

Voice Parameters in Children With Down Syndrome

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Summary: Down syndrome (DS) is the most frequent chromosomal disorder. Commonly, individuals with DS have difficulties with speech and show an unusual quality in the voice. Their phenotypic characteristics include general hypotonia and maxillary hypoplasia with relative macroglossia, and these contribute to particular acoustic alterations. Subjective perceptual and acoustic assessments of the voice (*Praat-4.1* software) were performed in 66 children with DS, 36 boys and 30 girls, aged 3 to 8 years. These data were compared with those of an age-matched group of children from the general population. Perceptual evaluations showed significant differences in the group of children with DS. The voice of children with DS presented a lower fundamental frequency (F_0) with elevated dispersion. The conjunction of frequencies for formants (F_1 and F_2) revealed a decreased distinction between the vowels, reflecting the loss of articulatory processing. The DS vocalic anatomical functional ratio represents the main distinctive parameter between the two groups studied, and it may be useful in conducting assessments.

Key Words: Down syndrome—Voice—Acoustical—Perceptual—Pediatrics.

INTRODUCTION

Down syndrome (DS) is the most common aneuploid disorder, with a frequency of 1/770 live births worldwide.¹ The life expectancy for these individuals now exceeds 50 years, attributable mainly to a more interventionist therapeutic approach.² The most direct consequence of the lengthening lifespan of persons with DS is the increased number of affected adults in the population, with several implications for service providers, namely, the need for special health support, educational, vocational, and social services, to achieve a better quality of life. Communication plays an increasingly greater role in everyday life in our society, reflecting the growing importance attributed to disorders of the voice, speech, and language. Thus, individuals with disabilities in speech are at great disadvantage in terms of cognitive, mental, and social development. The majority of individuals with DS have

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some difficulties with the processes of oral communication, and may present an unusual quality of voice, both having a possible negative social effect.³ The phenotypic characteristics of DS include mental retardation, general hypotonia, maxillary hypoplasia with a relative macroglossia, short neck, and obesity, all of which could contribute to particular acoustic alterations.¹

Perceptual studies describe the quality of voice of individuals with DS as husky and monotonous,⁴ and raucous and low pitched⁵ and, when compared with voices in the general population, these children's voices exhibit more breathiness, roughness, and nasality.⁶ The possible anatomical and physiological bases for the vocal phenotype remain largely undefined.

Providing objective assessment of voice quality has been emphasized by the increasing availability and use of methods of acoustic analysis for clinical diagnosis, monitoring therapeutic progress, speech therapy, and research. Nevertheless, assessment of pediatric vocal problems can be demanding. Fiberoptic endoscopy is often difficult in the uncooperative child, and the stroboscopic examination is technically challenging in any young patient.^{7,8}

Acoustic characteristics are culture- and language-dependent. Few reports have described the acoustic characteristics of the voices of children with DS, and they used very small numbers of patients.^{3,9} With respect to Portuguese-speaking populations, only one study (of Brazilian Portuguese) concerned adolescents.¹⁰ It is therefore useful to obtain more data about perceptual and acoustic parameters that may contribute to our knowledge of differences in vocal quality of children with DS. The main objective of this study was to assess vocal quality in a group of children with DS, compared with an age-matched control group. This evaluation included both objective acoustic measures and perceptual evaluation. To our knowledge, this is the first such study performed in Portuguese-speaking children with DS.

PATIENTS AND METHODS

Subjects

Recruitment of children with DS was done by mail to the main organizations working with this

group in Portugal. Two centers for clinical observation were involved, one in the south (Lisboa) and the other in the north (Porto). The criteria for inclusion were the following: (1) cytogenetic diagnosis of trisomy 21, (2) age between 4 and 8 years, (3) integration in general schools, (4) no evidence of significant hearing loss, (5) adequate cooperation, and (6) informed consent from their legal representatives. The children with DS in this age range are usually able to present good levels of collaboration, and they are still distant from presenting vocal alterations due to puberty.

Approval was obtained from the committees on ethics in research of all the institutions involved. Of the 106 children with DS, 66 (36 boys and 30 girls) with a mean age of 5.8 years were selected for inclusion in this study. An otolaryngologist examined all the children included in this study. All recent hearing tests were analyzed. The cooperating children who did not pass recent hearing tests underwent a hearing screening before collection of data. Only children with absence of significant hearing loss (thresholds lower than 40 dB for 500–4000 Hz) were included.

The control group without DS was composed of 204 children (104 boys and 100 girls) recruited at local schools, with a mean age of 5.7 years. According to their teachers, their developmental level was close to their chronological age. Children with nasal or laryngeal pathology and significant hearing loss were excluded.

Recording

To obtain vocal samples, the children were instructed to vocalize and sustain each of the five main Portuguese vowels (/a/, /e/, /i/, /o/, and /u/), for at least 3 seconds, at their most comfortable pitch and at a constant intensity. Each vowel was recorded at least five times. They also named a group of figures, presented on cards, applying the main phonetic sounds in Portuguese. Order and elements of tasks were randomized for each subject. Recordings were made in a quiet room with a high-quality tabletop microphone (Sony-V420; Sony Electronics, Inc., New York, NY) using a constant mouth-to-microphone distance of 10 cm and 45° off-axis positioning. This was achieved by using a fixed forehead rest and instructing children

to keep their foreheads against the device. Voice input was recorded using a digital audiotape recorder (Sony DAT TCD-D3; Sony Electronics, Inc.) sampled at 44,100 Hz with 16-bit resolution and stored in an IBM-compatible computer disk.¹¹ The middle stable portion of the sample was then extracted.

Acoustical analysis

Records of sampled vowels were analyzed by computer using the program *Praat* (Version 4.1). Acoustic analysis of the voices relied on a task presenting sustained vowels to avoid as far as possible any interference with the control of speech prosody and articulation. The parameters, calculated by *Praat* and displayed numerically and graphically, were each classified into one of seven groups of measurements: (1) fundamental frequency (F_0), including average F_0 , standard error (SE) of F_0 , variability coefficient ($SE/F_0 \times 100\%$), highest F_0 , and lowest F_0 ; (2) intensity (I) as well as average I and maximum I ; (3) frequency-perturbation, through absolute jitter and jitter %; (4) amplitude-perturbation, with shimmer dB and shimmer %; (5) evaluation of noise by harmonic-to-noise ratio (HNR); (6) evaluation of balance by spectral tilt (ST); and (7) formants (F), from F_1 to F_3 , and their bandwidths (Bw), from Bw₁ to Bw₃. The *Praat* program applies a Gaussian-like window and computes the linear predictive coding (LPC) coefficients using Burg's algorithm for each window of analysis.¹² Matches between values for frequencies of formants from computer analyses and those from the spectrogram were confirmed visually. Because the various measures of perturbation of pitch and amplitude exhibit high internal correlation, only some of the parameters for jitter and shimmer were considered for further analysis. Assuming that the high proximity of the first formant and second harmonic frequencies in vowels /i/ and /u/ removes reliability from the procedure, the ST was not considered for these vowels.¹³

Perceptual analysis

Perceptual analysis was done by a panel of two expert speech therapists, using a modified four-point scale of the GRBAS scale proposed by Hirano.¹⁴ The scale adapted to Portuguese as the RASAT scale was used¹⁵: "Rouquidão" (grade),

"Aspreza" (roughness), "Soprosidade" (breathiness), "Astenia" (asthenic speech), and "Tensão" (strained speech). The scale varies from 0, normal to 3, severe. For the sake of auditory-perceptual analysis, the same types of acoustic samples were blindly randomized across patients.

Statistical analysis

Descriptive statistics were obtained for data for both the patients with DS and the control group (non-Down syndrome). For overall comparison of the groups, a single factor (group) analysis of variance with two covariables (ANCOVA) was used for the different variables under study. Gender and age were selected as covariables, to withdraw from the analysis any error due to possible differences across gender and age. All statistical analysis was performed using *SPSS Version 13.0* for Windows (SPSS, Chicago, IL).

RESULTS

The vocal profiles of children with DS were compared with data from the group of children without DS. Although chronological age was used to compare the results of the two groups, it should be remembered that the developmental age is always lower in the group with DS.

Perceptual ratings

All the parameters for perceptual analysis showed significant differences ($P < 0.001$) between the two groups, although the difference was only slightly relevant for "asthenia" (asthenic speech) ($P < 0.01$) (Figure 1). Children in the group with DS functioned on a significantly lower level than did children in the control group.

Acoustical analysis

The results for the main acoustic parameters in the two groups studied are shown in Figures 2–4. Comparison of groups with adjustments for age and gender was performed for the obtained acoustic variables through ANCOVA. From this analysis, we found differences between groups to be very significant ($P < 0.001$) for all variables evaluated for each vowel studied, with the exception of average values of F_0 for vowel /u/, where no significant

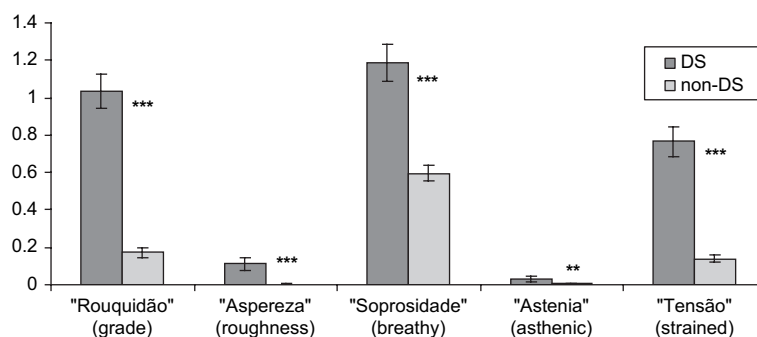


FIGURE 1. Perception evaluation variables (RASAT (four-point scale): mean and SE of mean of speech therapy evaluation data for DS and non-DS children group. Differences between each group, according to the Mann-Whitney test: ** $P < 0.01$; *** $P < 0.001$.

difference was found between the two groups. In general, children with DS exhibited a lower value for all the different variables of F_0 across all vowels except for the variability coefficient, superior in the group with DS (Figure 2). Mean values of individual measures of standard error (SE) of F_0 for children of both groups were compared. The values for SE for children with DS were significantly higher. Moreover, the values for SE decreased with age in the group without DS, something not observed in children with DS (Figure 3). Measurements of frequency-perturbation were also statistically different in the two groups, with higher values obtained in the group with DS (Figure 2).

Concerning measurements of intensity- and amplitude-perturbation, values for the group with DS were always greater than those for children without DS (Figure 2).

Measurements for noise evaluation showed significant differences for HNR, with lower values for children with DS, and the same was verified for ST (Figure 2).

The formants (F_1 to F_3) also exhibited significant differences between the two groups (Figures 2 and 4). Studying the relationship between values for F_1 and F_2 for the five vowels (vowel triangle) revealed that the group with DS had a more limited triangle than the control group, with less distinctive individual values (Figure 4). The relationship of values for F_1 between vowels /a/ versus /i/ and /u/ (studied to evaluate the effects of height of the tongue) and values for F_2 between vowels /i/ versus /u/ (studied to assess the positioning of the body of the tongue

along the horizontal plane) were also assessed, and it was shown that the two groups were statistically different in these (Table 1).

DISCUSSION

Children with DS frequently participate in speech therapy. Intervention with these patients' vocal patterns is important, as they may worsen their efficiency at communication in consequence of their voice deviations. Acoustic evaluation of the voice allows for quantitative analysis of changes that are subject to regression in these children. As better understanding of their deficits in motor control for speech is achieved, therapy can be more effectively designed to address those deficits. The present study was designed to achieve a more complete understanding of both the acoustic bases and the underlying anatomical and physiological bases of the differences that set the voices of many children with DS apart from the voices of normal counterparts.

Perceptual evaluation of the voice is essentially based on subjective assessment by the examiner, according to international standard protocols, and should be considered as a complementary evaluation.¹¹ Although in the present work *Praat* software was used to evaluate acoustics, the results can to a large extent be considered valid for comparison with results using MDVP. For discussion of this issue several available results were considered. For instance, Deliyski and colleagues¹⁶ showed that results obtained using *Praat* and MDVP have

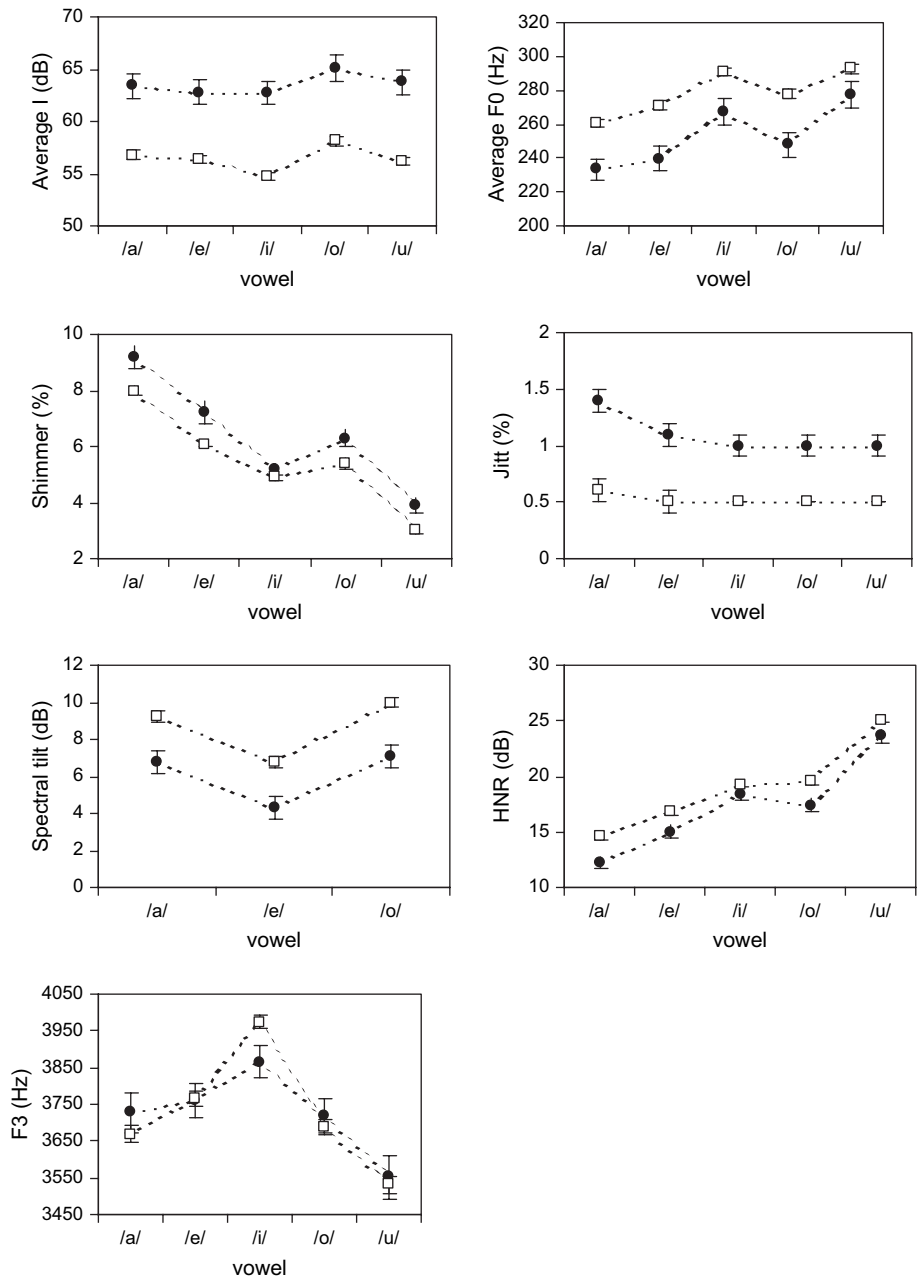


FIGURE 2. Mean and SE of mean of some acoustic variables of the voice of DS (●) and non-DS (□) children. Jitt (%), jitter (%); F_3 , third formant.

been in full agreement for measurements of amplitude-perturbation. Moreover, slight differences that may exist between these programs would only be identifiable under situations of very low signal-to-noise ratio. *Praat* uses a compensation of the auto-correlation function and is stricter in classifying vocal segments as such (0.45 threshold compared

to 0.29 threshold for MDVP).¹⁷ As all our experiments reported here were performed in quiet environments, and vowels were used as sampling sounds, it can therefore be considered that the two algorithms would provide similar results.

The vocal disorder in children with DS is well recognized during infancy, and is described as

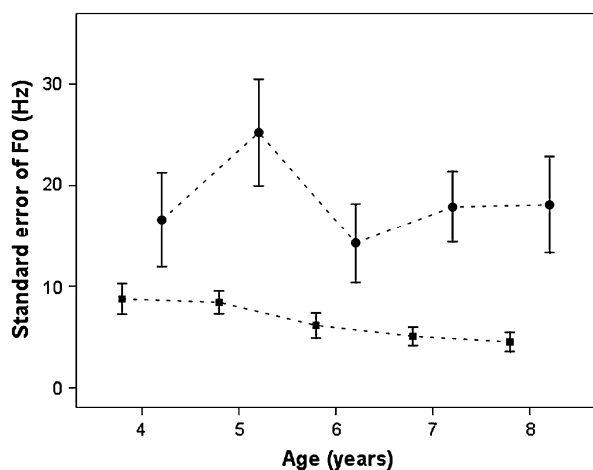


FIGURE 3. Evolution of SE of F_0 (Hz) in DS (●) and non-DS (■) children with age range.

a monotonous, rough, long-lasting cry, with an F_0 of about 60 to 70 Hz.^{8,18} Other studies have found that infants with DS begin their canonical babbling (an important developmental precursor to spoken language) 2 months later than normal, and show also a less stable pattern with increasing age than infants developing normally.¹⁹ Some investigators have described the perception of difference in vocal quality of children with DS as more strained, hoarse, and guttural,¹⁸ and as exhibiting more breathiness, roughness, and nasality⁶ and at a lower

pitch compared to voices of a group of normal children.²⁰ Perceptual evaluation of children with DS in the present study confirmed data from previous reports. These children presented a significantly higher degree of hoarseness, roughness, and breathiness, with strained and even asthenic voices, compared to voices of the control group (Figure 1).

Other reports, however, have showed no consensus for distinction of vocal quality in speakers with DS, such as higher values,⁹ and no differences³ in values for F_0 in children with DS, compared to voices of normal children of similar ages. In fact, the present study demonstrates that children with DS, compared to the control group, had a lower F_0 with higher values for SE, which may reflect the perceived vocal instability. These findings are probably related to the large variability of frequencies for F_0 typically presented in vocal disorders of neurologic origin. In neurologic disorders, there can be a lack of control of vocal fold tension, and this can affect vibration frequency.²¹ The instability of the voice may decrease the intelligibility of speech and present a negative psychodynamic concern, lowering the real capacities of the speaker.

A study of Brazilian Portuguese-speaking adolescents with DS confirmed that the pattern of vocal quality in 4- to 8-year-olds persisted with age.¹⁰ The voices of these subjects were characterized by vocal instability, nasality, and hoarseness. The present data provide evidence that values for SE decrease with age in the control group, but this does not occur in the group with DS. This is an important developmental aspect, since there is normally a gradual age-dependent decline in variability of performance until at least 11 to 12 years of age.²² If variability is taken as an index of maturation of motor control, then it appears that production of speech in children with DS lacks the improvement observed in the age-matched control group.

Current results support hypotheses by Honda²³ concerning the effects of position of the tongue on frequency of F_0 in vowels. As in the control group, children with DS exhibited higher mean values of F_0 for the vowels produced with the tongue in a high position, such as /i/ or /u/ (although these children maintained lower means than in the control group), and they had a lower mean F_0 for vowel /a/ (Figure 2). This suggests

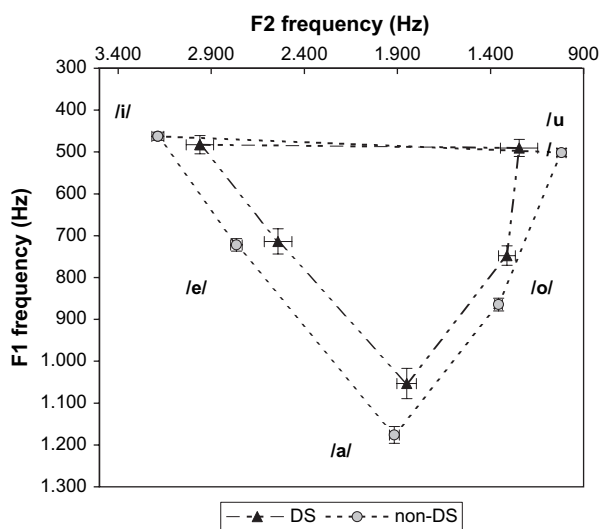


FIGURE 4. A plot of F_1 and F_2 frequencies (mean values and 95% confidence intervals) of vowels of two groups of Portuguese children: DS and non-DS.

TABLE 1. Comparison Between DS and Non-Down Syndrome (Non-DS) Relationship of F_1 Frequency Values Between Vowels /a/ and /i/ or /u/ (Tongue Vertical Movement) and of F_2 Frequency Values Between Vowels /i/ and /u/ (Horizontal Tongue Movement) (Mean \pm SE [sample size])

Variable	DS	Non-DS
F_1 /a/ / F_1 /i/ ***	2.24 \pm 0.52 (60)	2.60 \pm 0.51 (203)
F_1 /a/ / F_1 /u/ ***	2.20 \pm 0.53 (60)	2.38 \pm 0.41 (201)
F_2 /i/ / F_2 /u/ ***	2.58 \pm 0.55 (56)	3.22 \pm 0.50 (194)

Values according to t test for independent samples is *** $P < 0.001$.

that the functional effect of the connection between the tongue and the larynx, through the hyoid bone, is maintained in DS.

Measurements of frequency-perturbation in this study were also statistically significant, with higher values for the group with DS. According to Titze,²⁴ jitter varies mainly with the number and firing-rate of motor-units in the thyroarytenoid muscle. Increasing the number and the firing-rates of motor-units progressively smooths the force generated by the muscle, resulting in a decrease of perturbation. This was associated with higher F_0 and lower amounts of jitter. Therefore, it is possible that the lower values of F_0 observed in children with DS are related to the general muscular hypotonia characteristic of the syndrome. This impedes the existence of a more effective motor-unit function of the laryngeal muscles and presupposes a neuromuscular immaturity. It appears to be possible to improve these parameters by teaching the subjects, and through practice, to modify the parameters for twitch in laryngeal muscle, such as motor-unit recruitment and motor-unit firing-rate, with a consequent generation of smooth muscular force and an increase in phonatory stability.²⁵

In the present study, children with DS also exhibited higher values for measurements of amplitude-perturbation. These results are consistent with the findings of Pentz and Gilbert.³ Several studies^{26,27} have found that jitter and shimmer can be significantly correlated with the perception of abnormal vocal qualities, including hoarseness, breathiness, and harshness. Others find that the HNR value is the most sensitive indicator of changes in the vocal organ and is an excellent clinical parameter in the analysis of both vocal pathology and treatment.²⁸ In the present study, the group

with DS also presents a constant lower value for the quotient between harmonics and noise levels, across all vowels. This represents a higher degree of noise in the analyzed sample, compared with children in the normal group as reported by other investigators.³ ST is a measure that mainly assesses performance at the glottal source. This variable is calculated according to Praat in differences in band energy. A decreased value of ST in the group with DS is related to a breathier and more forced voice, compared to voices in the control group. Research into this area is still ongoing.

Analysis of formants

The well-described differences of shape and dimension of the vocal tract between the two groups studied may explain some of the variations in acoustic and perceptual assessments, mainly at the frequencies for formants. The degree of vowel opening associated with the lowering of the mandible and of the tongue has a direct relationship to frequency for F_1 , which increases with the opening of the mouth.²⁹ Therefore, analysis of the first formant (F_1) revealed that mean values were mainly low for the low-back vowels (/a/ and /o/) compared with those spoken by the control group (Figure 4). This may be related to the limited adjustment of vertical movement of the relatively large tongue in the undersized oral cavity characteristic of children with DS.

The new proposed parameter DS-VR

In the group with DS, excluding low-back vowels /a/ and /o/, the other three vowels showed substantial variation in values of frequency for F_2 (Figure 4). The mid-upper-front vowels /e/ and /i/ displayed an important decrease in mean values

for F_2 of about 8% and 7%, respectively. The upper-back vowel /u/ presented a large increase in values for frequency of F_2 of about 22%, which could be due to a limited range of tongue movement in the high-back position, probably related to maxillary hypoplasia and to decreased pharyngeal space.²⁹ Moreover, due to the opposite signs of these two variations in mean values of F_2 for vowels /i/ and /u/, joint evaluation by means of the ratio $F_{2\ /i/} / F_{2\ /u/}$ can show a cumulative decrease of about 20% (quotient 0.8) relative to children in the control group (Table 1). Inversely, it represents an increase of 25% (quotient 1.25) for children in the control group when compared with children in the group with DS.

This parameter, the variation in ratio of values of F_2 for the two extreme-upper vowels (/i/ and /u/), reveals itself as a strong candidate for distinguishing among voices in groups who have DS and children who do not. We call this new parameter the “DS vocalic anatomical functional ratio” (DS-VR). The DS-VR parameter should be very useful for monitoring therapeutic evolution in children with DS. We intend to research the potential application of DS-VR in daily clinical use, and for this purpose it will be very important to document the effectiveness of this variable throughout the age range.

Regarding the frequencies of F_2 and corresponding horizontal movement of the tongue with effects on the pharyngeal space, values obtained for vowels /e/ and /i/ may also reflect difficulty in effective anterior movement of the tongue resulting in decreased pharyngeal space.²⁹ The less functional forward movement of the tongue in children with DS does not allow enough laryngeal tension, and this was reflected in the lower F_0 found in the group with DS.³⁰ The differences in frequency of F_1 and F_2 for the five vowels spoken by both groups are well expressed in Figure 4. They have globally a similar pattern but the group with DS exhibits an inner disposition, as the strategy to produce the different vowels may be acquired in a less accurate way. The existence of smaller distinctions between the *loci* of representation of average values for pairs of frequencies (of F_1 , F_2) for the group with DS may cause reduced discrimination of vowels. These results probably indicate the

difficulty of neuromuscular adjustment in the vocal tract and difficulty in aerodynamic control during the production of vowels. On the other hand, increase of means of frequencies for F_3 in the group with DS, with the exception of vowels /e/ and /i/ that showed a reduction, is not totally correlated with the decreased space in the oral cavity behind the incisors²⁹ (Figure 2).

It may be important to assess the morphology of the vocal tract by magnetic resonance imaging to try to explain the observed acoustic differences between the two groups. Specifically, we would like to explain objectively why the five vowels are subdivided into two groups, namely, the back-low vowels (/a/ and /o/) that present substantial variations in frequencies for F_1 , and the others vowels (/i/, /e/, and /u/) in which the main variation is observed in frequencies for F_2 (Figure 4).

CONCLUSIONS

In conclusion, the voice in children with DS presented a lower frequency for F_0 with increased dispersion (SE), probably related to perceptual instability. All the measurements of perturbation and noise evaluation that we analyzed showed significantly higher values in children with DS compared to those for the control group. Otherwise, the conjunction of frequencies for F_1 and F_2 reveals decreased distinction among the vowels, reflecting the loss of discrimination of speech. The proposed DS-VR parameter represents the main distinctive parameter between the two groups studied, and it will probably be useful as a new therapeutic assessment. It must be emphasized that by applying this tool, DS-VR, we were able to show that it is possible to find and use some invariant features for children with DS, which contrasts with well-known phenotypic variation in the syndrome. The present data demonstrate characteristics of resonance of the vocal tract in children with DS and may help to define their functional vocal profile. These results appear to reflect the configuration of the vocal tract in children with DS and their developmental process in neuromuscular control of speech. The increased understanding of voice characteristics of children with DS should provide a basis for therapeutic intervention when its need is indicated.

Computer-assisted vocal analysis, here implemented by using *Praat* software, provides objective and reproducible acoustic measurements, and it is well tolerated by children as young as 4 years old. These attractive features are relevant to its application in a pediatric population, especially those with mental retardation.

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