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REVIEW ARTICLE

A systematic review of population-based gingival health studies among children and adolescents with autism spectrum disorder

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KEYWORDS

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Abstract The prevalence of autism spectrum disorder (ASD) is close to 1% in the United States of America and other countries**.** Special attention should be given to oral health in individuals with ASD as they are often affected by oral diseases. However, gingival health in children with ASD and adolescents is controversial in terms of the severity of disease and number of people affected.

Aim: To conduct a systematic review and meta-analysis to assess the gingival health status of children and adolescents with ASD.

Methods: The search was conducted using eight databases for articles that met the inclusion and exclusion criteria. This search produced 742 relevant papers, but only five with sufficient data on gingival and plaque indices were eligible for inclusion in this systematic review and meta-analysis.

Results: The homogeneity of the sample was tested using the Cohen Q test, which identified significant heterogeneity ($P < 0.0001$), indicating the use of the random effect's standard mean difference. Significantly higher gingival index and plaque index values were found in children and adolescents with ASD than in children without ASD.

Conclusion: Individuals with ASD need help and better access to oral healthcare. Further investigation is needed with regard to gingival health in individuals with ASD and caries risk assessment to understand how this disorder affects oral health. A standardized index for gingival health will help in the inclusion of more studies to assess gingival health in children and adolescents with ASD.

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Contents

1. Introduction

Autism, autistic disorders, Asperger's syndrome, and pervasive developmental disorders not otherwise specified comprise a heterogeneous group of neurodevelopmental disorders known as autism spectrum disorders (ASDs). ASD is not a disease but a syndrome with multiple interacting causes, both genetic (Folstein and Rosen-Sheidley, 2001; Bayou et al., 2008; Boyd et al., 2010; Landrigan, 2010) and non-genetic (Hamilton and Ventura, 2006). ASD is defined behaviorally as a syndrome involving the abnormal development of social and language skills, poor muscle tone, poor coordination, and visual and hearing impairment in some cases (American Psychiatric Association, 2013). In general, patients with ASD are irresponsive to environmental conditions and are inflexible to change. Self-injury, depression, anxiety, and unpredictable behaviors are some of the common symptoms. Children with ASD are reported to be more selective in their food and tactile sensitivity than unaffected children (Valicenti-McDermott et al., 2006).

The number of reported cases of ASD has increased in the last few years (Rapin, 1997), most likely due to changes in recent developments in diagnostic methods, awareness, referrals, and available facilities (Centers for Disease Control and Prevention, 2011). The prevalence of ASDs has reached 1% in the United States of America and similar values in other countries (American Psychiatric Association, 2013). It affects male individuals four times more often than female individuals and occurs in families of varying ethnic backgrounds and social classes (American Psychiatric Association, 2013).

While several research studies have been conducted from a medical point of view, studies on the oral health and dental needs of children with ASD have been insufficient. Few studies have discussed the dental needs and oral health status of individuals with ASD (Jaber, 2011; Kopycka-Kedzierawski and Auinger, 2008; Marshall et al., 2010). It has been reported that inconsistencies in communication and behavior have been reported to lead to deterioration of oral health, such as poor oral hygiene, increased caries risk, and different degrees of gingivitis (Fakroon et al., 2015; Jaber, 2011). Furthermore, some prescribed medications used to modify the behavior of individuals with ASD may have adverse side effects on oral and gingival health (e.g., gingival enlargement and xerostomia) (Murshid, 2014; Hafez et al., 2016). Studies also show that poor oral hygiene, periodontal diseases, bruxism, and tongue thrusting found in children with ASD may be caused by parental negligence, brought on by the difficulty of caring for children with ASD (El Khatib et al., 2014; Fakroon et al., 2015; Jaber, 2011).

A thorough search of the relevant literature shows limited studies describing the gingival condition in individuals with ASD. Some studies have reported differences in the prevalence of gingivitis compared to that in non-autistic individuals (DeMattei et al., 2007; Kopycka-Kedzierawski and Auinger, 2008; Stein et al., 2012), while other studies have reported no significant differences (Marshall et al., 2010; Loo et al., 2008; Kuter and Guler, 2019). The controversy in these findings demonstrates the need to conduct a comprehensive literature search to evaluate the gingival health status among a population diagnosed with ASDs. The collected information would provide a baseline for dental professionals to enable them to provide the necessary preventive measures and effective treatment. The aim of this study was to conduct a systematic review and *meta*-analysis to assess the prevalence of periodontal diseases in children and adolescents with autism compared to that in non-autistic individuals.

2. Methods

A comprehensive literature search was conducted using eight databases, systematically searching for qualified abstracts, posters from conference proceedings, or articles published in a peer-reviewed journal that reported and discussed the prevalence of gingival health status in children and young adults diagnosed with ASD. The eight databases were MEDLINE, PubMed (www.pubmed.gov), the Cochrane Library (https://www.cochranelibrary.com), Web of Science (http://www.isiknowledge.com), Controlled-Trial Database (http://controlled-trial.com), Clinical Trials-US National Institutes of Health (http://www.clinicaltrials.gov), National Institute for Health and Clinical Excellence (http://www.nice.org.uk), Virtual Health Library (https://bvsalud.org), and Saudi Digital Library (https://sdl.edu.sa/sdlportal/en/publishers.aspx).

The search terms used included autism, autistic, Asperger, ASD, pervasive, and pervasive developmental disorders in all combinations with the terms oral health, gingival health status, and gingival condition in all epidemiological studies (cross-sectional, case control, cohort, and clinical trials).

During the first stage, relevant publications were selected by topic, title, and abstract, (n = 742). All selected papers were screened, and whether they met the following inclusion criteria was assessed:

Table 1 Data of included studies.

Study, Author, Year	Country of Origin	N	Age (years)
Oral health status and behaviors of children with autism spectrum disorder: A case-control study. El Khatib et al., 2014	Alexandria, Egypt	Autistic: 100 Control: 100	3–13
Oral health among preschool children with autism spectrum disorders: A case-control study. Du et al., 2015	Hong Kong, China	Autistic: 347 Control: 347	2–7
Oral lesions and dental status of autistic children in Yemen: A case-control study. Al-Maweri et al., 2014	Sana'a, Yamen	Autistic: 42 Control: 84	5–16
Comparison of Gingival Health and Salivary Parameters among Autistic and Non-Autistic School Children in Riyadh. Diab et al., 2016	Riyadh, Saudi Arabia	Autistic: 50 Control: 50	4–15
Evaluation of oral health status and influential factors in children with autism. Onol et al., 2017	Isparta, Turkey	Autistic: 63 Control: 111	6–14

Table 2 Meta-Analysis for the outcome variables: Gingival Index and Plaque Index.

Gingival Index

Study	Autistic		Control		SMD	95% CI	t-value	p-value	Weight (%)	
	N1	Mean (SD)	N2	Mean(SD)					Fixed	Random
El Khatib et al., 2014	100	2(0.73)	100	1.4(0.8)	0.781	0.492 to 1.069			15.53	20.14
Du et al., 2015	347	0.37(0.29)	347	0.51(0.27)	−0.499	−0.650 to −0.348			55.95	20.58
Al-Maweri et al., 2014	42	1.36(0.84)	84	1.02(0.51)	0.529	0.152 to 0.907			9.12	19.74
Diab et al., 2016	50	1.82(0.65)	50	1.35(0.86)	0.608	0.205 to 1.011			8.05	19.61
Onol et al., 2017	63	1.91(0.56)	111	1.22(0.54)	1.255	0.918 to 1.592			11.37	19.93
Total (fixed effects)	602		692		0.0818	−0.0312 to0.195	1.420	0.156	100.00	100.00
Total (random effects)	602		692		0.528	−0.222 to 1.279	1.381	0.168	100.00	100.00
Test for heterogeneity: Q = 139.14;df = 4;p < 0.0001; I^2 = 97.13% (95% CI: 95.28% to 98.25%)										

Plaque Index

Study	Autistic		Control		SMD	95% CI	t-value	p-value	Weight (%)	
	N1	Mean (SD)	N2	Mean(SD)					Fixed	Random
El Khatib et al., 2014	100	2(0.73)	100	1.4(0.8)	0.807	0.518 to 1.096			15.78	20.13
Du et al., 2015	347	0.45(0.24)	347	0.6(0.27)	−0.587	−0.739 to −0.435			56.48	20.44
Al-Maweri et al., 2014	42	1.5(0.81)	84	1.05(0.51)	0.715	0.333 to 1.098			9.08	19.82
Diab et al., 2016	50	1.92(0.36)	50	1.44(0.43)	1.209	0.780 to 1.638			7.26	19.64
Onol et al., 2017	63	2.06(0.73)	111	1.24(0.54)	1.327	0.987 to 1.667			11.40	19.97
Total (fixed effects)	602		692		0.0999	−0.0143 to0.214	1.716	0.086	100.00	100.00
Total (random effects)	602		692		0.687	−0.201 to 1.574	1.518	0.129	100.00	100.00
Test for heterogeneity: Q = 188.96;df = 4;p < 0.0001; I^2 = 97.88% (95% CI: 96.68% to 98.65%)										

- Reporting gingival health status of individuals with ASD measured based on plaque index and gingival indices (cross-sectional, case control, cohort, and clinical trials).
- All age ranges especially children and adolescents.
- Both sexes.
- Written in English language.

Duplicate articles were excluded from the analysis. There were no restrictions on the year of publication or the type of study. Studies differed in the use of periodontal health indices, such as the Community Periodontal Index of Treatment Needs (CPITN) and Simplified Oral Hygiene Index (OHI-S). These were eventually excluded due to the different modalities of the measurements, which may not be comparable with other indices and will affect the outcomes. Finally, five studies (n = 5) met the inclusion criteria, provided applicable data, and were included in the systematic review and *meta*-analysis (Table 1).

The second stage focused on data collection. Each included study was systematically reviewed by three reviewers for data collection. The collected information included authors, year of publication, origin country of the study, sample size, and sex and age range of subjects. Outcomes and statistical results are displayed in Tables 1 and 2.

2.1. Data analysis

The *meta*-analysis of this systematic review was performed using MedCalc for Windows version 15.0 (MedCalc Software, Ostend, Belgium). The *meta*-analysis was applied for two quantitative (continuous) variables: gingival index (GI) and plaque index (PI). The mean and standard deviation were used to describe the two variables. As recommended by Cohen, the standardized mean difference (SMD) was used as a summary of pooled statistics with cut-off values of the pooled effect of

0.2, 0.5, and > 0.8 representing small, medium, and large effects, respectively. The statistical significance of the SMD was assessed using Student's *t*-test. Heterogeneity in the pooled data was identified using Cohran's Q (weighted sum of squares on a standardized scale) and I^2, which indicates the percentage of total variation across the studies. A cut-off value of $I^2 > 50\%$ was used to rule out higher levels of unexplained variability in the effect sizes. Both fixed-effect and random-effect models were used to obtain the pooled estimates. Statistical significance and precision of estimates were reported using $p \leq 0.05$, and 95% confidence intervals. Forest plots were used to graphically show the results (highlighting the overall effect using both fixed and random effect models) of the studies included in the *meta*-analysis. Funnel plots were used to identify the publication bias of studies that were included in the systematic review.

3. Results

3.1. Gingival index

For the GI, statistical significance was assessed by combining the difference in the mean values extracted from the five studies and compared between the ASD and control groups. There were no statistically significant differences in SMD values according to both fixed and random effect criteria. The Cochran's Q value (Q = 139.14, P < 0.0001) was statistically significant, and the I^2 value (97.13%) was high, which suggests heterogeneity among the five studies. Hence, the pooled SMD according to the random effects model was used to infer no significant difference in the mean values of GI between the two groups (SMD = − 0.528, t = 1.381, P = 0.168). The overall effect (0.528) was considered to be of medium size (Table 2).

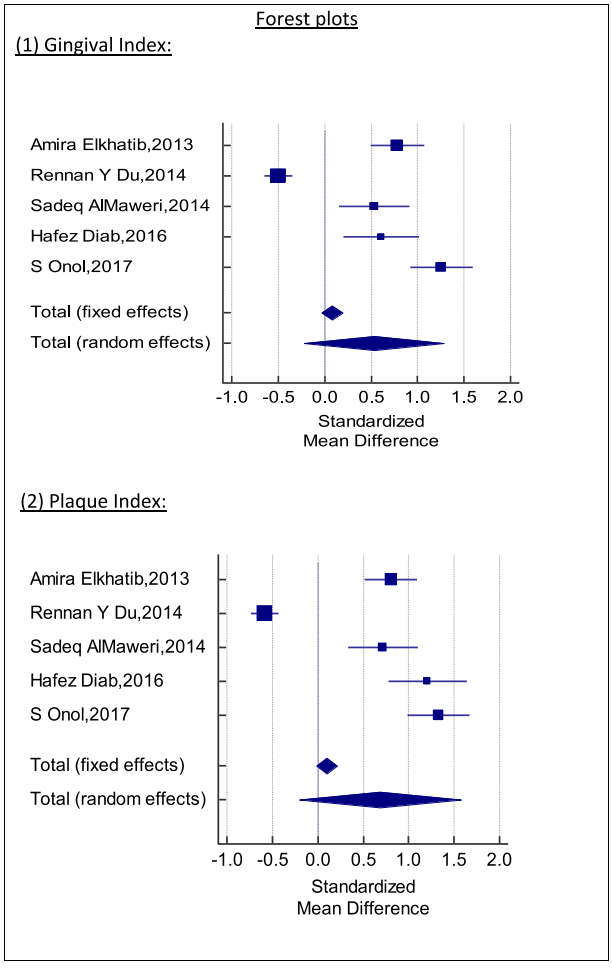

Fig. 1 Forest plots for included studies.

3.2. Plaque index

For the PI, the statistical significance was assessed by combining the differences in the mean values extracted from the five studies and compared between the ASD and control groups. There were no statistically significant differences in the SMD values based on both the fixed and random effect criteria. The Cochran's Q value (Q = 188.96, P < 0.0001) was statistically significant, and the I^2 value (97.88%) was high, which implies heterogeneity among the five studies. Hence, the pooled SMD according to the random effects model was used to infer no significant difference in the mean values of PI between the two groups (SMD = −0.687, t = 1.518, P = 0.129). The overall effect (0.687) was considered to be of medium size (Table 2).

The corresponding forest plots for GI and PI show the effect size of each study and the combined effect size according to fixed and random effects models (Fig. 1). The small number of studies found per the inclusion criteria and the variation in the sample size in each included study were major limitations in addressing the funnel plot dots.

4. Discussion

Due to the difficulty of dealing with ASD patients, especially children, treating them in a dental setting can be challenging. Therefore, individuals with ASD are more often affected by preventable oral diseases, such as periodontal disease (Du et al., 2015). Negligence, poor communication, and other behavioral conditions may also contribute to oral diseases (El Khatib et al., 2014). Access to oral healthcare, special management in dental clinics, and supervision during their daily oral hygiene routine are necessary for children with ASD (Shah et al., 2011). The lack of cooperation from dental clinics means that fewer ASD patients have access to oral healthcare (Stein et al., 2012). Dental teams should strive to create a comfortable environment for patients with ASD despite the added difficulty. These environments should be adaptable, using different auditory and visual pedagogies, as they are proven to provide superior cooperation from those diagnosed with ASD (Kopycka-Kedzierawski and Auinger, 1999). The Sensory Adapted Dental Environment (SADE) method was also reported to help reduce anxiety, pain, sensory discomfort, and distress (Cermak et al., 2015).

Children and adolescents with ASD had significantly higher GI and PI than children without ASD. These results were reported in all studies except for that of Du et al. (2015) who found GI in preschool children diagnosed with ASD was lower than that in non-autistic children. However, no significant differences were reported in the same study for PI, which is a sign of progression in dental disease. Poor oral hygiene is believed to be the cause of gingival disease, which is why studies that

reported higher GI and PI in individuals with ASD are significant. Medications, such as anticonvulsants, taken by children with ASD cause side effects that may contribute to gingival disease, as reported by ElKhatib et al. (2014). The variability of the environment of the control subjects in this study was another limitation to be considered. Control subjects in Diab et al. (2016) were attending dental clinics, whereas control subjects in the studies from ElKhatib et al. (2014), Al-Maweri et al. (2014), and Onol and Kırzıoğlu (2018) were conducted in schools, which implies that educated children are more aware of oral hygiene instructions.

The major limitation of this study was the small number of studies that followed our inclusion criteria for reporting periodontal health status, and the majority of studies mainly focused on dental caries. Another limitation was the multiple indices used to measure gingival health, which made it difficult to compare with the reported data.

5. Conclusions

Children and adolescents with ASD have poorer oral hygiene and higher rates of gingival disease than non-autistic individuals. Poor oral hygiene is believed to be the primary cause of gingival disease. Individuals with ASD need help and better access to oral healthcare. Educating dental teams and raising parents' awareness will assist in providing the highest standard of oral healthcare. Further investigations need to be done with regard to gingival health in ASD individuals, along with caries risk assessment, to understand the effect of the disorder on oral health. A standardized index for gingival health will help in the comparison of more studies to assess the prevalence of gingival health in children and adolescents with ASD.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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