

# Acoustic analysis of prosody in Sydenham's chorea

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## ABSTRACT

There are few studies of language and speech in patients with Sydenham's chorea (SC). We have done an acoustic analysis of fundamental frequency ( $F_0$ ), duration and intensity of declarative and interrogative sentences made by 20 SC patients, 20 patients with rheumatic fever (RF) without chorea, and compared them with 20 healthy age-matched controls (CO). Each group included 12 females. We found that there is no difference between the RF and CO groups in all studied parameters. Patients with SC, however, presented with a speech characterized by decreased  $F_0$  range (difference between minimum and maximum  $F_0$ ), shorter duration of sentences, and higher intensity of the first syllable of sentences. The findings were not influenced by the nature of the sentences (i.e., declarative or interrogative), but for all variables they were significantly more severe in males than females. In conclusion, we have demonstrated that patients with acute SC have an impairment of modulation of  $F_0$  and longer duration of emission of sentences, resulting in a monotone and slow speech. This pattern is similar to what has been described in other basal ganglia illnesses, such as Parkinson's disease, Huntington's disease and Wilson's disease.

**Key words:** Sydenham's chorea, chorea, rheumatic fever, speech, prosody.

## Análise acústica da prosódia em coréia de Sydenham

## RESUMO

Há poucos estudos sobre linguagem e fala em pacientes com coréia de Sydenham (CS). Fizemos uma análise acústica da frequência fundamental ( $F_0$ ), duração e intensidade de sentenças declarativas e interrogativas feitas por 20 pacientes com CS, 20 pacientes com febre reumática (FR) sem coréia, comparando-os com 20 controles saudáveis e pareados por idade (CO). Cada grupo incluiu 12 mulheres. Foi encontrado que não há diferença entre os grupos FR e CO quanto a todos parâmetros estudados. Pacientes com CS, contudo, apresentaram-se com fala caracterizada pela redução da variação de  $F_0$  (diferença entre  $F_0$  mínima e máxima), duração mais curta das sentenças e maior intensidade da primeira sílaba das sentenças. Os achados não foram influenciados pela natureza das sentenças (i.e., declarativas ou interrogativas), mas os achados foram mais evidentes em todas as variáveis em homens em contraste com mulheres. Em conclusão, demonstramos que pacientes com CS aguda têm prejuízo da modulação da  $F_0$  e duração mais longa da emissão de sentenças, resultando em fala mais lenta e monótona. Esse padrão é semelhante ao que tem sido descrito em outras feições dos núcleos da base, tais como doença de Parkinson, doença de Huntington e doença de Wilson.

**Palavras-chave:** coréia de Sydenham, coréia, febre reumática, fala, prosódia.

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Sydenham's chorea (SC), one of the major criteria of rheumatic fever (RF), is the most common cause of acute chorea in children worldwide and remains as an

endemic condition in developing and underdeveloped areas of the world<sup>1</sup>. SC presumably results from antibodies induced by  $\beta$ -hemolytic group-A Streptococcus

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which cross-react with basal ganglia epitopes<sup>2</sup>. Disruption of the striato-frontal motor circuit is associated with chorea and tics in SC<sup>1,3</sup>. There is also evidence of impairment of the basal ganglia circuit related to behavior control in SC, resulting in the development of obsessions, compulsions and other behavioral abnormalities in a significant proportion of patients<sup>4</sup>.

Prosody refers to non-verbal aspects of the speech, including intonation, rhythm, accent and temporal distribution. An acoustic investigation of prosody involves analysis of fundamental frequency,  $F_0$ , (physical correlate of melody), duration (physical correlate of articulation time) and intensity (physical correlate of the vocal energy employed by the speaker)<sup>5,6</sup>. Parkinson's disease (PD) is the disorder of the basal ganglia where prosody has been more comprehensively investigated. At least 80% of subjects with PD develop speech disorders over the course of their disease. The speech of PD patients is characterized by variable mean  $F_0$ , inability to keep a stable  $F_0$ , a smaller range of  $F_0$ , responsible for a monotone speech, decreased and imprecise vocal intensity, imprecise articulation, vocal tremor, and reduced speech velocity. These abnormalities may be present in early stages of the disease, their frequency and severity increase with progression of PD, and have little response to dopamine replacement therapy<sup>7-15</sup>. In contrast, speech has been scarcely studied in patients with chorea. Even in Huntington's disease (HD), the most common cause of hereditary chorea, there are just a few investigations of prosody. Gordon and Illes found decreased verbal fluency as well as reductions of melodic line, phrase length, and articulatory agility<sup>16</sup>. Studying eight patients with clinical diagnosis of HD, Ramig found that their sustained vowel phonation was characterized by abrupt drops in  $F_0$ , vocal arrests, reduced maximal vowel duration, and adductory and abductory phonatory disruption<sup>17</sup>. Another investigation demonstrated the existence of speech timing impairment in HD, with reduction in the duration of syllables, of pauses between phrases, and of sentences<sup>18</sup>. Hertrich and Ackermann showed that speech in HD is characterized by reduced  $F_0$ , range of modulation of intensity, velocity, and duration of the syllable accent<sup>19</sup>. These same authors studied speech timing in 13 subjects with HD, confirming the findings of Ludlow et al.<sup>18</sup>: all patients presented with increased variability of utterance duration and/or voice-onset-time. A subgroup had reduced speech time concomitant with overproportional lengthening of short vowels<sup>20</sup>.

To our knowledge, there has been no investigation of prosody in SC. In fact, speech and language have been poorly studied in SC. In a retrospective chart review of 240 cases, Nausieda et al.<sup>21</sup> report that 38.7% of their patients had dysarthria. More recently, we have demonstrat-

ed that patients with acute SC have decreased phonetic, but not semantic, verbal fluency in comparison with matched healthy controls<sup>22</sup>. As it is a hyperkinetic condition, one can hypothesize that patients with SC and HD share similar prosodic features. The aim of this study is to perform an acoustic analysis of speech of patients with SC to determine the features of their prosody.

## METHOD

We have enrolled three groups of subjects to participate in this study: patients with RF but without SC (Group RF), patients with acute SC (Group SC), and healthy controls (Group CO). The inclusion criteria for the RF were the Jones criteria; SC was diagnosed according to a Jones modified criteria (i.e., acute chorea with unknown cause despite comprehensive workout to rule out alternative explanation; history or previous Streptococcus infection, presence of concomitant carditis, elevated ASTO, and/or acute phase reactions further support the diagnosis)<sup>23</sup>. Acute SC was defined as first bout of chorea. For each SC patient, we recruited one RF patient and one control subject of same gender and age. All subjects, native speakers of Brazilian Portuguese, underwent audiological, laryngological and neurological evaluations.

The subjects were instructed to read and retain five declarative and five interrogative sentences, which later they would speak without reading them. Previously a study with children native speakers of Brazilian Portuguese showed that this method of induction reproduces prosodic features similar to spontaneous speech<sup>24</sup>. The sentences were recorded on a Digital Audio Tape Recorder (TCD-D8 Sony®). The unidirectional cardioid head microphone (HD-74 Leson®) was placed 20 cm in front of the mouth of the subject. The acoustic analysis of the statement was performed by the software WinPitch 1.93 (Philippe Martin®). The following prosody parameters were studied: maximum  $F_0$ , minimum  $F_0$ , difference between maximum and minimum  $F_0$  (tessitura); duration of sentences, number of syllables of sentences; and intensity of the initial syllable of the sentences. The statistical analysis used t-Student test as well as ANOVA test by the method of Tukey for multiple comparisons, with differences considered statistically significant if  $p < 0.05$ . The protocol was approved by the local ethics committee and all participants signed an informed consent.

## RESULTS

Each group comprised 20 subjects, with 12 females and 8 males. The mean age ± standard deviation for females and males in all groups were, respectively, 12.41 ± 2.72 years. All SC patients had generalized chorea without parkinsonian features. Subjects in the control and RF group had normal neurological examination, but all of

the latter had carditis. Below follow the results for each of the investigated prosody parameters. Since there was no difference between each of the studied sentences, the results are presented as mean of all declarative sentences and interrogative sentences. Moreover, as prosody is influenced by gender, the results are shown separately for females and males.

### F<sub>0</sub>

The mean±standard deviation of the tessitura of declarative sentences for females in the SC, RF and CO groups were 87.9±26.4, 148.7±42.2 and 159.8±46.8. These values among males were 67.7±33.2, 106.5±59.5 and 114.4±55.7. The figures for interrogative sentences in females were 125.5±34.6, 173.0±51.2 and 180.3±41.1, whereas in males the values were 107.1±49.3, 124.4±50.4 and 145.5±71.6. ANOVA and test of Tukey showed that for both types of sentences, males have smaller tessitura than females ( $p=0.002$  and  $p=0.008$ ). Similarly, this parameter was smaller in patients with CS in comparison with controls and RF patients ( $p=0.000$  and  $p=0.000$ ).

### Duration

The mean±standard deviation of duration of declarative sentences for females in the SC, RF and control groups were 1679.2±518.1 ms, 1320.8±344.1 ms and 1204.2±393.4 ms. These values among males were 1487.5±168.5 ms, 1150.0±187.1 ms and 1256.2±279.6 ms. The figures for interrogative sentences in females were 1675.0±376.3 ms, 1262.5±279.7 ms and 1279.2±270.9 ms whereas in males the values were 1575.0±218.76 ms, 1287.5±201.3 ms and 1250.0±260.5 ms. ANOVA test by the method of Tukey showed that for both types of sentences females produce sentences of longer duration than males ( $p=0.008$  and  $p=0.04$ ). Similarly, this parameter was longer in patients with CS in comparison with controls and RF patients ( $p=0.000$  and  $p=0.000$ ). The mean±standard deviation of number of syllables of declarative sentences for females in the SC, RF and control groups were 4.1±0.7, 5.4±0.2 and 4.8±0.9. These values among males were 4.8±0.2, 5.8±0.2 and 5.7±0.6. The figures for interrogative sentences in females were 4.2±0.7, 5.1±0.8 and 5.4±0.8 whereas in males the values were 4.3±0.5, 5.5±0.8 and 5.6±0.9. Statistical analysis with t-Student test showed that for females, the number of syllables was smaller in patients with CS in comparison with controls and RF patients for both declarative ( $p=0.01$  and  $p=0.001$ ) and interrogative sentences ( $p=0.04$  and  $p=0.001$ ). In contrast, female RF patients produced a greater number of syllables than controls ( $p=0.006$ ) in declarative sentences but a smaller number in interrogative sentences ( $p=0.02$ ). Similarly, males with SC produced a smaller number of syllables than controls and RF patients

for both declarative ( $p=0.001$  and  $p=0.001$ ) and interrogative sentences ( $p=0.04$  and  $p=0.001$ ). There was no difference, however, between controls and RF patients for both declarative ( $p=0.55$ ) and interrogative sentences ( $p=0.85$ ).

### Intensity

The mean±standard deviation of intensity of the first syllable of declarative sentences for females in the SC, RF and CO groups were 41.63±2.64 dB, 33.28±5.00 dB and 32.76±6.35 dB. The values of the SC group were significantly higher than the RF and CO group ( $p=0.000$  in both cases) but there was no difference between RF and CO ( $p=0.78$ ). The results for males were 39.70±5.10 dB, 31.7±5.05 dB and 30.80±3.51 dB. Again, patients with chorea had higher intensity than the RF and CO groups ( $p=0.000$  in both cases) whereas the comparison of RF and CO failed to identify a significant difference ( $p=0.52$ ). The figures for interrogative sentences in females were 42.30±2.82 dB, 34.10±4.62 dB and 33.31±5.37 dB. The comparison between the studied groups yielded results similar to those found for declarative sentences: the intensity in SC was higher than in RF and CO ( $p=0.000$ ,  $p=0.000$ ) and the latter two were similar ( $p=0.66$ ). For males the SC, RF, and CO values were, respectively, 41.12±8.08 dB, 35.18±5.35 dB and 33.22±3.51 dB. Again, SC patients had a higher intensity than RF and CO ( $p=0.000$  and  $p=0.000$ ) which were similar ( $p=0.14$ ).

### DISCUSSION

To our knowledge, this is the first study of prosody in SC. We have enrolled a relatively large number of patients with this condition whose demographic and clinical features (i.e., pediatric age of onset and over-representation of females) are typical of acute SC<sup>1,21</sup>. The inclusion of a group with RF but without SC is justified by the findings that some, although not all, studies have shown these patients have an increased frequency of behavioral abnormalities, such as obsessions, compulsions and hyperactivity, which are also found in SC<sup>1,4</sup>. Furthermore, we have shown that RF patients have mean titer of serum anti-basal ganglia antibodies (ABGA) higher than controls<sup>2</sup>. As ABGA are presumed to be responsible for the development of brain dysfunction in SC<sup>1</sup>, one may speculate that other non-motor features, such as prosody abnormalities, may be found in this particular group of subjects. It could be expected, then, that patients with RF without chorea had changes in prosody comparable to those found in the SC group. However, in contrast to this hypothesis, overall the acoustic analysis of speech of the subjects of our study failed to identify any significant change in prosody of patients with RF but without chorea. The only exceptions were the findings that RF female patients produced a greater number of syllables than con-

trols in declarative sentences but a smaller number in interrogative sentences. Interestingly, these findings were not reflected in the duration of sentences which failed to differentiate the RF and control groups. One possible explanation for only female RF patients having a smaller number of syllables than controls in declarative sentences is the greater susceptibility of females to develop SC<sup>1,21</sup>. According to this hypothesis, circulating ABGA antibodies have a greater potential to induce functional changes in women. It remains unclear, however, why, in contrast with what was expected, our female RF patients produced interrogative sentences with a greater number of syllables than control. Nevertheless, patients with acute SC have a consistent pattern of change of prosody on acoustic analysis: decreased tessitura, i.e., a limited range of change of F<sub>0</sub> both in declarative and interrogative sentences; increased intensity of the sentences; longer duration of sentences and decreased number of syllables, which leads to the conclusion that there was a higher number of pauses. The findings of duration of sentences and number of syllables confirm our previous investigation of a different cohort where we found decreased verbal fluency<sup>22</sup>. For all variables, the results were more evident in males.

From a perceptual point of view, our results show that patients with acute SC have dysarthria characterized by monotone character due to the inability to modulate F<sub>0</sub> and slow speech with longer duration of sentences and decreased number of syllables. These findings have a resemblance with results found in studies of speech of patients with HD. Gordon and Illes, for instance, described reduction of melodic line in their patients<sup>16</sup>. Similarly, Hertrich and Ackermann demonstrated that the difficulty to modulate the speech in HD is not only limited to F<sub>0</sub>, also involving intensity and duration<sup>19</sup>. We have not found, however, variability of speech parameters in our patients with SC as it has been described in some other reports in HD. In one of these studies, there were abrupt drops in F<sub>0</sub>, with adductory and abductory phonatory disruption<sup>17</sup>. Both Ludlow et al.<sup>18</sup> and Hertrich and Ackermann<sup>20</sup> have also found increased variability of utterance duration and/or voice-onset-time. Moreover, in their studies a subgroup of patients were found to have an explosive speech, as defined by reduced speech tempo on acoustic analysis. These findings have been interpreted by the authors as related to choreic movements of the muscles of the phonatory structures. Obviously, this difference may reflect the distinct underlying mechanisms in HD, a degenerative disease with widespread lesions in the brain, and SC, an auto-immune condition with dysfunction confined to the basal ganglia<sup>1</sup>. It is possible however, that methodological differences may account for at least some of the discrepancies. For instance, in one of these studies the author used analysis of sustained emission of a vowel<sup>17</sup>, whereas

we performed acoustic analysis of declarative and interrogative sentences which mimic spontaneous speech<sup>24</sup>.

An interesting point raised by our results is the similarity with features of prosody found in PD in some studies. Given the hyperkinetic nature of SC, one could expect to find features opposite to those seen in PD, the prototype of hypokinetic condition. Of note, even studies of patients with PD whose experimental paradigm differs from ours found results similar to those herein reported for SC. Ludlow et al.<sup>18</sup> found that both patients with HD and PD had problems in the control of sentence duration, although the deficits were more severe in the former. In another study, the authors investigated the temporal organization of the speech in HD and PD, asking subjects to speak a sentence 10 times. They found that all patient groups had low maximal speech rates and decreased variability of speech rate<sup>25</sup>. In our investigation of PD patients with a methodology similar to the present study, we found that in early to moderate stages of the illness the speech is characterized by decreased range of variation of F<sub>0</sub>, unchanged duration, higher intensity which, however, has a lower range of variation<sup>13,14</sup>. There are several potential explanations for the overall similarities of the prosody of PD, HD, and SC. The first hypothesis is the use of anti-dopaminergic agents in patients with chorea causing the development of drug-induced parkinsonism. Although studies of HD have not controlled for this variable, this is an unlikely explanation in our investigation since no one of our patients had any parkinsonian feature. The second possibility is the existence of nigro-striatal dysfunction both in HD and SC. As to the former, this has been well characterized<sup>1</sup>. In relation to SC, we have demonstrated that these patients have an increased risk of developing extrapyramidal side-effects of dopamine receptor blockers<sup>26</sup>. More recently, we found that a significant proportion of patients with remission of SC (disappearance of chorea after withdrawal of all medications) remain with persistent bradykinesia<sup>27</sup>. These findings support the notion of dysfunction of nigro-striatal system in SC. Finally, one may speculate that the basal ganglia role in the control of prosody is impaired in a similar manner regardless of the nature of the cause of the dysfunction, always resulting in a slow speech with decreased tessitura and increased intensity. This hypothesis is further supported by the finding of Volkmann et al that patients with Wilson's disease (WD) also present with this pattern of prosody<sup>25</sup>.

In conclusion, we have demonstrated that patients with acute SC have an impairment of modulation of F<sub>0</sub>, longer duration of emission of sentences as well as increased intensity of sentences, resulting in a monotone and slow speech. This pattern is similar to what has been described in other conditions with basal ganglia impairment, such as PD, HD and WD.

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