

ABSTRACT

The purpose of this epidemiological study was to determine the differences in the prevalence of caries between individuals with Down syndrome (DS) and their siblings. A sibling-matched, population-based and cross-sectional survey was performed. This study involved 138 (62%) children with DS and 86 (38%) siblings, aged 2–26. The children were compared in different subgroups: [2, 6], [6, 12], and [13, 26]. Data was gathered through the use of a complete questionnaire and clinical observation. Data analysis was performed by using SPSS® v.18.0 software with any p value $< .05$ considered as significant. The DS group presented a significantly higher percentage of children within the caries-free group: 72% versus 46% of the siblings group ($p < .001$). In the age gap [2, 6] the median value of DMFT was the same in both groups ($p = .918$). In the age gap [6, 12] the median value of DMFT in the DS group was 0 and in the siblings group was 1 ($p = .004$). In the age gap [13, 26] the median value of DMFT in the DS group was 0, whereas in the siblings group the median value was 3, which constitutes a significantly high difference ($p = .003$). The results of this study suggest that Portuguese children with DS have lower caries prevalence than their siblings.

KEY WORDS: dental caries, Down syndrome, oral pathology

Comparative study between dental caries prevalence of Down syndrome children and their siblings

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Introduction

Down syndrome (DS) or trisomy 21 represents the most common chromosomal abnormality associated with intellectual impairment.^{1,2}

Clinically, it is characterized by generalized hypotonia, neurological changes, structural cardiopathy, respiratory problems and a greater risk of infection, dental anomalies and orofacial dysmorphism, which therefore requires special attention in the dental treatment of these patients.²⁻⁵

DS children have specific orofacial features. The most common oral findings in these children include mouth breathing, open bite, macroglossia, fissured lips and tongue, delayed teeth eruption, missing and malformed teeth, microdontia, crowding, malocclusion and bruxism.⁶⁻⁹

Caries is the most common chronic disease of childhood, and can have a direct and devastating impact on the patients' overall health, especially for those with certain systemic health problems.^{10,11} Caries is a multifactorial infectious disease and there are a lot of different factors that affect its occurrence.⁸

The most commonly used indicator of caries experience is an index comprising disease and treatment markers, the decayed, missing, and filled teeth

(DMFT). This index should be analyzed together with factors such as diet, frequency of snacking, social status, oral health of close relatives, dental awareness and past dental history.^{5,12,13}

The oral health problems observed in these patients show some variances when compared to the general population. The majority of published studies has reported that patients with DS have lower rates of caries than those without DS.^{5,14,15} Nonetheless, several studies have found that people with and without DS share the same caries rates while other studies have reported higher caries rates in those with DS.^{12,16}

The main objective of this study was to characterize the caries prevalence in a Portuguese population with DS and to compare its distribution with their siblings.

Methods

The sibling-matched, population-based, and cross-sectional survey design used in this study was applied with the support of the Department of Dentistry of Oporto and Lisbon's Universities (Portugal). This study was approved by the ethical committee of the Department of Dentistry of University of Oporto. Consent was obtained according to the Declaration of Helsinki of 2002.

The recruitment of children with DS was done by mailing detailed study information to the main organizations working with this population in Portugal. Two centers were set up, one in the south, Lisbon, and the other one in the north, Oporto. The inclusion criteria implemented for the DS group were: (1) cytogenetic diagnosis of trisomy 21, (2) adequate cooperation, and (3) informed consent from the children's legal representatives. Study inclusion criteria for the control group were: (1) be a sibling of children with DS, (2) adequate cooperation, and (3) informed consent from the children's legal representatives. The control groups were sibling-matched based on closeness in age. DS children without siblings were also included. In principle, children who did not allow for a correct evaluation of the oral cavity would be excluded from the study but that did not happen as all children were collaborative. Therefore, no child was excluded.

The sample included 138 children with DS (62%) and 86 siblings of these children (38%), aged between 2 and 26 years old. The children were compared in different subgroups: [2, 6], [6, 12], and [13, 26].

The parents were told about the objectives of the study. Those who agreed to participate signed an informed consent form. The information was gathered through the use of a complete questionnaire and clinical observation. The questionnaire included socioeconomic questions concerning the subject, such as age, gender, and residential location.

The clinical examination of every participant was carried out by a certified dentist (one examiner) aided by two dental assistants. One dental assistant

was responsible for verifying the correct completion of forms, helping and supervising parents. The other dental assistant was in charge of filling clinical record after the dentist's observation of the children's oral cavities. To perform dental observation the dentist used an intramirror, a probe and compressed air. This observation was made with the help of artificial light. The observations registry was carried out tooth by tooth, through a geometric-type odontogram and the DMFT index was determined. According to the criteria and recommendations established by the World Health Organization (WHO),¹⁷ the study of the population of DMFT has epidemiological interest. No radiographic examination was undertaken.

For the socioeconomic characterization of the sample, this study adopted the International Classification of Graffar. All the data was grouped in three classes: high (class I and II), average (class III), and low (class IV and V).

The population with DS presents a delayed dental eruption of 1–2 years,^{7,18,19} therefore reducing the time exposure of teeth to caries' etiological factors. Bearing this fact in mind, and with the purpose of reducing statistical bias, the two groups in different age gaps were compared, to achieve a higher accuracy in the factor "permanence time in oral cavity."

To determine the risk factors associated with caries prevalence the study calculated Odd Ratios and the respective confidence intervals at 95% resorting to logistic regression. To adjust the odds ratio to the DMFT for the variables of group, sex, age (as continuous) and Graffar's social classification, the study applied the multivariable logistic regression with the Enter method adapted to these same variables.

To conduct a descriptive analysis of the sample the 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, independent tests were applied. These included the chi-square for hypotheses regarding the categorical variables and Student's *t*-test and the Mann–Whitney test for hypotheses concerning continuous variables with symmetrical and asymmetrical distributions, respectively. A *p*-value of .05 was considered to be statistically significant. The analysis was performed using the statistical analysis program SPSS®, version 18.0 (Statistical Package for Social Sciences).

Results

The sample included 224 children (138 with DS, 86 siblings) aging between 2 and 26 years old. Children from 15 districts of Portugal were observed.

The prevailing age group of the sample was that of 6–12 (48%) in both groups. The mean age of the total sample was 11 years old (10 years old for the DS group and 12 years old for the siblings group). Comparing the children's age groups percentages, they were very similar between the DS group and the siblings group (Table 1). Statistically, there was no significant difference ($p = .713$).

Approximately half of the sample was male (54%), with no significant statistical differences in the gender element in both groups ($p = .670$).

Only 195 of the children's legal representatives answered to the Graffar's social classification. In the total sample (DS and siblings), 145 (74%) belonged to a high socioeconomic background, 43 (22%) belonged to an average socioeconomic background and 7 (4%) belonged to a low socioeconomic background. The percentage of children in each social group was very similar in both groups (Table 1). Thus, statistically, there was no significant difference ($p = .350$).

Table 2 shows the data obtained from the clinical evaluations with regards 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 or greater than one. The DS group presented a significantly higher percentage of children within the caries-free group:

Table 1. Characterization of the sample.

	Total (n = 224)		DS (n = 138; 62%)		Siblings (n = 86; 38%)		p Value
	n	(%)	n	(%)	n	(%)	
Gender							
Male	121	(54)	73	(53)	48	(56)	.670 ^a
Female	103	(46)	65	(47)	38	(44)	
Age (years), med (min-max)	10	(2-26)	10	(2-20)	11	(2-26)	.170 ^b
Age (recoded)							
[2, 6]	37	(16)	25	(18)	12	(14)	.713 ^a
[6, 12]	107	(48)	65	(47)	42	(49)	
[13, 26]	80	(36)	48	(35)	32	(37)	
Graffar	n = 195		n = 119		n = 76		
Class I-II	145	(74)	84	(71)	61	(80)	.350 ^a
Class III	43	(22)	30	(25)	13	(17)	
Class IV-V	7	(4)	5	(4)	2	(3)	

med = Median; min = minimum; max = maximum.
^aIndependent chi-square test.
^bMann-Whitney test.

Table 2. Organized data for DMFT index evaluation.

DMFT	Total (n = 224)		DS (n = 138; 62%)		Siblings (n = 86; 38%)		p Value
	n	(%)	n	(%)	n	(%)	
0	139	(62)	99	(72)	40	(46)	<.001 ^a
≥1	85	(38)	39	(28)	46	(54)	

^aIndependent chi-square test.

Table 3. DMFT parameters.

	Total (n = 224)		DS (n = 138; 62%)		Siblings (n = 86; 38%)		p Value
	n	(%)	n	(%)	n	(%)	
Decayed teeth, med (min-max)	0	(0-15)	0	(0-8)	0	(0-15)	.008 ^a
Missing teeth, med (min-max)	0	(0-11)	0	(0-6)	0	(0-11)	<.001 ^a
Filled teeth, med (min-max)	0	(0-4)	0	(0-4)	0	(0-2)	.820 ^a

med = Median; min = minimum; max = maximum.
^aMann-Whitney test.

72% versus 46% of the siblings group ($p < .001$).

In the evaluation of the number of decayed teeth, it was possible to observe that the median value was 0 in both groups. Nevertheless, in the DS group the minimum and maximum values ranged from 0 to 8, whereas in the siblings group they ranged from 0 to 15. Thus, there was a statistically significant difference ($p = .008$). Concerning the number of filled

teeth, we also found a statistically significant difference. The DS group presented results of 0-6 teeth, whereas their siblings presented values of 0-11 filled teeth ($p < .001$). Concerning the missing teeth, there was no difference between the two groups ($p = .820$; Table 3).

When comparing the two groups, children with DS and their siblings, in the different age gaps (Table 4) no difference was found between DMFT in the

age gap [2, 6], being the median value 0 attributed to both groups ($p = .918$). In the age gap [6, 12], it was found that the median value of DMFT in children with DS was 0 and 1 in their siblings, with a highly significant difference ($p = .004$). In the age gap [13, 26], it was found that the median value of DMFT in children with DS was 0, whereas in the siblings group the median value was 3, which was a significantly high difference ($p = .003$).

The DS population is characterized by delayed tooth eruption (1-2 years) in both the primary and permanent dentitions.^{7,18,19} The fact that the teeth erupted later may also be a factor that affects a lower presence of cavities, taking into account that the teeth are thus subject to cariogenic factors over a shorter period of time. To match the time the teeth are exposed to cariogenic factors in the oral cavity 1 or 2 years were increased to the age gaps of children with DS, in relation to the siblings group, as can be verified in Tables 5 and 6.

Taking into account a delay of 1 year in dental eruption in the DS group, the siblings group of the age gap [2, 6] was compared with the DS children in the gap [3, 7] (Table 5). The results show that there were no differences between DMFT values ($p = .971$). In the siblings

Table 4. Organized data for DMFT index evaluation, by age gaps.

	Total (n = 224)		DS (n = 138; 62%)		Siblings (n = 86; 38%)		p Value
	med	(min-max)	med	(min-max)	med	(min-max)	
DMFT							
[2, 6]	0	(0-6)	0	(0-1)	0	(0-6)	.918 ^a
[6, 12]	0	(0-15)	0	(0-10)	1	(0-15)	.004 ^a
[13, 26]	1	(0-17)	0	(0-11)	3	(0-17)	.003 ^a

med = Median; min = minimum; max = maximum.
^aMann-Whitney test.

Table 5. Organized data for DMFT index evaluation, by age gaps, giving a year beforehand to the age gaps in DS children.

Age gaps						
Siblings	[2, 6] (n = 12)		[6, 12] (n = 42)		[13, 26] (n = 32)	
DMFT, med (min-max)	0	(0-6)	0	(0-15)	3	(0-17)
DS	[3, 7] (n = 23)		[7, 13] (n = 75)		[14, 26] (n = 31)	
DMFT, med (min-max)	0	(0-1)	0	(0-10)	0	(0-11)
p	.971		.023		.009 ^a	

med = Median; min = minimum; max = maximum.
^aMann-Whitney test.

Table 6. Organized data for DMFT index evaluation, by age gaps, giving 2 years beforehand to the age gaps in DS children.

Age gaps						
Siblings	[2, 6] (n = 12)		[6, 12] (n = 42)		[13, 26] (n = 32)	
DMFT, med (min-max)	0	(0-6)	1	(0-15)	3	(0-17)
DS	[4, 8] (n = 26)		[8, 14] (n = 71)		[15, 26] (n = 27)	
DMFT, med (min-max)	0	(0-6)	0	(0-10)	0	(0-11)
p	.612		.041		.007 ^a	

med = Median; min = minimum; max = maximum.
^aMann-Whitney test.

group of the age gap [6, 12] it was found that the median value was 0, with minimum and maximum values of 0-15 respectively, whereas in the DS group of the age gap [7, 13] the median value was 0, but with 10 as the maximum value. Thus, statistically, there was a significant difference ($p = .023$). The siblings aged [13, 26] presented a median value of DMFT = 3, whereas DS children aged [14, 26] presented a median value of DMFT = 0, with a highly significant difference ($p = .009$).

The previous exercise was repeated taking into account a delay of 2 years in dental eruption in DS children (Table 6). Comparing the siblings group in the age gap [2, 6] with the children with DS in the age gap [4, 8], the results show that the median value of DMFT was 0 for both groups, varying from 0 to 6. Thus, this difference was not statistically significant ($p = .612$). In the siblings group of the age gap [6, 12], the median value was 1, with minimum and maximum values of 0-15, respectively, whereas in

children with DS of the age gap [8, 14] the median value was 0, but with 10 as the maximum value. Thus, statistically, there was a significant difference noted ($p = .041$). The siblings aged [13, 26] presented a median value of DMFT = 3, whereas the DS children aged [15, 26] presented a median value of DMFT = 0, with a highly significant difference ($p = .007$).

To have a better approach to the potential differences in tooth eruption age, this study considered a logistic regression analyses with caries experience (DMFT yes/no) as the dependant variable, and group (DS vs. Siblings), gender, age, and Graffar's social classification as independent variables (Table 7). The DS group had a lower risk of having DMFT than the siblings group.

Discussion

The characterization of the level of oral health in a population is important in order to establish priorities concerning preventive and therapeutic activities. In health care planning it is essential to identify and quantify the necessities of the target population.

Although the majority of the children were from Oporto and Lisbon, children from 15 districts of Portugal were observed, which clearly shows the receptiveness of the children's parents to this study. The sample used in this study presented a good distribution with respect to age and sex parameters.

In this study, the prevalence of caries-free in the total sample was 62%. This value shows that caries prevalence was not significantly high, a result that may demonstrate that this population belongs, in general, to a high socioeconomic background. Generally speaking, a child growing up in a low socioeconomic background has underprivileged social, economic and educational conditions, which may limit access to professional health care services. Even so, in similar socioeconomic background, DS children presented a lower caries prevalence.

Vigild¹⁸ examined dental caries experience among institutionalized and noninstitutionalized intellectual disabled

Table 7. Logistic regression analyses with caries experience (DMFT yes/no) as the dependant variable, and group (Down syndrome vs. siblings), gender, age, Graffar's social classification as independent variables.

	Total (n = 224)		DMFT No (n = 139; 62%)		DMFT Yes (n = 85; 38%)		<i>p</i> ^a	OR ^a	IC 95%		OR ^b	IC 95%	
Group, n (%)													
DS	138	(62)	99	(72)	39	(28)	<.001	0.343	0.195;	0.601	0.324	0.168;	0.625
Siblings	86	(38)	40	(46)	46	(54)		1.000	–		1.000	–	
Gender, n (%)													
Male	121	(54)	76	(63)	45	(37)	.800	1.000	–		1.000	–	
Female	103	(46)	63	(61)	40	(39)		1.072	0.624;	1.842	1.160	0.607;	2.218
Age, med (min-max)	10	(2–26)	9	(2–25)	13	(2–26)	<.001 ^d	1.150	1.083;	1.221	1.152	1.076;	1.234
Graffar, n (%)													
Class I–II	145	(74)	93	(64)	52	(36)	.671	1.000	–		1.000	–	
Class III	43	(22)	24	(56)	19	(44)		1.416	0.709;	2.826	1.667	0.780;	3.563
Class IV–V	7	(4)	4	(57)	3	(43)		1.341	0.289;	6.225	1.197	0.211;	6.801
med = median; min = minimum; max = maximum.													
^a Independent chi-square test.													
^b OR = odds ratio univariate.													
^c OR = odds ratio adjusted to the group, gender, age (continuous), and Graffar's social classification variables.													
^d Mann–Whitney test.													

children with DS. Furthermore, caries were assessed in a control group of intellectual disabled individuals without DS. A total of 288 children, 6–19 years old, were examined. The prevalence of caries-free individuals was 40%. The study also confirmed that institutionalized DS subjects had a lower caries prevalence than those living at home, probably as a result of differences in the environment.

Brown and Cunningham²⁰ examined 80 institutionalized DS individuals (1–39 years) and they found that 44% of the sample was caries-free. Stabholz *et al.*²¹ examined the prevalence of dental caries in 32 DS children, aged 8–13 and they found 84% of them to be caries-free. Barnett *et al.*²² compared the prevalence rates of periodontitis and dental caries in 30 DS patients and 30 matched intellectual disability controls. The results revealed a greater prevalence of periodontitis and a lower prevalence of dental caries in DS patients compared to the intellectual disability controls. Morinushi *et al.*²³ evaluated the incidence of dental caries in 75 DS children in the age group of 2–18 years old. The frequency of all children who were caries-free was 46.1%

while in children under 5 years old that frequency rose to 61.4%.

When caries prevalence was evaluated in the age gaps, there were no differences recorded between both groups of the age gap [2, 6], which can be related to the reduced sample dimension of this age gap. When comparing the other age gaps, this study found that the DS group had a lower prevalence of dental caries.

It is well-known that caries increases with age¹¹ and in the siblings group the study showed that the median value of the DMFT increased with age, while in the DS group the median value remained the same (zero), even with aging.

This epidemiological study suggests that Portuguese children with DS have lower caries rates than children without DS. The literature attributes the low prevalence of caries in individuals with DS to factors such as eruptive pattern, higher prevalence of bruxism, dental morphology, salivary composition and differences in the composition of the microbiota.^{19,23,24}

The method used to detect dental caries was based on a clinical examina-

tion using a probe and mirror and on the DMFT index, according to WHO standards. According to several studies, this method is efficient for the detection of dental cavities, but not for noncavitated lesions. With the inclusion of noncavitated lesions, it may be possible to obtain a better idea of disease prevalence, resulting in a better understanding of treatment needs.⁵ However, the DMFT index approach was preferred due to its objectivity and the large sample size.

Even though the study included children from 15 different districts, the majority of them belonged to the surrounding areas of the only two observation locations, Oporto and Lisbon. As such, this might also be considered a limitation to the present study, as different observation locations might have allowed a wider representative group.

Conclusion

The results of this study suggest that Portuguese children with DS have lower caries rates than do children without DS.

Author Contributions

Viviana Macho: responsible for the conception and design, data collection and manuscript redaction; Miguel Palha: responsible for the critical revision of its contents; Ana Paula Macedo: responsible for the critical revision of its contents; Orquidea Ribeiro: responsible for the critical revision of its contents; Casimiro Andrade: responsible for the critical revision of its intellectual contents and final approver of the version to be published.

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