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Down syndrome: a risk factor for malocclusion severity?

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Abstract: The aims of the present study were to compare aspects related to malocclusion between individuals with Down syndrome (DS) and a control group, establish malocclusion severity, and identify determinant factors. A total of 120 individuals (60 with DS and 60 with no physical or mental impairment), were included in the study. Data were collected through interviews, analyses of the medical charts, and oral examinations. The criteria of the Dental Aesthetic Index were used for the diagnosis of malocclusion. Chi-square test ($p \leq 0.05$) and multivariate logistic regression were used for comparisons between the two groups and to determine the association between the dependent (malocclusion severity) and independent variables. Statistically significant differences were found between the two groups for the following variables: missing teeth, diastema, overjet, mandibular protrusion, anterior open bite, posterior crossbite, facial type, lip incompetence, and Angle classification. DS, a history of premature birth, and long face pattern were found to be associated with malocclusion severity. Individuals with DS exhibited more occlusal problems than those in the control group.

Keywords: Malocclusion; Down Syndrome; Orthodontics.

Introduction

Down syndrome (DS), also known as Trisomy 21, is the most common chromosomal disorder in humans, affecting 1–2 individuals out of every 1000 live births.[1,2,3] Patients with this syndrome exhibit cognitive impairment as well as bone growth disorders and generalized muscle hypotonia.[4,5] Moreover, oral health problems, such as inadequate oral hygiene, periodontal disease, caries, malocclusion, and tooth loss, are more prevalent in this group of individuals than in the general population.[6,7,8,9] Malocclusion, in particular, exerts a considerable negative impact on the quality of life, causing problems related to the performance of daily activities, such as speech, swallowing, and chewing, and discrimination based on physical appearance.[10,11]

Patients with DS exhibit alterations such as an abnormal positioning of the tongue, craniofacial deformities (reduction in maxilla and mandible size, and narrow oropharynx), dental alterations (number and size of teeth), and muscle disorders.[12,13,14] All these factors contribute toward the development of transversal and vertical alterations in the occlusion, such

as anterior open bite, posterior or anterior crossbite, and the proclination of the anterior teeth.[2,3]

The prevalence of malocclusion and associated factors have been assessed in individuals with DS.[8,12,13] However, a critical reading of the literature reveals that there is a gap in knowledge concerning the determinant factors associated with the malocclusion establishment and severity. Many studies have failed to address certain individual, social, and behavioral aspects, such as age, birth weight, a history of premature birth, breathing pattern, and facial type.[5,14,15] Moreover, methodological limitations, including insufficient sample size, inadequate data acquisition instruments, study designs with various sources of bias, and the use of only descriptive and comparative statistical analyses, have been encountered in the past. Thus, studies that can provide evidence with greater scientific weightage are of particular importance for the development of public policies aimed at preventive strategies and oral health promotion.

The aims of the present study were to compare aspects related to malocclusion between individuals with DS and a control group, establish malocclusion severity, and identify the determinant factors.

Methodology

The present study involved a sample made up of 120 individuals; 60 with DS (37 males and 23 females, average age of 14.73 years) and 60 with no physical or mental impairment but affected by different types of malocclusion (control group: 19 males and 41 females, average age of 12.18 years). The patients in the control group were randomly selected from individuals awaiting orthodontic treatment in the Course of Orthodontics specialization at the School of Dentistry in Itaúna, Brazil. Only those individuals who had not yet been submitted to orthodontic intervention were included in the study.

Data were collected through interviews, assessments of medical charts for the confirmatory diagnosis of DS, and oral examinations. Information pertaining to age, gender, mother's schooling, weight, and a history of premature birth were collected from children's mothers during the interview. Oral examinations were performed by an orthodontist. Prior to the assessment and diagnosis of malocclusion based on the criteria of the Dental Aesthetic Index (DAI), the orthodontist participated in a calibration exercise involving 12 individuals who did not belong to the main study and achieved high agreement values (maximal and minimal kappa values of 1.00 and 0.81, respectively). The DAI provides four outcome possibilities: mild malocclusion or absence of abnormality, for which treatment is not necessary (DAI ≤ 25); defined malocclusion, for which treatment is elective (DAI = 26-30); severe malocclusion, for which treatment is highly desirable (DAI = 31–35); and very severe or debilitating malocclusion, for which treatment is fundamental (DAI ≥ 36).[16] Because all participants exhibited some type of malocclusion, the DAI score was dichotomized as moderate (DAI ≤ 30) and severe (DAI > 30) malocclusion. Malocclusions were clinically categorized as Class I, Class II, or Class III, based on the Angle classification. During the clinical examination, posterior crossbite, facial type, and dentition (mixed or permanent) were also determined.

A lip competence examination was performed based on the method described by Ballard;[17] the mandible was in the physiological resting posture and lips were in the juxtaposition (sealed) with no contraction of the orbicular muscles of the mouth or mentalis. Lip incompetence was recorded when the individual required vigorous contractions of the orbicular muscles of the mouth and mentalis to achieve a lip seal. In children, lip incompetence and breathing patterns (nasal or mouth) were assessed during the clinical examination and by interviewing the mothers when the child was not aware of being observed, thereby revealing the inherent behavior.

Data analysis was performed using the Statistical Package for Social Sciences (SPSS for Windows, version 17.0, SPSS Inc., Chicago, USA) and included frequency distribution and association tests. Associations between the dependent (malocclusion severity) and independent variables (gender, age, birth weight, a history of premature birth, mother's schooling, breathing pattern, dentition type, and facial type) were determined using the chi-square test ($p \leq 0.05$). Variables with a p-value of ≤ 0.20 were incorporated into the multivariate logistic regression model (forward stepwise procedure). The study was approved by the Ethics Committee with the protocol number

0004.0.380.000–09. Parents/guardians signed the terms of informed consent authorizing their child's participation in the study.

Results

Individuals with DS had more number of missing teeth and a greater occurrence of mandibular protrusion, anterior open bite (mm), posterior crossbite, and lip incompetence. As shown in Table 1, individuals in the control group had a greater occurrence of diastema (≥ 2 mm) and overjet (≥ 4 mm). Differences were also found with regard to facial type and Angle classification between the two groups. Short face pattern and Angle Class III malocclusion were more frequent among the individuals with DS, whereas long face pattern and Angle Class II malocclusion were more frequent among the individuals in the control group (Table 1).

Malocclusion severity was greater among the individuals with DS than among those in the control group ($p = 0.028$). Considering individual, social, and behavioral factors, there was a greater frequency of severe malocclusion among individuals aged > 10 years and among those with a history of premature birth, lip incompetence, mouth breathing pattern, and long face pattern (Table 2). After the adjustment of the model, logistic regression revealed that DS, a history of premature birth, and facial type (long face) were associated with a greater malocclusion severity (Table 3), regardless of the age or gender.

Discussion

The majority of studies assessing factors associated with malocclusion in individuals with DS only provide descriptive and comparative statistical analyses and fail to determine malocclusion severity in these individuals.[8,12,13] For this reason, the control group was comprised of individuals without DS and seeking orthodontic treatment, rather than individuals unaffected by malocclusion or those with similar malocclusions.

The most frequent malocclusions among the individuals with DS were mandibular protrusion, anterior open bite, and posterior crossbite. These findings confirm the results of previous studies reporting high prevalence values for malocclusions stemming from vertical and transversal occlusal alterations.[2,3,14,18] Such alterations are associated with insufficient bone development, orofacial muscle hypotonia, and the positioning of the tongue.[2,14,19,20] Muscle hypotonia associated with a reduced volume of the oral cavity and characterized by a deep, atresic palate may lead to a tendency toward habitually projecting the tongue against the teeth or outside the mouth.[5,18,21] Abnormal function and the position of the tongue can affect tooth eruption causing anterior open bite, tooth alignment, and the arch shape.[12,22] However, these variables were not assessed in the present study. A pervious study[23] using the same control group identified that patients with cerebral palsy also had more anterior open bite, suggesting that muscle impairment in both disorders contributes toward malocclusion.

Patients with DS generally have a short face pattern and a reduced development of the middle third of the face, resulting in a Class III occlusal relation.[8,24,25] The present study corroborates these findings. However, patients with long face pattern also exhibited a greater chance of developing severe malocclusion. The long face pattern is associated with muscle hypotonia and a tendency toward the downward rotatation of the mandible, favoring the development of Angle Class II malocclusion.[26]

Altered breathing pattern is strongly associated with malocclusion.[20] Góis *et al*.[27] found that children with mouth breathing patterns have a 10-fold greater chance of exhibiting malocclusion compared with those with nasal breathing patterns. In the present study, mouth breathing was associated with both malocclusion prevalence and severity because 85% of the mouth breathing patients exhibited severe malocclusion. In the logistic regression analysis, however, this variable did not adjust to the model because most individuals with DS also exhibited mouth breathing patterns. Mouth breathing patterns are more frequent due to orofacial muscle hypotonia and an absence of the lip seal.[12]

Children with a history of premature birth had a greater chance of developing severe malocclusion. A recent systematic review reported scientific evidence for altered palatal morphology among children born prematurely but with contradictory findings.[28] However,

Table 1. Univariate analysis considering aspects related to malocclusion in the Down syndrome group and control group without mental impairment.

	Group		
	Control n (%)	Down syndrome n (%)	*P**
Missing teeth			
None	59 (60.2)	39 (39.8)	< 0.001
At least one	1 (4.5)	21 (95.5)	
Crowding			
None	31 (44.9)	38 (55.1)	0.196
1 or 2 segments	29 (56.9)	22 (43.1)	
Spacing			
None	34 (48.6)	36 (51.4)	0.711
1 or 2 segments	26 (52.0)	24 (48.0)	
Diastema (mm)			
< 2	47 (46.1)	55 (53.9)	0.041
≥ 2	13 (72.2)	5 (27.8)	
Maxillary irregularity (mm)			
< 2	48 (50.0)	48 (50.0)	1.000
≥ 2	12 (50.0)	12 (50.0)	
Mandibular Irregularity (mm)			
< 2	50 (48.1)	54 (51.9)	0.283
≥ 2	10 (62.5)	6 (37.5)	
Overjet (mm)			
< 4	46 (44.2)	58 (55.8)	0.001
≥ 4	14 (87.5)	2 (12.5)	
Mandibular protrusion			
No	59 (56.7)	45 (43.3)	< 0.001
Yes	1 (6.3)	15 (93.8)	
Anterior open bite (mm)			
< 2	57 (56.4)	44 (43.6)	0.001
≥ 2	3 (15.8)	16 (84.2)	
Posterior crossbite			
Absent	53 (65.4)	28 (34.6)	< 0.001
Present	7 (17.9)	32 (82.1)	
Facial type			
Short face	1 (4.2)	23 (95.8)	<0.001
Average	45 (58.4)	32 (41.6)	
Long face	14 (73.7)	5 (26.3)	
Lip incompetence			
No	44 (58.7)	31 (41.3)	0.014
Yes	16 (35.6)	29 (64.4)	
Angle classification			
Class I	31 (70.5)	13 (29.5)	< 0.001
Class II	25 (61.0)	16 (39.0)	
Class III	4 (11.4)	31 (88.6)	
Dentition			
Mixed	44 (55.0)	36 (45.0)	0.121
Permanent	16 (40.0)	24 (60.0)	

*Chi-square test ($p \leq 0.05$)

Table 2. Univariate analysis of association between malocclusion severity and individual, social, and behavioral variables.

	Malocclusion severity (DAI)		
	Moderate	Severe	*P**
Group			
Control	35 (58.3)	25 (41.7)	0.028
Down syndrome	23 (38.3)	37 (61.7)	
Sex			
Male	23 (41.1)	33 (58.9)	0.136
Female	35 (54.7)	29 (45.3)	
Age (y)			
≤10	37 (59.7)	25 (40.3)	0.010
>10	21 (36.2)	37 (63.8)	
Birth weight (g)			
≥ 2500	29 (44.6)	36 (55.4)	0.282
< 2500	9 (60.0)	6 (40.0)	
Premature birth			
No	52 (52.0)	48 (48.0)	0.030
Yes	4 (23.5)	13 (76.5)	
Mother's schooling (y)			
> 4	30 (46.9)	34 (53.1)	0.733
≤ 4	28 (50.0)	28 (50.0)	
Breathing			
Nasal	55 (55.0)	45 (45.0)	0.001
Mouth	3 (15.0)	17 (85.0)	
Lip incompetence			
No	43 (57.3)	32 (42.7)	0.011
Yes	15 (33.3)	30 (66.7)	
Dentition			
Mixed	43 (53.8)	37 (46.3)	0.093
Permanent	15 (37.5)	25 (62.5)	
Facial type			
Short face	10 (41.7)	14 (58.3)	0.014
Average	44 (57.1)	33 (42.9)	
Long face	4 (21.1)	15 (78.9)	

*Chi-square test ($p \leq 0.05$)

another well-designed controlled study suggested that prematurely born children exhibit more malocclusion characteristics and have greater requirement for orthodontic treatment than full-term born children.[29] These findings suggest that preterm children should be more closely monitored by orthodontists, who should be aware of the tendency toward severe malocclusion and the orthodontic treatment needs among such children.[29] Further longitudinal studies are required to address the issue as to whether premature births result in dentofacial alterations.

A recent study used the DAI to determine the degree of malocclusion in patients with DS and found that 83.2% of them had severe and very severe malocclusion.[30] In the present study, after adjusting for all potential variables related to malocclusion severity, it was found that DS is a risk factor for severe malocclusion. Therefore, the assessment and monitoring of individual, oral, social, and behavioral aspects is of considerable clinical importance to orthodontists. This is particularly true with regard to individuals with DS who receive little assistance

Table 3. Univariate and multivariate logistic regression analyses considering associations between malocclusion severity and independent variables.

	OR Non adjusted (95%CI)	P	OR adjusted (95%CI)	*P*
Group				
Control	1.00		1.00	
Down syndrome	2.25 (1.1-4.7)	0.029	4.24 (1.3-13.4)	0.014
Age (y)				
≤ 10	1.00		1.00	
> 10	2.61 (1.2-5.4)	0.011	2.69 (0.9-8.4)	0.086
Gender				
Male	1.00		1.00	
Female	0.58 (0.3-1.2)	0.138	0.58 (0.2-1.4)	0.233
Premature birth				
No	1.00		1.00	
Yes	3.52 (1.1-11.5)	0.038	4.11 (1.1-15.9)	0.041
Facial type				
Short face	1.00		1.00	
Average	0.54 (0.2-1.3)	0.188	2.15 (0.6-7.4)	0.226
Long face	2.68 (0.7-10.5)	0.158	13.1 (2.2-76.1)	0.004
Breathing				
Nasal	1.00			
Mouth	6.93 (1.9-25.1)	0.003	---	
Lip incompetence				
No	1.00	0.012	---	
Yes	2.69 (1.2-5.8)			
Dentition				
Mixed	1.00		1.00	
Permanent	1.94 (0.9-4.2)	0.095	1.08 (0.3-3.5)	0.898

and the professionals who treat such individuals are most often ill prepared to offer better care. Thus, the treatment of malocclusion for individuals with DS can lead to a considerable improvement in their quality of life.[18,20]

Conclusions

Vertical and transversal alterations in the occlusion, such as mandibular protrusion, anterior open bite, and posterior crossbite were significantly more frequent among the individuals with DS than those in the control group. The determinant factors associated with malocclusion severity were DS, a history of premature birth, and long face pattern.

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