

Prevalence and determinant factors of malocclusion in children with special needs

Ana Cristina Oliveira*, Saul Martins Paiva**, Milene Torres Martins**,
Cintia Silva Torres** and Isabela Almeida Pordeus**

Departments of* Social and Preventive Dentistry and **Pediatric Dentistry and Orthodontics, Faculty of Dentistry, Federal University of Minas Gerais, Minas Gerais, Brazil

Correspondence to: Dr Ana Cristina Oliveira, Department of Social and Preventive Dentistry, Faculty of Dentistry, Federal University of Minas Gerais, Rua Gentios, 40/1103 Coração de Jesus, Belo Horizonte, Minas Gerais 30380-490, Brazil. E-mail: anacboliveira@yahoo.com.br

SUMMARY Careful attention to malocclusion in children with special needs leads to a considerable improvement in the quality of life. The present study analysed the prevalence of malocclusion in children with Down syndrome (DS) and cerebral palsy (CP) as well as associations with individual, socio-economic, and behavioural factors.

A cross-sectional study was carried out that included 181 mothers and their children with DS and CP (aged 3–12 years) at two institutions for individuals with special needs in Rio de Janeiro, Brazil. Data were collected using a questionnaire administered to the mothers and a dental examination of the children. Clinical examination recorded the following: anterior/posterior crossbite and anterior openbite (AOB). The control variables were the mother's level of education as well as the gender and age of the child. Statistical analysis of the data was performed using the chi-square test and multiple logistic regression.

An anterior crossbite was present in 20.4 per cent, a posterior crossbite in 21.5 per cent, and an AOB in 29.8 per cent. The presence of DS, bottle feeding, and non-nutritive sucking habits for 24 months or more was determinant factors for an anterior crossbite and the presence of DS, bottle feeding and non-nutritive sucking habits for 24 months or more, and respiratory infection in the previous 6 months was determinant factors for a posterior crossbite. The presence of CP and non-nutritive sucking habits for 24 months or more was determinant factors for an AOB. Thus, the prevalence of malocclusion in children with special needs was associated with the type of disability, use of bottle feeding and non-nutritive sucking habits for 24 months or more, and respiratory infection in the previous 6 months.

Introduction

Individuals with Down syndrome (DS) and cerebral palsy (CP) are particularly prone to orofacial disorders. Systemic dysfunction in such individuals may predispose them to oral disease, which in turn may aggravate the systemic disease. Regular assessments, preferably by a team of specialists, should be performed in order to identify and prevent functional problems and disease processes (Ortega *et al.*, 2007; Asdaghi Mamaghani *et al.*, 2008; Oliveira *et al.*, 2008a).

Aesthetically, aspects such as bad breath, poorly positioned teeth, trauma, gingival bleeding, the habit of maintaining the mouth open, and drooling can either cause feelings of compassion in others or repugnance and discrimination, thereby accentuating attitudes of social rejection. Although oral disease and orofacial malformations are rarely life threatening, they can cause pain, infection, respiratory complications, and problems with mastication (Ackerman and Wiltshire, 1994; Tausche *et al.*, 2004; Kaye *et al.*, 2005; Oliveira *et al.*, 2008b).

A number of studies have stressed the considerable prevalence of malocclusion in individuals with CP and DS (Ackerman and Wiltshire, 1994; Onyeaso, 2003). In a study

involving 381 children with disabilities in South Africa, 74 per cent were found to be in need of orthodontic treatment (Ackerman and Wiltshire, 1994). Using the dental aesthetic index, 124 Nigerian children/adolescents with disabilities were investigated; 58 per cent had a significant treatment need (Onyeaso, 2003). Tausche *et al.* (2004), in German children without anomalies, using the Index of Orthodontic Treatment Need, found that the proportion of children in need of orthodontic treatment was 26 per cent.

Children with DS and CP have the habit of projecting the tongue against the teeth and out of the mouth and suffer frequent episodes of upper airway infection, which leads to a greater prevalence of mouth breathing and malocclusion (Mitchell *et al.*, 2003; Venail *et al.*, 2004; Chawla *et al.*, 2006; Ortega *et al.*, 2007). Children with the habit of maintaining their mouth open exhibit abnormal oromuscular movements and respiration, which compromises the coordination and articulation of the lips and cheeks during speech and swallowing (Zavaglia *et al.*, 2003; Korbmacher *et al.*, 2004; Ortega *et al.*, 2007). In a study in Brazil, children with a mouth breathing habit were found to have a 10-fold greater chance of exhibiting a malocclusion in

comparison with those with nasal breathing (Góis *et al.*, 2008).

The aim of the present study was to investigate the prevalence of an anterior crossbite, a posterior crossbite, or an anterior open bite (AOB) in children with DS and CP as well as any association with individual, socio-economic, and behavioural factors.

Subjects and methods

An observational cross-sectional study was carried out with a convenience sample of 181 mothers and their children with DS or CP between 3 and 12 years of age from two public healthcare institutions directed at the care of individuals with special needs in the city of Rio de Janeiro, Brazil. The study received approval from the Ethics Committee of the Oswaldo Cruz Foundation (FIOCRUZ).

The participants were asked to take part in the study while awaiting care at the institutions. The mothers individually received clarification regarding the study. Those who agreed to participate were asked to enter the medical office with their child, at which point they signed an informed consent, answered the questionnaire, and witnessed the researcher perform an oral examination of their child. The exclusion criteria were a child not accompanied by their mother, a history of previous orthodontic treatment, and simultaneous presence of the occlusal abnormalities investigated.

The questionnaire contained items addressing individual, socio-economic, and behavioural characteristics. Ethnic categorization was determined from physical traits using categories for the Brazilian population: white, black, mulatto, and yellow, adopting the criteria established by the IBGE (2000). Dental examinations were carried out by one author (ACO), with the help of a research assistant who recorded the data. Examinations were performed under natural light, using a disposable mouth mirror (Prisma®, São Paulo, Brazil) and a Community Periodontal Index probe (Golgran®, São Paulo, Brazil), also known as a ballpoint probe (WHO, 1997). The following malocclusions were diagnosed: anterior crossbite [lower incisors in front of the upper incisors (negative overjet/absent overbite)], posterior crossbite (posterior teeth of the upper arch displaced to the palatal region in relation to the lower teeth either unilaterally or bilaterally), and AOB [no contact between the upper and lower anterior teeth (absent overjet)]. Diagnoses were based on the criteria established by the WHO (1997).

Intra-examiner calibration, test-retest of the questionnaire, and pilot study were carried out prior to the main study. In order to determine intra-examiner variability, 35 children with DS and CP from a non-governmental organization that offers care to individuals with special needs in the city of Rio de Janeiro, Brazil, were examined and re-examined after a 10 day interval. These children did

not participate in the main study. Intra-examiner diagnostic agreement was considered very good (Rigby, 2000) as the Kappa values achieved for each clinical condition were AOB = 0.87, anterior crossbite = 0.74, and posterior crossbite = 0.92. In order to test the internal validity of the questionnaire, the testing and retesting of the instrument were carried out with the 35 mothers of the children who participated in the intra-examiner calibration step. The retest was carried out after a 10 day interval. The results of the test-retest agreement revealed Kappa values ranging from 0.73 to 1.00, which were considered very good to excellent (Rigby, 2000).

A pilot study was then carried out to test methodological procedures and data collection instruments. A sample of 30 pairs of mothers and children with DS or CP participated. This step confirmed the validity of the methodology to be employed.

After analysis of this preliminary phase, the main study was carried out. Data analysis was performed using the Statistical Package for Social Sciences (version 17.0 for Windows; SPSS Inc., Chicago, Illinois, USA). Univariate and bivariate analyses were carried out using chi-square ($P < 0.10$) and Fisher's exact tests. The dependent variables were the presence or absence of an anterior crossbite, a posterior crossbite, or an AOB. The independent variables were related to the individual, behavioural, and clinical characteristics of the sample. Multiple logistic regression was used in the multivariate analysis. The criterion for inclusion into the model was a significance value of less than 25 per cent in the outcome of the bivariate analysis ($P < 0.25$, backward stepwise procedure). Explanatory variables were also selected based on the theoretical/epidemiological model; although these variables may have achieved $P > 0.25$, they have a strong epidemiological relationship with the dependent variables.

Results

The overall sample comprised 181 mother/child pairs: 102 children with DS [46 girls (45.1 per cent) and 56 boys (54.9 per cent)]/56 children had white skin (54.9 per cent) and 46 were black or mulatto (45.1 per cent) and 79 children with CP [36 girls (45.6 per cent) and 43 boys (54.4 per cent)]/41 children had white skin (51.9 per cent) and 38 were black or mulatto (48.1 per cent). The diagnosis of DS and CP was obtained from the medical records at the institutions at which the children were treated. No child had a history of tongue reduction. A total of 21 mother/child pairs were excluded from the study: children with a previous orthodontic history (three with CP), those not accompanied by their mother (six with DS and two with CP), those with both an AOB and a posterior crossbite (three with DS and one with CP), and six mothers refused to participate due to a lack of time/disinterest or the child's refusal to be examined (four with DS and two with CP).

Figure 1 displays the distribution of the children with regard to the prevalence of malocclusion. Among the conditions evaluated, an AOB was the most prevalent (29.8 per cent).

Table 1 shows the results of the bivariate analysis for anterior crossbite, posterior crossbite, and AOB. Chi-square and Fisher's exact tests showed a statistical association between the presence or absence of an anterior crossbite, a posterior crossbite, or an AOB and the following variables: type of disability, breastfeeding, non-nutritive sucking habits, gender, and age of the child.

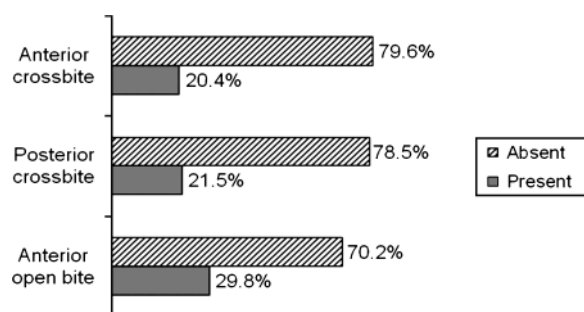


Figure 1 Distribution of children regarding the prevalence of malocclusion ($n = 181$).

The results of the multiple logistic regression analysis are shown in Table 2. Independent from the other variables analysed in the model, children with DS had a 16-fold greater chance of being diagnosed with an anterior crossbite [odds ratio (OR) = 16.41, 90 per cent confidence interval (90% CI): 5.61–48.00]. Moreover, children whose mothers reported the use of bottle feeding [OR = 1.88 (90% CI: 0.91–3.87)] or non-nutritive sucking habits for 24 months or more [OR = 2.07 (90% CI: 0.98–4.36)] had approximately a 2-fold greater chance of having an AOB.

Children with DS had a 2.25-fold greater risk of exhibiting a posterior crossbite [OR = 2.25 (90% CI: 1.09–4.64)]. Those whose mothers reported the use of bottle feeding or non-nutritive sucking habits for 24 months or more had a 2.35-fold [OR = 2.35 (90% CI: 1.18–4.67)] and 2.79-fold [OR = 2.79 (90% CI: 1.42–5.44)] greater chance of being diagnosed with a posterior crossbite. Children having a respiratory infection in the previous 6 months had a 2.47-fold greater chance of exhibiting a posterior crossbite [OR = 2.47 (90% CI: 0.89–6.79)].

After adjustment of the variables in the model, children diagnosed with CP had a 3-fold greater chance of having an AOB [OR = 2.84 (90% CI: 1.60–5.05)]. Those whose mothers reported non-nutritive sucking habits for 24 months or more had a 3.27-fold greater chance of exhibiting an AOB [OR = 3.27 (90% CI: 1.84–5.81)].

Table 1 Distribution of the sample according to the prevalence of crossbite and openbite and associated factors ($N = 181$).

Independents variables	<i>n</i>	Anterior crossbite		Posterior crossbite		Anterior open bite	
		<i>n</i> (%)	<i>P</i> value	<i>n</i> (%)	<i>P</i> value	<i>n</i> (%)	<i>P</i> value
Type of disability							
Down syndrome	102	34 (33.3)	<0.01*	28 (27.5)	0.02**	20 (19.6)	<0.01**
Cerebral palsy	79	3 (3.8)		11 (13.9)		34 (43.0)	
Breastfeeding							
Less than 6 months (or absent)	111	19 (17.1)	0.16**	27 (24.3)	0.268**	39 (35.1)	0.05**
6 months or more	70	18 (25.7)		12 (17.1)		15 (21.4)	
Use of bottle feeding							
24 months or more	95	20 (21.1)	0.83**	25 (26.3)	0.10**	33 (34.7)	0.13**
Less than 24 months (or absent)	86	17 (19.8)		14 (16.3)		21 (24.4)	
Non-nutritive sucking habits (sucking or finger biting)							
Yes	69	16 (23.2)	0.47**	21 (30.4)	0.02**	32 (46.4)	<0.01**
No	112	21 (18.8)		18 (16.1)		22 (19.6)	
Respiratory infection (less than 6 months)							
Yes	148	33 (22.3)	0.23*	35 (23.6)	0.16*	45 (30.4)	0.72**
No	33	4 (12.1)		4 (12.1)		9 (27.0)	
Gender							
Female	82	22 (26.8)	0.05**	19 (23.2)	0.62**	24 (29.3)	0.88**
Male	99	15 (15.2)		20 (20.2)		30 (30.3)	
Age (years)							
3–6	104	18 (17.3)	0.22**	15 (14.4)	<0.01**	34 (32.7)	0.32**
7–12	77	19 (24.7)		24 (31.2)		20 (26.0)	
Maternal schooling							
8 years or more of study	97	18 (18.6)	0.49**	20 (20.6)	0.74**	26 (26.8)	0.33**
Less than 8 years of study	84	19 (22.6)		19 (22.6)		28 (33.3)	

*Fisher's test (cell with value less than 5).

**Chi-square test.

Table 2 Risk factors for the prevalence of malocclusion in children with Down syndrome (DS) or cerebral palsy (CP) and the results of multiple logistic regression analysis ($n = 181$)*

Dependent variables	Independent variables	Odds ratio [90 per cent confidence interval (90% CI)] crude	Odds ratio (90% CI) adjusted*
Anterior crossbite	Type of disability (DS)	12.66 (3.72–43.11)	16.41 (5.61–48.00)
	Use of bottle feeding (24 months or more)	1.08 (0.52–2.23)	1.88 (0.91–3.87)
	Non-nutritive sucking habits (24 months or more)**	1.30 (0.62–2.72)	2.07 (0.98–4.36)
Posterior crossbite	Type of disability (DS)	2.33 (1.08–5.05)	2.25 (1.09–4.64)
	Use of bottle feeding (24 months or more)	1.83 (0.88–3.82)	2.35 (1.18–4.67)
	Non-nutritive sucking habits (24 months or more)**	2.28 (1.11–4.69)	2.79 (1.42–5.44)
	Respiratory infection (less than 6 months)	2.24 (0.73–6.82)	2.47 (0.89–6.79)
Anterior open bite	Type of disability (CP)	3.09 (1.60–5.98)	2.84 (1.60–5.05)
	Non-nutritive sucking habits (24 months or more)**	3.53 (1.82–6.87)	3.27 (1.84–5.81)

*Adjusted for control variables (gender and age).

**Sucking or finger biting.

Discussion

Malocclusion has a considerable impact on the lives of children with DS and CP, associated with problems in their daily activities, including discrimination due to their physical appearance and problems related to oral functions, such as swallowing and speech (Kaye *et al.*, 2005; Ortega *et al.*, 2007; Asdaghi Mamaghani *et al.*, 2008; Oliveira *et al.*, 2008b).

The results of the present study demonstrate that children with DS and CP had a considerable prevalence of an anterior crossbite (20.4 per cent), a posterior crossbite (21.5 per cent), and an AOB (29.8 per cent). These findings may be explained by the fact that children with DS and CP are breastfed less than those with normal development (Pisacane *et al.*, 2003) have a greater percentage of non-nutritive sucking habits (Ortega *et al.*, 2007) and exhibit physiological abnormalities that facilitate the appearance of malocclusions. Such abnormalities include the underdevelopment of the orofacial musculature (Mitchell *et al.*, 2003; Venail *et al.*, 2004; Chawla *et al.*, 2006; Ortega *et al.*, 2007; Asdaghi Mamaghani *et al.*, 2008).

Although the variables 'bottle feeding and non-nutritive sucking habits' were not selected for inclusion in the logistic regression model and the dependent variable 'anterior crossbite' for statistical reasons ($P > 0.25$), they were included in the model due to the fact that they exhibited a strong relationship with the prevalence of an anterior crossbite in previous studies (Leite-Cavalcanti *et al.*, 2007; Góis *et al.*, 2008). The same was true for the inclusion of 'breastfeeding' in the regression model for a posterior crossbite (Peres *et al.*, 2007; Leite-Cavalcanti *et al.*, 2007) and 'respiratory infection' in the regression model for an AOB (Góis *et al.*, 2008; Oliveira *et al.*, 2008b).

In the logistic regression analysis, the type of disability remained in the models for anterior crossbite, a posterior crossbite, and an AOB. The children with DS had a 16- and 2.2-fold greater chance of being diagnosed with an anterior or a posterior crossbite, respectively. The greater prevalence of these malocclusions in this population may be explained

by the characteristic abnormalities found with this syndrome, including obstruction of the pharynx (caused by macroglossia, narrow, high palate, micrognathia, and hypertrophy of the tonsils), functional problems (hypotonia, obesity, and desiccation of the mucous membranes), and nasal obstruction (reduced airway, hypertrophy of the adenoids, sinusitis, or rhinitis; Mitchell *et al.*, 2003; Venail *et al.*, 2004; Góis *et al.*, 2008). The presence of a protrusive hypotonic tongue leads to problems with sucking, speech, and mastication, as well as poor positioning of the teeth (Zavaglia *et al.*, 2003; Korbmacher *et al.*, 2004; Chawla *et al.*, 2006).

The children diagnosed with CP exhibited almost a 3-fold greater chance of having an AOB. This finding may be explained by the fact that muscle incompetence impairs lip seal in individuals with CP and leads to a systematic anterior posture of the tongue, facilitating the onset and maintenance of the habit of tongue interposition (Ortega *et al.*, 2007). The development of the jawbone, mimetic, and masticatory muscles is closely connected. Thus, deformations of the craniofacial region and jaws develop as a result of the disturbed or changed tonicity of the muscles (Asdaghi Mamaghani *et al.*, 2008). According to the above authors, from an orthodontic standpoint, this constitutes an aetiological basis for dentoalveolar and/or skeletal deviations.

The use of bottle feeding remained in the regression models for anterior and posterior crossbites, demonstrating that children diagnosed with these malocclusions were those whose mothers reported the use of bottle feeding for a period of 24 months or more. This finding corroborates those described in the literature, affirming that bottle-fed children have a greater chance of having a crossbite (Peres *et al.*, 2007; Góis *et al.*, 2008). The artificial nipple of the feeding bottle is made from more rigid material, which can force the interior of the oral cavity, causing inappropriate alignment of teeth and transverse growth of the palate, which are conditions that can lead to the development of a posterior crossbite (Drane, 1996).

Non-nutritive sucking habits remained in the models for anterior crossbite, posterior crossbite, and AOB, which corroborates the results of previous studies (Warren and Bishara, 2002; Cozza *et al.*, 2007; Leite-Cavalcanti *et al.*, 2007; Peres *et al.*, 2007; Heimer *et al.*, 2008). The imbalance between external and internal muscle forces as well as the increase in mandibular width and the tendency towards a reduction in maxillary width might have contributed to this occurrence (Warren and Bishara, 2002; Bishara *et al.*, 2006; Cozza *et al.*, 2007; Ortega *et al.*, 2007; Heimer *et al.*, 2008; Asdaghi Mamaghani *et al.*, 2008).

A considerable proportion of individuals with DS and CP have oral motor difficulties, such as problems with swallowing and chewing (Mizuno and Ueda, 2001; Tahmassebi and Curzon, 2003; Johnson *et al.*, 2004; Senner *et al.*, 2004; Kaye *et al.*, 2005). This may be due to difficulty in sucking as a result of the hypotonicity of the lips, perioral muscles, and masticatory muscles as well as deficiency in the movements of the tongue (Mizuno and Ueda, 2001; Ortega *et al.*, 2007). Thus, such children are generally breastfed less than those with normal development (Pisacane *et al.*, 2003; Tahmassebi and Curzon, 2003; Johnson *et al.*, 2004; Senner *et al.*, 2004) and have a greater prevalence of non-nutritive sucking habits (Ortega *et al.*, 2007). According to a number of authors, breastfeeding offers a protective effect as it promotes correct development of the jaws and strengthens the muscles involved in the process of sucking to obtain breast milk (Peres *et al.*, 2007).

In an attempt to diminish the occurrence of malocclusion in children with DS and CP, professional orientation for the establishment of good habits in the first years of life is essential. The children identified with one of the aforementioned malocclusions had a greater chance of being those whose mothers reported non-nutritive sucking habits for a period of more than 24 months. A number of authors associate the prevalence of malocclusion (especially a crossbite and AOB) in the primary and mixed dentition with non-nutritive sucking habits (Warren and Bishara, 2002; Cozza *et al.*, 2007; Leite-Cavalcanti *et al.*, 2007; Ortega *et al.*, 2007; Peres *et al.*, 2007; Heimer *et al.*, 2008). Among such habits, pacifier sucking has proven to be most strongly associated with malocclusion, especially if the habit persists beyond 2 years of age (Tomita *et al.*, 2000; Bishara *et al.*, 2006; Góis *et al.*, 2008).

Respiratory infection remained in the logistic regression model for posterior crossbite, confirming the strong relationship between these variables found in previous studies (Peres *et al.*, 2007; Góis *et al.*, 2008). This result highlights this variable as another determinant factor for the development of malocclusion among children with DS and CP as they exhibit systemic complications that generate a greater frequency of respiratory infection (Mitchell *et al.*, 2003; Venail *et al.*, 2004).

Although the present study design was strong, it has some limitations that should be addressed. A weakness of cross-sectional studies is the difficulty in establishing causal relationships from a cross-section in time (Kirkwood and

Stern, 2003). Moreover, the nutritive and non-nutritive oral habits were obtained from the reports of the mothers; as the data were not collected directly, they are subject to information and memory bias. Caution should be taken in order not to influence the respondents and avoid bias when interpreting the results (Martins *et al.*, 2008). In Brazil, it is difficult to obtain a representative sample of children/adolescents with DS as there is no governmental database that registers all individuals with this syndrome. Thus, the choice was made to use a convenience sample to identify the important aspects related to the presence of malocclusion in this population (Kirkwood and Stern, 2003). Internal validation of the study was ensured by the test-retest of the questionnaire and the intra-examiner calibration, which achieved good agreement (Rigby, 2000).

Children with special needs normally undergo dental examination at an older age (Asdaghi Mamaghani *et al.*, 2008; Oliveira *et al.*, 2008a). It is therefore fundamental that the care offered by health professionals is integral and multidisciplinary, especially in encouraging parents/guardians to seek dental care for younger children, when preventive procedures and education are still possible.

An appropriate approach to addressing these determinant factors can lead to a reduction in the prevalence of malocclusion in children with DS and CP, with a consequent beneficial effect on the development of this group. To improve the oral health of individuals with special needs, it is essential for public oral healthcare services to incorporate intervention methods directed at the prevention and treatment of malocclusions. It is therefore necessary to understand the panorama of dental needs of children with disabilities in order to ensure care that can help this proportion of the population overcome their difficulties and improve both their development and quality of life.

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