Original Article

Anterior Open Bite and Speech Disorders in Children with Down Syndrome

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ABSTRACT
Objective: To estimate the prevalence and to determine if there is an association between anterior open bite and the presence of speech disorders in a group of Mexican children with Down syndrome (DS).
Materials and Methods: The subjects were a group of Mexican children with Down syndrome (DG) and a control group (CG) of Mexican pediatric patients without disabilities matched by age. The children in both groups came from families having children with anterior open bite and children without it. A parental questionnaire, dental study casts, and a speech test were used to measure the studied variables. Data were analyzed using the chi-square test (χ^2 test), and one-way analyses of variance (ANOVA), followed by the Tukey post hoc test.
Results: Prevalence of anterior open bite was 31.6% in the DG and 22.8% in the CG. The total speech errors by omissions, substitutions, distortions, and additions indicated that there were significant differences between both groups ($F = 31.68$, $P < .001$). In general, no significant difference in speech disorders was observed between the DG and the CG regardless of the presence of anterior open bite.
Conclusions: No association existed between speech disorders and anterior open bite in the samples studied.

KEY WORDS: Down syndrome; Speech; Language; Malocclusion; Anterior open bite; Disability; Disabled populations

INTRODUCTION

Down syndrome (DS), or trisomy 21, is the most frequent chromosome disorder manifested in newborns. Worldwide, the risk for this chromosomal aberration has not changed substantially over the last decades—one out of 600–750 neonates has this genetic abnormality.[1,2]

Speech and DS

The speech of most children with typical development is fully intelligible at age 4 years, even though their phonological systems are not yet complete.[3] However, speech and language are a major problem for many children with DS. Even those who are relatively capable in other areas of life may have great difficulty in communicating with people who do not know them well. This can bring about a restriction of opportunities for full integration and participation in society.[4]

There is considerable individual variation in the delay of language acquisition in children with DS,[5,6] but only about 5% of these children have extremely limited speech and must rely on a small number of single words and signs.[7] Their early vocabulary development is delayed by an average 8–9 months in comparison with typically developing children.[5]

Language acquisition of children with DS is probably affected by both development delay and specific disorders in certain language areas.[8] The phonological system of those children is influenced by several factors that can create difficulties in perceiving and producing speech.[3] These factors include: neurological development of the language,[9] language learning environment,[3,8] hearing loss,[3] subtypes of DS,[2] motor co-

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ordination and timing deficit,[7] and orofacial characteristics.[3,7,10]

Orofacial Characteristics

Children with DS frequently have differences in the oral-facial structures and in the functional movements that affect speech production.[7] Persons with DS have skeletal and muscular systems that differ from those individuals without DS. The skeletal system is characterized by absent or deficient bone growth and a smaller oral cavity. The muscular system is characterized by absent and extra muscles in the facial region, a large muscular tongue, and hypotonicity. Any of these factors is likely to influence motor movements associated with speech and to negatively impact the articulatory and phonatory abilities of children with DS.[3] The reduced muscle tone in the lips and cheeks contributes to an imbalance of forces on the teeth, with the force of the tongue having a greater influence. In turn, this contributes to the open bite often seen in children with DS. On the other hand, the primary skeletal abnormality affecting the orofacial structures in DS is an underdevelopment or hypoplasia of the midface region. In many persons this hypoplasia also causes a prognathic Class III occlusal relationship which, in turn, contributes to an open bite.[10]

Anterior Open Bite

Anterior open bite probably affects less than 5% of children.[11] Teeth play an important role in speech production, but the relationship between tooth position and speech remains controversial. The ability of patients to adapt their speech to compensate for abnormal tooth position is recognized, but the mechanisms for this adaptation remain incompletely understood. While certain irregularities show a relationship with speech disorders, this does not appear to correlate with the severity of the malocclusion. Therefore, tooth position may play a role in articulatory speech disorders which, although not the most severe, represent 50% to 60% of all speech disorders.[12]

Anterior open bite is, however, the most frequent type of malocclusion involved in phonological disorders. Nicolá and Jonathan[12] stated that 63% of the cases with open bite have an alteration in speech. They also mentioned that Bernstein[13] concluded that the defects in speech are not related to any malocclusion except anterior open bite. The precise influence of each factor is difficult to determine and may vary from one child to another.[3,6] In some cases, their speech remains unintelligible throughout childhood and adolescence.[3] Studies reported in the literature have tried to determine if the malocclusion, specifically the open bite, could be associated with a higher difficulty in the articulation of the speech in children with DS.

This study aimed to estimate the prevalence of anterior open bite and to determine whether there is an association between this type of malocclusion and the presence of speech disorders in a group of Mexican children with DS (DG) and a control group of Mexican pediatric patients without disabilities (CG).

MATERIALS AND METHODS

Ethical Implications

The design and conduct of this study complied with the regulations for the protection of human subjects stipulated by the organizations where data collection took place.

Environment

Using a case-control study design, we recruited 57 children, aged 3 to 15 years, with DS studying at The John Langdon Down Foundation in 2003. The Foundation is a not-for-profit organization located in Mexico City that offers programs delivering psychopedagogical support and advice to persons with DS and their families. To participate in this study, volunteers provided verbal consent to an oral examination, including parental questionnaire, dental impressions, and a speech test. Their guardians/parents provided written consent.

The 57 students had no orthodontic appliances, nor were they wearing oral prostheses. They had no history of previous orthodontic therapy, and they were free of conditions such as autism, cerebral palsy, intellectual disability more than mild, sinusitis, asthma, cleft lip and palate, or hearing loss more than 15 dB. By an ethical implication all the children had a history of previous speech therapy. They had the eight incisors. This group (DG) was further divided into two subgroups: Subgroup 1 included children with DS without anterior open bite and Subgroup 2 included children with DS with anterior open bite.

Study children were matched (1:1) by age to participants in the CG who did not have disabilities. The 57 children from the CG were outpatients seeking dental care at the admissions clinic of the Dental School, Universidad Nacional. The CG participants were free of systemic diseases and chronic medication use, had no orthodontic appliances or oral prostheses, and had no history of previous orthodontic therapy or speech therapy. They had eight incisors erupted. Children in the CG agreed to participate in the study as part of the preliminary evaluation undertaken by all first-time patients attending the admission clinic. The CG was also divided into two subgroups: Subgroup 3 included

Table 1. Frequencies and Percent of Children From the Four Subgroups With Speech Disorders by Error Type[a]

	Subgroup 1 (n = 39)		Subgroup 2 (n = 18)				Subgroup 3 (n = 44)		Subgroup 4 (n = 13)			
	F	%	F	%	χ^2	*P*	F	%	F	%	χ^2	*P*
Omissions	39	(100)	18	(100)	0.02	>.05	3	(6.8)	1	(7.7)	0.32	>.05
Substitutions	29	(74.4)	15	(83.3)	0.78	>.05	9	(20.5)	2	(15.4)	0.43	>.05
Distortions	11	(28.2)	4	(22.2)	0.22	>.05	5	(11.4)	0	(0)	2.56	>.05
Additions	16	(41.0)	7	(38.9)	4.59	<.05	1	(2.3)	1	(7.7)	0.79	>.05

[a] F indicates frequency.

children without disabilities and without anterior open bite. Subgroup 4 included children without disabilities, but with anterior open bite.

In both groups, all the children were from the same geographic area and belonged to the same socioeconomic status. They were all monolingual speakers of Spanish. Their guardians/parents were native Spanish speakers, and the mothers of the children had the same level of education (bachelor's degree). The information was obtained from each child's medical history. A parental questionnaire also collected information regarding medical history.

Variables Collected

We diagnosed anterior open bite using the dental study casts. Anterior open bite was defined as the absence of contact and existence of a vertical space between the maxillary and mandibular incisors in centric relation. Because people with DS frequently have a Class III malocclusion, the children included in this sample were considered with open bite regardless of the presence or absence of anterior crossbite. The examiner (a pediatric dentist) performed standardization training by evaluating 25 casts of patients not included in the study population reexamined 7 days after the first assessment. The kappa value (intraexaminer reliability) was 1.00.

A speech test was done to determine the phonetic errors. The phonological analysis used in this study was the same employed routinely by The John Langdon Down Foundation. A speech therapist and a pediatric dentist showed 58 pictures of common objects to each child which he or she was required to name. The two examiners recorded the phonological errors. A total of 5 vowel sounds: /a/, /e/, /i/, /o/, and /u/; 18 single consonant sounds: /b/, /ch/, /d/, /f/, /g/, /j/, /k/, /l/, /m/, /n/, /ñ/, /p/, /r/, /rr/, /s/, /t/, /x/, and /y/; 8 homosyllabic consonant sounds: /bl/, /fl/, /gl/, /kl/, /pl/, /br/, /dr/, and /tr/; 17 heterosyllabic consonant sounds: /kt/, /ld/, /ls/, /lt/, /mb/, /mp/, /nf/, /nj/, /nt/, /ny/, /rb/, /rs/, /rm/, /rt/, /sb/, /sk/, and /st/; and 9 diphthong sounds: /ai/, /au/, /ei/, /ia/, /ie/, /io/, /iu/, /ua/, and /ue/ were studied.

The single consonants were analyzed taking into account three word positions: initial, medial, and final. Error types analyzed were omissions, substitutions, distortions, and additions of phonemes. A word was considered correct if it was named or repeated without any omission, substitution, distortion, or addition of a sound. The standardization of the two examiners was carried out by evaluating 25 patients not included in the study population, on two occasions. To apply the speech test, the kappa value for the intraexaminer reliability obtained by the pediatric dentist was 0.93, and by the speech therapist was 0.95. The kappa value for the interexaminer reliability was 0.93.

Statistical Analysis

We used SPSS (version 12.0) for data analysis. To estimate whether there was a significant difference between the prevalence of anterior open bite in both groups, as well as in the percentages by error type of the children of each subgroup, χ^2 test was applied. Because error means by types and phonemes were also calculated, one-way analyses of variance (ANOVA) followed by the Tukey post hoc test were used to determine whether there were statistically significant differences among subgroups.

RESULTS

The DG consisted of 57 children with a mean age of 8.7 ± 3.0 years (range 3.0–15.0 years). The CG included 57 children with a mean age of 8.6 ± 3.0 years (range 3.0–15.0 years). The groups were divided into two subgroups respectively, as follows: Subgroup 1 included 39 children with DS without anterior open bite; Subgroup 2 included 18 children with DS with anterior open bite; Subgroup 3 consisted of 44 children without disabilities and without anterior open bite; and finally, Subgroup 4 included 13 children without disabilities with anterior open bite.

Prevalence of Anterior Open Bite

In the DG 18 (31.6%) children showed anterior open bite, whereas in the CG 13 (22.8%) children presented

Table 2. Mean, Standard Deviation, and Statistically Significant Difference of Error Type Among the Four Studied Subgroups[a]

	Mean (SD) by Subgroups							
	1		2		3		4	
Error Type	n = 39		n = 18		n = 44		n = 13	
Omission of								
Vowels	31.3	(52.2)	20.7	(45.3)	.0	(.0)	1	(.5)
Initial single consonants	15.8	(15.7)	13.6	(14.3)	6.8E-02	(.3)	.0	(.0)
Medial single consonants	15.6	(20.6)	12.2	(17.6)	2.3E-02	(.1)	.1	(.5)
Final single consonants	6.1	(4.9)	5.4	(4.1)	.2	(.6)	.1	(.5)
Homosyllabic consonants	6.6	(3.7)	6.8	(2.8)	2.3E-02	(.1)	.0	(.0)
Heterosyllabic consonants	14.0	(6.3)	14.6	(4.7)	.0	(.0)	.0	(.7)
Diphthongs	6.7	(3.3)	6.9	(2.9)	.0	(.0)	.0	(.0)
Total omissions	97.1	(101.5)	80.3	(86.3)	.27	(1.2)	.5	(1.7)
Substitution of								
Vowels	5	(.9)	1.4	(2.1)	.0	(.0)	7.7E-02	(.3)
Initial single consonants	6.8	(6.2)	7.0	(5.4)	.2	(.5)	.5	(1.7)
Medial single consonants	7.0	(6.7)	8.4	(5.6)	.2	(.5)	.3	(.8)
Final single consonants	.6	(.9)	.3	(.6)	6.8E-02	(.2)	.0	(.0)
Homosyllabic consonants	2.3	(2.5)	2.1	(2.1)	.3	(1.3)	.3	(1.1)
Heterosyllabic consonants	1.3	(2.3)	2.2	(2.3)	.6	(2.2)	.5	(1.7)
Diphthongs	.5	(1.7)	.2	(.5)	9.1E-02	(.4)	7.7E-02	(.3)
Total substitutions	19.0	(16.9)	21.6	(12.4)	1.4	(4.3)	1.7	(3.2)
Distortion of								
Vowels	2.7E-02	(.3)	.0	(.0)	.0	(.0)	.0	(.0)
Initial single consonants	.3	(.7)	.1	(.3)	.1	(.4)	.0	(.0)
Medial single consonants	.2	(.6)	.3	(.6)	4.5E-02	(.2)	.0	(.0)
Final single consonants	5.1E-02	(.2)	.0	(.0)	2.2E-02	(.1)	.0	(.0)
Homosyllabic consonants	.0	(.0)	.0	(.0)	2.2E-02	(.1)	.0	(.0)
Heterosyllabic consonants	5.1E-02	(.2)	5.6E-02	(.2)	4.5E-02	(.2)	.0	(.0)
Diphthongs	.0	(.0)	.0	(.0)	.0	(.0)	.0	(.0)
Total distortions	.7	(1.4)	.4	(.9)	.2	(.7)	.0	(.0)
Addition of								
Vowels	5.1E-02	(.3)	5.6E-02	(.2)	.0	(.0)	.0	(.0)
Initial single consonants	.4	(1.0)	.4	(.8)	.0	(.0)	.0	(.0)
Medial single consonants	.1	(.4)	5.6E-02	(.2)	2.3E-02	(.1)	7.7E-02	(.3)
Final single consonants	.3	(.5)	.2	(.4)	.0	(.0)	.0	(.0)
Homosyllabic consonants	5.1E-02	(.3)	.0	(.0)	.0	(.0)	.0	(.0)
Heterosyllabic consonants	.0	(.0)	.0	(.0)	.0	(.0)	.0	(.0)
Diphthongs	5.1E-02	(.3)	5.6E-02	(.2)	.0	(.0)	.0	(.0)
Total additions	1.0	(1.8)	.7	(1.1)	2.3E-02	(.1)	7.7E-02	(.3)
Absolute total of errors	117.8	(90.2)	103.1	(76.5)	1.9	(5.7)	2.2	(7.7)

[a] Subgroup 1 includes children with Down syndrome (DG) without open bite; Subgroup 2, DG with open bite; Subgroup 3, children of the control group (CG) without open bite; Subgroup 4, CG with open bite.

this malocclusion. There was a significant difference between them ($\chi^2 = 24.6$; $P < .001$).

Speech Disorders Associated With Anterior Open Bite

The percentages of the children from the four subgroups that presented some error type can be seen in Table 1. When comparing the percentages to determine whether any significant differences existed between the subgroups of the children with DS and the children without disabilities, we observed a significant difference only in the presence of additions in the children of the Subgroups 1 and 2 ($\chi^2 = 4.59$; $P < .05$).

The means by error types that occurred in the children of the four subgroups are shown in Table 2. These mean values were compared, through the one-way ANOVA test and the Tukey post hoc test, and marked with an asterisk when there was any statistically significant difference between two subgroups. There was a significant difference among the subgroups of different groups in all the cases except in the substitution of the heterosyllabic consonants ($F = 2.72$; $P = .068$) and of diphthongs ($F = 1.23$; $P = .302$). When comparing means of error type by omissions and substitutions, we found that the two subgroups belonging to the CG (Subgroups 3 and 4) manifested the lower means.

Table 2. Extended

ANOVA		Tukey Post Hoc Test					
F	(*P*)	1–3	1–4	2–3	2–4	1–2	3–4
6.25	(<.001)	*	*				
18.69	(<.001)	*	*	*	*		
10.50	(<.001)	*	*	*			
28.93	(<.001)	*	*	*	*		
71.24	(<.001)	*	*	*	*		
112.93	(<.001)	*	*	*	*		
84.36	(<.001)	*	*	*	*		
17.12	(<.001)	*	*	*	*		
9.31	(<.001)		*	*	*		
22.79	(<.001)	*	*	*	*		
24.67	(<.001)	*	*	*	*		
5.89	(<.001)	*	*				
9.39	(<.001)	*	*				
2.72	(.068)						
1.23	(.302)						
24.30	(<.001)	*	*	*	*		
1.17	(.326)						
2.20	(.092)						
2.12	(.102)						
.59	(.622)						
.52	(.667)						
.23	(.876)						
—	—	—					
2.42	(.070)						
.60	(.617)						
3.72	(.014)	*					
.94	(.426)						
5.57	(<.001)	*	*				
.63	(.594)						
—	—	—					
1.02	(.386)						
5.33	(.002)	*					
31.68	(<.001)	*	*	*	*		

On the other hand, we observed no significant differences between the subgroups when analyzing the error type by distortion ($F = 2.42$; $P = .070$). Finally, when we compared the means by additions made in each of the four subgroups, significant differences were present only by additions of initial single consonants between the subgroups of children without anterior open bite (Subgroups 1 and 3) ($F = 3.72$; $P = .014$), as well as by additions of final single consonants among the children with DS without anterior open bite (Subgroup 1), and the children of the two subgroups belonging to the CG (Subgroups 3 and 4) ($F = 5.57$; $P < .001$). In all the cases the lower means were presented in the subgroups of the CG (Subgroups 3 and 4).

DISCUSSION

Some methodological limitations apply to the present study. Results cannot be extrapolated to the overall population with Down syndrome in Mexico, in particular because this is a highly detailed study in a small, well-defined population, but also because no reliable Down syndrome national figures are available. Furthermore, the student population at The John Langdon Down Foundation probably represents a population whose needs are met with the highly-structured, well-documented educational and medical programs the Foundation offers. Other children with Down syndrome may have less advantageous oral health or educational situations or diagnoses.

It has been proposed that matching children in terms of the level of language comprehension is the most appropriate strategy given the current knowledge on language development in DS.[8] However, currently there is no specific protocol for assessing articulation and phonology in children with DS.[7] Nevertheless, the most common technique for investigating the quality of speech sound production is subjective analysis of test syllable words, or text passages by speech pathologists. There are some disadvantages. For example, each speech sound production is a unique, fundamentally nonreproducible event, and the analysis depends on the hearing abilities, training, and subjective assessment of the examiner. On the other hand, speech intelligibility can be judged by someone who is receiving and interpreting a spoken message. The disadvantages can be avoided by having recordings evaluated by several examiners.[14] In the present study, two calibrated examiners (a language therapist and a pediatric dentist) applied the phonological test. Some studies have reported that phonological process analyses have highlighted similarities between children with DS and those with typical development.[3,15]

The frequency and nature of omissions and substitutions in DS speakers has been documented in several studies, but data on distortions and additions are virtually nonexistent.[15] In the present work, we included an analysis of distortions and additions.

Studies reported in the literature have been carried out in which (1) the main alterations of speech have been observed in children with DS, without taking into account the malocclusion, and (2) the possible association between the alteration of speech and the malocclusion in general, or the open bite specifically, has been evaluated only in infantile populations without DS.

Regarding the first type of study, Dodd[16] reported that children with DS made twice as many articulation errors and almost twice as many inconsistent substitutions as did children with mental retardation matched for mental age. In the current study, we did not include children matched by mental age, but we observed that in the DG the highest means of errors due to substitutions were presented in the simple consonants when affected phonemes were located in the initial and medial positions of the words. In our study children with DS made fewer substitutions in the vowels and in the diphthongs than in the consonants.

With regard to the second type of studies, Tomita et al[17] reported that in 2139 boys and girls aged 3–5 years without DS, the malocclusion did not show any influence on speech difficulties. However, Stahl and Grabowski[18] reported that open bite was the malocclusion most frequently associated with dysfunction in speech in 8864 preschool and school-aged children without DS. Our results agree with those obtained by Tomita et al,[17] since we did not observe significant differences between the subgroups of the CG children when they made omissions, substitutions, distortions, or additions of the phonemes. The same occurred with the DG, except that the children of this group with anterior open bite substituted the vowels significantly more often than did the children with DS and without this malocclusion.

Khinda and Grewal[19] found a strong association between the open bite and the articulation disorders in children aged 7–16 years without DS. Contrary to what Nicolá and Jonathan[12] reported, we did not find that some phonemes were significantly affected by the presence of anterior open bite in the CG.

Baños et al[20] reported that in 50 Mexican children, aged 5 years, without DS with language delayed, 32% showed open bite. Of the 32%, 16% manifested substitutions, 12% omissions, and 4% distortions of phonemes.

The prevalence of anterior open bite reported by Baños et al[20] was similar to that found in both groups in the present study. In the CG the anterior open bite was present in 22.8% of the cases, whereas in the DG this malocclusion was detected in 31.6%. Of the children without disabilities who had anterior open bite, 7.7% presented omissions, 15.4% substitutions, and 7.7% additions, with no case of distortions. In general, our results are similar to those reported by Baños et al.[20]

CONCLUSIONS

- The prevalence of anterior open bite is significantly higher in the DG than in the CG.
- The anterior open bite was not associated with speech disorders either in children with DS or in children without disabilities.
- Early orthodontic treatment of children with DS and open bite may not be justified for the improvement of speech, but there are several other dental conditions for which early treatment may be indicated.

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