

## *Short report*

# Aphasia with elation, hypermusia, musicophilia and compulsive whistling

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**SUMMARY** A musically naive patient with dominant fronto-temporal and anterior parietal infarct developed transcortical mixed aphasia. From early convalescence, he exhibited elated mood with hyperprosody and repetitive, spontaneous whistling and whistling in response to questions. He often spontaneously sang without error in pitch, melody, rhythm and lyrics, and spent long periods of time listening to music. His behaviour progressively improved in parallel with very good recovery of verbal skills. Musicality and singing are rarely tested at the bedside. Preservation of these abilities in aphasics might portend eventual recovery.

Preservation of singing and of musical abilities in musically educated or naive patients suffering from aphasia has been known for over a century.<sup>1</sup> Many reports therefore exist of cases of aphasia with and without amusia which include transcortical aphasias.<sup>2,3</sup> This patient is unusual because he developed exaggerated behaviour of repetitive singing and whistling useless in terms of meaningful substitution communication. He also exhibited a non-fluctuating elated mood and musicophilia.

*Case report.* This 57-year-old right-handed Latin man was admitted to the hospital because of acute onset of severe headache, nausea, slurred speech and vertigo. His past medical history was unremarkable and he never had visual or hearing difficulties. He enjoyed music and dancing but could not read or write music or play a musical instrument. On admission, blood pressure was 200/102 mm t/g, pulse was regular at 76 per minute and he was afebrile. Routine laboratory studies, EEG, CT and radioisotopic brain scans, and complete neurological examination were normal. Spinal fluid was bloody. Cerebral arteriography disclosed a 1.6 cm diameter aneurysm arising from the origin of the left posterior communicating artery. Surgical muscle wrapping of the aneurysm was performed. One day after opera-

tion, the patient developed right horizontal gaze paresis without hemianopsia, right hemiplegia and transcortical mixed aphasia. Babinski sign was positive on the right; Gerstmann's syndrome was not present. Cerebral arteriography revealed severe spasm of the left middle and anterior cerebral arteries with early vascular changes suggestive of an infarct. CT scan 2 weeks after operation showed a large area of low attenuation involving the anterolateral aspect of the left frontal lobe, the left anterior temporal lobe, and the left anterior parietal lobe. A few days after the onset of aphasia, he began spontaneously to sing Spanish songs; he often whistled spontaneously or in response to spoken or written questions. Detailed communication assessment was performed several weeks after surgery. By this time, right horizontal gaze paresis was still present but the hemiplegia had improved and he remained with right-handed preference. His mood and affect were elated without evidence of crying or laughing outbursts. Auditory comprehension of spoken words was poor. There were marked delays in identifying objects by function and name. Monaural and binaural hearing was preserved with good recognition of familiar sounds. There was no evidence of auditory hallucinations. Visual perception was normal. No hemianopsia was present. Reading was moderately impaired with no consistent comprehension of prepositions or object-function phrases. In verbal output examination, he had word-finding difficulties, dysfluencies and phonemic errors. He very frequently whistled instead of attempting to answer with phonemes, but he could not whistle familiar songs to command and could not communicate by means of whistling familiar songs. He did poorly in sentence completion testing. Repetition was excellent. He spontaneously sang Spanish songs without prompting with excellent pitch,

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melody, rhythm, lyrics and emotional intonation. He could tap, hum, whistle and sing along. He could write, and sequentially recite the days of the week or months of the year. Although he could write some intelligible words but not sentences, he did not write the names of any objects or words presented in whole word dictation or words orderly spelled in an intelligible way. Emotional intonation of speech (prosody), spontaneous facial emotional expression, gesturing and pantomimia were exaggerated. Pseudobulbar palsy was not present. After continuous speech therapy, in 28-month followup his entire behaviour pattern has improved, paralleling his great improvement in verbal communication skills.

## Discussion

Attempts to classify the amusias or to localise the areas of the brain involved in musical abilities are often conflicting.<sup>4</sup> Nevertheless, there is considerable evidence presented in favour of the hypothesis that the "minor" non-dominant hemisphere is the site for emotions<sup>5</sup> and music.<sup>6-8</sup> Patients submitted to total dominant hemispherectomy who promptly regained their ability to sing<sup>9</sup> further support this hypothesis in terms of this anatomical location for musical abilities. Equally, carotid injections of sodium amytal generally demonstrate that normal individuals rendered temporarily aphasic by the barbiturate can sing well, while injections into the non-dominant side affects singing, leaving spoken language intact.<sup>10</sup> Dichotic listening of melodies in normal subjects suggests a greater role of the right hemisphere in musical perception,<sup>11</sup> but this role is reversed in musicians who shift musical perception to the dominant hemisphere.<sup>12</sup> It is further postulated that the left hemisphere has a role in sequential arrangement, analysis,<sup>12</sup> tempo, rhythm,<sup>13</sup> and initiation of singing,<sup>14</sup> while automatic melody perception and intonation in a more global mode is executed by the right hemisphere.<sup>12</sup> This patient's clinical findings seem to support the greater role of the non-dominant hemisphere in music, somehow normally dormant and "released" by dominant hemispheric damage. Emotional characterisation of language (prosody) in stroke patients has been shown to be a function of the right hemisphere.<sup>15</sup> The patient herein described exhibited language and musical hyperprosody paralleling his elated mood and affect. Monrad-Krohn<sup>16</sup> considered the dysprosodies as an after-effect of aphasia occurring during recovery. They seemed, therefore, to be associated more with language functions than with musical faculties. A patient reported later, however, developed expressive amusia without aphasia but with dysprosody.<sup>6</sup> Ustvedt<sup>17</sup> noted that musical performance is integrated to emotions and speculated that a subcortical (hypothalamic) locus for music

exists. Recently this integration has been experimentally demonstrated.<sup>18</sup> Although once held as a property of the dominant anterior central convolution,<sup>19</sup> whistling, along with humming, is considered part of the expressive musical language. Pathological mood alterations are common in stroke patients and aphasics.<sup>20,21</sup> Classically, patients with left hemispherical injury are reported to exhibit depressive-catastrophic reactions, contrasted with "indifferent" reactions to the extent of exhibiting excessive jocularity in those suffering from injuries of the minor hemisphere.<sup>22</sup> Benson<sup>21</sup> divided the aphasics into two groups: "concerned" aphasics having non-fluent speech, anterior lesions and features of frustration and depression; and "unconcerned" aphasics with poor comprehension, posterior lesions, and usually indifferent, good humoured or euphoric, and sometimes paranoid behaviour. A recent study<sup>20</sup> comparing stroke and trauma patients concluded that the presence of a frontal lesion was the cause of depression in stroke patients. Alteration of mood of the patient described here resembles more closely the mood observed in "unconcerned" aphasics. This is unusual because he had transcortical mixed aphasia rather than Wernicke's aphasia, and CT showed greater damage of the dominant frontal lobe with the right hemisphere remaining intact. Even more unusual was his unexplained musicophilia.

Because singing and musicality in aphasics are not frequently tested by clinicians, and patients with aphasia usually do not exhibit the spontaneous hypermusicality exhibited by this patient, it is probable that many cases of aphasia without amusia or with preserved singing abilities are missed. This pitfall may lead to failure to refer these patients to a language therapist with knowledge of melodic intonation treatment in aphasics.<sup>23</sup> Preservation of this musical ability in aphasics might portend eventual recovery.

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