Isolated dysarthria due to extracerebellar lacunar stroke: a central monoparesis of the tongue

Peter P Urban, Susanne Wicht, Hanns Ch Hopf, Susanne Fleischer, Otmar Nickel

Abstract

Objectives—The pathophysiology of dysarthria can preferentially be studied in patients with the rare lacunar stroke syndrome of “isolated dysarthria”.

Methods—A single study was carried out on seven consecutive patients with sudden onset of isolated dysarthria due to single ischaemic lesion. The localisation of the lesion was identified using MRI. The corticolingual, corticoroctal, and corticospinal tract functions were investigated using transcranial magnetic stimulation. Corticopontocerebellar tract function was assessed using 99mTc hexamethylpropylene amine oxime-sound emission computed tomography (HMPAO-SPECT) in six patients. Sensory functions were evaluated clinically and by somatosensoy evoked potentials.

Results—Brain MRI showed the lesions to be located in the corona radiata (n=4) and the internal capsule (n=2). No morphological lesion was identified in one patient. Corticollingual tract function was impaired in all patients. In four patients with additional cortico-orofacial tract dysfunction, dysarthria did not differ from that in patients with isolated corticollingual tract dysfunction. Corticospinal tract functions were normal in all patients. HMPAO-SPECT showed no cerebellar diaschisis, suggesting unimpaired corticopontocerebellar tract function. Sensory functions were not affected.

Conclusion— Interruption of the corticollingual pathways to the tongue is crucial in the pathogenesis of isolated dysarthria after extracerebellar lacunar stroke. (J Neurol Neurosurg Psychiatry 1999;66:495–501)

Keywords: dysarthria; lacunar stroke; corticobulbar tract; magnetic evoked potentials; SPECT

Dysarthria is common in cerebral lesions of different origin and location. However, even in patients in whom the site of the lesion was apparent from imaging studies, a definite conclusion on the involvement or sparing of individual fibre tracts could not be reached due to the close proximity and varying location of the tracts so that the nature of dysarthria has never been clear. Dysarthria due to stroke is most often associated with other neurological deficits such as hemiparesis, hemiataxia, clumsiness of one hand, central facial paresis, and tongue deviation. To exclude interferences due to accompanying neurological deficits, we selected patients with isolated dysarthria due to a small singular ischaemic lesion. This lacunar syndrome is exceedingly rare, as shown in a recent consecutive series of 227 patients with lacunar infarction in whom isolated dysarthria was noted in only 0.4%. In seven patients with isolated dysarthria we functionally tested the relevance of major pathways involved in speech production.

Patients and methods

We report on seven consecutive patients with sudden onset of dysarthria in the absence of other previous or current neurological signs or symptoms. The clinical findings in each patient, including the risk factors for stroke, are summarised in table 1. Dysarthria was diagnosed on the basis of auditory-perceptual presentation and confirmed by two experienced speech therapists. Speech function was assessed using a neurophonetic test battery (modified from Ziegler et al). Articulation was evaluated on the basis of various samples—that is, spontaneous speech, repetition of sentences and words, reading a short story, and rapid iteration of syllables (/pa/,/ta/,/ka/). The examination of laryngeal function included laryngoscopy, stroboscopy, and perceptual examination of voice quality, voice stability, pitch, and loudness. Sustained realisation of vowels and fricatives and repetition of sentences of increasing length provided information on respiratory support. Speech tempo was measured based on the syllable repetition rate per second using a sound spectrograph (CSL 4300; Kay Elemetrics Corp, Pine Brook, NJ, USA).

The localisation of the lesion was identified by MRI. Horizontal and coronal or sagittal planes were obtained with conventional spin echo techniques using a 0.5 or 1.5 Tesla tomograph (Philips T5/ACS). All images were T1 and T2 weighted and gadolinium enhanced. Slice thickness was 5 mm without gaps.

The atlases published by Matsui and Hirano and Nieuwenhuys et al were used as anatomical references.

The corticolingual projections were examined by activating the tongue muscles using transcranial magnetic stimulation (TMS) and...
recording the compound muscle action potentials (CMAPS) at either half of the tongue. Two pairs of Ag/AgCl surface disc electrodes at an interelectrode distance of 18 mm were mounted on a spoon shaped metacrylate device adapted to the oral cavity. The electrodes were placed above the lateral dorsum of the tongue. Slight contraction of the tongue muscles was achieved by gently pressing the dorsum of the tongue against the mouthpiece.

The cortico-orofacial projections were investigated by activating the orofacial muscles using TMS and recording the CMAP of the buccinator muscles at either side of the face. We used pairs of Ag/AgCl surface disc electrodes embedded at a distance of 18 mm in a specially designed fork shaped metacrylate device which was adapted to the oral vestibulum. The electrodes were in contact with the insides of the cheeks. Slight contraction of the buccinator muscles was achieved by pursing the lips.

Filter settings for CMAP recordings were 20–2000 Hz. A Magstim 200S (Novametrix, Whitland, Dyfed, UK) and a circular coil (mean diameter 9 cm) with a peak magnetic field of 2.0 Tesla were used for TMS.

For cortical stimulation the centre of the coil was positioned tangentially, 4–6 cm (tongue) and 1–2 cm (buccinator muscle) lateral to the vertex, at the vertex (upper limbs) and 4 cm in front of the vertex (lower limbs). On stimulation of the left (right) hemisphere, side “A” (“B”) was viewed from above. Stimulation strength was increased stepwise during slight preinnervation until stable latencies were achieved. Out of four recorded responses the shortest onset latency (total conduction time, TCT) and largest amplitude (peak to peak) of the CMAP were measured.

A detailed description of lingual and facial recording techniques and normative data have been published elsewhere.7–10 Sensation in the oral cavity was tested with pinprick, touch, two point discrimination, and stereognosis (using stimuli of different shape (a cube, ball, or ring)) as suggested by Ringel and Ewanowski11 and Ringel et al.12 SSEPs were elicited at the median nerve using a standard technique outlined in the IFCN committee guidelines.13 SPECT imaging was performed on six patients of this series. After the patients had rested in a dark and silent room for a period of 20 minutes, 550 MBq 99mTc-hexamethylamineoxime (HMPAO) were administered intravenously. After another 10 minute period the patient was placed in the supine position with the head fixed in an adjustable head holder and the images were obtained. Special care was taken to avoid head tilting. A double head rotating gamma camera (Picker, Prism 2000) interfaced to a computer (Picker, Odyssey) with a 20% symmetric energy window centred on the 140 keV peak was used. A total of 120 20 s images were obtained over a 360 degree circular revolution (step and shoot paradigm), using a low energy, high resolution parallel hole collimator. The average radius of rotation was 17 cm. The resolution of the system was 12 mm and expressed as full width at half maximum at the centre of the field of view and at a depth of 15 cm from the camera face. A total of 2–4 million total counts were collected over a period of 25 minutes. The images were acquired in a 64×64 matrix. One pixel (5.8 mm) thick transverse oblique slices were reconstructed in parallel to the orbitomeatal line using a low pass filter (Butterworth). Attenuation correction was performed (Chang algorithm) with an attenuation coefficient of 0.13/cm. To compare the tracer uptake between both cerebellar hemispheres from each slice of the cerebellum, a region of interest (ROI) was drawn around the contour of one hemisphere. On mirroring the ROI onto the other hemisphere with the midline serving as the axis, count rates could be acquired within identical regions. To avoid partial volume effects, the upper and the lower slices of the cerebellum were not considered for analysis. ROIs were drawn for each slice at the transverse oblique level to allow better comparison with...
Figure 1  (A) MRI lacunar infarct in the right corona radiata (left side of the figure, patient 1). (B) MEP to the orofacial muscles, showing normal contralateral responses after bilateral cortical stimulation. (C) MEP to the tongue muscles, showing absent responses at both halves of the tongue after magnetic stimulation over the affected right hemisphere (right side of the figure).
Figure 2  (A) MRI lacunar infarct in the left corona radiata (right side of the figure, patient 3). (B) MEP to the orofacial muscles, showing normal contralateral responses after bilateral cortical stimulation. (C) MEP to the tongue muscles, showing delayed responses at both halves of the tongue (L>R) after magnetic stimulation over the affected left hemisphere (left side of the figure).
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MRI. The left/right ratio was calculated for the count rate of each slice and as a global count rate for the entire cerebellar hemispheres (adding the count rate of all slices). The global left/right cerebellar asymmetry in normal subjects (n=9) was established at a mean of 1.00 (SD 0.07) and a range from 0.82 to 1.18 (mean ± 2.5 SD).14 For ethical reasons we therefore abstained from establishing normal values in the present study.

Informed consent for this study was obtained from all participants and the study was approved by the local ethics committee (Landesärztekammer Rheinland-Pfalz).

Results

All patients presented with sudden onset of speech difficulties. Dysarthria was characterised by slurring with imprecise articulation and the patients reported a “thick” tongue. Articulatory movements and speech rate were mildly slowed, showing a mean syllable repetition rate of 4.7 syllables/s (normal rate; 6 syllables/s). Modulation of pitch and intensity were reduced. Scanning, explosive, or dysprosodic speech was absent. The degree of dysarthria was mild to moderate in most cases; no patient had unintelligible speech. The voice was breathy, sometimes pressed, and slightly hoarse. Laryngoscopy showed normal vocal fold motility.

The neurological examination showed no deficits unrelated to dysarthria: three patients (patients 1, 4, 6) showed slight tongue deviation and five patients (patients 1, 2, 4, 5, 7) demonstrated mild facial paresis of the central type. None of the patients showed palatal weakness. However, only one patient showed no clinical deficit beside dysarthria. Sensory function of the oral cavity was unimpaired. Median nerve SSEPs were normal in all patients. Dysarthria cleared within 2 weeks to 6 months (median: 1.2 months).

In all patients TMS of the lesion side (above the affected hemisphere) showed absent (patients 1, 2, 5, 6, 7) or delayed (patients 3, 4) corticolingual responses at both halves of the tongue (table 2). TMS evoked orofacial responses contralateral to the lesion side were absent in three patients (patients 2, 5, 7), delayed in one (patient 6) and present in three. TMS of the peripheral hypoglossal and facial nerve elicited biphasic CMAPs at PCT within the normal range in all patients showing normal peripheral impulse conduction. The corticospinal projections to the upper and lower limbs were normal in all patients.

HMPAO-SPECT (table 3) performed in six of seven patients showed no significant side differences in global tracer uptake between the cerebellar hemispheres compared with control data.14 For the left/right ratio of single slices, there was no focal reduction in tracer uptake within the cerebellar hemisphere.

Brain MRI showed single lesions in the corona radiata (n=4) and genu and posterior limb of the internal capsule (n=2) without other lacunar infarctions (table 1). In patient 5 no morphological lesion was identified.

No differences in dysarthria were noted with regard to the location and size of the lesion or the side of the affected hemisphere. We found no correlation between the neurophysiological abnormalities (absence of delay of responses, isolated corticocolingual or cortico-orofacial involvement) and the severity or auditory presentation of dysarthria.

Discussion

Dysarthria in association with facial and lingual hemiparesis due to lesions established at necropsy in the genu of the internal capsule were described as early as about 100 years ago (for a historical review see Bogousslavsky and Regli®). However, due to the close proximity and varying location of fibre tracts no definite conclusions concerning the nature of dysarthria could be achieved.

The pathophysiology of dysarthria may be derived from focal brain lesions, especially in lacunar stroke, compared with large, widespread, or multifocal disorders such as brain tumour (rarely limited to one anatomical structure; oedema, and microscopic tumour spread make an anatomic-functional correlation more difficult than for discrete infarctions), haemorrhage (leads to mass effects and dissection of blood in neighbouring structures), and degenerative pathological conditions—for example, Parkinson’s and Huntington’s disease. The last disorders may also be associated with dysarthria although the fact that several central nervous system structures are involved makes it difficult here to identify the fibre tracts attributable to dysarthria. Unilateral lacunar infarcts thus represent a disorder allowing the correlation of clinical signs and symptoms with anatomical structures and functional testing. We therefore investigated seven patients with isolated dysarthria due to lacunar stroke over a three year period.

Impaired articulation is one of the most prominent features of dysarthria. As the tongue and orofacial muscles are the most important articulators19 20 we investigated the corticolingual and cortico-orofacial pathways using TMS. Corticocolingual fibres project bilaterally from either hemisphere to the hypoglossal nuclei whereas cortico-orofacial fibres project predominantly to the contralateral subnuclei.19 20 The degree of limb muscle paresis correlates with an increase in latency and decrease in amplitude of the TMS muscle response in ischaemic cerebral lesions.22 23 These parameters reflect the degree of functional impairment of the fast conducting large diameter pyramidal fibres.24 The absence of a response is associated with a more severe lesion than a delayed response. Because amplitudes of the TMS evoked potentials show a wide inter-individual variation,25 26 which also applies to tongue and orofacial muscle responses, only absent or delayed (>mean±2.5 SD) responses were considered as abnormal. Control values were obtained from 43 healthy subjects.

The characteristics of dysarthria in our patients were almost identical. Dysarthria was mild to moderate. No differences in dysarthria...
were noted with regard to the location and size of the lesion and the side of the affected hemisphere. The most common features were imprecise articulation, a mildly slowed speech rate, and a slightly monotonous voice. These auditory findings are similar to those reported previously for central motor impairment due to hemispheric infarction. The uniformity of speech abnormality found in the described locations is consistent with a common pathophysiological basis.

The common abnormality in all patients with dysarthria was involvement of the corticolingual projections as disclosed by TMS. However, only three (patients 1, 4, 6) out of seven patients had clinical signs of a tongue movement disorder (fig 1A–C). One patient (patient 3) showed dysarthria without any further clinical deficits. In this patient, TMS of the left motor cortex showed delayed responses at both halves of the tongue due to lacunar infarction in the left corona radiata (fig 2A–C). In four patients with additional cortico-orofacial tract involvement speech performance was not more significantly or differently disturbed, suggesting that impairment of the corticoro-facial tract does not necessarily contribute to the development of dysarthria in lacunar stroke. This assumption was recently confirmed in a patient with an isolated corticoro-facial tract lesion, who did not show dysarthria.

Normal TMS results of the peripheral hypoglossal nerve and the absence of clinical signs at peripheral facial and hypoglossal nerve lesions in our patients indicate that the observed conduction abnormalities after cortical stimulation must be attributed to the central lesions shown by MRI. The lesions were located within the pyramidal tract, between the lower motor cortex and the genu and the posterior limb of the internal capsule. The left hemisphere (six patients) was more often affected than the right hemisphere (one patient). This confirms previous findings, that dysarthria is not restricted to left sided lesions only, but to a lesser degree also occurs in right sided lesions. In one patient (patient 5) TMS showed corticofacial and corticolingual tract involvement although MRI did not identify a morphological lesion. However, negative MRI findings of lacunar infarction are not uncommon, reflecting the still limited ability of MRI in detecting small lacunar lesions.

The detection of a larger number of subclinical corticollingual tract lesions than of subclinical cortico-orofacial tract lesions by TMS may be due to the bilateral symmetric tongue innervation which clinically masked the sequel of a lesion in one hemisphere, whereas the predominantly unilateral (contralateral) innervation of the lower facial muscles was more often associated with clinically apparent paresis. The not infrequently found coincidence of corticollingual and corticofacial tract involvement likely results from the close proximity of both fibre tracts along their entire course from motor cortex to brainstem. Most ischaemic lesions therefore affect not only the corticollingual pathway. Both factors may explain the clinical finding that dysarthria due to the unilateral vascular lesion of one cerebral hemisphere is most often associated with the clinical finding of facial, but not of unilateral tongue weakness.

Sensory deficits are not the only factors related to dysarthria, which is confirmed by the fact that the introraoral sensory functions and median nerve SSEPs were undisturbed in all our patients. Because dysarthria has been attributed to corticopontocerebellar tract involvement, we performed HMPAO-SPECT, which is associated with a reduced tracer uptake of one cerebellar hemisphere (cerebellar diaschisis) in the presence of a cortical system responsible for the entire motor innervation to the tongue muscles, whose precise and highly coordinate interactions are required for the production of different sounds. We conclude that impairment of the central lingual motor subsystem is a major factor accountable for imprecise articulation in dysarthric patients. Due to the somatotopically arranged pyramidal tract fibres, partial motor deficits may occur with lacunar lesions in the corona radiata and internal capsule leading to a central monoparesis of the face, upper limb, and lower limb. We have now six central monoparesis of the tongue may also occur, clinically presenting as isolated dysarthria.

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