CLINICAL SCIENCE

Caries in Portuguese children with Down syndrome

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OBJECTIVES: Oral health in Down syndrome children has some peculiar aspects that must be considered in the follow-up of these patients. This study focuses on characterizing the environmental and host factors associated with dental caries in Portuguese children with and without Down syndrome.

METHODS: A sibling-matched, population-based, cross-sectional survey was performed.

RESULTS: Down syndrome children presented a significantly greater percentage of children without caries, 78% *vs.* 58% of non-Down syndrome siblings. This difference in the DMFT index (number of decayed, missing and filled teeth) essentially reflects data obtained from treated teeth, for which 91% of children with Down syndrome had never had a tooth treated *vs.* 67% of siblings. This result was statistically significant, whereas results for decayed and lost teeth did not differ between Down syndrome children and their unaffected siblings. Additionally, in Down syndrome children, a delayed eruption of the second molar occurs. Down syndrome children and their siblings have similar oral hygiene habits, but a higher percentage of Down syndrome children visit a dentist before the age of three years, in comparison to their siblings. Bruxism was also more common in Down syndrome children compared to their siblings.

CONCLUSIONS: Our results show that Portuguese children with Down syndrome have lower caries rates than children without Down syndrome. This reduced prevalence may be associated with the parents' greater concern about oral health care in Down syndrome children, resulting in their taking them sooner to visit a dentist, as well as to a higher bruxism prevalence and delayed tooth eruption.

KEYWORDS: Down syndrome; Caries; Pediatrics; Prevention; Dentistry.

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INTRODUCTION

Down syndrome (DS), also called trisomy 21, is a congenital genetic disorder characterized by a triplication of material on chromosome 21. A number of medical problems, including cardiac defects, gastrointestinal anomalies, and autoimmune and dermatological disorders, as well as a significantly increased risk of acute leukemia (most pronounced in children less than five years old), have been found to be associated to DS.[1,2] The oral health problems observed in these patients show some differences compared to in the general population. A majority of published studies have reported that patients with DS have lower rates of caries than those without DS,[2-4] although several studies have found that people with and without DS share the same caries rates[5,6] and other studies have reported higher caries rates in those with DS.[5,6]

The most commonly used indicator of caries experience is an index comprising disease and treatment markers, the DMFT (Decayed, Missing and Filled Teeth). This index should be analyzed together with factors such as diet, frequency of snacking, social status, oral health in close relatives, dental awareness and past dental history.[2,6] In individuals with DS, a more accurate assessment of caries risk is likely to be obtained by also examining specific morphological and host abnormalities.[2,6,7]

The DS population is characterized by delayed and unsequenced tooth eruption in both the primary and permanent dentitions, hypodontia and microdontia.[2,7] Missing teeth contribute frequently to malocclusion, most often an open bite.[2,7] An anterior open bite and posterior cross-bite are other notable findings. Crowns may also be short, small and conical.[2,7] Bruxism (tooth-grinding) is a behavioral manifestation displayed by some children with or without DS and may further contribute to alterations in tooth morphology and mineralization.[1,2,8]

Underdevelopment of the bones of the midface, compared to those of the mandible (midface hypoplasia), is common, causing a shortened palate in the anteroposterior dimension.[2,7] However, the dental arch shape does not differ significantly from age- and sex-matched children without

DS.[8,9] The small palate causes a relative enlargement of the tongue (macroglossia), so the tongue increases its pressure against the teeth.[2,7] The tongue has poor tone, as do the oral muscles and muscles of mastication.[2,7] This poor tone is exacerbated by a pronounced open bite, largely a result of midface hypoplasia, which often causes an open-mouth posture and tongue protrusion.[2,7] Mouth breathing ensues from difficulty breathing through narrow and sometimes occluded nasal passages and is worsened by frequent upper respiratory tract infections. These skeletal and soft-tissue features may contribute to increased drooling, angular cheilitis, a dry mouth and an increased prevalence and severity of fissured lower lips and tongue.[2,9-11]

Some dental morphological anomalies previously described may be protective against caries formation. Delayed eruption and small, spaced teeth theoretically reduce caries risk by reducing the opportunity for food to stagnate between the teeth and reducing smooth surface area for colonization by cariogenic bacteria. However, if DS patients have previously had increased amounts of fermentable carbohydrates, food stagnation and poor oral hygiene associated with a dry oral cavity due to mouth breathing, then their risk may be higher than that of other children.[2,7]

Thus, the aim of this study was to characterize the environmental and host factors associated with dental caries in Portuguese children with and without DS. For that purpose, a sibling-matched, population-based, cross-sectional survey design was used.

MATERIAL AND METHODS

1. Study design and sample

A sibling-matched, population-based, cross-sectional survey design was used with the support of the Department of Dentistry of Porto University (Portugal). All DS children between 6 and 18 years of age included in a national database were invited to participate in the present study. The controls were sibling-matched based on closeness in age. The final sample consisted of 45 Caucasian sibling pairs. Data were gathered through the use of a complete questionnaire and clinical observation. The clinical examination of all participants was conducted by a medical dentist (one examiner), aided by a dental assistant. The assistant was responsible for verifying the correct completion of forms, helping and supervising parents, and filling in the clinical record after observation of the oral cavities of the children. The sample included patients with trisomy 21 and their non-affected siblings, aged 6 to 18 years, who were of both sexes and were cooperative. To calculate age, we considered the last birthday. We excluded patients without trisomy 21, uncooperative children, children more than 18 years old and those receiving antimicrobial medication in the last three weeks.

This study was approved by the ethical committee of the Department of Dentistry of Porto University. Consent was obtained according to the Declaration of Helsinki of 2002.

2. Variables

The questionnaire included socio-demographic questions concerning the subject, such as age, gender and residential location. Also, general personal and familial health status and deleterious habits were assessed. In addition, the DMFT index was determined. Other pertinent variables concerning diet, the use of fluoride elixirs at home, daily tooth brushing, and the child's first dental visit were assessed in the clinical observation.

3. Statistics

For a descriptive analysis of the sample, appropriate summary statistics were applied. The categorical variables were described using absolute and relative frequencies (%), and continuous variables were described using mean and standard deviation or median, minimum, and maximum, depending on whether their distribution was symmetric or asymmetric. When appropriate, independence tests were applied, including the chi-square or Fisher's exact test to test hypotheses regarding the categorical variables and Student's t test and the Mann-Whitney test to test hypotheses concerning continuous variables with symmetrical and asymmetrical distributions, respectively. A p-value of <0.05 was considered to be statistically significant. The analysis was performed using the statistical analysis program SPSS®, version 17.0 (Statistical Package for Social Sciences).

RESULTS

1. Sample characterization

We studied 90 participants: 45 (50%) children with DS and 45 (50%) unaffected siblings of these children closest in age.

1.1. Gender and age distribution. Table 1 shows the gender and age distribution of the sample. With respect to gender, the sample included 54% male patients and 46% female patients, and no statistically significant difference was found between the group of DS patients and their siblings. Age is an important variable in epidemiological surveys of caries: the older the participant is, the longer the exposure of teeth to cariogenic factors tends to be. The predominant age group in this study was 10 to 15 years (41%), with the median being 13 years. There were no statistically significant differences between the ages of the children with DS and those of their siblings.

1.2. Demographic distribution. Children from 15 Portuguese districts were called to participate in this study. Children from 10 districts of Portugal (60% north, 20% center and 20% south) were observed, demonstrating good adherence of the parents or legal guardians of these children. Among all the districts, Porto (n = 20) and Famalicão (n = 6) were the cities with the most participants.

1.3. Parents' education. The prevalence of caries is affected by dietary patterns, oral hygiene habits and different attitudes toward health, which are interconnected with the socio-economic and cultural status of the populations. Within the populations of the parents accompanying the children, 22% had the minimum compulsory education (9 years of school), 36% had less

Table 1 - Age and gender characterization of the sample.

	Total (n = 90)		DS (n = 45)		Siblings (n = 45)		*p*-value
Gender, *n (%)*							
Male	49	(54)	22	(49)	27	(60)	*0.290**
Female	41	(46)	23	(51)	18	(40)	
Age (years), *med (min-max)*	13	(6-20)	13	(6-20)	13	(6-18)	*0.827***
Age (years), *n (%)*							
(6, 10)	19	(21)	10	(22)	9	(20)	*0.682**
(10, 15)	37	(41)	20	(44)	17	(38)	
(15, 18)	34	(38)	15	(33)	19	(42)	

*Independent chi-square test;
**Mann-Whitney test.

education (4 or 6 years of school), and 42% had more education (12 or more years of school).

1.4. Family history and pregnancy. We investigated possible problems occurring during pregnancy (e.g., risk of miscarriage, placental abruption, etc.) as well as medication used during pregnancy. Additionally, smoking habits and hereditary diseases were also assessed. None of these factors were shown to be associated with the occurrence of DS, given that there was no statistical significance between the two groups.

1.5. Personal history — childhood illnesses. Of all the childhood diseases researched (e.g., polio, rickets, measles, pertussis, diphtheria, and mumps), only the occurrence of chicken pox differed between the groups, affecting 71% of DS children and 90% of their siblings ($p = 0.038$).

2. Deleterious habits

Bruxism was found in a higher proportion of the DS children (23%), compared with the siblings (2%), and this difference was statistically significant ($p = 0.004$). With regard to other deleterious habits, such as thumb-sucking and onychophagy, no statically significant differences were found.

3. Dietary habits

The ingestion of a cariogenic diet, evaluated based on acidic and sweet foods, was found to be similar between DS children and their siblings. The percentage of children that ate acidic food once a week or less was 98% for the DS group and 93% for their siblings, whereas the percentage of children that ate sweets once a week or less was 100% for the DS group and 95% for their siblings.

4. Oral hygiene habits

Most of the children brushed their teeth one to three times a day, i.e., 100% of the DS group and 98% of their siblings. Twice a day was the most frequent pattern, being practiced by 65% of the DS children and 47% of their siblings. If tooth-brushing occurred once a day (19% of DS children and 28% of siblings), it was most frequently performed at night (98% of DS children and 93% of siblings). The most common toothbrush bristles used were medium strength in both groups: 80% of DS children and 89% of their siblings. Eighty-four percent of children in both groups used fluoride elixirs. No statistically significant differences were found for these data between the two groups.

All participating children visited a dentist for the first time before the age of 12. However, a higher percentage of DS children visited a dentist before three years of age (77%) in comparison to their siblings (34%, $p<0.001$).

5. Clinical evaluation

Table 2 represents the data obtained from the clinical evaluations with regard to the DMFT index. The children were divided into those with a DMFT index equal to zero, considered caries-free, and those with a DMFT index equal to or greater than one. Interestingly, the DS group presented a significantly higher percentage of children within the caries-free group: 78% *vs.* 58% of siblings ($p = 0.042$). This difference in the DMFT index essentially reflects the data obtained for treated teeth, in which 91% of children with DS had never had a tooth treated, while 67% of the siblings had never had a tooth treated. This result was statistically significant ($p = 0.004$), whereas the results obtained for decayed and lost teeth did not differ between DS children and their siblings ($p>0.05$).

Table 2 - Organized data for DMFS index evaluation.

	Total (n = 90)		DS (n = 45)		Siblings (n = 45)		*p*-value
	n	(%)	n	(%)	n	(%)	
Decayed, n (%)							
0	75	(83)	39	(87)	36	(80)	*0.396**
≥1	15	(17)	6	(13)	9	(20)	
Lost, n (%)							
0	85	(94)	42	(93)	43	(96)	*1.000****
≥1	5	(6)	3	(7)	2	(4)	
Treated, n (%)							
0	71	(79)	41	(91)	30	(67)	*0.004**
≥1	19	(21)	4	(9)	15	(33)	
DMFS, n (%)							
0	61	(68)	35	(78)	26	(58)	*0.042**
≥1	29	(32)	10	(22)	19	(42)	

*Independent chi-square test;
***Fisher's exact test.

Additionally, during the clinical evaluation, it was possible to note that, in children with DS, there was delayed eruption of the second molar. The proportions of children with no erupted teeth within the DS group *vs.* among their siblings were as follows: tooth 17, 64% *vs.* 40% ($p = 0.02$); tooth 27, 69% *vs.* 44% ($p = 0.019$); tooth 37, 64% *vs.* 42% ($p = 0.035$); and tooth 47, 60% *vs.* 40% ($p = 0.058$).

DISCUSSION

DS is the one of the most common causes of genetic intellectual disability in the industrialized world, with an incidence of 10 to 12 per thousand in Portugal.[8] Despite this observation, our current understanding of the oral health problems experienced by this group is inadequate.[8,12-14] Our epidemiological study was able to demonstrate that Portuguese children with DS have lower caries rates than do children without DS.

The sample used in the present study presented a good distribution with respect to age, sex and race parameters. Regarding geographic distribution, our sample was more concentrated in the north of Portugal, given that the clinical evaluation was performed in the city of Porto. Most of the families that composed our sample belonged to the medium socio-economic level. The fact that the socio-economic level of the parents of these children was good may explain the satisfactory participation rate of the families in this study. The children's cooperation was one of the inclusion criteria, even though the clinical evaluation was particularly difficult for those children with DS to tolerate.

Regarding personal history and childhood illness, the only difference observed between the DS children and their siblings was a lower prevalence of chicken pox in DS children. This lower prevalence could be due to the fact that these children grew up in a more protected environment; however, chicken pox prevalence and caries development do not seem to be related.

In the present study, the method used to detect dental caries was based on a clinical examination with a probe and mirror and on the DMFT index, according to World Health Organization (WHO) standards. According to several studies, this method is efficient for the detection of dental cavities, but

not for non-cavitated lesions.[15-20] With the inclusion of non-cavitated lesions, it may be possible to obtain a better idea of disease prevalence, resulting in a better understanding of treatment needs.[2,21,22] However, the DMFT index approach was preferred due to its objectivity and the large sample sizes.

Interestingly, our data clearly show that Portuguese children with DS have lower rates of caries than do children without DS, given that the DS group, in comparison to their siblings, presented a significantly higher percentage of children within the caries-free group who had never had a tooth treated. This difference could be due to several factors (e.g., social-economic status, diet, oral hygiene, specific morphology), as referred to in the introduction.

The prevalence of tooth decay is linked to socio-economic and cultural factors of the population. In this study, the controls were the siblings of DS children, avoiding skewed results due to differing socio-economic status.

Regarding dietary habits, many epidemiological studies have linked the incidence caries and food habits, identifying both protective and aggravating factors. Consistency and viscosity, the cooking process, and even the time of ingestion of food containing simple carbohydrates changes the food's cariogenicity.[8,16,18-20] The present study investigated in qualitative terms the dietary habits of the sample population. Interestingly, the consumption of sweet or acidic food was similar between the DS children and their siblings. Therefore, diet cannot be responsible for the differences found regarding caries rates in these two groups.

Different oral hygiene habits between DS children and their unaffected siblings may also explain the difference in caries rates, but in our study both groups had the same frequency of brushing, used the same kinds of brushes, and both used fluoride elixirs. However, some studies have reported that cleaning of the interproximal surface of the teeth in children with DS is easier than in people without DS due to the common presence of microdontia with diastema.[2,20] In addition, several studies have found that 50% of young children and 23% of older children with DS were incapable of brushing their teeth alone and required parental help, making efficient brushing more likely in children with DS than in children without DS. However, is difficult to prove that this fact is responsible for the difference found in the DMFT index between DS children and their siblings. Nevertheless, the present study found that Portuguese DS children visit a dentist sooner than do children without DS. This fact may indicate that parents are more concerned about oral health care in this special group of children.

Two other factors revealed by this study that could justify the lower caries rates were that DS children had a higher prevalence of bruxism and that they had delayed tooth eruption. Higher bruxism rates may be explained by the fact that children with DS have more occlusion abnormalities, have flaccid support ligaments of the temporo-mandibular joint and, normally live in a state of chronic anxiety.[7,16-18] This tooth friction induces smoother occlusal surfaces, allowing better tongue self-cleaning and also facilitating oral hygiene.[2] Delayed tooth eruption is in agreement with previous studies finding that the eruption of both the temporary dentition and the permanent teeth are delayed 6 to 18 months in DS children.[2,20,22] The fact that the teeth erupted later may also be a factor in explaining the lower presence of cavities, taking into account that the teeth are thus subject to cariogenic factors over a shorter time period.

CONCLUSION

Our epidemiological study was able to demonstrate that Portuguese children with DS have lower caries rates than do children without DS. This reduced caries prevalence in DS children may be associated with parents' greater concern about oral health care in DS children, resulting in their taking them sooner to visit a dentist, as well as with the higher bruxism prevalence and delayed tooth eruption in DS children.

Nevertheless, more studies are necessary to identify other protective factors against caries in these patients.

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