Oral health status of Italian children with Autism Spectrum Disorder

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DOI 10.23804/ejpd.2021.22.03.12

Abstract

Aim Autism Spectrum Disorder (ASD) is characterised by communication deficits and repetitive unusual behaviours. The behaviour guidance of these children represents a challenge for the dental team. The aim of the study was to evaluate the oral health status in a group of Italian children with ASD.

Materials and methods Study Design: Sixty-four Italian children with ASD and 64 controls were included. Data were collected by means of questionnaires and clinical examinations.

Results Dental trauma (p=0.007), bruxism (p=0.001) and biting objects habit (p=0.021) were more frequent in the study group; fluoride exposure was lower (p=0.001) (chi-square test). The mean plaque index was 1.48 ±0.75 in the study group and 0.81±0.56 in the control group (p=0.001; Mann-Whitney U test); the mean dmft/DMFT was 3.00 ±1.2 and 2.3 ±1.8 in the study group and 1.8 ±1.1 and 1.0 ±1.1 in the control group (p<0.001; Mann-Whitney U test). Anterior open bite was more frequent in the study group (p=0.013; Chi-square test). No significant differences were found for enamel defects, molar relationship, posterior crossbite and deep bite. Significantly more children with ASD showed a negative behaviour (80% vs 35%: p =0.001; Chi-square test).

Conclusion Children with ASD have a poorer oral health status than healthy children. The early establishment of a home dental hygiene should be encouraged.

KEYWORDS Autism spectrum disorder; Oral health.

Introduction

Autism spectrum disorder (ASD) is a term used to describe a wide range of early-appearing social communication deficits and repetitive sensory-motor behaviours associated with a genetic component and other causes. The disorder is characterised by social communication deficits and repetitive, unusual sensory-motor behaviours. Because there are no reliable biomarkers, the diagnosis is clinical [Khan et al., 2012]. The global prevalence of ASD was estimated around 1% [Elsabbagh et al., 2012]. ASD is associated with coexisting conditions that can have a negative impact on the wellbeing of the child. Coexisting conditions include other neurodevelopmental disorders, epilepsy, intellectual disability, learning difficulties, speech and language delays, tics, sleeping problems, restricted and rigid food choices, obesity and gastrointestinal symptoms [Lord et al., 2018]. The American Psychiatric Association's Diagnostic and Statistical Manual of Mental Disorders (DSM)-5 criteria [APA, 2013] include: deficits in social-emotional reciprocity (e.g. reduced sharing of interests, emotions, or affectivity), deficits in non-verbal communicative behaviours (e.g. abnormalities in eye contact and body language, or deficits in understanding and use of gestures), deficits in developing, maintaining, and understanding relationships (e.g. difficulties adjusting behaviour to suit various social contexts). Due to these characteristics, the behaviour guidance of children with ASD in the dental setting can be very challenging for the dental team leading in many cases to the delivery of the dental treatment under general anaesthesia [Capp et al., 2010].

Previous studies were conducted to assess the oral health status of children with ASD, leading to conflictual data, particularly regarding the dental caries experience: some authors reported a lower caries experience [Kuter and Guler, 2019; Mattei et al. 2007; Namal et al. 2007] while other supported the hypothesis that ASD may be considered a risk factor for dental caries [Suhaib et al., 2019; Marshall et al., 2010]. To the best of our knowledge, the recent available literature on this topic does not include studies conducted in Europe.

The aim of this study was to describe the oral health status of Italian children with ASD in comparison with healthy children.

Material and methods

This study was conducted on outpatients attending the Special Needs Dentistry and Paediatric Dentistry Unit, Department of Biomedical and Neuromotor Sciences, University of Bologna, Italy. Children with a documented diagnosis of ASD were referred from the Paediatric Units of St. Orsola-Malpighi Polyclinic, Department of Medical and Surgical Sciences, University of Bologna, Italy.

All the children with ASD attending the Unit of Special Needs Dentistry for a first dental visit between January 2015 and March 2018 were recruited. As a control group, the same number of Italian healthy children were included. The following exclusion criteria were applied: medical condition associated with oral diseases; unable to cope with an oral examination; dental prophylaxis in the previous 6 months; history of orthodontic treatment. The assumptions made for sample size calculation were: caries prevalence in healthy children = 45% [Loo et al. 2008]; caries prevalence in children with ASD = 70% [Campus et al. 2007]; alpha-error = 5%, beta-error = 20%.

The estimated sample size per group was 60 children.

The investigators were two dentists trained in special needs dentistry and paediatric dentistry. The local Ethics Committee of the Bologna University Hospital Authority St. Orsola-Malpighi Polyclinic (Bologna, Italy) approved this study (PG. N 0019293). In full accordance with the ethical principles of the Helsinki

Declaration, the written informed consent for participation and publication was obtained from the parent/guardian of the subject before enrolment.

Data collection: medical and dental history

A parent filled in a medical history form, a dental form and the "WHO Oral Health Questionnaire" regarding oral hygiene, fluoride intake, diet and oral habits [WHO, 2013].

Oral examination

Each individual was examined on a dental chair by one investigator with a dental mirror and a WHO periodontal probe. A mouth-opener was used if needed.

Oral and orthodontic diagnoses were based on clinical findings. The Silness and Loe Plaque Index (PI) was used for oral hygiene [Silness and Loe, 1964], and the decay, missing, filled teeth Index for dental caries of the primary dentition (dmft) and of the permanent dentition (DMFT) [WHO, 2013]. The dental history was used to identify teeth extracted for caries or missing for other reasons (agenesis, dental trauma). The modified index of developmental defects of enamel (mDDE) was used to assess dental enamel defects [FDI, 1992]. Regarding occlusion, the following clinical variables were evaluated: relationship between the maxillary and mandibular first permanent molars (Angle's classification system); relationship between the upper and lower dental arch in the three planes of space (sagittal, vertical, transverse). The behaviour of each child was recorded using the Frankl scale into four categories from definitely negative (grade 1) to definitely positive (grade 4) [Frankl et al., 1962].

Training and calibration

The 2 investigators were trained and calibrated in the application of PI and dmft/DMFT Index on a group of 10 children with special health care needs at the Special Needs Dentistry Unit. Using Cohen's kappa statistics, inter-examiner agreement was reported as good (0.78 PI) and very good (0.86 dmft/DMFT). Intra-examiner reliability of the method, tested by one investigator repeating the assessment in 5 children after 2 hours, was reported as good (0.75 PI) and very good (0.85 dmft/DMFT).

Statistical analysis

The statistical analysis was performed using the Software Package for the Social Sciences (SPSS for Windows, version 23.0, SPSS Inc, Chicago, IL, USA). Descriptive statistics were first performed. Continuous variables were tested for distribution using the Kolmogorov-Smirnov test. Between-groups comparisons would be performed with the two-sample t-test if data were normally distributed; the Mann-Whitney U test would be used if data were not normally distributed. The χ^2 test was used for the categorical variables. For analysis purposes, the behaviour of each child was dichotomised as positive ("definitely positive" and "positive" categories of the Frankl scale) and negative ("definitely negative" and "negative" categories of the Frankl scale). A p-value of less than 0.05 was considered to be statistically significant.

Results

One child with ASD was excluded due to a congenital heart disease and 4 because unable to cope with the oral examination. Sixty-four children with ASD were included in the study group: forty-two were males (66%), 22 were females (34%), mean age 9.0 ±2.9 years. Sixty-four Italian healthy children were included in the control group: thirty-seven were males (58%), 27 were females (42%), mean age 8.4 ±3.0 years. No significant differences according to gender (p= 0.363; χ^2 test) and age (p= 0.252; Mann-Whitney U test) were found between the groups.

Medical and dental history

As reported by the parents, the median age of the first dental visit was 6 years (range:3-10) in the study group and 5 years (range:3-8) in the control group. The difference between the two groups was statistically significant (p< 0.001; Mann-Whitney U test). Sixteen children (23%) in the study group and 35 (55%) in the control group had at least one dental recall during the past 12 months (p< 0.001; χ^2 test). None of the subjects with ASD was able to brush his/her teeth autonomously and the oral hygiene was carried out by the parents. Detailed data are shown in Table 1.

Oral hygiene, dental caries and dental enamel defects

The mean PI was 1.48 ±0.75 in the study group and 0.81±0.56 in the control group; the difference between the two groups was statistically significant (p=0.001, Mann-Whitney U test). In the study group, oral hygiene was rated as good (PI 0-1) in 27 (42%) children with ADS, fair (PI 1–2) in 29 (45%), poor (PI 2-3) in 8 (13%). In the control group, oral hygiene was rated as good (PI 0–1) in 40 (73%) children, fair (PI 1–2) in 23 (36%) poor in 1 (1%). The difference between the two groups was statistically significant (p= 0.013; χ^2 test). The mean dmft was 3.00 ±1.2 (d=2.5; m=0.3; f=0.2) in the study group and 1.8 ±1.1 (d=1.0; m=0.1; f=0.7) in the control group. The difference between the two groups was statistically significant (p<0.001; Mann-Whitney U test). A significantly higher number of healthy children had fillings in the primary teeth (p= 0.034; Mann-Whitney U test). The mean DMFT was 2.3 ±1.8 (D=1.6; M=0.2; F=0.5) in the study group and 1.0±1.1(D=0.5; M=0; F=0.5) in the control group. The difference between the two groups was statistically significant (p<0.001, Mann-Whitney U test). A significantly higher number of healthy children had fillings in the permanent teeth (p=0.021; Mann-Whitney U test). Two children (3%) with ADS had one extracted first permanent molar. Twenty-one children with ASD (32%) were caries free and 42 healthy children (66%); the difference between the two groups was statistically significant (p<0.001, χ^2 test). Developmental defects of enamel (hypomineralisation) were found in 8 children (13%) in the study group (demarcated opacity) and 12 (19%) in the control group (10 demarcated opacity; 2 diffuse opacity); the difference between the two groups was not statistically significant (p= 0.232, χ^2 test).

Occlusion

The Angle's classification system, based on the relationship between the maxillary and mandibular first permanent molars, was not applicable in 15 children of the study group and 14 of the control group due to the absence of the first permanent molars during the primary/mixed dentition. Class I malocclusion was found in 34 children (70%) of the study group and 38 (76%) of the control group. Class II malocclusion was found in 13 children (26%) of the study group and 9 (18%) of the control group. Class III malocclusion was found in 2 children (4%) of the study group and 3 (6%) of the control group. The difference among the groups was not statistically significant (p= 0.565, χ^2 test). A posterior crossbite was found in 9 children (14%) of the study group and 10 (17%) of the control group. The difference between the two groups was not statistically significant (p= 0.565, χ^2 test). An anterior open bite was found in 12 children

	study group n (%)	control group n (%)	p-value
Taking medication[1]	11 (17)	1 (2)	0.002*
Allergy[1]	1 (2)	2 (3)	0.559
Medications during the first [2] years[1]	29 (45)	19 (30)	0.067
Breastfeeding[1]	25 (39)	51 (80)	0.001*
Formula feeding[1]	39 (61)	13 (20)	0.001*
Medical pathologies[1] - Epilepsy	7 (11)	1 (2)	0.028*
General anaesthesia[1]	25 (39)	1 (2)	0.001*
Frequency of toothbrushing[2] - Several times per week - At least Once a day - Twice a day	 11 (17) 24 (38) 29 (45)	 6 (9) 24 (48) 34 (53)	 0.293
Frequency of intake of sugary snacks/ beverages[2] - ≤ 1/day -≤ 2/day -≥ 3/day	 28 (44) 19 (30) 17 (26)	 6 (9) 19 (30) 39 (61)	 0.001*
Fluoride exposure[1] - Toothpaste - Drops/tablet - Both	 9 (14) 6 (9) 1 (1)	 54 (84) 1 (2) 9 (14)	 0.001*
Dental trauma[1]	33 (52)	19 (29)	0.007*
Oral habits[1]: - non-nutritive sucking - oral breathing - bruxism - biting objects - no chewing activity	 20 (31) 7 (11) 47 (73) 9 (14) 8 (13)	 13 (20) 13 (20) 25 (39) 0 1 (2)	 0.225 0.224 0.001* 0.021* 0.038*

**statistically significant*

[1]Chi-square test

[2]Mann-Whitney U test

TABLE 1 medical Medical and dental hystory among of children with ASD and healthy children.

(19%) of the study group and 3 (5%) of the control group. The difference between the two groups was statistically significant (p= 0.013, χ^2 test). A deep bite was found in 9 children (14%) of the study group and 10 (17%) of the control group. The difference between the two groups was not statistically significant (p= 0.803, χ^2 test).

Behaviour

According to the Frankl behaviour rating scale, 51 children (80%) of the study group and 3 (5%) of the control group showed a negative behaviour. The difference between the two groups was statistically significant (p= 0.001, χ^2 test).

Discussion and conclusion

This study represents the first data collection on the oro-dental features of children with ASD in Italy. The medical history showed that significantly more children with ASD took medications. This difference is due to comorbidities, such as hyperactivity, anxiety and epilepsy. In the present study, 11% of the children with ASD suffer from epilepsy. All seizure types can be associated with autism, with two peaks at the age of onset: during the early childhood and during the adolescence [Lord et al., 2018]. Epilepsy has a great impact on the daily life of the affected individuals and always requires a medical treatment. In a previous study, Bagattoni et al. [2017] found a high frequency (30%) of traumatic dental injuries in the ASD subgroup in a group of Italian children and adolescents with special health care needs. The difference between the ASD subgroup and the other subgroups was significant. In the present study, the dental history confirmed this finding. Al-Sehaibany [2018] found a prevalence of dental trauma of 25.7% among 257 Saudi preschool children with ASD and 16.3% among healthy children (p=0.012). Children with ASD had higher rates of dental trauma during daily activities, falling while walking or during episodes of self-harm while in the control group during leisure activities. Habibe et al. [2016] found a frequency of dental trauma of 39.3% among 122 Brazilian children and adolescents with ASD with no statistically significant difference compared to the healthy children. Du et al. [2015] found a similar frequency of dental trauma in children with and without autism. Children with ASD were often described as hyperactive; poor muscle tone, poor motor planning, toe walking and deficits in motor coordination are also common findings in people with ASD increasing the risk of DT. Despite conflicting data, it is conceivable that individuals with ADS are at greater risk of dental trauma and that epilepsy may be an additional risk factor.

Conflicting data exist in the medical literature regarding the prevalence of dental caries in individuals with ASD. Some authors reported a lower prevalence of dental caries in children with ADS compared to healthy children [Kuter and Guler, 2019; Mattei et al., 2007; Namal et al., 2007]. In contrast, Marshall et al. [2010] considered autism as an indicator of high caries risk. A recent systematic review and meta-analysis [da Silva et al., 2017] reported a pooled caries prevalence of 60.6%. Our study showed a statistically significant difference in caries experience between the two groups. In children with ADS, the caries experience was related to the exposure to many predisposing factors: formula feeding was more common than breastfeeding, the PI was significantly higher and the first dental visit significantly delayed. Most children with ASD did not have an adequate fluoride exposure: only 14% used a fluoride toothpaste. Du et al. [2015] found that many children with ADS refuse the use of toothpaste during

toothbrushing or that the brushing time is shorter, reducing the exposure to topical fluoride. Rada [2010] showed that some parents had concerns about the safety of topical fluoride in their children with ADS. In contrast with the high caries experience, our results showed a lower intake of sugar containing snacks/beverage in children with ADS compared to healthy children. According to our results, Namal et al. [2007] found a lower consumption of cariogenic foods and less snacking. As recently suggested by Ferrazzano et al. [2020], ASD may be considered an indicator of high caries risk and oral hygiene may be the most influential risk factor associated with new caries lesions.

The mean PI was significantly higher in the study group and none of the children was able to brush his/her teeth autonomously. In agreement with our results, Onol and Kırzıoğlu [2018] reported a higher PI in children with ASD than in healthy children: (2.06 ± 0.73 vs 1.24 ± 0.54; p< 0.001). These results may be explained by the lack of cooperation and the parents' difficulty in carrying out an effective toothbrushing. In a prospective study conducted on 15 children with ASD (aged 5–13 years-old) and their parents, pictures illustrating a correct toothbrushing technique were placed in the room where oral hygiene was usually performed. After 12 months, the amount of visible dental plaque was significantly lower and those parents/caregivers who previously found it difficult/very difficult to maintain a good oral hygiene for their child, after the study, were able to do that [Pilebro and Bäckman, 2005]. El Khatib et al. [2014] suggested that the families of children with ASD are often exhausted by the need for constant supervision, so parents give less attention to the oral hygiene and seek dental care. The delay in the first dental visit found in our study group supports this hypothesis. Not surprisingly, the median age of the first dental visit was also high in the control group. In a previous study in the same geographic area, 126 paediatricians were interviewed regarding their expertise and attitude for promoting oral health: 66% encouraged the first dental visit after 4 years of age [Bagattoni et al. 2016].

Regarding the oral habits, bruxism, biting objects and no chewing activity were significantly more frequent in children with ADS. In particular, bruxism was found in 73% of the study group. Studies have reported that bruxism is frequent among children with ASD [Mattei et al., 2007; Onol and Kırzıoğlu 2018]. Onol and Kırzıoğlu [2018] found that bruxism was less frequent in children with ASD who started their special education before the age of three years. Regarding the occlusion, as reported by Onol and Kırzıoğlu [2018], we did not find any significant difference in molar relationship between the two groups. In contrast, we found a higher frequency of anterior open bite in the study group than in the control group. The habit of biting objects may be associated with this malocclusion.

In the present study, 80% of the children with ASD showed a negative behaviour. The same finding was showed by Onol and Kırzıoğlu [2018]. As reported by Ferrazzano et al. [2020], children with ADS are prone to agitation and emotional dysregulation; hypersensitivity to sensory input and short attention span make it difficult for dentists to examine and treat children with ASD. This may be the reason of the higher number of untreated dental caries reported in both dentitions, in addition to less access to dental recalls. Murshid [2011] described the parents' inadequate knowledge about successful methods of behaviour management in the dental setting as the main reason for a poor access to dental care, in addition to the inadequate training of the general dentists and the lack of proper facilities.

The results from the present study suggests the need for more information, provided by paediatricians and dentists, regarding the importance of the oral health promotion in children with ASD. Paediatricians should encourage parents to seek dental care for periodic dental recalls and reassure them regarding the need and safety of topical fluoride exposure. The early establishment of a home dental hygiene is necessary to promote a positive relationship between the paediatric dentist and the child and to decrease the treatment needs leading to fewer opportunities for negative experiences and for the use of general anaesthesia. A fluoride toothpaste and additional fluoride treatments, as in-office fluoride varnish, are recommended for all the children with ASD. Since oral hygiene was the major risk factor for dental caries, frequent dental hygiene recalls are advisable. The use of pit and fissure sealants is strongly encouraged to prevent dental caries of the permanent teeth.

In the present study, data regarding the medical and dental history were collected through the use of a self-administered questionnaire filled by parents. The WHO recommends the use of simplified structured questionnaires for the collection of self-assessed data on oral health and risk factors in children and adolescents. Both questionnaires have been pilot-tested in a range of countries across the world. However, social desirability bias and memory bias may have affected the responses.

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