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【 CASE REPORT 】

Acute Ischemic Stroke due to Undifferentiated Sarcoma: A Case Report and Literature Review

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Abstract:
Tumor emboli due to a sarcoma are usually confirmed by an autopsy or operative findings. A sarcoma embolus in an acute stroke patient is rare. We herein report a 37-year-old man with acute stroke caused by internal carotid artery occlusion who underwent embolectomy. A histopathological analysis of an embolus obtained with a mechanical retriever device was diagnosed as undifferentiated sarcoma. This is the first case of extracardiac sarcoma extraction via mechanical retrieval performed during intervention for acute ischemic stroke. A histopathologic evaluation with embolectomy is important for diagnosing tumor emboli.

Key words: mechanical embolectomy, tumor embolus, sarcoma

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Introduction

Stroke caused by large-vessel occlusion due to tumor emboli is uncommon. Tumor emboli are usually confirmed by an autopsy or operative findings (1). A few reported patients with stroke due to tumor emboli were diagnosed by a histological examination after mechanical endovascular embolectomy. Verifying an extracardiac tumor embolus, especially sarcoma, is very rare.

Case Report

A 37-year-old right-handed man experiencing migraine presented with sudden-onset left-sided limb weakness and was immediately admitted to our hospital. He had left hemiplegia with neglect, mild dysarthria, and a National Institute of Health Stroke Scale (NIHSS) score of 13. Magnetic resonance imaging of the brain revealed acute infarcts in the right middle cerebral artery area. Subsequent magnetic resonance angiography and carotid duplex scanning showed an occluded right internal carotid artery. Intravenous recombinant tissue plasminogen activator (0.6 mg/kg) was administered 120 minutes after symptom onset, without neurological improvement. Endovascular treatment was chosen because his symptoms suggested a clinical radiological mismatch (Figure A and B).

Cerebral angiography showed right internal carotid artery proximal occlusion. Mechanical endovascular embolectomy with 3 passes of a Trevo® ProVue Retriever 4×20 mm (Stryker, Kalamazoo, USA) was successful, achieving partial recanalization with thrombolysis in cerebral infarction (TICI) grade 2b (Figure C-E).

A pathological examination of the embolus revealed atypical cells with pleomorphic nuclei (Figure F) that were immunohistochemically positive only for vimentin and desmin (Figure G), suggesting undifferentiated sarcoma.

Transesophageal echocardiography showed no cardiac tumor. Positron emission tomography-computed tomography revealed the uptake at the aortic root, indicating that the embolus originated from an occult aortic sarcoma (Figure H). One month after the stroke, his NIHSS score was 4, and his modified Rankin Scale score was 2. He was discharged on day 54 and later prescribed proton therapy for the sarcoma.

Discussion

To our knowledge, this is the first case of extracardiac undifferentiated sarcoma extraction via mechanical retrieval performed during acute ischemic stroke intervention. Undif-

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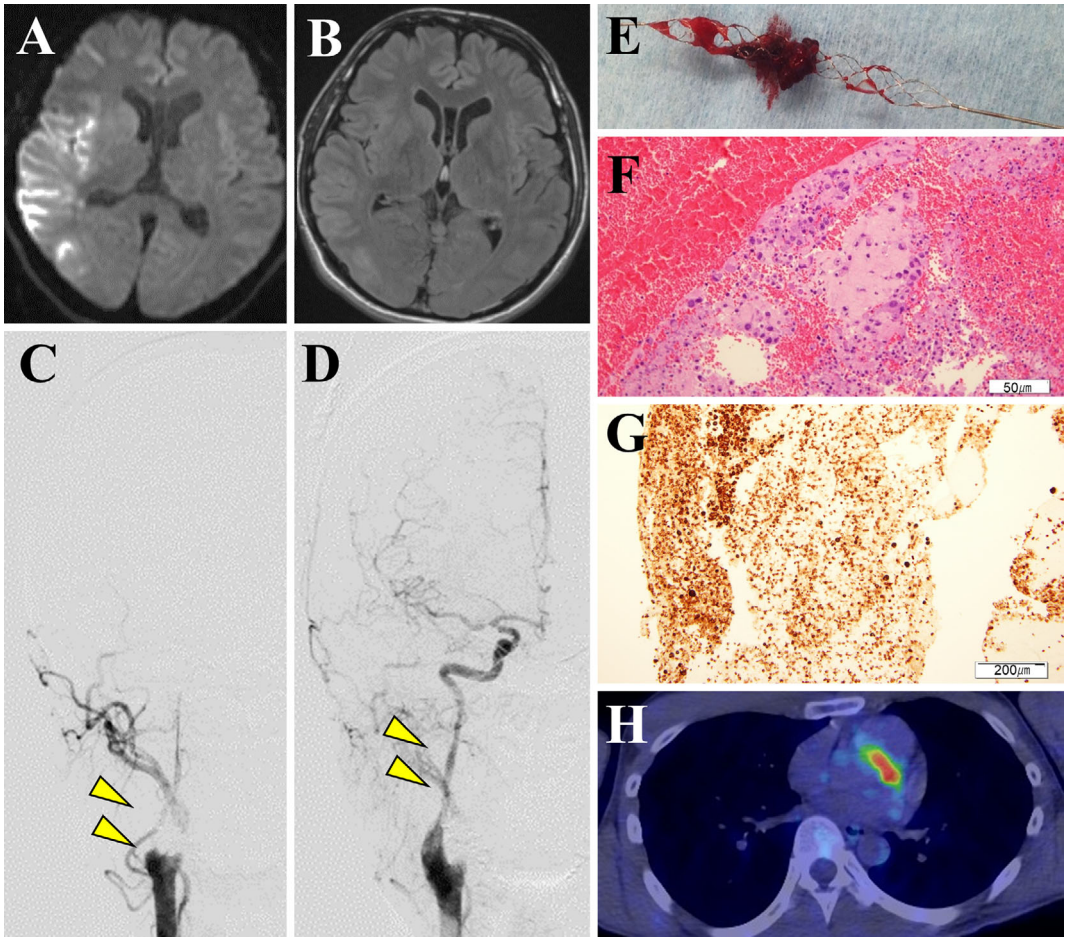

Figure. **(A) Pre-procedural diffusion-weighted imaging shows right middle cerebral artery area infarcts. (B) T2-fluid attenuated inversion recovery imaging suggested a clinical radiological mismatch. (C) Right common carotid artery angiography shows occlusion of the right internal carotid artery. (D) Successful recanalization (TICI grade 2b). (E) Specimen on the mechanical clot retriever. (F) The retrieved embolus contains atypical cells with pleomorphic nuclei (Hematoxylin and Eosin staining). (G) The immunohistochemical findings were positive for vimentin. (H) Positron emission tomography-computed tomography revealed the uptake at the aortic root.**

ferentiated sarcomas are rare and are often diagnosed by an autopsy. In our case, a histopathological analysis of the embolus obtained from the mechanical retriever device was diagnostic of an undifferentiated sarcoma. Thus, mechanical embolectomy may be useful for evaluating tumor-induced acute large-vessel occlusions in order to obtain a histological diagnosis.

Several trials have demonstrated the benefit of endovascular therapy for acute stroke patients with large-vessel occlusion (2), although the embolectomy-retrieved emboli were not sufficiently investigated histopathologically. As the appearance of the embolus in our case could not be macroscopically distinguished from that of a common thrombus, it was difficult to diagnose it as a tumor embolus without a routine pathological examination. As reports of tumor emboli extraction due to mechanical retrieval in acute ischemic stroke are important, we explored recently reported stroke patients with tumor emboli treated with endovascular embolectomy (Table). Of the 14 cases, 10 (71%) were of cardiac origin, 6 of which (60%) were cardiac myxomas. Cardiac origin is among the most frequent origins of intracranial tumor emboli (3). The occlusion sites were treated with clot retrieval (n=10) and aspiration embolectomy (n=5) devices. Altogether, 12 (85%) cases achieved successful recanalization (TICI 2b-3). The extracardiac tumors included breast cancer, melanoma, and lung cancer (4-6). In these cases, the primary lesion had already been confirmed before embolectomy, whereas in our case, it was diagnosed after embolectomy.

In conclusion, a histopathological evaluation following embolectomy is important for diagnosing tumor emboli. Although embolectomy for tumor emboli appears to be effective and relatively safe, further studies are needed to verify our results.

Ethics approval was provided by the Ethics Committee of Ichinomiya Nishi Hospital, Japan.

The authors state that they have no Conflict of Interest (COI).

Table. Summary of Reported Cases with Histological Confirmation of Cardiac and Non-cardiac Tumor Causing Stroke with Endovascular Embolectomy.

Case	Tumor types	Primary origin	Age/ sex	Initial NIHSS	Site of occlusion	Treatment modalities	Recanalization	Clinical outcome	Reference
Cardiac tumor									
1	Papillary fibroelastoma	Unknown	64/ M	16	Right M1	Solitaire®	TICI 3	NIHSS 3	[7]
2	Myxoma	Unknown	45/ NA	22	Left M1	TPA + Solitaire® + Trevo® + intracranial stent	TICI 0	NIHSS 19, mRS 4	[8]
3	Myxoma	Unknown	34/ NA	26	Left M1, A2	Solitaire®	TICI 3	NIHSS 3, mRS 2	
4	Myxoma	Unknown	46/ M	NA	Left carotid T	TPA+Forced suction thrombectomy (Optimo® + Penumbra®)	TICI 2b	Left eye blindness	[9]
5	Myxoma	Unknown	34/ M	9	Right M1 proximal	TPA + Merci®	TICI 3	mRS 2	[10]
6	Papillary fibroelastoma	Unknown	62/ M	24	Left M1	TPA + Solitaire®	TICI 2b	NIHSS 10, mRS 3	[11]
7	Unclassified sarcoma	Known	55/ M	22	Right distal ICA	Penumbra®	TICI 3	Improved (details unknown)	[12]
8	Myxoma	Unknown	4/ M	16	Left M1	TPA+Solitaire®	TICI 3	Only mild weakness	[13]
9	Myxoma	Unknown	70/ M	11	Left carotid T, M1,M2	TPA + Penumbra® + Trevo®	TICI 2b	NIHSS 1, mRS 2	[14]
10	Papillary fibroelastoma	Unknown	75/ M	18	Left M1	Penumbra®	TICI 3	Improved (details unknown)	[15]
Non-cardiac tumor									
11	Breast tumor	Known	62/ F	19	Left PCoA, M1, A1	Merci®	No flow through the MCA	Motor aphasia, right hemiplegia	[4]
12	Melanoma	Known	22/ F	4	Left MCA	Forced suction thrombectomy	TICI 3	NIHSS 1	[5]
13	Lung adenocarcinoma	Known	69/ F	16	Right M1	Mechanical clot retrieval	TICI 2b or 3	Death	[6]
14	Undifferentiated sarcoma	Unknown	37/ M	13	Right proximal ICA	TPA + Trevo®	TICI 2b	NIHSS 4, mRS 2	Present case

F: female, ICA: internal carotid artery, NA: not available, NIHSS: National Institute of Health Stroke Scale, M: male, MCA: middle cerebral artery, mRS: modified Rankin Scale, PCoA: posterior communicating artery, TICI: Thrombolysis in Cerebral Infarction, TPA: tissue plasminogen activator

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