Novel transient alien limb phenomenon heralding a diabetic hyperosmolar non-ketotic state with leukoaraiosis: a video presentation

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DESCRIPTION

A 76-year-old right-handed non-diabetic woman presented with involuntary upper left limb movements for 2 h. The patient described the limb as having a will of its own. The movements occurred intermittently for 7 days, with each episode lasting 10–20 min. Segment 1 (video 1) showed levitation of the left hand with right hand restraint. Segment 2 displayed the left hand thumping the chest, with neck abrasions. Segment 3, 1 week later demonstrated neck strangulation. Segment 4, 2 weeks later exhibited normality. Physical examination revealed apraxia and dystonia but no parkinsonism. The Montreal cognitive assessment score was 25/30 due to executive impairment. The patient was dyslipidaemic with hypertensive heart disease. However, the blood sugar was 589 mg/dL, glycated haemoglobin was 11.2% and serum osmolality was increased at 329 mOsm/kg water without ketonuria. Other routine blood investigations were normal. Intravenous insulin and fluids were given. MRI performed on admission showed diffuse brain atrophy on T1 (figure 1A); extensive high signals in the periventricular white matter (leukoaraiosis) in axial T2 and fluid-attenuated inversion recovery imaging (figure 1B); normal diffusion-weighted imaging (figure 1C) and apparent diffusion coefficient (figure 1D). Sagittal T1 MRI view showing an intact corpus callosum (figure 1E).

Figure 1 MRI performed on admission: (A) Axial T1-weighted MRI showing diffuse cerebral atrophy. (B) Axial T2 MRI view showing extensive periventricular hyperintensities (leukoaraiosis; see arrows). (C) Axial T2 fluid-attenuated inversion recovery MRI showing leukoaraiosis consistent with small vessel ischaemic changes. (D) Axial diffusion-weighted MRI without hyperintensities suggestive of an infarct. (E) Sagittal T1 MRI view showing an intact corpus callosum (see arrow).
(figure 1B,C); no hyperintensities suggestive of stroke on diffusion-weighted imaging axial view (figure 1D) and intact corpus callosum in sagittal T1 view (figure 1E). MR angiography and EEG were normal. At 5 weeks, repeat MRI sequences showed no new changes. The patient remained well 3 months later on oral hypoglycaemics, with no suggestion of corticobasal syndrome.1 Alien limb phenomenon has many clinical and radiological correlations, including leukoaraiosis.2 Damage of brain neuronal connectivity has been postulated as the pathogenic mechanism.3

**Learning points**

- These intermittent, slow and fast jerky movements accompanied by self-inflicted injuries due to strangling, grasping, thumping with lifting and dropping the left upper limb met the classic definition of alien limb phenomenon, with unwilled and uncontrollable but seemingly purposeful movements of an upper or lower limb.
- Although dyskinesias, focal epilepsy, hemichorea-hemiballismus, asterixis and tremors have been associated with a diabetic hyperosmolar nonketotic state (DHNKS), the alien limb phenomenon has not been linked to DHNKS before.
- The pathogenesis of DHNKS-induced alien limb phenomenon is unknown but it probably implies disruption of the brain network causing loss of inhibitory tone.

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**REFERENCES**

