## **ARTICLE IN PRESS**

### Case Studies

### Effective ADAPT Thrombectomy in a Patient with Acute Stroke due to Cardiac Papillary Elastofibroma: Histological Thrombus Confirmation

Francesco Biraschi, MD,\* Francesco Diana, MD,† Francesco Alesini, MD,‡ Giulio Guidetti, MD,\* and Simone Peschillo, MD, PhD\*

> A 75-year-old man with hypertension and atrial fibrillation was admitted to our emergency room with right-sided hemiplegia and complete aphasia (National Institutes of Health Stroke Scale [NIHSS] score = 18). A noncontrast computed tomography scan showed a slight hypodensity in the left insular region and a bright hyperdense sign in the M1 tract of the left middle cerebral artery (MCA). Angio-CT confirmed an occlusion of the M1 tract of the MCA. Magnetic resonance diffusion-weighted imaging/perfusion-weighted imaging was obtained and revealed a mismatch in the left parietal cortical region. Complete revascularization was achieved by thromboaspiration with the A Direct ASPIRATION first PASS TECHNIQUE (ADAPT) technique. Histological examination of the embolic material revealed its nonthrombotic nature: cardiac embolic papillary elastofibroma (PEF). At discharge, good recovery of right-side hemiplegia was observed. This case report is the second in literature in which a histological confirmed cardiac embolic PEF is reported as a cause of embolic stroke. PEF is a rare but potentially treatable cause of embolic stroke. Understanding the nature of the embolic material would help in choosing the best revascularization approach. Key Words: Stroke-mechanical thrombectomy-papillary elastofibroma-cardiac embolism-MR perfusion—ADAPT—thrombus.

> © 2016 National Stroke Association. Published by Elsevier Inc. All rights reserved.

From the \*Department of Neurology and Psychiatry, Endovascular Neurosurgery/Interventional Neuroradiology, Sapienza University of Rome, Rome, Italy; †Department of Radiology, Sapienza University of Rome, Rome, Italy; and ‡Department of Oncological, Radiological and Anatomopathological Sciences, Sapienza University of Rome, Rome, Italy.

Received June 23, 2016; revision received July 12, 2016; accepted July 17, 2016.

Address correspondence to Francesco Biraschi, MD, Department of Neurology and Psychiatry, Endovascular Neurosurgery/ Interventional Neuroradiology, Sapienza University of Rome, Policlinico Umberto I, Viale del Policlinico 155, Rome 00100, Italy. E-mail: f.biraschi@gmail.com.

1052-3057/\$ - see front matter

### **Case Presentation**

A 75-year-old man with hypertension and atrial fibrillation was admitted to our emergency room with rightsided hemiplegia and complete aphasia (National Institutes of Health Stroke Scale [NIHSS] score = 18). The patient was found on the ground by the relatives early in the morning. The patient was last seen well on the night before he was admitted to the hospital. The patient was under a well conducted anticoagulation therapy with apixaban. At the emergency room, the noncontrast computed tomography scan (Fig 1, A) showed a slight hypodensity in the left insular region and a bright hyperdense sign on the M1 tract of the left middle cerebral artery (MCA) (Fig 1, B). Angio-CT confirmed an occlusion of the M1 tract in the left MCA. No intravenous (IV) recombinant

 $<sup>\</sup>ensuremath{\textcircled{}}$  © 2016 National Stroke Association. Published by Elsevier Inc. All rights reserved.

http://dx.doi.org/10.1016/j.jstrokecerebrovasdis.2016.07.026

# **ARTICLE IN PRESS**

#### F. BIRASCHI ET AL.



**Figure 1.** Noncontrast computed tomography demonstrates a slight hypodensity in the left insular region (A) and a very bright hyperdense sign in the left middle cerebral artery (B). Angio-CT confirmed an occlusion of the left middle cerebral artery (C).

tissue plasminogen activator was performed due to the unknown onset time of symptoms. To evaluate the opportunity for a mechanical revascularization, a magnetic resonance imaging scan was obtained. Diffusion-weighted imaging sequences showed a hyperintensity in the left insular region while the mean transit time was increased in the left parietal cortical region, suggesting a mismatch in that region. No fluid-attenuated inversion recovery changes were observed (Fig 2). In the angiosuite, M1 occlusion was confirmed by the presence of collaterals coming from the left anterior cerebral artery and from temporal branches of the left MCA (Fig 3, A). Complete revascularization was achieved by thromboaspiration (NeuronMax, ACE64, 3MAX; Penumbra, Alameda, CA) (Fig 3, B) in 15 minutes from groin puncture with a thrombolysis in cerebral infarction score of 3. The material removed after the aspiration is shown in Figure 4. Macroscopic evaluation revealed a 1-cm white-pink hard



**Figure 2.** Magnetic resonance study with axial T2 fluid-attenuated inversion recovery (A), diffusion-weighted images (B), and mean transit time (C) showing a mismatch in the left posterior parietal region.



**Figure 3.** Conventional angiography. Anteroposterior and lateral views of the left internal carotid artery injection before (A) and after (B) treatment.

tissue-like material. On histological examination, the hematoxylin/eosin stained slide showed a solid amorphous eosinophilic tissue, in which there were some calcium depositions, intermingled with some peripheral blood cells, such as red cells and monocytes. At the edge of the lesion, there were some pseudopapillary (fingerlike) projections on the surface of which CD34 immunostain showed the presence of some endothelial cells and the Weigert–van Gieson histochemical stain revealed some elastic fibers. No epithelial cells were found (immunohistochemistry for cytokeratin AE1/AE3 was negative).



**Figure 4.** (A) Macroscopic view of the embolic material aspirated. (B) Histological examination: left: pseudopapillary fronds with calcific axis that are considered to be the plate of the papillary fibroelastoma from which the thrombus has been detached; middle: at the edge of papillary fronds there were some elastic fibers retine (arrow—Veigert Van Gieson stain, 10×); right: pseudopapillary (fingerlike) projections lined by some endothelial cells (CD34 immunostain, 10×).

### ADAPT THROMBECTOMY IN A PATIENT WITH ACUTE STROKE

This complex of morphological, histochemical, and immunohistochemical data referred to a thrombus characterized by papillary fronds lined by the endothelium and elastic fibers that are considered to be the plate of the ultrasound referred papillary fibroelastoma from which the thrombus has been detached.

#### Discussion

To our knowledge, this is the second case in literature with histological confirmation of an embolic papillary elastofibroma (PEF) causing an ischemic stroke treated with mechanical thrombectomy.<sup>1</sup> Histologically confirmed tumor fragments have been isolated from the embolic material from coronary, pulmonary, and peripheral arteries, but not from cerebral embolism.<sup>2</sup> Five cases are reported in literature in which PEF is supposed to be the stroke etiology treated with both IV<sup>3,4</sup> and intraarterial recombinant tissue plasminogen activator,<sup>5,6</sup> and 2 of these cases are associated with mechanical disruption,<sup>67</sup> but none of the 5 cases has the histological confirmation of the embolic material. In these cases, an indirect diagnosis of the embolic nature of the thombus was made by histological analysis after cardiac PEF removal. It is long debated about the nature of the embolic material in these patients. Embolization may occur from the fragile papillary fronds of the tumor itself or from a thrombus formed within the tumor context. Our histological examination suggests that the nature of the emboli was a wide calcified mass surrounded by fibrin with a papillary base of implant. Thus, we can affirm with certainty that the embolization material was represented by a calcified mass that has come off the PEF and occluded the M1 segment of the left MCA. The very calcific nature of the thrombi was the reason why oral anticoagulant therapy was not useful in preventing cardioembolism. The nature of the embolic material has a relevant impact on therapeutic strategies to achieve recanalization. When embolization is from the thrombotic material surrounding the mass, treatment with IV alteplase could be useful<sup>3,4</sup>; on the other hand, when the embolizing material is a fragment from the tumor, fibrinolytic therapy is probably not useful. In these cases, mechanical recanalization could be the best effective treatment option for PEF embolism.<sup>7</sup> In our case, thromboaspiration has been proven to be feasible and efficient in restoring flow after a complete M1 occlusion. In our opinion thromboaspiration should be the first-line choice compared to other mechanical techniques when hard-calcified tissue fragments characterize the embolic material. Theoretically, indeed, it seems to

be difficult to pass through a hard tissue-like material coming from the tumor mass or a calcification, open a stent-retriever, and remove the thrombus. Currently, there are no tests that could predict the nature, either thrombotic or tumoral, of the embolic materials in patients with PEF. In our case, noncontrast computed tomography provided helpful information showing a very bright hyperdense sign in the MCA (156 Hounsfield units [HU]) that suggested the very calcific nature of the thrombus, then confirmed by histological examination. A recent systematic review, which correlates the imaging and histopathology of thrombi in acute ischemic stroke with etiology and outcome, highlights how patients with favorable angiographic outcome had a significantly higher mean clot HU than those with a poor angiographic outcome.8 These results suggest how understanding the nature of the embolic material dramatically would help in achieving recanalization. Thus, we suggest a routine histological examination of all clots removed during thrombectomy procedures to obtain a correlation between neuroimages, histological analysis, and patient outcome.

### References

- 1. Tejada J, Galiana A, Balboa Ó, et al. Mechanical endovascular procedure for the treatment of acute ischemic stroke caused by total detachment of a papillary fibroelastoma. J Neurointerv Surg 2014;6:e37.
- Gowda RM, Khan IA, Nair CK, et al. Cardiac papillary fibroelastoma: a comprehensive analysis of 725 cases. Am Heart J 2003;146:404-410.
- 3. Matijevic V, Poljakovic Z, Ilic I, et al. Cardiac papillary fibroelastoma: source of cerebral embolism treated with intravenous thrombolysis. J Stroke Cerebrovasc Dis 2011;20:485-487.
- Corrado G, Panisi P, Checcarelli N, et al. An unusual cause of ischemic stroke with successful thrombolysis. J Stroke Cerebrovasc Dis 2013;22:e691-e692.
- Palazzuoli A, Bruni F, Pasqui A, et al. Cardiac papillary fibroelastoma: an unusual source of cerebral embolism solved with local fibrinolysis. Neurol Sci 2002;22:469-472.
- Ljevak J, Mišmaš A, Bazina A, et al. An infrequent type of stroke with an unusual cause and successful therapy: basilar artery occlusion caused by a cardiac papillary fibroelastoma recanalized 12 hours after onset. Intern Med 2013;52:277-279.
- Santos AF, Pinto J, Raos V, et al. Stroke and cardiac elastofibroma: mechanical thrombectomy after thrombolytic therapy. J Stroke Cerebrovasc Dis 2014;23:1262-1264.
- Brinjikji W, Duffy S, Burrows A, et al. Correlation imaging and histopathology of thrombi in acute ischemic stroke with etiology and outcome: a systematic review. J Neurointerv Surg 2016;doi:10.1136/neurintsurg-2016-012391; [Epub 10 May 2016].