Unusual presentation of more common disease/injury

The striatocapsular infarction and its aftermaths

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Ischaemic stroke syndromes in the vascular territory of middle cerebral artery may have atypical presentation and radiographic findings because of the variable anatomy of that artery. Therefore, misdiagnosis of these syndromes as neoplastic or infectious processes is not uncommon. This case describes a 69-year-old comatose woman who was referred to us as having 'a brain tumour with massive surrounding oedema.' Further work-up revealed that she had a large left-sided lenticular nuclear infarction with some extension into the surrounding areas—the striatocapsular infarction.

BACKGROUND

Isolated and large lenticular infarction is a rare form of middle cerebral artery (MCA) occlusion that might present in a variety of ways; this case was referred to us as a brain tumour. Physicians interested in vascular neurology should be familiar with this form of ischaemic stroke so as not to misdiagnose the case, and should spread this 'message' to others.

CASE PRESENTATION

A 69-year-old woman was referred to our accident and emergency from a rural hospital for further management of brain tumour-associated coma. We received the patient approximately 20 h after she developed this coma. The patient's daughter stated that her mother was neither hypertensive nor diabetic and that she developed a progressive loss of consciousness over a matter of 5 h. On admission, her blood pressure was 170/100 mm Hg with a regular heart rate of 90 bpm and a respiratory rate of 20 cycles per minute; she was a febrile. Examination revealed a Glasgow Coma Scale of 3 and a right extensor planter. A non-contrast brain CT scan, which was done 10 h ago in the referring rural hospital showed 'an elongated deep left hemispheric hypodense area that is suggestive of brain oedema,' as the report stated, but no film was provided. The airway, breathing, and circulation resuscitation started, routine blood tests were ordered and a chest x-ray and abdominal ultrasound were arranged. Meanwhile, cerebral anti-oedema measures with mannitol infusion and controlled hyperventilation were started. The next day we did a non-contrast brain CT scan; the whole left lenticular nucleus and part of the caudate appeared hypodense, creating a triangular/coma-shaped area, and the overlying insula and cortex were normal looking. The same day, a transthoracic echocardiographic examination was done and, apart from mild diastolic dysfunction, the examination was unremarkable. Carotid Doppler studies detected mild bilateral stenosis at the origin of both internal carotids; however, no percentage was given. Her blood cell counts were within their normal reference range.

INVESTIGATIONS

Brain MRI with contrast was done on day 4 of admission; the left lenticular nucleus (putamen and globus pallidus) and the head of caudate appeared infarcted with some extension into the surrounding areas; the adjacent thalamus and the overlying cortical areas seemed to be spared (figure 1–4).

DIFFERENTIAL DIAGNOSIS

The location of the radiographical abnormality and its 'shape' may suggest a neoplastic or an infectious process. We took the opinion of our colleagues and postgraduate trainees after providing them with a brief history of the patient and examining her brain MRI imaging; none of them had suggested an arterial cause and only one considered a venous system occlusion. The majority pointed towards a malignant tumour and few suggested an encephalitic process.

OUTCOME AND FOLLOW-UP

We stopped mannitol infusion and controlled hyperventilation, and aspirin, atorvastatin, metoprolol and enalapril were started after doing the MRI (ie, on day 4 of admission). On day 6, the patient spontaneously opened her eyes and on day 7 both eyes were voluntarily opened and she fully moved her left arm and leg. Although she responded to orders appropriately (raise your left arm, close your eyes, put your left arm above your head, etc), she did not say a word or utter a sound. She was discharged after 10 days, prescribed the same medical treatment and physiotherapy was arranged.

DISCUSSION

The MCA is the largest branch of the internal carotid artery and is the continuation of this artery into the Sylvian fissure; the MCA starts just below the medial part of the anterior perforated substance. This artery is the source of blood supply of most of the lateral surface of the cerebral hemisphere in addition to deep structures (white and grey matters) of the frontal and parietal lobes.¹ In terms of vascular anatomy, the MCA is divided into three 'segments': the proximal,

Figure 1 Non-contrast brain CT scan of the patient at the level of the basal ganglia approximately 2 days after developing coma. Note the left-sided deep hemispheric hypodense area that fits the site of the lenticular nucleus. The referring physician considered this an oedema surrounding a malignant mass.

Figure 2 T1-weighted MRI with gadolinium of the same patient 4 days after admission. A hypointense coma-shaped area that did not take the contrast; the infarcted area consisted of putamen, globus pallidus, anterior limb of internal capsule and head of caudate.

Sylvian and the distal segments. The penetrating lenticulostriate arteries arise from the posterior and superior aspect of the 'proximal' segment; the adjacent corona radiate, external capsule, claustrum, putamen, parts of the globus pallidus, body of the caudate nucleus and the upper parts of

Figure 3 An axial T2-weighted brain MRI image of the patient at the level of the basal ganglia. Note the heterogeneous hyperintense signals at the left basal ganglia; the overlying cortex and the adjacent thalamus are spared.

Figure 4 Coronal T2 FLAIR brain MRI image of the patient; note that the abnormal signal of the T2-weighted image did not suppress on the FLAIR film. The left basal ganglia have the bulk of the abnormal signal intensity; note the extension of the signal into the core of the left temporal lobe. The cortical areas and thalamus are not involved.

the anterior and posterior limbs of the internal capsule are nourished by these penetrating arteries. The Sylvian segment gives rise to branches in the Sylvian fissure that supplies the insula of Reil. Then, the distal segment of the stem may face one of three paths: it may bifurcate into two branches in 78% of individuals, it may trifurcate into three arteries in 12% of individuals or it may divide into multiple trunks with variable sizes and length.² Therefore, the commonest event is that the MCA divides into anterior and posterior branches that supply most of the lateral surface of the cerebral hemisphere. Because of this variable vascular anatomy, the clinical picture of MCA occlusion is highly variable and may be isolated to a specific area—for example, complete proximal main stem occlusion, deep arterial territory infarction or superficial cortical (anterior or posterior) syndromes.^{3 4}

Acute unilateral ischaemic stroke confined to the lenticular nucleus (putamen and globus pallidus) is a rare event comprising 1.6% of the deep MCA territory ischaemia. The overall clinical pictures of isolated deep MCA ischaemic strokes are usually dominated by damage to the internal capsular structures rather than the damaged lenticular

nucleus; therefore, faciobrachiorural paresis is the most prominent feature in most patients.⁵ Ghika *et al*⁶ and Moulin *et al*⁷ reported pure infarctions limited to the internal capsule with a multitude of presentations; however, pure ischaemic stroke confined to the lenticular nucleus remains poorly defined. According to Giroud *et al*,⁸ the presentation of lenticular nuclear damage may include a cognitive-behavioural syndrome (mainly from pallidal lesions) or a motor syndrome (resulting from putamenal damage). However, this distinction is blurred in most cases by the variable vascular supply and efficiency of the collaterals.

Our patient developed an acute ischaemic infarction involving the left putamen, globus pallidus, part of the caudate nucleus and the anterior limb of internal capsule—a coma-shaped area on brain CT scan or MRI. Most likely, this has resulted from occlusion of the lateral striate branches of the proximal segment of the MCA ending with the so-called striato-capsular syndrome.⁹ This syndrome comprises hemiparesis (usually affects the arm more than the leg) and cortical signs (from damage to the subcortical fibres connecting the cortex and deep structures) such as non-fluent aphasia, neglect, apraxia and so forth.⁹

The patient's speech disturbance is clinically consistent with an expressive type of dysphasia. Very rarely, the speech disturbance of striato-capsular infarctions may manifest itself as a mild limb weakness but with severe dysarthria.⁹ However, Bhata and Marsden¹⁰ suggested that abulia is the commonest behavioural abnormality in patients with focal basal ganglia lesions; this might easily be considered a dysphasic component of the symptomatology. Nadeau and Crosson¹¹ critically reviewed the available literature of 'subcortical aphasia'. They concluded that the language defects of striatocapsular infarctions are due to sustained hypoperfusion of the overlying cortex that is not visible on the conventional imaging studies. Conventional four-vessel cerebral angiography is a very valuable tool in defining the vascular occlusion and its anatomy; unfortunately, the lack of expertise in our hospital radiology department with regard to this invasive investigation cancelled the use of this option.

We believe that the resultant cytotoxic oedema of the infarcted tissue has resulted in a mass effect and impingement of the underlying thalamus and upper midbrain with secondary dysfunction of the ascending reticular activating system; the possible cause behind this patient's coma. However, this is unlikely to develop within a 'few hours' of the vascular occlusion. The patient's daughter insisted that her mother did not complain of any weakness and the family did not notice any abnormalities in their mother's speech; the only notion was that the patient's consciousness progressively deteriorated to the point of reaching coma within a few hours. We question the duration of the patient's initial presentation; we believe that the duration of the deterioration of her consciousness is much longer.

The patient came back after 2 weeks for a scheduled follow-up visit. She demonstrated a right-sided pyramidal weakness of grade 3 and an extensor planter. During the visit, she did utter a few sounds but no single word. During this visit, we noticed right-sided spasticity. We specifically asked the family if there was any 'dystonic' posturing or any abnormal movement of the right arm/leg during the past 2 weeks and the reply was no. Giroud, Bhatia and

Marsden^{8 10 12} noticed that dystonia was the commonest acute, subacute or delayed manifestation of lenticular damage. Our patient did not develop such a manifestation and this observation is consistent with that of Russmann and colleagues⁵; none of their 13 patients with pure lenticular infarctions developed any form of movement disorders.

Learning points

- ▶ The MCA is a medium-sized artery that supplies most of the hemispheric structures, both superficial and deep.
- ▶ The variable vascular anatomy of the MCA may result (when occluded) in a multitude of hemispheric ischaemic strokes syndromes with atypical presentations and brain imaging findings. The inexperienced physician might easily misdiagnose these syndromes as neoplastic or infectious process.
- ▶ The occurrence of 'sudden' or rapidly progressive neurological dysfunction(s) should always prompt the physician to search for a vascular cause, even if the brain imaging findings are atypical.
- ▶ Isolated ischaemic infarction of the lenticular nucleus and part of the caudate is a very rare event resulting from occlusion of the lateral striate arteries; because of the location and shape of the damaged area, an alternative diagnosis, other than stroke, usually comes into mind.
- ▶ Pure lenticular infarction may not commonly result in dystonia in contrast to what is mentioned in previous literature.

Competing interests None.

Patient consent Obtained.

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