Limb-Shaking Transient Ischemic Attack due to Stenosis of the Middle Cerebral Artery
- Case Report -

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We present a case of a 51-year-old man with frequent left limb-shaking transient ischemic attacks due to severe stenosis of the right proximal middle cerebral artery (MCA). The involuntary movement was provoked by walking and it was relieved by adopting a squatting posture. Initially, we tried antiepileptic medication, but it did not eliminate the involuntary movement. Cerebral angiography and single photon emission computed tomography demonstrated decreased perfusion and reserve capacity of the right cerebral hemisphere which correspond to stenotic proximal MCA territory. After superficial temporal artery to MCA anastomosis, the decreased reserve capacity was restored and his limb-shaking attack completely disappeared. His history of radiotherapy for craniopharyngioma, in addition to hypercholesterolemia and diabetes mellitus, seems to have contributed to gradual stenosis of right MCA.

KEY WORDS: Limb-shaking TIA · STA-MCA anastomosis · Radiotherapy · MCA stenosis.

Introduction

The involuntary limb-shaking is a rare symptom in obstructive cerebrovascular disease, but is recognized as a type of transient ischemic attack (TIA), which is influenced by postural change1,2,7,8,10,13. It is sometimes mistaken for focal seizure, because of its similar appearance, and this may lead to antiepileptic agent misuse1,13. But it differs from focal seizure in that it involves only the extremities, with no face or trunk involvement, no tonic contraction or tonic-clonic movement1,8 and no epileptiform pattern on EEG1,7-9,15.

In many cases, cerebral angiography showed definite carotid stenosis on the side opposite the movement disorder, and the focal cerebral ischemia is likely to be pathogenetic1,2,4,7,8,10,13. Their symptoms improve after surgical revascularization1,2,7,8,13.

In this rare ischemic symptom, the carotid artery is recognized as the most common site of occlusive lesion1,8.

We present a case of limb-shaking TIA resulting from proximal middle cerebral artery (MCA) stenosis after cranial irradiation, which is very unique in its stenosis site and radiotherapy history. His involuntary limb-shaking movements are strongly suspected of resulting from radiation induced MCA stenosis.

Case

A 51-year-old, right-handed man visited hospital because of involuntary limb-shaking movements of the left arm and leg. Over 8 months, he had experienced many episodes, which initiated after several minutes of walking, and which ceased on squatting or sitting. This involuntary movement made him difficult to walk alone or to balance himself. The movement always involved the left arm and leg and never involved the face, trunk or right limbs. During such episodes, his consciousness was clear and he experienced no accompanied motor weakness or speaking difficulty. He had no prior seizure history, and his symptom had no other triggering factors except walking. On admission, he had no abnormal neurologic findings but he was receiving medication for hypothyroidism, hypercholesterolemia and diabetes mellitus (DM) which was well controlled.

Fig. 1. Preoperative axial T2 weighted magnetic resonance image(A) showing cerebromalacia in the right frontal lobe (arrowhead). Magnetic resonance angiography(B) showing segmental narrowing of the right M1 (arrowhead) and faint trace of M2.
Eighteen years previously, he had suprasellar craniopharyngioma, which presented with visual disturbance and polyuria. After transcranial operation, he received 5,425cGy of radiotherapy for 8 weeks. The tumor remitted completely and no other sequela remained except for the hypopituitarism.

At first, he was misdiagnosed as having suffered from focal seizure, because of its seizure-like appearance. Therefore, an antiepileptic agent was tried, but it had no effect on his symptom, and the EEG showed no epileptiform activity.

The magnetic resonance image MRI revealed right frontal cerebromalacia Fig. 1A and magnetic resonance angiography MRA revealed multiple stenosis of the right MCA, the right internal cerebral artery ICA and the basilar artery Fig. 1B.

Cerebral angiography demonstrated multiple stenosis in the right MCA, the bilateral anterior cerebral artery ACA, the bilateral posterior cerebral artery PCA, the bilateral ICA and the basilar artery, and the degree of stenosis was most severe in the right MCA. The right MCA was almost occluded and its distal part was perfused through multiple fine collateral channels Fig. 2A, B. SPECT demonstrated both perfusion and reserve capacity decrease in the whole right cerebral hemisphere Fig. 3A, C.

We believed that his involuntary movement was due to TIA caused by right MCA stenosis, and right STA-MCA anastomosis was performed. No perioperative problem occurred. Seven days postoperatively, cerebral angiography demonstrated abundant vascular supply in the right cerebral hemisphere via the patent STA-MCA bypass Fig. 2D, E and his involuntary limb-shaking movement disappeared. Nine days postoperatively, however, left hemiparesis developed. He also had suffered from transient left hemiparesis following cerebral angiography preoperatively. The diffusion MRI demonstrated acute cerebral infarction in the right parietal lobe. But at that time of discharge from hospital, he recovered sufficiently to walk alone.

One year later, SPECT showed improved vascular reserve in the right parietal and frontal lobe Fig. 3B, D and his hemiparesis was completely recovered.

**Discussion**

**Involuntary limb-shaking TIA**

The limb-shaking TIA is the rare clinical symptom in occulsive cerebrovascular disease, although it has been described in several reports and its pathophysiology is relatively well-known. It is ascribed to the focal cerebral ischemia caused by cerebral hypoperfusion and a loss of autoregulation, usually resulting from cerebrovascular stenosis.

Although involuntary movement of this case was similar to focal seizure in appearance, it lacked the features of focal seizure in several aspects. It always involved the left arm and leg, never involved right limbs, face or trunk, never impaired consciousness, and never spread to tonic or tonic-clonic movement. These were the same characteristics of limb-shaking TIA as described in earlier reports. At first, his symptoms were misdiagnosed as focal seizure and an antiepileptic agent was tried. Such mistakes have been reported on several occasions due to the rarity of the symptoms and a similarity with focal seizure, in terms of appearance. Generally, limb-shaking TIA shows a normal EEG, which differentiates it from focal seizure.
His involuntary movement was initiated by walking and ceased on squatting or sitting, and was, therefore, closely related with postural change. These characteristics are also similar to those cases previous reported.1,4,8,13,15 Tatemichi15 reported that this may be attributed to a transient decrease in perfusion pressure and cerebral blood flow caused by postural change.

The right MCA stenosis was most prominent by cerebral angiography and MRI demonstrated that the cerebromalacia was in the right hemisphere. Such results suggested that his symptom was due to right MCA stenosis. The SPECT finding of decreased perfusion and vascular reserve also supported this suggestion.

After the STA-MCA anastomosis, his symptom improved and the SPECT study confirmed improved vascular reserve. These results are compatible with similar reports1,2,7,8,13 and proved that his symptom was due to TIA.

Consequently, we concluded that his involuntary limb-shaking movements were caused by TIA resulting from stenosis of the right MCA because: 1) his symptom had the same feature as other limb-shaking TIs reported upon1,2,7,9,15; 2) the MCA stenosis was angiographically documented, 3) SPECT showed decreased perfusion and vascular reserve, and 4) his symptoms improved after surgical revascularization.

However, in most reports1,2,7,8,13, the most common stenosis or occlusion site, which induce limb-shaking TIs, is carotid artery area. The MCA stenosis as the cause of this type of TIA is very rare. Interestingly, most of reported cases of MCA stenosis had previously received radiotherapy1,3,6,14.

**Radiation induced MCA stenosis**

The biologic effect of radiation on brain tissue has been extensively studied and many cases of radiation induced vascular stenosis or occlusion have been reported1,3,5,7,11,12,14. Radiation induces progression of the atherosclerosis, fibrosis of the vascular wall, regression of the elastic tissue and of the vascular wall, and hyperplasia of the fibroblast, finally leading the vessels to stenosis or occlusion5,7. It has been reported that these changes are limited in irradiated area1,12, and atherosclerosis is believed to be accelerated by hypercholesterolemia11. In the present case, no vessel biopsy was done, but considering his hypercholesterolemia, he may had been more sensitive to radiation.

Carotid artery stenosis after neck irradiation has also been described in many reports5,7, and the latency between radiotherapy and symptom onset was found to vary greatly, from 6 months to 27 years5,6,11. Many cases of cerebrovascular stenosis after paraseellar irradiation have also been reported5,6. Bitzer3 analyzed 41 cases and reported that MCA stenosis developed after paraseellar irradiation in many cases.

In the present case, the patient had been exposed to 54Gy of radiation, which was not an insignificant dose considering that only 10Gy of radiation has been reported to induce stenosis1,4, and the 18 years of latency between radiotherapy and symptom onset is not so long in comparison with other cases1,5,6,11. Accordingly, we concluded that previous radiotherapy may had played an major role in the formation of MCA stenosis, in addition to the contribution by hypercholesterolemia and DM.

**Conclusion**

In this case, involuntary limb-shaking was not focal seizure but unusual TIA caused by stenosis of the MCA. This symptom was ameliorated by STA-MCA anastomosis. Previous radiotherapy is strongly suspected to be the major cause of this MCA stenosis.

**References**