

Functional oral self-mutilation in children and adolescents: a scoping review

Automutilação oral funcional entre crianças e adolescentes: uma revisão de escopo

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ABSTRACT

Objective: To present characteristics of publications in the worldwide scientific literature produced on the topic of functional oral self-mutilation in children and adolescents. *Methods:* This study is a scoping review. The search was made in the following databases: MEDLINE/PubMed, BVS/LILACS, Web of Science and PsycNet. We used the Kappa test for the agreement analysis. The data were analyzed through descriptive statistics presenting both absolute and relative frequencies. *Results:* 18 articles have been selected, all of which were published in English between the years of 1968 and 2017 in 4 different continents. Most of them were case reports and were conducted in hospitals and universities involved male children. The research area with the highest number of studies was Dentistry, followed by Medicine and Psychology, with clinical examinations as their most utilized diagnostic strategy. *Conclusions:* The literature on the subject is still quite scarce and has a low level of scientific evidence, which highlights the need for studies with high levels of scientific evidence in order to reach a better understanding of matters that are inherent to this scoping review's topic.

Keywords: child, adolescent, self-mutilation, self-injurious behavior, nonsuicidal self-injury.

RESUMO

Objetivo: Apresentar as características das publicações científica produzidas na literatura mundial sobre o tema da automutilação oral funcional em crianças e adolescentes. *Métodos:* Trata-se de uma revisão de escopo. A busca foi feita nas seguintes bases de dados: MEDLINE/PubMed, BVS/LILACS, Web of Science e PsycNet. Foi usado o teste Kappa para a análise de concordância. Os dados foram analisados por meio de estatística descritiva apresentando frequências absolutas e relativas. *Resultados:* Foram selecionados 18 artigos, todos publicados em inglês entre os anos de 1968 e 2017 em 4 continentes diferentes. A maioria dos artigos tratavam-se de relatos de casos e foram realizados em hospitais e universidades envolvendo crianças do sexo masculino. A área de pesquisa com maior número de estudos foi a Odontologia, seguida da Medicina e da Psicologia, sendo o exame clínico a estratégia diagnóstica mais utilizada. *Conclusões:* A literatura sobre o tema ainda é bastante escassa e com baixo nível de evidência científica, o que evidencia a necessidade de estudos com alto nível de evidência científica para melhor compreensão dos assuntos inerentes ao tema desta revisão de escopo.

Palavras-chave: criança, adolescente, automutilação, comportamento autolesivo, autolesão não suicida.

1 INTRODUCTION

Self-mutilation is defined as deliberate damage that is inflicted upon one's own body without suicidal intentions^{1, 2}. Oral and perioral structures may be traumatized by this sort of behavior, which involves the biting of lips, the jugal mucosa^{1, 3} and the tongue's lateral surfaces^{1, 4} as well as the autoextraction of teeth⁵.

Lesions occur mainly due to the introduction of fingernails or foreign objects, such as pencils or pens, into the oral cavity or due to biting the oral tissues. Clinically, they manifest in the form of swelling or ulcers coupled with gum recession. Thus they appear in the form of factitious ulcers, factitious gingivitis, factitious periodontitis and autoextraction of teeth^{6, 7, 2, 8}.

Self-mutilation can affect all types of individuals, regardless of age, gender, socioeconomic status or level of education^{9, 10, 11, 12}. However, a higher incidence of self-inflicted lesions has been reported among females and adolescents^{13, 6, 12}. And gingival oral self-mutilation is most commonly observed among girls up to 12 years of age^{6, 2}.

With respect to the possible etiology of self-mutilation, it can be broadly classified into two categories: organic and functional^{10, 12, 14}. Self-mutilation is said to be organic when it affects comatose patients, patients who have genetic syndromes, biochemical or enzymatic deficiencies, patients with congenital insensitivity to pain, as well as patients who may suffer from a heterogeneous group of neurological disorders and other alterations, such as autism, mental retardation, hereditary sensory neuropathies, congenital infectious diseases and epilepsy^{12, 15, 16}.

Functional self-mutilation, on the other hand, is found in physically healthy patients without detectable syndromes or genetic alterations¹². The lesions are consciously provoked and are a response to certain stimuli. In other words, they may either be used to draw attention, as a method of manipulation, as a means of asking for help, of gaining control over a situation or even as a way to pass the time in cases of social isolation^{15, 16}.

In these cases an emotional component such as parental divorce, school problems, the birth or death of a sibling, or an unhappy family environment may be present¹² and self-mutilation is known to be performed by patients without any associated comorbidities¹⁴.

Functional self-mutilation can be further classified into 3 subcategories: (1) lesions that are superimposed on a pre-existing one; (2) lesions that are secondary to a destructive habit; (3) lesions of unknown or complex etiology, often including a psychological component¹².

This study's aim is to map the literature on the topic of functional oral self-mutilation in children and adolescents and provide an opportunity to identify types and sources of evidence and gaps in research.

2 METHODS

This is a scoping review that used the Population, Concept and Context (PCC) strategy to orient its data collection. This methodology helps identify the key topics: Population, Concept, and Context and was therefore adopted to delineate the scoping review's research question. A *Scoping Review* study, according to the review method proposed by the Joanna Briggs Institute (JBI) (2015)¹⁷, allows one to map key concepts, clarify areas of research and identify knowledge gaps.

In this review, the Population was represented by children and adolescents, the Concept encompassed oral self-mutilation in cases where either an emotional factor or factors of an emotional nature were present, and the Context did not apply in this case. Thus, reconciling the key topics of the PCC with the aims of this review, the research question was constituted as: "What are the characteristics of the publications in the scientific literature on functional oral self-mutilation in children and adolescents?"

2.1 PUBLICATION SEARCH METHODS

The search for the scientific publications was conducted in periodicals indexed in the MEDLINE/PubMed, BVS/ LILACS, Web of Science, and PsycNet databases. The search carried out in August 2021 used the keywords: "child", "children", "adolescent", "teenager", "oral self-mutilation" – in Portuguese, the terms "*automutilação*" and "*comportamento de automutilação*" were used. The inclusion criteria for the articles were as follows: oral self-mutilation when a psychological factor is the cause of the behavior, in all languages. The exclusion criteria were: oral self-mutilation that is resulting from the following factors: 1. comatose patients; 2. genetic syndromes; 3. biochemical or enzymatic deficiencies; 4. neurological disorders (autism, mental retardation, hereditary sensory neuropathies, congenital infectious diseases and epilepsy); cases in which oral self-mutilation is due to cultural practices; suicide; articles that were inaccessible.

2.2 SELECTION CRITERIA AND PROCEDURES

The included articles were separated according to each database and categorized in a Microsoft Excel database using the items: 'Author', 'Year of publication', 'Title' and 'Abstract'. The selection conducted by means of reading the titles and abstracts aimed to verify whether the articles corresponded to the research question or not, and it was carried out independently by two researchers who performed an initial calibration. Data were processed using *SPSS software*, version 25. The Kappa test ($p=0.933$) was used for the agreement analysis.

The disagreements were discussed and analyzed. The complete reading of the titles and abstracts by both raters obtained the Kappa value of $p=0.905$ (*almostperfect*).

Finally, the pre-selected articles were read in their entirety with the aim of identifying their relevance to the research more accurately and determining whether the inclusion and exclusion criteria were being considered.

2.3 PROCEDURE FOR DATA EXTRACTION AND SUMMARIZATION

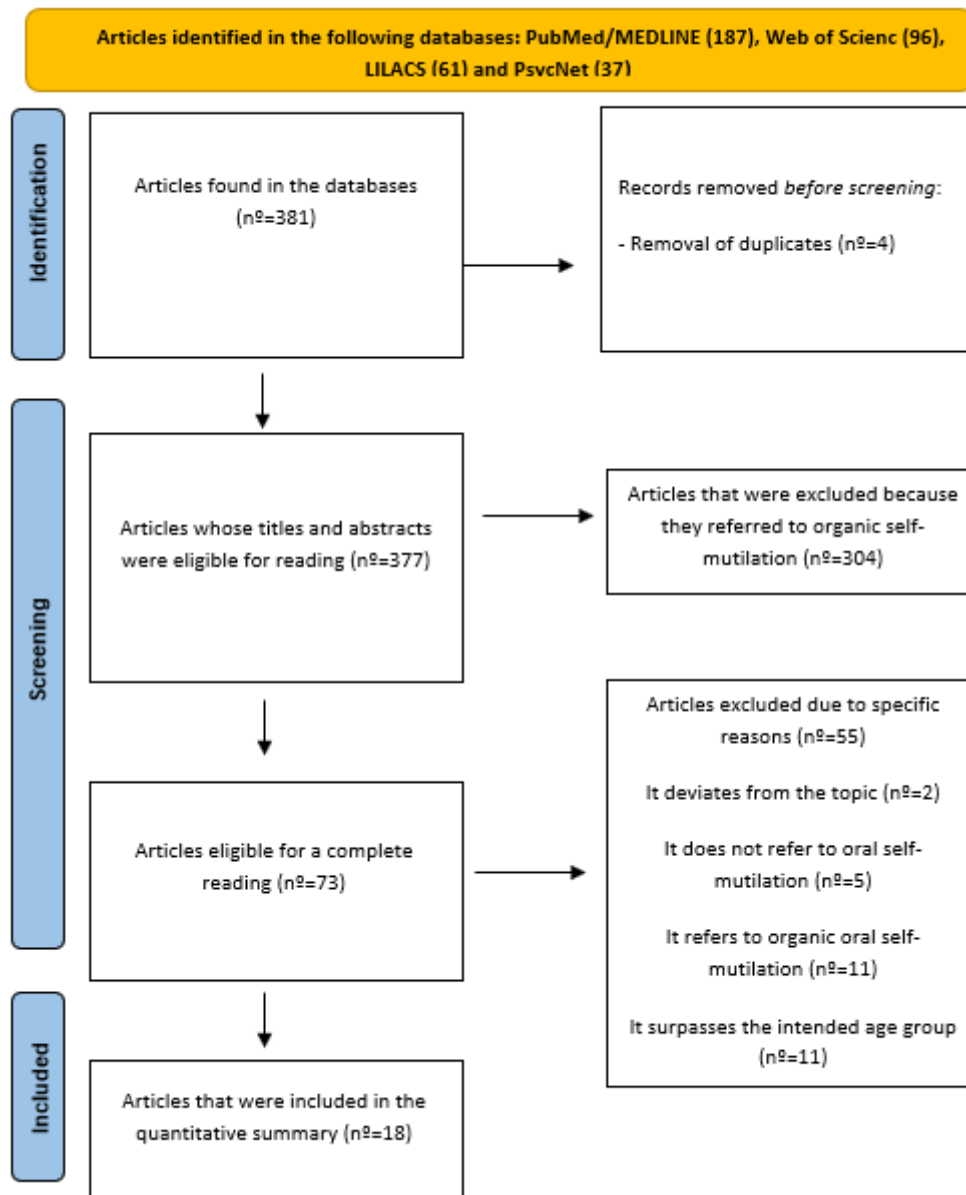
In this last step, the most relevant data concerning the scope of the topic were extracted for further processing and data analysis. Data extraction was performed by a single reviewer through feeding the database. It was constituted in Microsoft Excel as a table, according to the following data: 1. identification number; 2. authors; 3. year of publication; 4. database; 5. title; 6. abstract; 7. type of self-mutilation; 8. classification; 9. language; 10. country; 11. population; 12. age group; 13. gender; 14. sample size; 15. study plan; 16. site; 17. area of concentration; 18. instrument used; 19. main results.

The identification number was registered sequentially according to the order of data collection (Pubmed, Web of Science, Lilacs, PsycNet) and the reading of titles and abstracts in a Microsoft Excel database. The Degree of Recommendation and the Level of Evidence were analyzed and classified according to Phillips et al.³⁵. The data were analyzed by means of descriptive statistics, showing both absolute and relative frequencies. The Kappa test was used to determine the agreement among the raters. *SPSS software*, version 25, was used for the analyses.

3 RESULTS

Eighteen articles have been selected through the search process presented in the PRISMA flowchart (Figure 1).

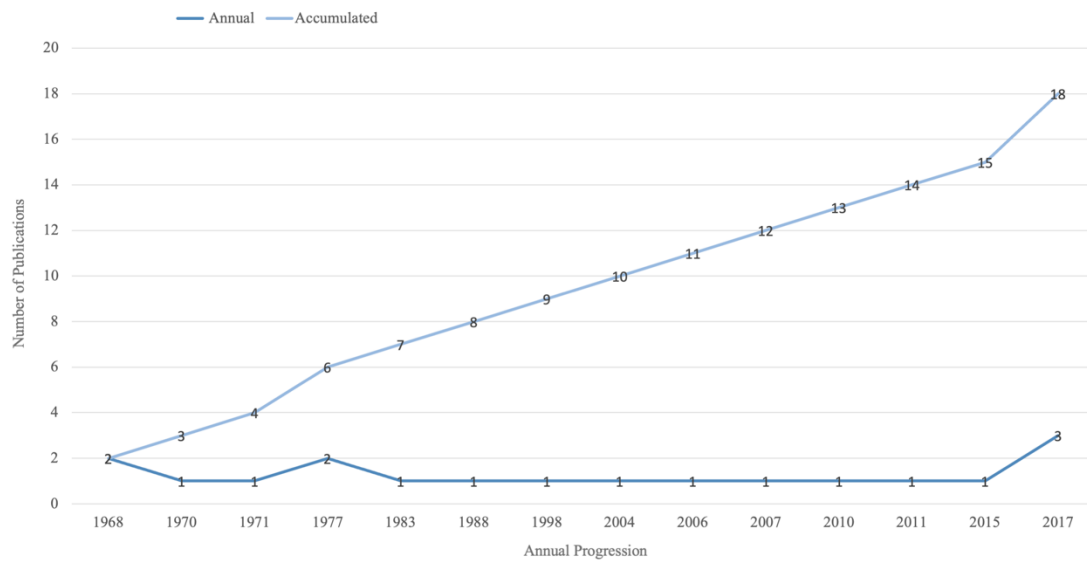
Figure 1: Diagram of the process of inclusion and exclusion of studies, according to Preferred Reporting Items for Systematic Review and Meta-Analyses (Prisma 2020)18.



Source: authors

Language-wise, all the selected articles (18) were published in English. Regarding the year of publication, it was found that between 1968 and 2017, 1 to 3 articles were published in different years (Figure 2).

Figure 2: Distribution of scientific publications on functional oral self-mutilation in children and adolescents between 1968 and 2017.



Source: authors

In Table 1, we find the general information of the articles included in the scoping review^{19, 20, 21, 12, 22, 23, 24, 25, 26, 27, 2, 28, 29, 30, 31, 32, 33, 34}.

Table 1: General characteristics of the selected articles (n°=18)

Characteristics	n°	(%)
Study Plan		
Case-Control	2	(11.11)
Case Report	14	(77.78)
Cohort	1	(5.56)
Case Series	1	(5.56)
Study Site		
School	2	(11.11)
Hospital	7	(38.89)
School Clinic (Universities)	7	(38.89)
Private Clinic	1	(5.56)
School and Orphanage	1	(5.56)
Sample Size		
≤100	15	(83.33) ¹
101 - 500	1	(5.56)
>1000	2	(11.11)
Age Group		
Children	8	(44.44)
Adolescents	7	(38.89)
Both	3	(16.67)

Diagnostic Strategy for functional oral self-mutilation		
Clinical Examination	9	(50.00)
Clinical Examination and questionnaire	1	(5.56)
Clinical and Histopathological Examination	4	(22.22)
Clinical and Psychological Examination	1	(5.56)
Clinical and Radiographic Examination	1	(5.56)
Reports from Family Members	1	(5.56)
Observation	1	(5.56)
Continent		
The Americas	2	(18.18)
Europe	5	(45.45)
Africa	1	(9.09)
Asia	3	(27.27)
Area of Research		
Dentistry	16	(88.89)
Medicine	1	(5.56)
Psychology	1	(5.56)

¹Comprises from 1 to 17 clinical case reports
Source: authors

As for the country of publication, England stood out in this sample with 4 publications over the years, followed by the United States with 3 published papers, Brazil and Denmark with 2 papers, and other countries (Saudi Arabia, South Korea, Germany, Turkey, South Africa, India, and Croatia) with 1 production on the subject.

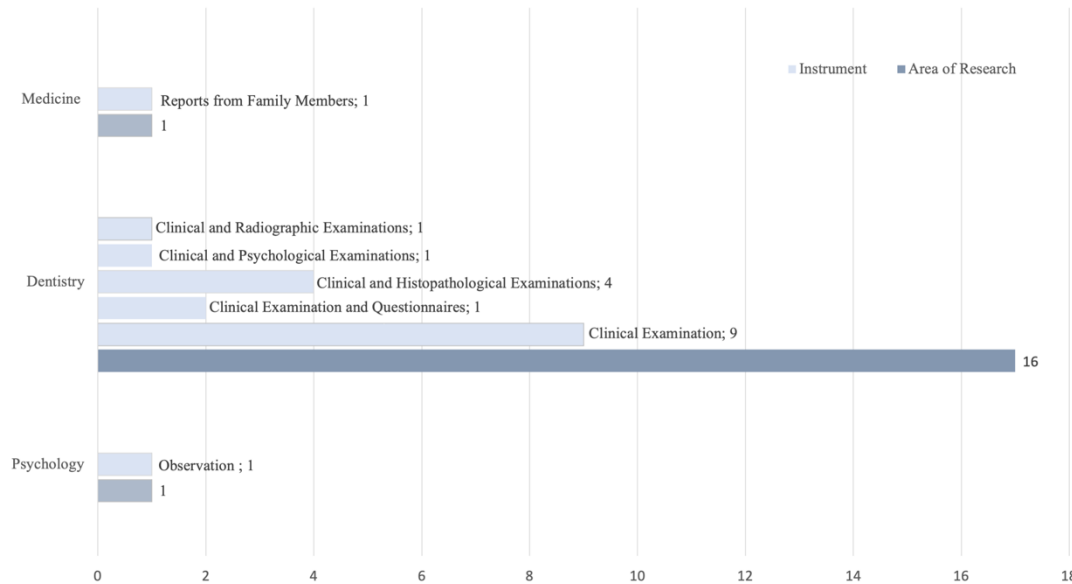
The age range and gender of the population of the studies included in the scoping review were also analyzed. Thus, more studies with children than with adolescents were conducted (Table 1). Regarding the sex distribution of the studies, (38.89%) were male; (27.78%) were female; (27.78%) were studies that involved components of both sexes; and (5.56%) did not report the sex of the individual involved in the study.

From observing these studies' plan and the composition of their samples, it seems that the observational studies had their sample sizes defined as: 1 case control (n°=180); 1 case series (n°=17); 1 prospective case control (n°= 1255); 1 cohort (n°= 2377).

When relating the area of knowledge with the instrument used for assessing the issue, it has been found that in the area of Dentistry, clinical examination was the most used type of diagnostic instrument, followed by clinical and histopathological examination, clinical examination and questionnaire, clinical and psychological examination, and clinical and radiographic examination. In the area of Medicine, in this sample, the diagnostic tool was "reports from family members" – a result that proved similar to the area of Psychology, which used the criterion "observation" to diagnose functional oral self-mutilation in children and

adolescents with the habit of compulsively biting their lips. The relation between the data referring to each area of research, as well as the instrument used for the diagnosis, is described in Figure 3.

Figure 3: Relation between each area of research and the instrument used for the diagnosis of functional oral self-mutilation.



Source: authors

The articles were submitted to analysis in terms of quality and classified by their Level of Scientific Evidence (LE), according to the classification developed by the *Oxford Centre for Evidence-Based Medicine*³⁵. Thus, according to the analysis of the articles, most of the studies (78.95%) were classified as LE C. It is important to highlight that the most frequent type of study among those included in this scoping review was that of case reports (78.95%). Regarding the Grade of Recommendation, one study had GR A; two articles had GR B; and most of the studies (78.95%) had GR C. Table 2 presents the complete result of the quality analysis and the study types of the included articles.

Table 2: Levels of Scientific Evidence and Grades of Recommendation of the articles, according to the *Oxford Centre for Evidence-based Medicine*³⁵.

Types of Study											
LE	SRRCT	RCT/CS	CS	SRCS	CS	ES	SRCCS	CCS	CR	EO	TOTAL
1A	0	0	0	0	0	0	0	0	0	0	0
1B	0	1	0	0	0	0	0	0	0	0	1
1C	0	0	0	0	0	0	0	0	0	0	0
2A	0	0	0	0	0	0	0	0	0	0	0
2B	0	0	0	0	0	0	0	0	0	0	0
2C	0	0	0	0	0	0	0	0	0	0	0
3	0	0	0	0	0	0	0	0	0	0	0
3B	0	0	0	0	0	0	0	2	0	0	2
4	0	0	0	0	0	0	0	0	15	0	15
5	0	0	0	0	0	0	0	0	0	0	0
TOTAL LE	0	1	0	0	0	0	0	2	15	0	18
TOTAL GR	1			2			15				
	A			B			C				

LE: Level of Evidence; GR: Grade of Recommendation; SRRCT: Systematic Review of Randomized Clinical Trials; RCT/CS: Randomized Clinical Trials/Validated Cohort Studies; CS: Case Series; SRCS: Systematic Review of Cohort Studies; CS: Cohort Studies; ES: Ecological Studies; SRCCS: Systematic Review of Case-Control Studies; CCS: Case-Control Studies; CR: Case Reports; EO: Expert Opinion.

Source: authors

4 DISCUSSION

The first point that must be analyzed concerns the low scientific productivity over the years on the topic of functional oral self-mutilation in children and adolescents, despite its relevant social importance. Based on the data of this research, there seems to be a slight tendency towards the increase in the number of works in this area in the last decade, although this is inconsistent data. This may reflect the low quality level of the studies, thereby showing the lack of an appropriate instrument to assess the idiopathic cases of oral self-mutilation, and consequently, of methodologically standardized study plans.

Another fact that is pertinent to this discussion concerns the language of publication used, considering that out of all the articles included in the scoping review, (100%) of them were published in English. This could be attributed to two factors: if we observe the geographical distribution of the countries that have conducted the studies, we can verify that most of them are English-speaking countries – despite the fact that Brazil, a Portuguese-speaking country, stands out in this list. However, this is justified by a global trend that seeks to establish English as an international language in science³⁶, since papers published in this language are more cited and therefore have a greater scientific reach, which enables them to disseminate knowledge on a larger scale.

Oral self-mutilation is not uncommon. A literature search conducted in the PubMed database using the term “oral self-mutilation” found 296 articles¹². However, only 14 of these

articles described cases of functional oral self-mutilation in pediatric patients. In this scoping review, as we have expanded the databases and population, 381 articles were found. Of these, (4.72%) dealt with functional oral self-mutilation in children and adolescents.

Regarding its prevalence, self-mutilation can affect all types of people^{9, 10, 12}, but adolescents are the most affected by it^{13, 37, 11}. In cases of oral self-mutilation the gingival area is the most frequently affected one and this is usually the case with female children¹². In this regard, the present review found that more studies were developed with children than with adolescents; studies which only involved children in their study population represented (44.44%), while more studies were developed with males (38.89%) compared to those involving females or individuals from both sexes.

Most of the literature on the subject of oral self-mutilation published in English comes in the form of case reports, which hinders the diagnosis and consequently the elimination of other pathological diseases, as well as the selection of therapeutic options²². This finding corroborates with our result, since 15 (78.95%) of the papers had a study plan that referred either to case reports or case series.

As we looked into the literature in the search for instruments that have been used to assess this scoping review's topic, we found that the clinical picture of a lesion caused by cheek bites, for example, was remotely described in the years 1962 and 1964 in the form of a case report^{38, 39}. Conversely, Hjrting-Hansen and Holst evaluated oral self-mutilation lesions by bites on the oral mucosa from the standpoint of the association between clinical and histopathological examinations²⁷. Hildebrand et al.¹² established a diagnosis of functional oral self-mutilation after an intraoral clinical examination and a complete interview with the patient and her parents. Depending on the case and the area of research, the approach to diagnosis may differ. In fact, there is no consensus in the literature on which instrument should be used to assess oral self-mutilation in children and adolescents.

The focus of this review, which is the topic of functional oral self-mutilation, tends to be pertinent to Dentistry, given its specificity. However, other areas of research also tend to address this issue. In this regard, for example, Marneros et al.²⁵ have documented in a Clinical Psychiatry journal a case of penis and tongue self-amputation in an adolescent following the use of natural drugs – a medical approach that also involves functional oral self-mutilation. Also, based on observation, which is a tool widely used by Psychology, Mestre et al.⁴⁰ emphasize that the importance of observation and systematic recording of behavior lies in how much society can benefit from a thorough description of a given behavior, since it allows professionals from different areas to discuss an observed phenomenon using its main

characteristics as their basis. One example of this would be the elaboration of the Diagnostic and Statistical Manual of Mental Disorders (DSM), created by the North American Psychiatric Association in 1952 and now updated into DSM V – 2013⁴¹. Using this method, Lyon²³ has reported a successful case of psychological therapy for chronic lip-biting in an adolescent girl using behavioral techniques.

Moreover, one of the difficulties in studying this topic is found in the heterogeneity of terminology in the literature^{42, 43}, as there are several terms used as synonyms for self-injurious conduct, for instance: self-injury, self-damage, self-mutilation, self-harm, cutting, caving, parasuicide, self-lesioning behavior without suicidal intent, suicidal behavior^{44, 43}.

5 CONCLUSION

The selected studies have led us to the conclusion that the literature on functional oral self-mutilation in children and adolescents is still scarce. Most studies on this topic are case reports, and therefore have a low level of scientific evidence. The literature also does not present a specific instrument to help with the diagnosis. Since the 1960s until the present day, clinical examination associated with a medical history has been considered the main strategy used for cases of functional oral self-mutilation. Finally, studies with a high level of scientific evidence are necessary to better understand issues that are inherent to the topic of this scoping review.

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