

Articles

Post-Stroke Lingual Dystonia: Clinical Description and Neuroimaging Findings

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Abstract

Background: Lingual dystonia is extremely rare following stroke. We describe clinical features and neuroimaging findings in a series of 11 patients (seven acute and four chronic) with post-stroke lingual dystonia and review the literature.

Methods: This was a case series using a preformed structured proforma and review of literature using a PubMed search.

Results: In our case series, all patients had dysarthria as a presenting symptom. Seven patients had acute presentation (six had an ischemic infarct and one had thalamic hemorrhage) and four had chronic presentation (all had infarct). All patients except one had small infarcts, with the majority of them in the basal ganglia and subcortical white matter regions. Additional chronic ischemic lesions were seen in all patients with acute presentation. The majority of the patients with acute (five out of seven; 71.42%) presentation had left-sided involvement on imaging. We could identify only one case of acute post-stroke lingual dystonia following the PubMed search. Three other cases of post-stroke lingual dystonia with chronic presentation have been described; however, these were associated with oromandibular or cranial dystonia.

Discussion: Our results, based on brain lesions, suggest that all lingual dystonia patients with acute infarcts had underlying chronic infarcts. Overall, more left-sided than right-sided strokes were observed with post-stroke lingual movement disorders including dystonia; however, the data were not significant (p=1). All patients had dysarthria, with only one having mild tongue weakness and only four having facial weakness. This suggests that the lingual dystonia was responsible for the dysarthria rather than weakness in these patients.

Keywords: Dystonia, magnetic resonance images, stroke

Citation: Pandey S, Tater P. Post-stroke lingual dystonia: clinical description and neuroimaging findings. Tremor Other Hyperkinet Mov. 2018; 8. doi: 10.7916/ D8RB8NJC

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Editor: Elan D. Louis, Yale University, USA

Received: October 14, 2018 Accepted: November 27, 2018 Published: January 8, 2019

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Funding: None.

Financial Disclosures: None.

Conflicts of Interest: The authors report no conflict of interest.

Ethics Statement: This study was performed in accordance with the ethical standards detailed in the Declaration of Helsinki. The authors' institutional ethics committee has approved this study and all patients have provided written informed consent.

Introduction

Primary lingual dystonia is rare and mostly occurs in association with oromandibular dystonia secondary to varied causes.¹ Tardive dystonia is the most common cause; others include neurodegenerative disorders such as pantothenate kinase-associated neurodegeneration, neuroacanthocytosis, Wilson's disease, neurodevelopmental disorders like Rett's syndrome, metabolic disorders like Lesch–Nyhan syndrome, post-varicella infection, polycythemia rubra vera, brain damage secondary to anoxia, post-traumatic, non-DYT1 primary generalized dystonia, primary lingual dystonia induced on speaking, paroxysmal episodic focal dystonic spasms, and rarely stroke.¹ In the series published by Esper et al.² of 17

patients with lingual protrusion dystonia, 41% were secondary to medications, 18% heredodegenerative and post-encephalitic, 12% generalized dystonia, and 29% focal primary lingual dystonia, but none was secondary to stroke. Abnormal involuntary movement disorders develop in 1–4% of stroke victims.³ Some of these disorders occur immediately after acute stroke, some can develop later, and others represent delayed-onset progressive movement disorders.⁴ They are reported most commonly secondary to basal ganglia and thalamic strokes.⁵

To the best of our knowledge, only one case of lingual dystonia following acute stroke has been reported.⁶ Two other chronic cases of post-stroke oromandibular dystonia with lingual dystonia have been

reported.^{7,8} Another case of post-stroke cranial lingual dystonia was reported; however, the onset was not described in the paper.⁹ Here, we describe a case series of 11 patients who developed lingual dystonia following stroke. We have also searched the available literature for studies describing involuntary tongue movements following stroke and their anatomical localization, with the main focus on lingual dystonia.

Methods

We evaluated 11 patients with post-stroke lingual dystonia who attended our movement disorder clinic at a tertiary care center as a part of a research project "A clinical study of post-stroke movement disorders". The study was approved by our institutional ethics committee (IEC) (IEC number: F.11/IEC/MAMC/10/04). Written informed consent was obtained from all participating individuals according to the IEC guidelines. Detailed history and clinical examination of the patients were performed using a preformed structured proforma. Information regarding demographic details of the patients, time of symptom onset, time to onset of lingual dystonia, stroke risk factors, history of recurrent stroke, recovery status post stroke, history

of drug intake prior to onset of lingual dystonia, description of tongue movement by the patient, use of sensory trick, whether induced on speaking, tongue protrusion or retraction dystonia and functions affected because of this, such as eating, drinking, swallowing, breathing, speech, singing, and any salivary drooling or history of tongue bite were noted. During clinical examination, the tongue movements of each patient were videotaped for 3-5 minutes. All patients signed informed consent to be videotaped and for the publication of their videotapes. None of the patients in our study had a history of exposure to neuroleptic agents including antiepileptic drugs. All patients other than the one (case 7) showing hemorrhage on non-contrast computed tomography (CT) of the head underwent magnetic resonance imaging (MRI) (Figure 1). We assessed the topography of the lesions according to the anatomical structures involved (cortical, subcortical white matter, basal ganglia, thalamus, subthalamus, brainstem, and cerebellum) for uniformity. All patients received treatment for stroke according to the standard guidelines. For lingual dystonia, oral pharmacological drugs (trihexphenidyl, clonazepam, tetrabenazine in isolation or combination) were used

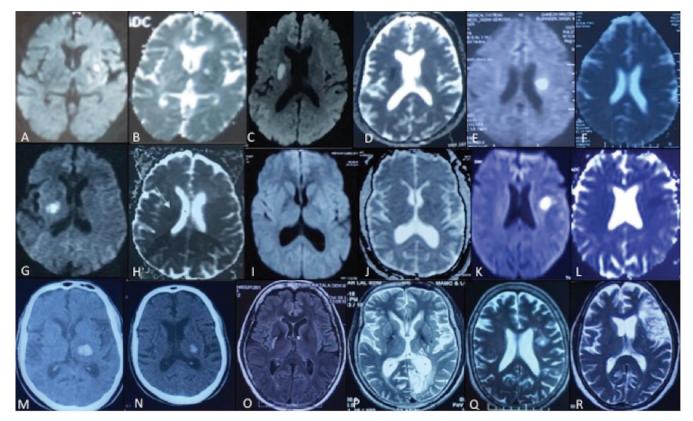


Figure 1. Diffusion-weighted and Apparent Diffusion Coefficient Sequences of Magnetic Resonance Images of Brain. (A,B) Case 1: acute infarct in left basal ganglia. (C,D) Case 2: acute infarct in right basal ganglia, right corona radiata, and subcortical white matter. (E,F) Case 3: acute infarct in left basal ganglia, left corona radiata and subcortical white matter. (K,L) Case 4: acute infarct in the right basal ganglia and adjacent white matter. (I,J) Case 5: acute infarct in the left basal ganglia and adjacent white matter. (K,L) Case 6: acute infarct in the left corona radiata. (M,N) Case 7: left thalamic hemorrhage. Non-contrast computed tomography of head. (O) Case 8: chronic lacunar infarcts in the right basal ganglia and right parietal region. Axial T2 fluid-attenuated inversion recovery image. (P–R) Axial T2-weighted sequences of magnetic resonance images of brain. (P) Case 9: chronic infarcts in left posterior cerebral artery region, left thalamus and lacunar infarcts in bilateral basal ganglia). (Q) Case 10: chronic infarcts in left frontal subcortical region. (R) Case 11: chronic infarct of left middle cerebral artery region.

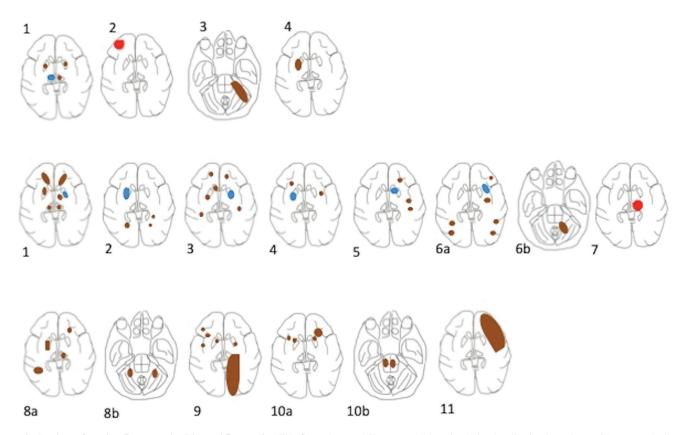


Figure 2. Lesions Causing Post-stroke Lingual Dystonia. This figure (top row) illustrates the imaging lesion localization in patients with post-stroke lingual dystonia described in the literature. Case 1 depicts acute post-stroke lingual dystonia, cases 2 and 3 are patients with chronic post-stroke oromandibular (lingual) dystonia, and case 4 is a patient with chronic post-stroke cranial lingual dystonia. The bottom two rows illustrate the imaging lesion localization in our patients with post-stroke abnormal lingual movements. Cases 1–7 are acute post-stroke lingual dystonia patients whereas cases 8–11 are chronic post-stroke lingual dystonia patients. Red lesions represent hemorrhagic stroke and blue lesions represent acute ischemic stroke whereas brown represents chronic ischemic stroke.

for all the patients and three patients (one acute and two chronic) also received injection of onabotulinum toxin A. Response to therapy was based on the questionnaire and physician interview and categorized as excellent, moderate, mild, or none at 3 months' follow-up.

We also selected cases of post-stroke lingual movement disorders reported in the literature (PubMed search) using search terms "(Stroke) and (lingual movement disorders, lingual dystonia, lingual tremor, lingual chorea, and lingual myoclonus)" in English in June 2018. We generated 31 articles using "Stroke and lingual movement disorders", seven articles using "Stroke and lingual dystonia", seven articles using "Stroke and lingual tremor", two articles using "Stroke and lingual chorea", and five articles using "Stroke and lingual myoclonus". After excluding duplications, 20 cases (one had acute lingual dystonia) of post-stroke lingual movement disorders were identified in the literature. Lesion locations, as indicated in the original publications of post-stroke lingual dystonia, were traced; a representative slice of each lesion is illustrated in Figure 2 (Top row). A similar representative slice of each lesion for our 11 cases is also illustrated in Figure 2 (Bottom two panels) for comparison.

Results

Case description

Case 1. A 70-year-old female presented to us with a 1-day history of sudden onset dysarthria, irrelevant talking, and impaired comprehension. She had a past history of recurrent stroke in the previous 2 years in the form of left hemiparesis with slurring of speech. On clinical examination, she had lingual dystonia, and on protrusion, tongue tremor was also seen. She was also examined during speaking, eating, drinking, swallowing, and breathing. Her speech was dysarthric, and she had difficulty in eating. MRI of the brain revealed acute infarct in the left basal ganglia region with bilateral chronic infarcts (Figure 1A,B). She was treated with tablets of trihexphenidyl (2 mg twice daily for 15 days), tetrabenazine (25 mg twice daily for 1 month), and clonazepam (0.25 mg once daily for 1 month). At 3 months' follow-up, she had a moderate improvement in lingual dystonia (Video 1, case 1).

Case 2. A 54-year-old male presented to us with a 7-day history of sudden-onset slurring of speech while driving his vehicle. On examination, he had lingual dystonia at rest. He was also examined



Video 1. Post-stroke Lingual Dystonia. Lingual dystonia cases 1-11.

during speaking, eating, drinking, swallowing, breathing, and singing. His speech was dysarthric, and he had difficulty in singing. MRI of the brain revealed acute infarcts involving right basal ganglia, right corona radiate, and subcortical white matter changes in the frontoparietal and periventricular regions (Figure 1C,D). He was treated with tablets of trihexphenidyl (2 mg three times daily for 1 month), and clonazepam (0.25 mg once daily for 2 months). At 3 months' follow-up there was mild improvement in his symptoms (Video 1, case 2).

Case 3. A 40-year-old male presented to us with a 5-day history of sudden-onset slurring of speech, twisting of tongue, and facial deviation. On examination, he had severe lingual dystonia at rest and mild dystonia on protrusion. He was also examined during speaking, eating, drinking, swallowing, breathing, and singing. His speech was dysarthric, and he had difficulty in eating and singing. MRI of the brain showed acute infarct in the left basal ganglia, left corona radiate, and bilateral subcortical white matter with bilateral chronic lacunar infarcts (Figure 1E,F). He was treated with trihexphenidyl tablets (2 mg three times daily for 2 months), clonazepam (0.25 mg once daily for 2 months), and tetrabenazine (12.5 mg thrice daily for 1 month). At the 3-month follow-up there was an excellent response in his lingual dystonia (Video 1, case 3).

Case 4. A 59-year-old male presented to us with sudden-onset slurring of speech, and left-sided brachiofacial weakness for 7 days. On examination, he had lingual dystonia at rest. He was also examined during speaking, eating, drinking, swallowing, and breathing. His speech was dysarthric, and he had difficulty in eating. MRI of the brain revealed acute infarct involving the right basal ganglia and periventricular white matter changes (Figure 1G, H). He was treated with trihexphenidyl tablets (2 mg twice daily for 1 month) and tetrabenazine (25 mg thrice daily for 1 month) and there was a moderate response in tongue dystonia at the 3-month follow up (Video 1, case 4).

Case 5. A 41-year-old male presented with sudden-onset difficulty in speaking for 2 days. On examination, there was severe lingual dystonia at rest and mild dystonia on protrusion. He was also examined during speaking, eating, drinking, swallowing, breathing, and singing. His speech was dysarthric, and he had difficulty in eating and singing. MRI of the brain showed acute infarct in the left basal ganglia region

and chronic infarcts in the left frontoparietal cortical-subcortical regions (Figure 11,J). He was treated with trihexphenydyl tablets (2 mg twice daily for 15 days) and clonazepam (0.25 mg once daily for 1 month) and one session of injection of onabotulinum toxin A (20 U). He had a moderate response in lingual dystonia at the 3-month follow-up (Video 1, case 5).

Case 6. A 45-year-old female presented to us with a history of suddenonset dysarthria, and equal dysphagia to solids and liquids for 4 days. Four months previously she had sudden-onset left hemiparesis, which had recovered partially. On clinical examination, the tongue showed dystonic posturing on rest and protrusion. She was also examined during speaking, eating, drinking, swallowing, and breathing. Her speech was dysarthric, and she had difficulty in eating, drinking, and swallowing. MRI of the brain revealed acute infarct in the left corona radiata region with bilateral chronic lacunar infarcts (Figure 1K,L). She was treated with trihexphenidyl (2 mg twice daily for 2 months), clonazepam (0.25 mg once daily for 1 month), and tetrabenazine (25 mg thrice daily for 1 month). At the 3-month follow-up there was an excellent response in her lingual dystonia (Video 1, case 6).

Case 7. A 55-year-old male presented with a history of sudden-onset slurring of speech and right-sided hemiplegia with altered behavior for 3 days. On clinical examination, he had severe lingual dystonia at rest, which was mild on protrusion. He was also examined during speaking, eating, drinking, and swallowing. His speech was dysarthric, and he had difficulty in eating. His CT scan of the brain revealed left thalamic hemorrhage (Figure 1M,N). He was treated with tetrabenazine tablets (25 mg three times daily for 2 weeks). After 2 weeks, he was discharged from hospital in a stable condition, but died at his home following a myocardial infarction (Video 1, case 7).

Case 8. A 62-year-old female presented to us with a history of left hemiparesis and slurring of speech 2 years ago. On clinical examination, she had severe tongue dystonia at rest and mild dystonia on protrusion. She was also examined during speaking, eating, drinking, swallowing, breathing, and singing. Her speech was dysarthric, and she had difficulty in eating and singing. Her brain MRI showed chronic infarcts in the bilateral cerebellum, left thalamus, right basal ganglia, right posterior frontal and parietal areas, and left corona radiata regions (Figure 1O). She was treated with trihexphenidyl tablets (2 mg twice daily for 1 month), clonazepam (0.25 mg once daily for 1 month), and tetrabenazine (25 mg thrice daily for 2 months) and injection onabotulinum toxin A (20 U). At the 3-month follow-up, she had a moderate response in in her symptoms (Video 1, case 8).

Case 9. A 62-year-old male presented to us with a 5-month history of right hemiparesis and dysarthria. Tongue examination showed dystonia at rest and on protrusion with tremors. He was also examined during speaking, eating, drinking, swallowing, and breathing. His speech was dysarthric, and he had difficulty in eating. MRI of the brain showed chronic infarcts in bilateral basal ganglia and predominantly right temporoparietal regions with white matter hyperintensities in bilateral

periventricular areas (Figure 1P). He was treated with trihexphenidyl tablets (2 mg twice daily for 1 month), clonazepam (0.25 mg once daily for 2 months), tetrabenazine (25 mg twice daily for 15 days), and levodopa+carbidopa (125 mg thrice daily for 3 months). At the 3-month follow-up, he had a mild response in the symptoms of lingual dystonia (Video 1, case 9).

Case 10. A 59-year-old female on treatment for type 2 diabetes mellitus presented with a history of sudden-onset dysarthria and right-sided brachiofacial weakness for 18 months. On examination, she had sustained dystonic posturing of the tongue at rest with tremor. She was also examined during speaking, eating, drinking, swallowing, breathing, and singing. Her speech was dysarthric, and she had difficulty in eating and singing. MRI of the brain revealed chronic infarcts in the left frontoparietal, bilateral pons, basal ganglia and right frontal subcortical regions (Figure 1Q). She was treated with trihexphenidyl tablets (2 mg twice daily for 2 months), tetrabenazine (25 mg twice daily for 1 month), and injection of onabotulinum toxin A (20 U). At the 3-month follow-up, she had mild improvement in symptoms of tongue dystonia (Video 1, case 10).

Case 11. A 65-year-old female presented with a 4-month history of right-sided hemiparesis and slurring of speech. Her weakness improved gradually over a month, but dysarthria persisted. On examination, she had intermittent dystonic posturing of the tongue at rest and protrusion. She was also examined during speaking, eating, drinking, swallowing, and breathing. Her speech was dysarthric, and she had difficulty in eating. Her brain MRI showed chronic infarct in the left middle cerebral artery territory with hemorrhagic transformation (Figure 1R). She was treated with trihexphenidyl tablets (2 mg twice daily for 3 months), and tetrabenazine (25 mg twice daily for 2 months) causing a mild improvement in her symptoms at the 3-month follow-up (Video 1, case 11).

Results

We evaluated 11 patients with lingual dystonia following stroke (Tables 1 and 2). Seven patients presented with acute stroke and four patients presented with chronic stroke. All patients had dysarthria as a presenting symptom and only one patient with acute stroke had dysphagia. The risk factors for stroke included diabetes mellitus, hypertension, smoking, alcohol, tobacco chewing, and heart diseases (rheumatic heart disease, dilated cardiomyopathy, and cardiac apical aneurysm).

Acute stroke patients (Table 3). Seven patients (five males, two females) presented with acute stroke and their age ranged from 40 to 70 years $(52 \pm 4.08 \text{ years mean} \pm \text{standard deviation [SD]})$. On neuroimaging all patients had an acute ischemic infarct except one, who had left thalamic hematoma. Previous history of stroke was present in two patients. The time to onset of stroke and presentation to the hospital ranged from 1 to 7 days (mean \pm SD, 4.14 ± 0.88 days). All seven patients had lingual dystonia at rest and five patients had dystonia on protrusion also. Tongue tremor was present in only

three patients. Focal neurological deficit was present in six patients, which included hemiparesis (n=3), brachiofacial weakness (n=2), upper limb dystonic tremor (n=2), laterocollis (n=2), upper limb weakness (1), and upper limb dystonia (n=1). Acute cerebrovascular lesions were seen in all seven patients, of whom two patients had basal ganglia involvement alone, one had involvement of subcortical white matter alone, three patients had involvement of both basal ganglia and subcortical white matter, and one had thalamic hematoma. All patients had left-sided lesions, except two who had right-sided involvement. All patients with acute ischemic lesions also had underlying chronic ischemic lesions (Table 4). At the 3-month follow-up, excellent response was seen in two patients, moderate response was seen in three patients, mild response was seen in one patient, and one patient with thalamic hematoma died.

Chronic presentation (Table 3). Four patients (one male and three females) presented with lingual dystonia following chronic ischemic infarcts and their mean age ranged from 59 to 65 years (mean \pm SD 61.75 ± 1.25 years). The time to onset of symptoms and presentation to the hospital ranged from four to 24 months (mean \pm SD: 12.75 \pm 4.92 months). All four patients had lingual dystonia at rest and three patients had dystonia on protrusion also. Tongue tremor was present in all patients. Focal neurological deficit was present in three patients, which included hemiparesis (n=2), brachiofacial weakness (n=1), mild tongue weakness (1), and upper limb dystonia (n=1). One patient with right-sided hemiparesis also had alexia, upper limb dystonic tremor, short shuffling gait, and postural instability. Chronic infarcts were seen in all patients, which involved cortical regions (n = 4), subcortical white matter (n=4), basal ganglia (n=3), thalamus (n=2), brainstem (1), and cerebellum (n=1). Three patients had bilateral involvement (predominantly left sided) and one patient had left-sided cortical and subcortical involvement. At the 3-month follow-up, a moderate response was seen in one patient and three patients had a mild response only.

Cases selected from literature search (Table 5). We identified 20 cases of post-stroke lingual movement disorders following a PubMed search using the search terms described in Methods.⁷⁻²⁰ Thirteen cases had acute presentation and seven had a chronic presentation. Rippling tongue movements were the most common acute movement disorders described in five cases, followed by myoclonus (n=3), dyskinesia (n=2), tremor (n=2), and dystonia (n=1). Chronic post-stroke lingual movement disorders included oromandibular dystonia, including lingual dystonia (n=2), dyskinesia (n=2), cranial lingual dystonia (n=1), chorea (n=1), and rhythmic movement (n=1). Lingual dystonia following acute stroke (right thalamic infarction) was reported in only one case (Kim et al.⁶). This patient had multiple old ischemic lesions in the bilateral thalami also. Two other chronic cases of post-stroke (left anterior inferior cerebellar infarct and right basal ganglia infarct) oromandibular dystonia with lingual dystonia have been reported (Akin et al.,⁷ Brissaud et al.⁸). Another case of post-stroke (right frontal hematoma) cranial lingual dystonia was reported, but the onset was not described in the paper (Alarcón et al.⁹). The majority

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	Case Number	Age and Sex	Handedness	Risk Factors for Stroke	Days of Onset	Lingual Dystonia	Tongue Tremor	Neuroimaging (All MRI Except Case 7)	Outcome
	1.	70 years, Female	Right	None	1 day	Severe on rest and mild on protrusion	On protrusion	Acute infarct in the left basal ganglia region with bilateral chronic infarcts	Moderate
	બં	54 years, Male	Right	HTN, chronic smoker and alcohol intake	7 days	Severe on rest	Absent	Acute infarcts involving right basal ganglia, right corona radiata and subcortical white matter changes in fronto-parietal and periventricular regions	Mild
	сi	40 years, Male	Right	NTH	5 days	Severe on rest and mild on protrusion	Absent	Acute infarct in left basal ganglia region, left corona radiata and adjacent subcortical white matter with bilateral chronic lacunar infarcts	Excellent
	4.	59 years, Male	Right	HTN, DM	7 days	Only on rest	Absent	Acute infarct involving the right basal ganglia and periventricular white matter changes	Moderate
	Ċ	41 years, Male	Right	DM	2 days	Severe on rest and mild on protrusion	On protrusion	Acute infarct in the left basal ganglia region and chronic infarcts in left frontoparietal cortical-subcortical regions	Moderate
	6.	45 years, Female	Right	DM, HTN, Dilated cardiomyopathy	4 days	Both rest and protrusion	On protrusion	Acute infarct in left corona radiata regions with bilateral chronic lacunar infarcts	Excellent
	7.	55 years, Male	Right	Chronic bidi smoker	3 days	Severe on rest and mild on protrusion	Absent	CT scan of brain revealed left thalamic hemorrhage	Patient died
	α	62 years, Female	Right	Rheumatic heart disease	24 months	Severe on rest and mild on protrusion	Tremor on protrusion	Chronic infarcts in bilateral cerebellum, left thalamus, right basal ganglia, right posterior frontal and parietal areas, and left corona radiata regions	Moderate
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Case Number	Age and Sex	Handedness	Risk Factors for Stroke	Days of Onset	Lingual Dystonia	Tongue Tremor	Neuroimaging (All MRI Except Case 7)	Outcome
6	61 years, Male	Right	Alcohol	5 months	Both on rest and protrusion	Tremor on rest and protrusion	Chronic infarcts in bilateral basal ganglia and predominantly right temporo-parietal regions with white matter hyperintensities in bilateral periventricular areas	Mild
10.	59 years, Female	Right	DM	18 months	Only on rest	Tremor on rest and protrusion	Chronic infarcts in left fronto- parietal, bilateral pons, basal ganglia and right frontal subcortical regions	Mild
11.	65 years, Female	Right	HTN,Cardiac apical aneurysm	4 months	Severe on rest and mild on protrusion	Tremor on protrusion	Chronic infarct in left middle cerebral artery territory with haemorrhagic transformation	Mild

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(eight out of 12) of the patients with acute post-stroke lingual movement disorder other than dystonia had left-sided involvement, two had right-sided involvement, and another two had bilateral involvement. Cortical involvement was seen in the majority (10/12) of the patients, and two had thalamic involvement. Patients with chronic post-stroke lingual movement disorder other than dystonia (n=4) had brainstem (n=2), basal ganglia (n=1), and subcortical involvement (n=1). Tongue tremor was present in two patients with acute left frontoparietal infarct.

Discussion

In our case series, 11 patients presented with post-stroke lingual dystonia; seven had acute presentation and four had a chronic presentation. Acute cerebrovascular lesions were seen in seven patients out of which two had basal ganglia involvement and three had both basal ganglia and subcortical involvement. Further, one patient had an acute lesion involving the subcortical white matter and one had thalamic involvement. Usually secondary dystonia develops weeks to months after the acute stroke, reflecting the excessive neuronal reorganization during recovery from motor weakness or sensory loss. Kim et al.⁶ reported a case of lingual dystonia secondary to acute infarct in the posterolateral thalamus, which occurred at onset of symptoms. However, there were multiple old ischemic lesions in the bilateral thalami. In our cases all patients with acute ischemic lesions had chronic subcortical infarcts (Tables 1 and 4). In patients with chronic presentation, there were lesions in the subcortical white matter and basal ganglia in the majority of the patients (three out of four, 75%). The above facts suggest that pre-existing ischemic lesions, which could be asymptomatic, likely contribute to the generation of lingual dystonia. Kim et al.⁶ hypothesized that an acute infarction in the right thalamus might have generated lingual dystonia in concert with underlying ischemic lesions in the contralateral thalamus. He also proposed that disinhibition of the thalamo-cortical pathways results in dystonia.⁴ Alarcón et al.⁹ reported a patient with cranial lingual dystonia following right frontal hematoma, but the onset of symptoms and details of dystonia were not available in the paper. Two chronic cases of oromandibular dystonia with lingual dystonia have also been reported.^{7,8} None of our lingual dystonia patients had associated cranial or oromandibular dystonia.

Another interesting observation in our case series was left-sided lesions in the majority (five out of seven) of the patients with acute presentation. Three patients with chronic presentation also had bilateral involvement with predominant left-sided involvement and one patient had only left-sided lesions. The majority (eight out of 12) of the patients identified through literature searches with acute post-stroke lingual movement disorder had left-sided involvement. There have been reports in the past stating the dominance of the cerebral hemispheres in the control of tongue movements. Watanabe et al.²¹ published a study in 2004, which used functional MRI techniques to study the role of the parietal cortex in spatial tongue movements and found that there was greater activation of the left inferior parietal lobule when tongue movements were performed. Funk et al.²² reported

Case Number	Age and Sex	Neurological Examination Findings
1.	70 years, Female	Left hemiparesis (MRC power grade 4-/5)
2.	54 years, Male	Normal motor power, left upper limb dystonic tremors, and right laterocollis
3.	40 years, Male	Right sided brachiofacial weakness (MRC power grade 4/5), right upper limb dystonia, laterocollis
4.	59 years, Male	Left sided brachiofacial weakness (MRC power grade 4/5)
5.	41 years, Male	Right upper limb weakness (MRC power grade 4+/5)
6.	45 years, Female	Left hemiparesis and dysphagia (MRC power grade 4-/5), left upper limb dystonic tremors
7.	55 years, Male	Right sided hemiplegia (MRC power grade 2/5)
8.	62 years, Female	Normal
9.	61 years, Male	Alexia, residual right hemiparesis (MRC power grade $4+/5$), brisk reflexes, right upper limb dystonia and dystonic tremors, short shuffling gait and postural instability
10.	59 years, Female	Right sided brachiofacial weakness (MRC power grade 4/5)
11.	65 years, Female	Right sided hemiparesis (MRC power grade 4-/5) including mild weakness of face and tongue
Abbreviation	: MRC: Medical resear	ch council.

Table 2. Neurological Examination in Post-stroke Lingual Dystonia Patients

Table 3. A Comparison of Demographic and Clinical Details of Acute and Chronic Post-stroke Lingual Dystonia Patients

	Acute Presentation	Chronic Presentation
Number of patients	7	4
Male: Female	5:2	1:3
Age	40–70 (Mean \pm SD: 52 $\pm4.08)$ years	59–65 (Mean \pm SD: 61.75 ±1.25) years
Duration of illness	1–7 (Mean \pm SD: 4.14 \pm .88) days	4–24 (Mean \pm SD: 12.75 \pm 4.92) months
Tongue dystonia at rest	7/7 (100%)	4/4 (100%)
Tongue dystonia at protrusion	5/7 (71.42%)	3/4 (75%)
Tongue tremor	3/7 (42.85%)	4/4 (100%)
Type of stroke		
Ischemic	6	4
Hemorrhagic	1	0
Neuroimaging findings	Acute lesion: 5 had left sided and 2 had right sided involvement	3 had bilateral and 1 had left side involvement
Outcome		
Excellent	2	0
Moderate	3	1
Mild	1	3
Died	1	0

Case	Imaging	Haemorrhage/			Subcortical White	ıl White	(
		Infarct	Cortical	cal	Matter Including Corona Radiata	cluding adiata	Basal Ganglia	l ia	Thalamus	SUI	Subthalamus		Brainstem		Cerebellum	lum
			Г	_ ≃	Γ	R	Г	2	Г	≃	L R		L I	 ≃	Г	2
	MRI	Acute and chronic infarct	Ch	Ch	Ch	Ch	AcCh	Ch	Ch	Ch	1		1		I	
5	MRI	Acute and chronic infarct	T		Ch	Ac, Ch		Ac	I		T		I		I	
3	MRI	Acute and chronic infarct	ſ		Ac, Ch	Ch	Ac	Ch	I		I	T			I	
4	MRI	Acute and chronic infarct	I		Ch	Ac, Ch		Ac	I		1				I	
21	MRI	Acute and chronic infarct	Ch	I	Ch		Ac		I		I		I		I	
9	MRI	Acute and chronic infarct	Ch	Ch	Ac, Ch	Ch	I		I		1		1		Ch	
7	NCCT	Haemorrhage	I		I		I		Ac		I		I		I	
œ	MRI	Chronic infarct		Ch	Ch	Ch		Ch	Ch		I		I		Ch	Ch
6	MRI	Chronic infarct	Ch	Ch	Ch	Ch	Ch	Ch	Ch		I		I		I	
10	MRI	Chronic infarct	Ch		Ch	Ch	Ch	Ch	I		I	0	Ch C	Ch		
11	MRI	Chronic infarct	Ch		Ch		I		I		I	,	I		I	
Abbrevia	tions: Ac: acute,	Abbreviations: Ac: acute, Ch: Chronic, MRI: magnetic resonance imaging, NCCT: non contrast computed tomography.	nagnetic	resonan	ce imaging, l	VCCT: non (contrast co	mputed	tomograp	hy.						

Table 4. Neuroimaging Findings in Acute and Chronic Patients with Post-stroke Lingual Dystonia

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Author/Year	Serial Number of Cases	Number of Cases	Age/Sex	Type of Movement	Lesion Site/Type	Acute/ Chronic
Dewey 1989	_	One	63 years, male	Oro-mandibular lingual dyskinesia,	Left Thalamic haemorrhage	Acute
O Combarros 1990	С	One	68 years, female	Oral dyskinesia (lingual dyskinesia)	Bilateral thalamo-capsular infarction	Acute
Jagota 2010	ŝ	One	26 years, female	Lingual myoclonus	Bilateral cortical frontotemporal and insular region ischemic infarct	Acute
Salazar 2012	4	One	35 years, male	Palatal and lingual tremor	Left frontoparietal region (left Middle cerebral artery ischemic infarct)	Acute
Pandey 2015	5	One	42 years, male	Tongue tremor	Left frontoparietal infarct	Acute
Saito 2016	6-10	Five	75–84 years 4 females, one male	Rippling tongue movements	4 with left primary motor cortex ischemic infarct and 1 with right primary motor cortex ischemic infarct	Acute
Rao 2018	11–12	Two	75 years, one male and one female	Lingual myoclonus	Left frontal opercular ischemic infarct, Right high frontal region ischemic infarct	Acute
Reilly 1992	13	One	50 years, female	Lingual chorea	Bilateral paramedian thalamic infarct (left>right)	Chronic
Fabiano 2000	14	One	64 years, female	Rhythmic tongue movements	Ischemic infarct in centrum semiovales and cerebral peduncles	Chronic
Lee 2005	15	One	82 years, female	Lingual dyskinesia	Right ventrolateral portion of mid and superior Pontine infarct	Chronic
Duffey 2007	16	One	64 years, male	Palatal tremor, lingual and jaw dyskinesia	Ventral pons and bilateral cerebellar infarcts	Chronic
Kim 2009	17	One	70 years, male	Lingual dystonia	Right thalamic infarction	Acute
Alarcon 2004	18	One	36 years, male	Cranial lingual dystonia	Right frontal haematoma	Chronic
Akin 2014	19	One	64 years, female	Oro-mandibular dystonia (lingual dystonia)	Left Anterior Inferior Cerebellar infarct	Chronic
Brissaud 2016	20	One	17 months old, female	Oro-mandibular dystonia (lingual dystonia)	Right basal ganglia infarct	Chronic

dominance of the left hemisphere for horizontal tongue movements in right-handed individuals.²² There was a study by Urban et al.²³ of patients with stroke who had dysarthria; this study evaluated the function of corticolingual and cortico-orofacial fibers using transcranial magnetic stimulation. Out of 18 patients they studied, 12 had left-sided lacunar infarcts. In another study of 255 pure motor stroke patients, it was observed that 65% had left-sided lesions with 47% having lacunar infarcts.²⁴ Seventy-four patients had dysarthria, which was secondary to lacunar stroke in maximum patients, and most had facial weakness as well. In our study all patients had dysarthria, with only one having mild tongue weakness and only four patients having facial weakness. This suggests that the lingual dystonia was likely responsible for the speech abnormality rather than weakness in these patients.

Urban et al.²⁵ reported a series of seven patients of isolated dysarthria secondary to lacunar infarct (corona radiata n=4 and internal capsule n=2), where patients had involvement of corticolingual fibers more than cortico-orofacial fibers. However, none of these patients had involvement of corticopontocerebellar fibers. Three of the seven patients had a slight tongue weakness. All but one patient in their series also had left-sided lesions. This study also hypothesized that dysarthria was seen secondary to focal discrete lesions involving corticolingual fibers rather than large lesions and could occur regardless of involvement of corticopontocerebellar involvement. In our study all except one patient (case 4, chronic infarct) had small infarcts (<15 mm) instead of large cortical lesions, thus further emphasizing the point that discrete fibers control individual muscles and finer tongue movements, and produce lingual dystonia at rest in all and on protrusion in some.

Tongue tremor on protrusion was present in all patients (four out of four, 100%) with chronic presentation; however, it was only present in three patients with acute stroke (three out of seven, 42.85%). The majority of the patients had subcortical lesions, except two chronic patients (cases 3 and 4) who also had additional left cortical lesions. Only two cases (Table 5) of tongue tremor have been reported previously and both had acute left-sided cortical infarct (Salazar and Miller,¹⁹ Pandey et al.¹⁶). Tremor rapidly improved in these cases and it was hypothesized that the cortical infarction temporarily produced dysfunction of its subcortical connections within the dentato-rubro-olivary pathway.

There are some inherent limitations in our study. First, the majority of our patients had ischemic infarct, which may be due to referral bias: being a tertiary care center, we may see more ischemic infarct patients than hemorrhagic stroke patients. Second, examining and categorizing tongue movements can be sometimes difficult, and subtle movements can be missed if not looked at carefully. Also, there is no clear-cut definition or diagnostic criteria presently for lingual dystonia causing diagnostic uncertainty in some cases. Third, the ability to hypothesize regarding localization appears to be limited by the heterogeneity of infarct location. Fourth, the treatment given to the patients was not uniform; therefore, the outcome may have differed.

Despite these limitations, we found a good number of patients with acute post-stroke lingual dystonia, including one patient who had acute thalamic hematoma. This raises the question of whether this phenomenon is more common than previously thought. In our case series, all lingual dystonia patients with acute infarcts had underlying chronic infarcts. The, majority of the patients had basal ganglia and subcortical white matter involvement, which may play an important role in the generation of lingual dystonia. Overall more left-sided than right-sided strokes were observed with post-stroke lingual movement disorders including dystonia; however, the data were not significant (p=1, Fisher's exact test). The presence of small infarcts (<15 mm) in our patients also highlights that discrete lesions may be required to bring out these manifestations, which strategically involve the individual fibers controlling the various tongue movements. This hypothesis should be tested using lesional studies in animal models. In our study, all patients had dysarthria, with only one having mild tongue weakness and only four patients having facial weakness. This suggests that lingual dystonia was likely responsible for the dysarthria rather than weakness in these patients.

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