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Case Report

CASE REPORT

Dysarthria-clumsy hand syndrome: The mystery of internal capsule anatomy

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Abstract

Lacunar acute ischemic stroke is a common neurological emergency worldwide. The clinical presentation varies widely depending upon the location of the stroke. Dysarthria-clumsy hand syndrome is a distinct clinical entity which is characterized by difficulty speaking due to weakness of the muscles of articulation and/or phonation and clumsiness in the upper extremity without significant weakness. It is classically described in lacunar strokes involving the splenium/genu of internal capsule or basis pontis. It is rarely reported in strokes involving the other areas of brain. We report a case of dysarthria-clumsy hand syndrome in a patient who developed lacunar acute ischemic stroke in the posterior aspect of the posterior limb of right internal capsule. He was also found out to have coarctation of the descending thoracic aorta, ectasia of the ascending aorta and an aneurysm in the left middle cerebral artery at its bifurcation. The significance of these coexisting vascular anomalies is unknown.

Key words: Dysarthria clumsy hand syndrome; internal capsule; lacunar stroke; basis pontis; coarctation of aorta; aneurysm

Introduction

Dysarthria-clumsy hand syndrome is a relatively rare clinical presentation of acute stroke. It is characterized by difficulty speaking due to weakness involving the muscles of articulation and/or phonation, including the muscles of tongue, pharynx and larynx. It also involves clumsiness of upper extremity (corresponding to the site of stroke) without weakness. There may be corresponding unilateral upper motor neuron facial weakness. Neurological examination in the domains of cognition/ language, motor and coordination of the lower extremities and motor power involving both the upper extremities are generally normal.

The exact proportion of acute ischemic stroke patients presenting with dysarthria clumsy hand syndrome across the globe is not known. However, as per a report, dysarthria clumsy hand syndrome accounted for 1.6% of all acute stroke patients and 6.1% of all lacunar syndromes [1]. It is more likely to be a presentation of lacunar stroke in the genu of the internal capsule or basis pontis [2,3], both of which in an early computerized tomography scan of brain that is done in the Emergency Department may be normal. The importance lies in the recognition of this distinct clinical entity as a possible presentation of acute ischemic stroke since this can be easily confused with a functional speech disorder/intoxication [4], particularly in a hyperacute setting.

Case report

A 46-year-old gentleman presented to the Emergency Department with acute onset mild slurring of speech and swallowing difficulty. He was apparently all right the previous night when he went to bed and woke up the next morning with the symptoms. He denied any visual or auditory disturbances. He denied any weakness or incoordination of the limbs. He did not report gait instability.

His past history was significant for diabetes mellitus, hypertension and dyslipidemia for which he was taking regular medications.

In the Emergency Department, apart from mild dysarthria and dysphagia due to weakness of the pharyngeal and lingual muscles the other neurological examination was grossly unremarkable.

Urgent computerized tomography scan of the brain showed old infarct in the left cerebellum. There were no features of acute large territorial infarct or hemorrhage. Computerized tomography angiography of the brain and neck showed hypoplastic left vertebral artery, fetal bilateral posterior cerebral arteries. The vessels of the anterior circulation were unremarkable. Incidentally, the coarctation of the descending thoracic aorta and ectasia of the ascending

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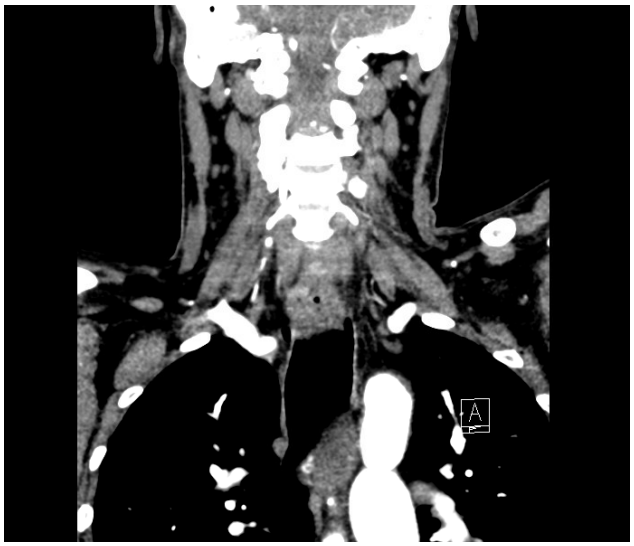

Figure 1. CT angiogram showing coarctation of decending aorta

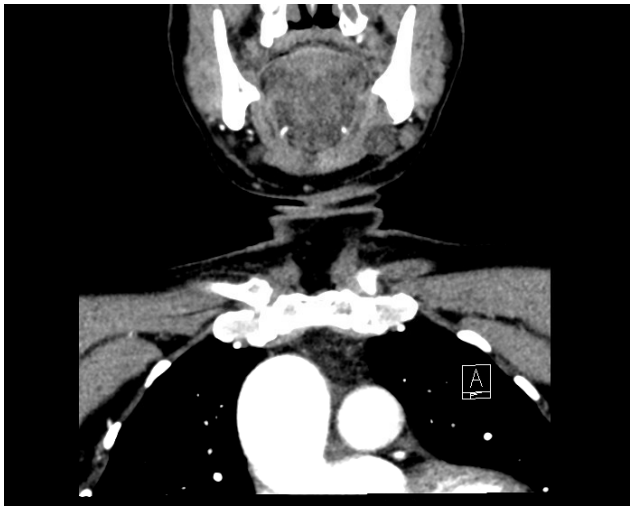

Figure 2. CT angiogram showing ectasia of ascending aorta

aorta were noted (fig. 1 and 2 respectively).

MRI brain showed acute lacunar infarct in the right internal capsule, in the posterior aspect of its posterior limb (fig. 3).

There was also evidence of multiple old infarcts involving the left thalamus and left internal capsule (fig. 4).

There were no recent or old infarcts in the pons. MR angiography of brain revealed an incidental left middle cerebral artery aneurysm of 4-5 mm near its bifurcation (fig. 5).

During the course in his hospital stay his neurological functions worsened. The next day he developed left facial weakness and clumsiness in his left upper limb. The dysarthria worsened to an extent that he was not able to pronounce a single word. His dysphagia also worsened which necessitated insertion of nasogastric tube. On neurological examination, he had profound weakness of the muscles of the tongue, pharynx and larynx. However he had no abnormal neurological findings in his both lower limbs and his gait was normal.

With regard to the MCA aneurysm and coarctation of aorta, he was asymptomatic and never sought medical attention for the same.

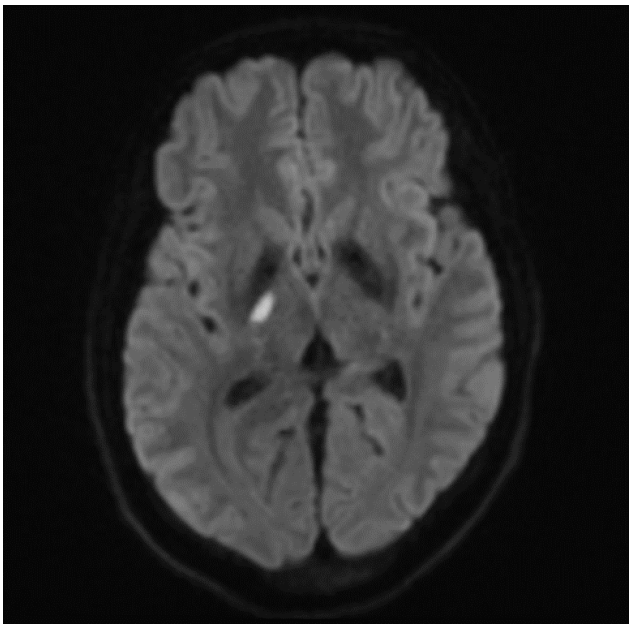

Figure 3. MRI diffusion weighted image showing acute lacunar infarction in right internal capsule

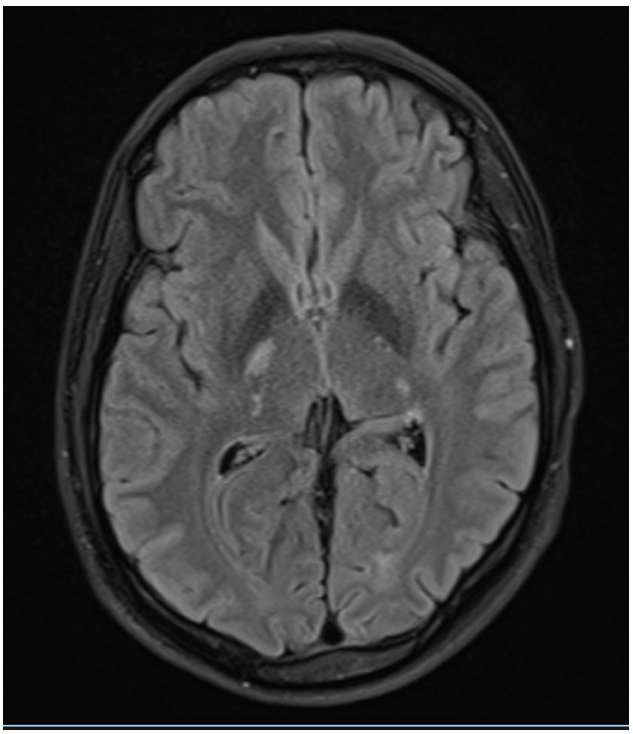

Figure 4. MRI FLAIR image showing multiple lold lacunar infarcts

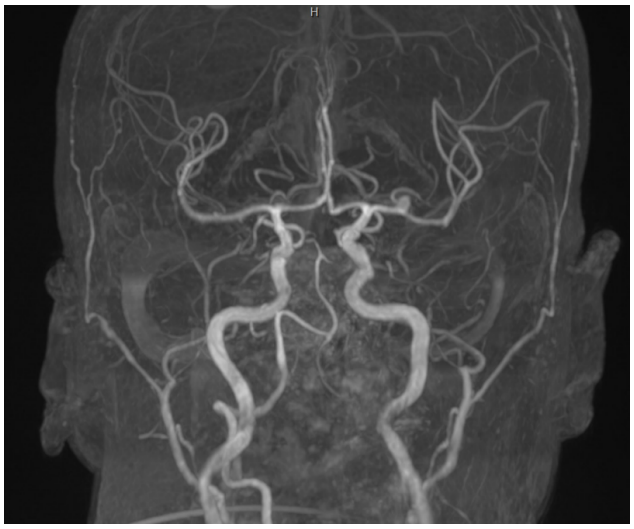

Figure 5. MR Angigraphy showing left middle cerebral artery aneurysm

Discussion

There are three key issues to be discussed in this case, viz. anatomy of internal capsule; anatomical localization of dysarthria clumsy hand syndrome; and possible association between the vascular anomalies and the variation in the internal capsule anatomy in a given individual.

Internal capsule is a deep cerebral white matter substrate that is anatomically divided into three parts viz. anterior limb, genu and posterior limb. The anterior limb of internal capsule is medially related to caudate nucleus and laterally to the lentiform nucleus. The posterior limb of internal capsule is medially related to thalamus and laterally to the lentiform nucleus.

The anterior limb of internal capsule consists of fibers of anterior thalamic radiation and fronto pontine fibers [6]. It also contains transversely communicating fibers between caudate nucleus and putamen [7]. The posterior limb of the internal capsule contains fibers of the posterior thalamic radiation, corticospinal tract, corticorubral tract, and corticopontine tract [6]. The anterior half of the posterior limb contains the corticospinal tract, corticorubral tract, and corticopontine tract. The corticospinal tract originates from the primary motor cortex and premotor areas. Fibers from the premotor areas are situated rostrally to fibers from the primary motor cortex [8]. The genu of the internal capsule consists of the fibers of superior thalamic radiation and corticobulbar tract [7]. The anatomical variations in the composition of the fibers in the internal capsule is not well understood yet. Diffusion tensor imaging and fibre tractography would be of help in that regard.

Dysarthria clumsy hand syndrome is classically described as a lacunar syndrome. The best recognised association of dysarthria clumsy hand syndrome has been with lacunes of the anterior limb of the internal capsule [9]. Lacunar stroke involving the genu of the internal capsule or also known to cause dysarthria clumsy hand syndrome [3]. Basis pontis is another location of lacunar stroke which is known to cause dysarthria clumsy hand syndrome [2]. Corona radiata, basal ganglia and thalamus are uncommon sites associated with dysarthria clumsy hand syndrome [1]. To the best of our knowledge, lacunar stroke involving the posterior aspect of the posterior limb of internal capsule is not mentioned in the literature to cause dysarthria clumsy hand syndrome. Two possible explanations in this particular case are,

A normal anatomical variation in the constituting fibers of internal capsule.

Pattern of neural plasticity and synaptic connections established after the recurrent lacunar strokes as seen in the MRI of the patient showing chronic ischemic changes that made this patient susceptible to develop dysarthria clumsy hand syndrome after a stroke in the posterior limb of internal capsule.

There is sufficient data in the literature to support the association between intracranial aneurysms and coarctation of aorta [10]. But there is no literature mentioning the association between the vascular anomalies present in this patient and the anatomical variation in the composition of the fibers of internal capsule (if any). More studies to answer these questions may be needed.

Conclusion

Dysarthria clumsy hand syndrome is a distinct clinical entity that is commonly associated with lacunar syndromes. The pathology may be localized to anterior limb or genu of internal capsule or the basis pontis. Lacunar strokes in the basal ganglia, corona radiata and thalamus may also present with dysarthria clumsy hand syndrome. However lacunar stroke in the posterior aspect of the posterior limb of internal capsule causing dysarthria clumsy hand syndrome is not described in the literature. The possible explanation in this case may be due to normal anatomical variation in the constitution fibers of internal capsule. Moreover, the concurrent presence of vascular anomalies in this patient may be a coincidence or an association.

Declarations

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Competing interests

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Patient consent for publication

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Ethical approval

Not applicable.

Informed consent

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