Systematic review

Prevalence of oral disease and treatment types proposed to children affected by Autistic Spectrum Disorder in Pediatric Dentistry: a Systematic Review

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Abstract

Purpose. To investigate the prevalence of dental caries and periodontal disease in children with ASD, and to analyse the necessity of treatment and the prevalence of using general anaesthesia in order to perform it.

Materials and Methods. A search was performed covering the last 10 years utilising the following databases: Pubmed, Scopus, Medline, BASE, Science Citation Index, Science Direct, Web of Science. Four reviewers evaluated each study. Review findings were summarised using the PRISMA Statement for reporting. Thirteen articles were included in this systematic review.

Results. When analysing the articles selected, the evidence turned out did not show a common DFMT and dmft for the groups of children affected by Autism Spectrum Disorder considered. When compared to group of unaffected children, groups of ASD children not always showed a higher prevalence of caries but always higher Periodontal Indexes (PI and GI), resulting in higher prevalence of periodontal disease. Where the treatment was performed and taken into consideration, there was a high incidence of necessity of General Anaesthesia due to the lack of collaboration of the children.

Conclusion. The high prevalence of treatment under general anaesthesia and the often-reported negative behaviour evidence how there is a lack of protocols specifically designed for these patients, in order to better improve their collaboration and subsequently their oral health and so additional strategies for a preventive care should be applied for these patients. Clin Ter 2020; 171 (3):e275-282. doi: 10.7417/CT.2020.2226

Key words: pediatric dentistry, autistic spectrum disorder, dental approach, dental treatment

Introduction

Autism spectrum disorder (ASD) is a complex developmental condition that involves persistent challenges in social interaction, speech and nonverbal communication, and restricted/repetitive behaviour. The effects of ASD and the severity of symptoms differ from person to person (1).

ASD is usually first diagnosed in early childhood (around 2-3-year-old) with many of the most-obvious symptoms. Some children, however, develop normally until toddlerhood and then stop acquiring skills or lose previously gained ones (2). Furthermore, autism is 3 to 4 times more recurrent if compared to 30 years ago. According to the National Center on Birth Defects and Developmental Disabilities, CDC, one in 59 children is estimated to suffer from autism and it is also three to four times more common in boys than in girls, as the latter, with ASD, exhibit less obvious signs compared to boys (3). A report from the World Health Organization stated that while some people with ASD can live independently, others have severe disabilities and require life-long care and support. Evidence-based psychosocial interventions, such as behavioural treatment and parent skills training programmes, can reduce difficulties in communication and social behaviour, with a positive impact on well-being and quality of life for people with ASD and their caregivers. Interventions for people with ASD need to be accompanied by broader actions for making physical, social and attitudinal environments more accessible, inclusive and supportive (18) Globally, access to services and support for people with ASD is inadequate. That is why this work is focused on the dental treatment of autistic children, in order to point out how difficult is for professionals and families to find the best way to do it and give innovative solutions to that.

Normal daily activities and/or activities such as tooth brushing and getting dressed as well as understanding the behaviour of other individuals, constitute a constant challenge also for parents.

Dental care, for patients with such disorders, requires special procedures and adaptations. It has been proved, that some patients with moderate or slight intellectual disorders can be successfully treated through paying particular attention to the procedures even if they adapt slowly and with difficulty to them (6). Autistic children, if compared to other patients with psychiatric disorders, show a greater obstacle to deal with, which is anxiety. A dental surgery is full of potential frightful stimuli, due to the peculiar environment and equipment on show (12). This may result in uncooperative behaviour such as crying or physical and verbal

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aggression so ad to avoid treatment. In these children, it is often more difficult to interpret, beforehand, the signs of fear and anxiety and thereby to prevent the loss of cooperation, but it is possible to carry out dental care when such anxiety signs are recognized and duly considered. People affected by intellectual disorders have significantly more untreated caries and a higher prevalence of periodontitis and gingivitis compared to the average population (14,17).

The aim of this systematic review was to investigate the prevalence of dental caries and periodontal disease in individuals with ASD, taking into consideration children and young adults. Moreover, the goal is to analyse the necessity of treatment and the prevalence of using general anaesthesia in order to perform it.

Strategy of research

The question of research

The systematic review of literature was performed identifying the main question of the research through which the current literature is investigated: Which is the incidence of caries and periodontal disease in autistic children and how the prevalent treatment is performed? How are behaviour and collaboration evaluated?

PICOs method

Thereafter, in order to more clearly identify the main topics on which concentrate the research on the databases, the above-mentioned question of research was defined through PICOs method. This is one of the methods through which a clinical question can be correctly formulated and derives from the Evidence Based Medicine (EBM) approach, used to write systematic reviews. According to this model, in creating the question, the totality of the following aspects should be considered:

- **P**: Population-Problem (e.g. Age, Gender, Ethnic group);
- I: Intervention (exposition to a risk factor, risky behaviour, prognostic factor);
- C: Control-Comparison (e.g. a placebo);
- **O**: Outcomes (risk of disease, accuracy of the diagnosis, occurrence tax of adverse condition);
- S: Study design.

Criteria of inclusion and exclusion

After the research question was defined according to PICOs method, inclusion and exclusion criteria were identified, in order to outline the research area of the systematic review.

The inclusion criteria defined were:

- a) Articles in English;
- b) Availability of full text;
- c) Children population from 0 to 18 suffering from ASD;
- d) Articles using DMFT as index of prevalence of caries and treatment;
- e) Articles analysing periodontal status of the patients;
- a) Articles analysing how the treatment is performed;
- g) Articles until 10 years back (2008-2019).

The exclusion criteria defined were:

- a) Absence of English language;
- b) No full text available;
- Adult population and children population not affected by ASD;
- d) Articles not relevant to the research question;
- e) Articles older than 10 years.

Keywords and research string

The keywords that have been used to properly investigate the subject of the research were: "Autism spectrum disorder", "Autistic Disorder", "Autism", "Dental care", "Oral health Management", "Oral hygiene", "Dental treatment", "dmf", "DMFT".

The first group of words, "Autism spectrum disorder", "Autistic Disorder", "Autism", regards the population to which the research is aimed. They were used with the Boolean operator OR in order to specifically indicate the problematic researched.

The second group of words, "Dental care", "Oral health Management", "Oral hygiene", "Dental treatment", regards the scientific area investigated and the type of intervention on the population.

Furthermore, a third group of words was added to specifically identify the articles containing the data concerning "dmf" and "DMFT".

In the end, the three groups were connected with the Boolean operator AND in order to produce the final search string: ("Autism spectrum disorder" OR "Autistic Disorder" OR "Autism") AND ("Dental care" OR "Oral health Management" OR "Oral hygiene" OR "Dental treatment") AND ("dmf" OR "DMFT").

Literature review

Literature review was made in the period between December 2018 and April 2019.

The databased used were Pubmed, Scopus, Medline, BASE, Science Citation Index, Science Direct, Web of Science.

The articles that came out were 69. After the DMFT OR dmf filter, the articles found became 21.

Selection of the articles and PRISMA method

What is PRISMA method?

An accurate writing of a systematic review is based on a standardized work protocol: in this review PRISMA (Preferred Reporting Items for Systematic reviews and Meta-Analyses) method has been used.

This method was used to select the studies to be included in the systematic review; both PRISMA statement and PRISMA flow chart have been used.

The first one includes a check-list of essential items to be taken into consideration in order to get a methodological precision in the systematic review and reduce the risk of bias, while the second one is useful to map the number of the articles identified through four phases: identification, screening, eligibility and inclusion.

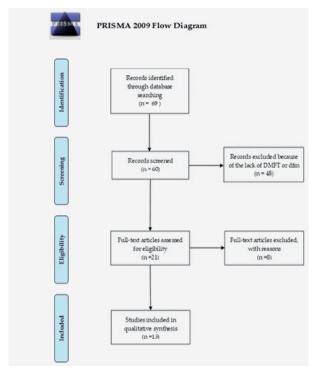


Fig. 1. Prisma Flow Chart

The whole process of selection and inclusion in the systematic review has been carried out by the author GZ and DC and is represented by the flow-chart below (Fig. 1).

The first phase of the process, that corresponds to identification, consists in identifying the final search string and to its application to the databases above mentioned inserting the several methodological filters and following the inclusion/ exclusion criteria.

In this first phase 69 articles were identified. In the second phase, the one of screening, after reading the title of the 69 articles identified, 9 articles were excluded (60 articles remaining) and after reading the abstracts 48 more were excluded because they didn't satisfy the inclusion criteria regarding the DMFT or dmf, identifying in the end **21 relevant articles**. In the third phase, the one of eligibility, 8 more articles were excluded because either the sample was not homogeneous either the articles were already systematic reviews. The 13 articles remaining were carefully read and evaluated by the author of the systematic review in order to verify the effective pertinence to the question of research and to the inclusion and exclusion criteria. The final phase of inclusion is still composed by 13 articles, after they have been evaluated to be pertinent to the topic after reading the full text.

Articles examined and results

The 13 articles examined, and part of the review, are summarized in the following Table 1.

When analysing the articles selected, the evidence turned out did not show a common DFMT for the groups of children affected by Autism Spectrum Disorder considered. When compared to group of unaffected children, groups of ASD children not always showed a higher incidence of caries, sometimes it was higher, (1,4,11,20) sometimes lower (7,9,13,22).

Furthermore, because of the variety and not homogeneity of the samples it was not possible to obtain a common DMFT, but what we can say is that in many samples it was shown to be increasing with age (7,10,21,22).

Figure 2 shows how various was the incidence of caries found in literature and, when available, a comparison with non-affected group part of the trials analysed.

The same evidence found for the DMFT can be shown comparing the dmft of the samples taken into consideration in the articles.

No common dmft was found, nor higher incidence in ASD group was demonstrated when compared to unaffected

Again, it was demonstrated to be increasing with age.

Figure 3 shows the distribution of dmft in the articles analysed in the review.

On the other side, what is clear in examining the articles in this review, was that the periodontal status in children affected by Autism Spectrum Disorder, was always pathologic

When compared to healthy children, children affected by Autism were demonstrated to have higher Periodontal Indexes (PI and GI), resulting in higher incidence of periodontal disease (1,7,22).

Figure 4 clearly shows the comparison between the groups.

Even when not compared to healthy children, in the literature analysed children affected by autism showed low levels of Oral Hygiene, high prevalence of gingivitis and high Simplified Oral Hygiene Index (OHI-S) (10,15,16).

Figure 5 illustrates the number of articles over the total analysed that report this data: just 2 articles over 12 did not take into consideration the level of Oral Hygiene (4,7); one over 12 showed an average Oral Hygiene Index of (2.19 21); all the others reported high periodontal indexes, gingivitis or high OHI-S.

Another important information that needs to be taken into consideration in this analysis is how all the treatment in this study groups were performed.

It is widely known how difficult can be collaborating with an Autistic child on the dental chair in order to perform even a simple treatment or just an examination; and this is confirmed by the high percentage of treatments that needed to be performed under general anaesthesia in the articles examined.

In some of the articles it was just taken into consideration a first examination, but in four of them, where the treatment was performed and taken into consideration, there was a high incidence of necessity of General Anaesthesia due to the lack of collaboration of the children (7,10,13,21).

"In a 10-year follow up, patients resisted efforts to establish personal contact with dental staff. Furthermore, sedative techniques were ineffective because of atypical response patterns. The use of general anaesthesia was the only solution to provide necessary dental care" (16).

Figure 6 shows the percentage of treatment performed Under General Anaesthesia in the articles analysed.

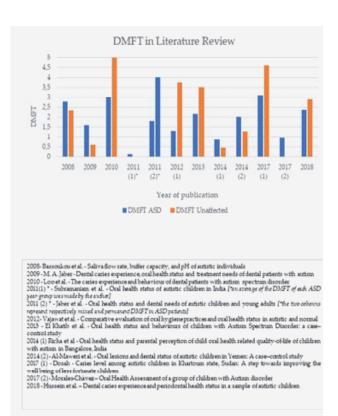
Finally, another important factor to be taken into consideration, is the key of this study: the behaviour.

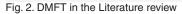
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Table 1. Articles examined in the Literature review

Article	Sample	dmf	DMFT	Periodontal status	Treatment	Cooperation
Bassoukou et al Saliva flow rate, buffer capacity, and pH of autistic individuals(2008) ⁴	25 ASD boys 25 unaff. boys	1.67 (±2.89) mean dmft ASD 1.75 (±2.87) mean dfmt unaffected	primary dentition 2.00±2.83 ASD 1.79±3.07unaff. mixed/permanent 2.00±2.20 ASD 3.00±3.10unaff. → 2.77±3.25 ASD →2.33±2.89 unaff	Not available	Not available	17 out of 25 ASD used drugs to control symptoms of behaviour
M. A. Jaber - Dental caries experience, oral health status and treatment needs of dental pa- tients with autism (2009) ¹¹	61 ASD patients 61unaffected patients	ASD median dmft 0.80+-0.2 unaffected median dmft 0.30+-0.3	ASD median dmft 1.6+0.64 unaffected median DMFT 0.6+0.29	ASD 3.3%b good OH 59% poor OH 37.8% fair OH 97%gingiv. 59% unaffected good OH	Restorative ASD 9.2% (30% UGA) 26.9% unaff.	10 year follow-up, pt. no established contact with the dental staff. Sedative techniques were ineffective. Use of GA was the only solution.
Loo et al The caries experience and behaviour of dental patients with autism spectrum disorder(2010) ¹³	395 ASD children 386 unaffec- ted children	Not available	ASD median DMFT=3 unaffected group median DMFT=5	ASD patients have a lower hygiene level when compared to patients without autism.	UGA: - 37.2% ASD - 29.8% unaff. Restorative or surgical treat.: -ASD 51,6% -Unaff. 38,1%	Cooperation (Frankl scale) ++ : ASD 9.2%; Unaff. 46.6% -/:ASD55,20%; Unaff.25,40%
Subramaniam et al Oral health status of autistic children in India(2011) ²¹	106 ASD children	4-5 y: 2.33±3.57 6-8 y: 1.77±2.43 9-12 y: 1.47±2.09 >13 y: 0.29±1.07	4-5 y: 0.08+-0.28 6-8y: 0.06+-0.36 9-12y: 0.33+-0.69 >13y: 0.1+-0.58	Average Oral Hygiene Index 2.19	6 pt. UGA 5 pt. sedation 27 no treat. for lack of collaboration	Cooperation (Frankl) +: 34.9% -/: 65%
Jaber et al Oral health status and dental needs of autistic children and young adults(2011) ¹⁰	61 autistic children	2.2 ± 1.77 increasing with age	1.8 ± 1.67 mixed 4.0 ± 1.44 permanent	100% had gingivitis poor OH 59% fair OH 37.8% good OH 3.2%	GA required 30% of the time	5 excluded because uncooperative. 10 year follow-up, pt. no established contact with the dental staff. Sedati- ve techniques were ineffective. Use of GA was the only solution.
Vajawat et al. – Comparative evalua- tion of oral hygiene practi- ces and oral health status in autistic and normal indivi- duals (2012) ²²	117 autistic patients 126 unaffected individuals	Not avai- lable	ASD median DMFT= 1.2966 unaffected median DMFT=3.736 increasing with age-	PI: ASD= 1.3039 Unaff.= 1.0015 GI: ASD =1.0015 Unaff.= 1.0015 PERIO DISEASE: ASD >>> Unaff.	Not available	Not available
Chadha et al Dental Survey of Institutionalized Children with Autistic Disorder (2012) ¹⁵	35 ASD children	Not avai- lable	range from 0 to 6 DMFT mean deft 6.4	OHIS poor 14 fair 14 good 7	Not available	Uncooperative children were excluded from the study
El Khatib et al Oral health status and behaviours of children with Autism Spectrum Disorder: a case—control study (2013) ⁷	100 ASD children 100 unaffec- ted children	ASD: 3.532±4.57 Unaff.: 3.562±3.86	ASD: Mixed 0.93± 1.58 Perm. 3.4±4.54 Unaffected: Mixed 0.79±1.26 Perm. 3.50±3.63"	PI: ASD = 2.02±0.73 Unaff.= 1.40 ±0.80 GI: ASD = 2.00±0.73 Unaff.= 1.40 ±0.80	GA: ASD = 39,4% Unaff. = 1% Sedation: ASD = 24,5% Unaff. = 6,2% TSD: ASD = 30,9% Unaff.=85,6%	Cooperation (Frankl) ++:ASD14%;Unaff.48% +: ASD 38%;Unaff.39% -:ASD 37%;Unaff. 11% : ASD 11%;Unaff. 2%
Richa et al - Oral health status and parental perception of child oral health related quality-of-life of children with autism in Bangalore, India(2014) ²⁰	135 ASD children 135 unaffec- ted children	ASD: 1.40±2.48 Unaff.: 0.46 ± 0.58;	ASD: 0.86 ± 1.22 Unaff.: 0.46±1.06	OHI-S highly superior in ASD ASD 2.07 ±0.83 Unaff. 0.59± 1.28	Not available	Not available

Al-Maweri et al Oral lesions and dental status of autistic children in Yemen: A case–control study (2014)¹	42 ASD children 84 unaffected children	ASD = 5.23±2.34 Unaffected = 4.06±2.98 increasing with age	Caries prevalence: ASD: 100% Unaff.: 90.5% DMFT ASD = 2.00±2.18 Unaff.=1.27±1.77	PI: ASD = 1.5 Unaffected = 1.05 GI: ASD = 1.37 Unaffected = 1.02	Not available	Uncooperative children were excluded from the study
Dosah - Caries level among autistic children in Khartoum state, Sudan: A step towards improving the well being of less fortunate children (2017) ⁷	44 ASD children 88 unaffected children	Not avai- lable	ASD = 3.09 Unaffected=4.59	Not available	Not available	Not available
12. Morales-Chávez – Oral Health Assessment of a group of children with Autism disorder(2017) ¹⁶	96 ASD children	2.41	0.96	OHI-S=3.4 83.3% with gingivitis 59.41% with calculus	Significant % of the autistic no collabor, UGA.	"A significant percentage of the autistic patients do not collaborate with the treatment and so it becomes necessary to put them UGA"
Hussein et al. – Dental caries experience and periodontal health status in a sample of autistic children (2018) ⁹	44 ASD children 44 unaffected children	ASD = 11.57 ±11.41 Unaffected = 15.36± 12.81	ASD = 2.36±3.86 Unaff.=2.89±3.78	~ 100% ASD gingivitis, a smaller % in the unaff. group OHI-S: ASD=1.69±0.806 Unaff.=1.291±0.89	Not available	Not available





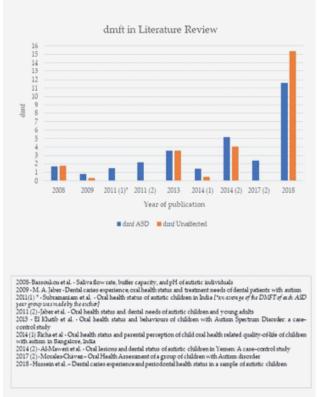


Fig. 3. dmft in the Literature review

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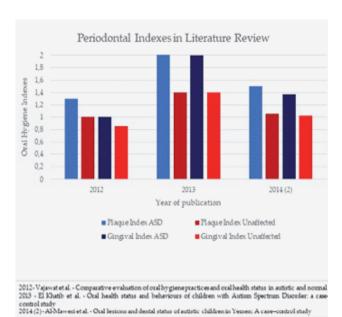


Fig. 4. periodontal indexes in the Literature review

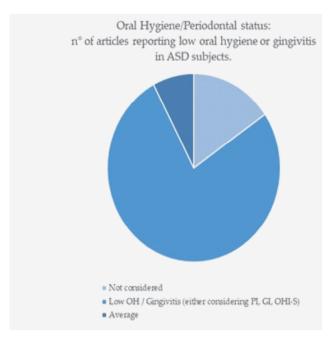


Fig. 5. periodontal status in the Literature review

It has been already mentioned how difficult is to perform a dental treatment on autistic children and how the chosen solution is often the general anaesthesia. This happens because the characteristic behaviour of an autistic child is different from the one of the other children, it lacks collaboration, of interaction with the operator.

Some authors in the articles included in this review have reported the behaviour of autistic children examined, using the Frankl scale (5), that divides the grades of behaviour in 4 classes (Table 2).

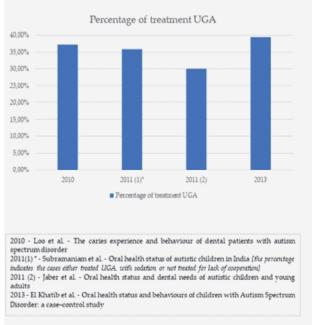


Fig. 6. percentage of treatment UGS in the Literature review

Table 2. Frankl behavioural rating scale

1		Definitely negative. Refusal of treatment, forceful crying, fearfulness, or any other overt evidence of extreme negativism.
2	-	Negative. Reluctance to accept treatment, uncooperative, some evidence of negative attitude but not pronounced (sullen, withdrawn).
3	+	Positive. Acceptance of treatment; cautious behaviour at times; willingness to comply with the dentist, at times with reservation, but patient follows the dentist's directions cooperatively.
4	++	Definitely positive. Good rapport with the dentist, interest in the dental procedures, laughter and enjoyment.

As predictable, Subramaniam et al. (21) reported a negative or definitely negative behaviour in 65% of the cases with ASD, Loo et al. (13) reported 55.6% of cases with negative or definitely negative behaviour over the total ASD children examined and El Khatib et al. (8) a 48% of cases in the same class of behaviour.

Even when the authors did not use the Frankl scale to report the behaviour, a lack of collaboration and cooperation that brought the subject either to be treated under general anaesthesia or to be excluded from the study, was always mentioned and remarked (1, 10, 11, 15, 16).

Discussion

Many patients with ASD show significant difficulty in daily life activities relative to their cognitive skills (23); the developmental deficit of executive functioning in autism is highly diversified with huge individual variation (24).

For this kind of subjects, the possibility of being treated with the Occupational therapy approach during childhood represents a better chance for a better social life, avoiding difficulties in work activities and potential psychiatric comorbidities (25).

Many children have a family history of autoimmune diseases (psoriasis, rheumatoid arthritis, celiac disease, Takayasu's arteritis), allergies and food intolerances, suggesting an involvement of the immune system in autism etiopathogenesis (26). Children and adolescents diagnosed with (ASD) are thought to be more vulnerable to oral diseases. This is due to their increased barriers to dental care services (27).

To summarize the analysis above reported, we can say that Autism spectrum disorder strongly affects oral health status and from the studies analysed, a clear high incidence of periodontal disease, but also a high caries incidence, was pointed out.

This is more likely due to the difficulty that children affected by Autism Spectrum Disorder have to maintain a good oral hygiene and to be periodically examined and eventually treated.

The high prevalence of treatment under general anaesthesia and the often-reported negative behaviour evidence how there is a lack of protocols specifically designed for these patients, in order to better improve their collaboration and subsequently their oral health.

Conclusions

According to recent guidelines for oral health prevention in childhood, individual additional strategies for a preventive care should be applied for these patients (19). All the health providers, family and caregivers should be involved with the goal of being aware, informed and motivated on oral health issues, and a better access system to the dental care structure, both logistic, professional and economical should be assured (28-29).

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