

Article

Oral Health Status of Children with Autism in Central Italy

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Abstract: Children with autism spectrum disorder (ASD) have significantly higher prevalence and caries severity compared to the average population. Knowledge about the oral health indices of children with this mental disorder is key to designing efficient plans of intervention. This paper reports the results of a study on the oral health status of children with ASD in central Italy. This is the first study of this type in Italy. The sample consists of 229 autistic children aged between 5 and 14 years, attending the Unit of Special Needs Policlinico Umberto I in Rome. Each patient received an intraoral examination to investigate decayed, missing, and filled teeth as well as periodontal status. Information on demographic attributes, dietary habits, medical history, and child's cooperativeness at the first visit was also recorded. Of the participants, 79.26% presented signs of gingivitis and about 90% of them had plaque. Caries prevalence was 66.38%. The average of the total number of decayed, missing, and filled teeth in the permanent and primary dentition was 2.91. Among the factors considered, only dietary habits and the periodontal indices showed statistically significant association with caries prevalence and caries severity. Despite the selection bias, that prevents us to interpret the results presented as epidemiological evidence, our study suggests that children with ASD in central Italy represent a population at risk.

Keywords: autism spectrum disorder; caries prevalence; oral prevention; special need patients

1. Introduction

Autism spectrum disorder (ASD) is a complex developmental condition that involves persistent challenges in social interaction, speech and nonverbal communication, as well as in restricted and repetitive behavior. It is more common in males than in females (with a ratio of 4:1) and is usually first diagnosed in early childhood (around 2 or 3 years old) when many of the most obvious symptoms occur [1]. The effects of ASD and the severity of symptoms differ from person to person. While some people with ASD can live independently, others have severe disabilities and require life-long care and support [2].

Based on epidemiological studies, the prevalence of ASD appears to be increasing globally over the past 50 years [2–5]. There are many possible explanations for this increase, including better diagnostic tools, improved reporting, a greater propensity for families to accept the pathology, a greater presence in the areas of support structures and associations, and changes in health policies.

The reported ASD prevalence varies substantially across studies with values ranging from 1.9/10,000 to 169/10,000 for USA, Europe, Canada, Asia, and Australia populations [6–9]. This variability may be due to the complexity of diagnosis in relation to the various clinical situations [10] and various methodological factors, such as differences in the diagnostic method, in the size of the sample examined, and in the age groups considered [11].

As far as Italy is concerned, prevalence rates estimated from local surveys on patients attended to by the National Health System (NHS) in Emilia-Romagna, Piemonte, and Toscana regions range from 15/10,000 to 80/10,000 [12].

Despite the differences in the reported prevalence, all these studies clearly indicate that ASD is a pathology with a strong social impact and important public health implications. In this respect, oral health problems represent a key component. Normal daily activities, such as tooth brushing as well as understanding the behavior of other individuals, constitute a constant challenge for children affected by ASD [13–18]. Several studies indicate that, due to these difficulties in maintaining good oral hygiene and being periodically examined and eventually treated, children with ASD have significantly more untreated caries and a higher prevalence of periodontitis and gingivitis as well as higher values of the total number of decayed, missing, and filled teeth in the primary dentition and in the permanent dentition (denoted by DMFT/dmft) compared to the average population [19–27]. In addition, dental care, for patients with moderate or slight intellectual disorders, requires special procedures and adaptations [28]. Autistic children, compared to other patients with psychiatric disorders, have an additional difficulty with which to deal, namely anxiety. A dental office is full of potentially frightful stimuli, due to the peculiar environment and equipment used. Evidence-based psychosocial interventions, such as behavioral treatment and parenting skills training programs, can reduce difficulties in communication and social behavior, with a positive impact on well-being and quality of life for persons with ASD and their interaction with caregivers [13,16,28]. These types of interventions, that require special training programs for the dentists operating with children affected by ASD, represent an important economic load for families and/or national public health systems.

In Italy, the NHS guarantees people with ADS free access to early diagnosis and individualized treatment, using methods and tools based on the most advanced scientific evidence (Ordinary supplement to the “Official Gazette” no. 65, March 18, 2017). A correct assessment of the economic load and of the resources needed for this plan of intervention, which is key for optimal decision-making, requires the knowledge and surveillance, at the regional level, of the oral health indices of ASD patients. No study of this type is available for Italy. This work is meant as a first step in this direction and it is in line with similar works performed for other countries [19–27].

The main goal is to create an epidemiological oral care profile for children with ASD aged between 5 and 14 years attending the Unit of Pediatric Dentistry of the Policlinico Umberto I Hospital in Rome (Italy). The Unit is a leading center and a reference site in central Italy for pediatric dentistry for children with ASD. Oral health status was investigated in terms of caries severity and periodontal health.

2. Materials and Methods

2.1. Study Design

This study was conducted using a cross-sectional design. Data were collected between June 2013 and November 2018 and analyzed to evaluate the oral health status of children with autism spectrum disorder. The study received formal review and approval by the Board of the Department of Oral and Maxillo-Facial Science, Unit of Pediatric Dentistry, of the Policlinico Umberto I Hospital, Rome, Italy (N. 15/2020 Prot. n. 0000216 del 06/02/2020). The study complied with the STROBE statement.

2.2. Study Population

Children aged between 5 and 14 years, attending for the first time the Unit of Special Needs Policlinico Umberto I in Rome, who were diagnosed with autism spectrum disorder according to the

International Classification of Diseases (ICD-10) [29] criteria by a qualified professional were included in this study. The exclusion criteria included the following: patients suffering from other diseases known to influence dental caries or the severity of periodontal disease such as Down's syndrome and diabetes; patients with no formal ASD diagnosis; and patients whose parents did not give consent to the study. The response rate was 100%, and a total of 229 children with ASD were included in this study. Each patient received a complete oral and periodontal examination using codes and criteria as described by the World Health Organization (WHO).

2.3. Data Collection

Data were collected by a single International Caries Detection and Assessment System (ICDASTM) calibrated researcher, having a degree in dentistry with clinical and epidemiological experience (training was conducted by a core member of the International Caries Detection and Assessment System Coordinating Committee). Prior to the study, the researcher completed a six-month training period with a qualified autism therapist to acquire the relational skills necessary for an effective communication and interaction with the children participating in the study. Intra-examiner reliability was assessed according to WHO guidelines [30]. Parents gave consent to the study and it was performed in accordance with the Declaration of Helsinki.

Kappa statistics was used to determine intra-examiner reliability. The kappa test was adopted on a tooth-by-tooth basis in order to check the intra-examiner agreement in caries diagnosis according to the methodology described by Peres et al. (2001). A kappa value of 0.82 was obtained.

Information on demographic attributes (gender, age, nationality), dietary habits, medical history (type of delivery, vaccines, medications, previous experiences of general anesthesia) was obtained from a face-to-face interview with the parents of the children using a questionnaire especially designed for this study. Clinical data (caries severity, gingival status, plaque accumulation) were obtained from the clinical examination at the Unit of Special Needs of the Policlinico Umberto I in Rome according to the World Oral Health Survey [30]. The Frankl scale, a reliable behavior rating system in common use in both clinical dentistry and research in ASD patients [31], was used to measure the level of cooperativeness at the first dental visits. Each child was classified, by a dentist experienced in treating ASD children, in one of the four categories of the Frankl scale: definitely negative, negative, positive, and definitely positive (for details, see Table 1).

Table 1. Descriptive statistics (frequency tables) for the 12 factors considered in the study. N_{tot} = total number of responses for each factor. For the Frankl scale, the rating is as follows. Definitely negative: Refusal of treatment, crying forcefully, fearful, or any other evidence of extreme negativism; Negative: Reluctance to accept treatment, uncooperative, some evidence of negative attitude but not pronounced, i.e., sudden withdrawal; Positive: Acceptance of treatment, at times cautious, willingness to comply with the dentist, at times with reserve, but patient follows the dentist's directions cooperatively; Definitely positive. Good relationship with the dentist; interested in the dental procedures; laughing and enjoying the situation.

Gender ($N_{tot} = 229$)	%	Medications ($N_{tot} = 217$)	%
Male	75.98%	Yes	27.19%
Female	24.02%	No	72.81%
Age ($N_{tot} = 229$)		Prev. anesthesia ($N_{tot} = 224$)	
5	9.61%	Yes	8.04%
6	10.48%	No	91.96%
7	10.92%	Plaque Index ($N_{tot} = 218$)	
8	14.85%	No plaque	10.09%
9	15.72%	Plaque detectable with the periodontal probe	28.90%
10	12.23%	Plaque visible to the naked eye at the gingival margin	46.33%
11	5.68%	Abundant accumulation of Calculus beyond the margin	14.68%
12	6.11%		
13	2.61%		
14			

Table 1. Cont.

Nationality (N _{tot} = 229)		Gingival Index (N _{tot} = 218)	
Italian	93.01%	Healthy	20.74%
Foreign	6.99%	Bleeding	33.64%
		Tartar	45.62%
Diet (N _{tot} = 215)		Frankl scale rating (N _{tot} = 229)	
Varied diet (unrestricted)		Definitely Negative	49.78%
Diet rich in sugar (unrestricted)	33.95%	Negative	37.12%
Restricted diet (for intolerances)	6.05%	Positive	13.10%
		Definitely positive	0.00%
Natural birth (N _{tot} = 202)		All vaccines (N _{tot} = 191)	
Yes	46.00%	Yes	93.70%
No	53.96%	No	6.30%

The sum of decayed, missing, and filled teeth in the primary dentition (dmft) and in the permanent dentition (DMFT), denoted by DMFT/dmft, was used to determine caries severity. Caries prevalence was defined as the proportion of patients with a positive dental caries history, i.e., with a positive value of DMFT/dmft. Plaque and gingival indices (with four and three levels, respectively) were used to assess the general prevalence of periodontal health according to the World Oral Health Survey.

Each patient was visited in a separate cubicle in the pediatric dentistry clinic to reduce anxiety and stress resulting from being close to other patients and from the noises produced by dental instruments used by colleagues operating nearby (in the clinic there were 10 non-separate dental chairs placed in an open environment). When, on the first visit, the patient's lack of collaboration made it impossible to perform an intraoral examination, closed visits were scheduled to desensitize the patient to the environment and make them familiar with the operator. In order to facilitate the communication and increase the patient's cooperation, illustrative detachable images on a rigid panel were used according to the Alternative Augmentative Communication method (sample images are available as supplementary material) [32–34].

An intraoral examination of soft and hard tissues was carried out using disposable gloves and a sterile first-aid kit consisting of a mirror, a probe, and sterile gauze. When the cooperativeness of the patients made it possible, the reading of an orthopantomogram X-ray was used for the diagnosis of carious lesions (not detectable on the extraoral clinical examination) and possible anomalies in the number and position of the dental elements. The data collected were recorded in the patient's medical record.

2.4. Data Analysis

Data were analyzed using the free software environment for statistical computing and graphics R, version 3.6.1 (R Foundation for Statistical Computing, Vienna, Austria) [35]. The descriptive analysis included frequency tables, as a display of univariate distributions of the factors considered in the study, and a histogram of total DMFT/dmft and position metrics of total DMFT/dmft (mean, median, first and third quartiles), as summary statistics of the distribution of caries severity. The relationship between caries prevalence and the factors of interest (demographic attributes, dietary habits, medical history, periodontal indices, and level of cooperativeness) was investigated using Fisher's exact test. Due to the high skewness, and therefore non-normality, of the DMFT/dmft data (see Figure 1), the study of the relation between caries severity and risk factors was performed using the Kruskal–Wallis test, which is a one-way ANOVA on ranks.

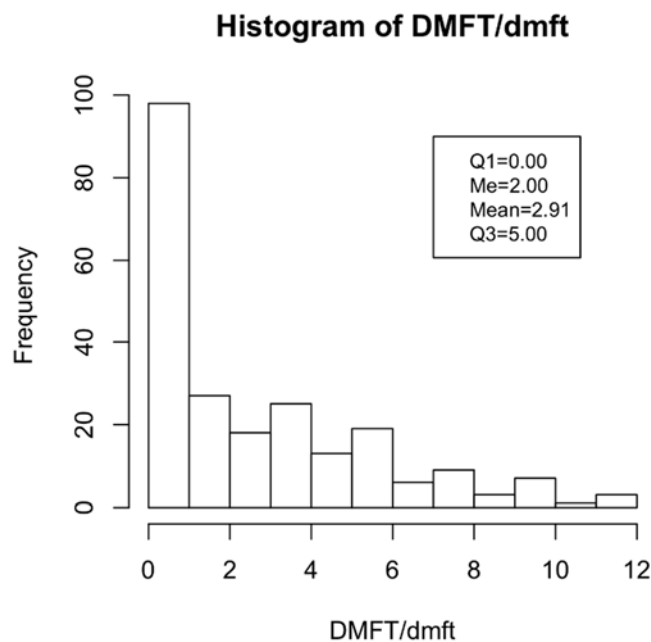


Figure 1. Histogram of the total number of decayed, missing, and filled teeth in the primary and permanent dentition (denoted by DMFT/dmft) for the 229 children in the study. Q1 = First quartile, Me = Median, Q3 = Third quartile.

3. Results

The distribution of the 12 factors considered in the study is presented in Table 1 as frequency tables. The total sample size is 229. However, complete information is available only for the demographic attributes (gender, age, and nationality) and the level of cooperativeness (measured as the Frankl scale rating). For the other factors, the percentage of missing values is quite heterogeneous, ranging from 2% to 17%. Approximately three-quarters of the sample are male (76%). The average age is 8.8 years. Most of the children (42.36%) are aged (in years) in the range of 8–10. The rest are almost equally spread in the ranges of 5–7 (31.01%) and 11–14 (26.63%). The vast majority of the participants are Italian (93.01%), have followed the vaccination plan established by the NHS (93.70%) and, at the time of the first visit, did not have previous experience of general anesthesia (91.96%). About one-third of the sample (27.19%) takes medications regularly or temporarily, and about half (46.00%) were born by natural childbirth. In terms of dietary habits, 60% of the participants have a diet rich in sugar and about one-third (34%) have a varied diet. In both cases, no diet restrictions (for intolerances or other reasons) are present.

The scores of the participants related to plaque and gingival indices suggest a poor periodontal health. Of the participants, 79.26% have signs of gingivitis (bleeding and/or gingival tartar) and about 60% of them either have plaque visible to the naked eye at the gingival margin or abundant accumulation of calculus beyond the margin. A small percentage has no plaque (10.09%) or healthy gums (20.74%).

As expected, the level of cooperativeness (at the first dental visits) for the vast majority (87%) of ASD children in the study was rated, according to the Frankl scale, as definitely negative (49.78%) or negative (37.12%). None of the children's behavior was rated as definitely positive.

Caries prevalence for the observed sample is 66.38% (152 out of the 229 participants have non-zero DMFT/dmft). The distribution of DMFT/dmft (see Figure 1) is very skewed. The average DMFT/dmft is 2.91 with a median of 2.00. The maximum observed DMFT/dmft value is 12. Among the 12 factors considered in the study, only the dietary habits and the periodontal indices (GI and PI) show statistically significant association with caries prevalence (see Tables 2 and 3) and with caries severity (see Tables 4

and 5). As one would expect, a diet rich in sugar and a high score in the GI and PI indices (i.e., poor periodontal health) are associated with higher prevalence and caries severity.

Table 2. Caries prevalence in relation to background and behavioral variables. N = total number of participants in each category; n = number of participants with positive DMFT/dmft in each category. *p*-values < 0.05, corresponding to statistically significant differences, are marked with an asterisk and the corresponding variable is highlighted in bold.

Variables	Caries Prevalence (n/N)	<i>p</i> -Value
Gender		
Male	66% (114/174)	0.890
Female	69% (38/55)	
Age		
5–7	70% (50/71)	0.233
8–10	69% (67/97)	
11–14	57% (35/61)	
Medications		
Yes	61% (36/59)	0.421
No	68% (107/158)	
Diet		
Free and varied diet	42% (31/73)	<0.001 *
Free diet rich in sugars	81% (104/129)	
Restriction for intolerances	62% (8/13)	
Natural birth		
Yes	62% (58/93)	0.555
No	67% (73/109)	

Table 3. Caries prevalence in relation to Gingival Index, Plaque Index, and ASD severity (measured as the Frankl scale rating). N = total number of participants in each category; n = number of participants with positive DMFT/dmft in each category. *p*-values < 0.05, corresponding to statistically significant differences, are marked with an asterisk and the corresponding variable is highlighted in bold.

Variables	Caries Prevalence (n/N)	<i>p</i> -Value
Gingival Index		
Healthy	49% (22/45)	<0.001 *
Bleeding	63% (46/73)	
Tartar	80% (79/99)	
Plaque Index		
No plaque	27% (6/22)	<0.001 *
Plaque detectable with the periodontal probe	54% (34/63)	
Plaque visible to the naked eye at the gingival margin	79% (80/101)	
Abundant accumulation of calculus beyond the margin	88% (28/32)	
Frankl scale		
Definitely Negative	62% (71/114)	0.194
Negative	67% (57/85)	
Positive	80% (24/30)	
Definitely positive	-	

Table 4. Caries severity in relation to background and behavioral variables. *p*-values < 0.05, corresponding to statistically significant differences, are marked with an asterisk and the corresponding variable is highlighted in bold.

Variables		Mean DMFT/dmft	<i>p</i> -Value
Gender			
Male		2.988	0.703
Female		2.655	
Age			
5–7		3.014	0.121
8–10		3.258	
11–14		2.229	
Medications			
Yes		2.559	0.170
No		3.101	
Diet			
Free and varied diet		1.384	<0.001 *
Free diet rich in sugars		3.930	
Restriction for intolerances		2.615	
Natural birth			
Yes		2.731	0.375
No		3.000	

Table 5. Caries severity in relation to Gingival Index, Plaque Index, and ASD severity (measured as the Frankl scale rating). *p*-values < 0.05, corresponding to statistically significant differences, are marked with an asterisk and the corresponding variable is highlighted in bold.

Variables		Mean DMFT/dmft	<i>p</i> -Value
Gingival Index			
Healthy		1.467	<0.001 *
Bleeding		2.986	
Tartar		3.717	
Plaque Index			
No plaque		0.636	<0.001 *
Plaque detectable with the periodontal probe		1.778	
Plaque visible to the naked eye at the gingival margin		3.693	
Abundant accumulation of calculus beyond the margin		4.875	
Frankl scale			
Definitely Negative		2.693	0.092
Negative		2.824	
Positive		3.967	
Definitely positive		-	

4. Discussion

To the best of our knowledge, this is the first study that provides information on the oral health status of children aged 5 to 14 years with ASD in central Italy (and more in general in Italy). The observed male/female ratio (which is approximately 4:1) is in agreement with values reported by the WHO based on which ASD is 3 to 4 times more frequent in males than in females [1]. The overall caries prevalence in our study is 66.4%, smaller than the one reported in similar studies in South Africa and United Arab Emirates [20,27] (where a prevalence up to 85% was observed) but (more than 20%) larger than the prevalence that appears in other studies for Turkey and India [22]. In terms of caries severity, the average DMFT/dmft score for our sample (2.91) is substantially smaller than the one obtained in comparable studies conducted in Africa [24,27] (where average values of DMFT/dmft of 4.79 and 7.23 have been found), slightly smaller than the one reported in a similar study in Turkey [36] and slightly larger than those obtained in studies on autism in Asian populations [20,21,36–38] (for which

an average value between 2.01 and 2.4 has been reported). We found that both caries prevalence and caries severity have a statistically significant association with the type of diet and the periodontal indices PI and GI of the autistic patient but, contrary to the findings of similar studies [12,36–38], a not statistically significant association with age and gender was found. No statistical association between autism severity (as measured by the Frankl scale rating) was found. This may be due to the fact that the vast majority of children with ASD (87% in our case) are not cooperative and thus the Frankl rating exhibits little variation in our sample, and to the presence of potential confounding variables (the subgroups of children with different Frankl rating values may not be homogeneous with respect to factors, such as dietary habits, relevant for caries prevalence and caries severity). In any case, the high percentage of non-cooperative children may explain the high prevalence and caries severity observed in our sample and represents a defining feature of children with ASD in comparison to the population of children without mental disorders.

These results should be interpreted with caution due to two important limitations of our study, namely selection bias and the age range considered. Our sample, in fact, consists of autistic children from a pediatric clinic that usually attracts patients with dental needs. Thus, the results of the present study can be indicative of the dental needs of patients with autism but cannot be used as epidemiological evidence. It should be said, however, that we expect the selection bias to be not too serious. Parents of children with ASD, in general, are members of local or national autism associations. Although the first group of children who entered the study in June 2013 had dental needs, after a few months following the beginning of the study, many parents of children with ASD, independently of the children's oral status, joined the study because of word of mouth in the autism parental associations.

On the other hand, the age range of the participants in the study (5–14 years) implied the inclusion of patients with primary, mixed, and permanent dentition whose caries prevalence can vary significantly. With these limitations in mind, a comparison with previous results in the literature can be attempted.

Some studies [37] have reported that the consumption of food/drinks rich in sugar in the population of children with ASD is comparable with the corresponding population not affected by this pathology. In our case, however, and in line with the values reported in other studies [39] a large proportion of the children in the sample (60%) has a diet rich in sugars and this proportion is substantially larger than the one reported in the non-autistic children population. For example, a study conducted in one province of the Lazio region [39] reports that only 17% of the total sample eat sugary foods every day. At a national level, a study conducted on eating habits in primary schools shows that 41% of the sample usually consume sugary drinks [40]. The difference can be explained by the fact that sweets have a comforting value for children with ASD and that a parent tends to satisfy the child's sugar demand when the management of a certain situation becomes problematic. An unbalanced diet rich in sugar combined with the difficulty of managing oral hygiene at home, however, inevitably leads to worse PI and GI indices (especially for patients taking medications, which in our case represent about one-third of the sample) [41]. It is not surprising that the percentages of children in our sample with absence of plaque (10.09%) and with healthy gums (20.74%) are substantially smaller than the corresponding values for the non-autistic Italian population in the same age range. For example, a study conducted in one province of the Lazio region [41] reported percentages of children with no plaque and healthy gums about three times higher (32.9% and 70%, respectively). A similar study for the southern Italy reports that 70% of the children in the sample have good periodontal health [42]. The comparatively high values of the PI and GI indices observed in our sample prove how difficult it is to maintain good oral hygiene in autistic children, due to the lack of collaboration and sometimes of independence of these children in performing daily procedures like brushing their teeth. Fundamental to reversing this trend would be the inter-professional collaboration between the team of therapists and neuropsychiatrists (who treat behavioral problems of patients with ASD), and the dentists and hygienists. Such a collaboration would make it possible to include notions of food habits and oral hygiene in the programmed teaching of behavior and appropriate rules during the therapies [43].

Finally, while some authors have reported that caries experience in children with ASD is comparable or even less severe than in control groups [20–23,26,37], others [13] suggest a worsening of caries experience associated with ASD. In our case, a rigorous comparison is not possible. Our sample did not include a control group, and epidemiological studies conducted in Italy to assess caries experience in children either nationwide [44–46] or regionally [47,48] target very specific populations (children aged 4, 5, or 12 years) and are not suitable for comparison with our results given the limited sample size for these age groups (see Table 1).

5. Conclusions

Our study, despite its limitations, suggests that school children (aged between 5 and 14) in central Italy affected by ASD represent a population at risk, with high caries prevalence and caries severity. The worsening of periodontal health due to scarce domestic hygiene and food habits for this population, together with the low level of cooperativeness, already highlighted in some of the existing literature for similar populations, stress the need for establishing specific preventive and intervention programs for children with ASD. One important aspect of such a program should include a collaboration between therapists/neuropsychiatrists and dentists/hygienists. This collaboration could have very beneficial effects in terms of improving dietary and oral hygiene habits of the children and would allow an early diagnosis and treatment of their oral health problems, drastically reducing the need for general anesthesia.

There are no other epidemiological studies on caries severities of autistic children in Italy that are available to confirm the findings reported in the present work. It is hoped that a data collection network will be set up to better understand the need for treatment and outpatient management to enhance the existing approach to dental care of patients with ASD, taking into account the communicative and behavioral difficulties that these patients present [49].

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Ethical Approval: All procedures performed in studies involving human participants were carried out in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

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