children and adolescents, based on previously published and planned methodological criteria. Overall, 15 observational studies (14 retrospective cohorts and 1 cross-sectional study) were included, totaling 276,712 biopsies from different socioeconomic and geographic settings (7 Brazilian studies, 2 Thai studies, 1 Iraqi study, 1 Chilean study, 1 US study, 1 British study, 1 Australian

study and 1 Taiwanese study). The overall prevalence of mucocele observed in children and adolescents, using the random effect model, was 1.77% (95% CI 1.19 to 2.65%; I2 = 99%, n = 276,712 biopsied samples), with a low risk of bias associated with all primary studies included. Furthermore, in relation to the place in which manifestations linked to mucoceles are identified, it was noticed that the lower lip is more frequently affected (reported in 7 of the 15 primary studies included). In addition, it was noted that two studies evaluated characteristics of elementary lesions undergoing biopsy, such as size, appearance, morphology and associated signs.

Recently, the assessment of multiple oral health conditions in pediatric and juvenile populations has become essential, especially in developed countries or countries that have an integrative national health system and well-established oral health policies. In this context, the assessment of epidemiological indicators of oral pathologies, especially mucocele, is essential in order to satisfactorily achieve development and rational planning of therapeutic, diagnostic and diagnostic measures related to this disease.

Notoriously, the prevalence of mucoceles observed in children and adolescents is low, considering the wide spectrum of oral diseases reported in the population evaluated. Among the oral pathologies most commonly found in children and adolescents, the high prevalence of cavitation (caries lesions), malocclusions, gingival alterations and hyperdontia can be highlighted (Kassebaum et al., 2015). Other oral conditions and oral pathologies are more commonly reported in health institutions when compared to mucoceles, such as temporomandibular disorders (prevalence between 7.3% to 30.4%) and dental trauma (17.5%), which complement the wide spectrum of conditions that affect children and adolescents (Christidis et al., 2019). Any persistent edema of unknown etiology in the salivary glands requires urgent investigation to rule out the possibility of benign or malignant neoplasia (Jones, Franklin, 2006).

In patients with erosive lichen planus there is a possibility that the inflammatory process may play a role in the pathogenesis of recurrent mucoceles, as the lymphocytic infiltrate can block the accessory gland duct, which would induce its rupture and subepithelial mucus leakage (Bermejo et al., 1999). In addition, smaller lesions bring less discomfort to the patient because they are more superficial, located in layers of tissue that are less vascularized and less rich in nerve structures, whereas large lesions can impair speech or chewing.

As there are no painful symptoms, it is usually the professional who detects the lesion in a routine oral exam. However, even with a low prevalence of mucoceles in children and adolescents worldwide, it is essential to establish an early and accurate diagnosis guided by a properly qualified dental surgeon, in order to avoid possible complications related to this disorder of the salivary glands.

The vast majority of oral diseases are confined to the oral tissues, but several underlying systemic conditions can present with signs and symptoms in the oral cavity. It is known that incidences of pathological conditions of the mouth and perioral structures are common in the oral cavity of children (Welbury et al., 2012). It is also noteworthy that benign and malignant neoplasms of various origins can develop in oral structures, in addition, the process of proliferation in the oral cavity has been an interesting field of study, as they can have a reactionary or neoplastic nature (Bahadure et al., 2012).

There are reports that benign lesions of the salivary gland can lead to retention or extravasation of mucus. Unlike the salivary duct cyst, the mucocele is not a true cyst as it does not have an epithelial lining (Neville et al., 1998; Sugerman et al., 2000).

The vast majority of mucoceles (85% to 95%) are due to extravasation, in addition, approximately 5% are due to mucus retention (Saza et al., 1982; Javali, Bhagwati, 2016).

The factors presented above are also highlighted by authors who show that in a series of Brazilian cases of 104 patients, 50% were under 20 years of age and 34.6% were under 15 years of age (Nico et al., 2008)

These factors are common in children and young adults, because younger people are more likely to experience trauma that induces mucin shedding (Neville et al., 1998; Shafer et al., 1987).

However, mucoceles have been reported in patients of all ages, including newborns and the elderly, in addition to a higher incidence also found in females, a fact that goes against studies that report no sex predilection (Harrison, 1975; Bodner 1991; Bermejo et al., 2013)

A retrospective study published in 2014, which included 56 hospital patients admitted between 2010 and 2011 showed that the prevalence of oral mucoceles was significantly higher in patients aged 15 to 24 years (65.52%), with additionally presenting irregularity of manifestation between different genres (More et al., 2014).

In most cases, minor salivary gland mucoceles are located in the lower lip mucosa, buccal mucosa, soft palate, and in the retromolar trigone region (Sugermann et al., 2000; Regesi et al., 2008; Bermejo t al., 2013). Furthermore, despite its wide distribution, the prevalence depends on its location: lower lip mucoceles are the most common, followed by lingual mucoceles and floor of mouth mucoceles (Neville et al., 1998; Garcia 2012; Shafer et al., 1987).

However, the occurrence of these nodular lesions is expected to be present in any region of the oral mucosa surface surrounding the accessory salivary glands. A common example of this varied distribution is predicted in multiple manuscripts previously published in the scientific literature, which over-described prevalence of exophytic lesions on the tongue, soft palate/mouth floor, and/or with unknown distribution (Gupta et al., 2007).

Among the included studies, it was found that most of the prevalence of mucoceles in children and adolescents were related to the brazilian scenario (7 studies). Only two studies reported in a detailed and systematic way the elementary characteristics of the oral lesions identified in pediatric and adolescent patients evaluated in the included observational studies. However, when present, the anatomopathological characterizations of oral mucocele lesions were reported as nodular, smooth-surfaced, homogeneously colored lesions, measuring between 0.9 and 1.97 cm.

As an additional description applicable to future studies on the subject, the existence of an exophytic lesion with a symmetrical pink color and with regular edges and slightly solid texture should be mentioned. Furthermore, during physical assessment, it is noticed that the nodules are not bleachable during application of local pressure (Botticelli et al., 2021).

As secondary outcomes and correlated to the morpho pathological characterization of these lesions, it is also important to assess trans luminescence during application of direct light, to differentiate superficial mucoceles (translucent bluish coloration) from deep mucoceles (pink coloration due to local angiogenic processes) (Adachi et al., 2011). Among the main limitations present in this study, we emphasize primarily that articles from other languages and inaccessible articles were not included, which may have limited the number of articles selected. In addition, the absence of critical and relevant primary and secondary outcomes for patients, such as the elementary characterization of the lesions presented by patients, recurrence rate of oral lesions after excision, complications related to dental treatment and multivariable correlations between socioeconomic, racial and prognosis profiles of patients, makes the subject still susceptible to multiple investigative scientific evaluations.

4. Conclusion

Thus, this study contributes to the literature on the manifestations of oral lesions such as mucoceles in children and adolescents, briefly identifying aspects that still need to be analyzed in successive studies. It is suggested that future studies consider the classification of the particularities of each clinical presentation of patients seen in dental offices, in order to establish better morphological and anatomical definitions of the lesions concerning mucoceles. In addition, studies that seek to standardize variables - related to age group, time interval, geographic region, genetic formation of the population, examined locations and classification of diseases into subgroups - are also important. Considering the global analysis of the included studies, it was found that, the pooled prevalence of mucoceles was 1.77% (CI: 1.19% - 2.65%).

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