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Y Numaguchi, R T Higashida, J M Abernathy and J C Pisarello

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Correspondence

Abbreviated Reports

Balloon Embolization in a Carotid-Cavernous Fistula in Fibromuscular Dysplasia

Fibromuscular dysplasia (FMD) of the cervicocephalic arteries is a well-known entity [1–4]. The association of FMD with aneurysms or dissection of intracranial or extracranial arteries is well documented; however, association of FMD with a carotid-cavernous fistula is rare. We describe a patient with FMD of both internal carotid arteries associated with a unilateral direct carotid-cavernous fistula that was treated successfully with detachable balloons.

Case Report

A 69-year-old woman presented with a history of 6 weeks of left-sided headaches. Two weeks before admission she developed redness in her left eye, which progressed in intensity and severity and resulted in total blindness 3 days before admission. Bilateral common carotid arteriography demonstrated a direct carotid-cavernous fistula on the left. The characteristic "string of beads" appearance representing FMD was also noted in the cervical portion of the internal carotid artery bilaterally (Figs. 1 and 2). Vertebral arteriography, with transient compression of the left common carotid artery and rapid filming, showed that the fistula was filled via a small entry site from the proximal portion of the cavernous segment of the internal carotid artery (Fig. 3). A Hieshima [4] detachable silicone balloon, 1.4 × 4.0 mm uninflated, was then flow-directed into the cavernous segment of the carotid artery. However, selective placement of the balloon into the cavernous sinus was not successful because of the small size of the fistula. Therefore, test occlusion of the internal carotid artery was performed, followed by an intra-arterial trapping procedure with two detachable balloons placed proximal and distal to the fistula. An immediate postembolization left common carotid angiogram showed complete occlusion of the internal carotid artery and a left vertebral angiogram showed that the fistula could no longer be seen (Fig. 4). The patient has done well in the 2 months since the procedure, with marked improvement in her proptosis and chemosis involving the left eye.

Discussion

Angiographic, clinical, and genetic features of FMD involving the cervicocephalic arteries were fully described by Mettinger and Ericson [2]. According to them, more than 400 such cases of FMD were reported, frequently in association with intracranial aneurysms (ranging from 39–51%); however, the association of cervicocephalic FMD with fistulae or arteriovenous malformations was relatively rare. Furthermore, only six patients with FMD reportedly had a carotid-cavernous fistula [1, 3–5]. The cause of the carotid-cavernous fistulae in those patients was most likely associated with rupture of an aneurysm or a dissection of the carotid artery [3].

Ideally, the fistula should be occluded by using single or multiple balloons with preservation of the arterial flow [4–6]. This can be especially difficult or even impossible when the fistula is small (as in our patient). In such instances, the intra-arterial trapping technique with detachable balloons can be rationalized, if the patient can tolerate carotid occlusion. However, it is ideal to save the carotid artery since these fistulae in FMD may be bilateral. Either a direct surgical approach or a retrograde venous approach to the cavernous sinus may be considered as alternative treatments [4]. The latter approach was not attempted in our patient because the inferior petrosal sinus was very small.

Little information has been documented regarding fistula size in patients with FMD and its successful treatment [3–5]. Hieshima et al. [4] recently reported four cases of FMD; two involving spontaneous carotid-cavernous fistulae and two involving vertebral arteriovenous fistulae. They pointed out the technical difficulty in treating fistulae associated with FMD because of the increased incidence of stenotic segments of the involved arteries and the small size of these fistulae orifices. We encountered similar technical difficulty during embolization of our patient. The fistulae arising with FMD may indeed be smaller in comparison with the direct traumatic carotid-cavernous fistulae that occur in most cases, making them technically more difficult to treat.

Yuji Numaguchi
Tulane University Medical Center
New Orleans, LA 70112
Randall T. Higashida
University of California School of Medicine
San Francisco, CA 94143
John M. Abernathy
J. Carlos Pisarello
East Jefferson General Hospital
New Orleans, LA 70011

Fig. 1.—Right common carotid angiogram, lateral projection. Characteristic "string of beads" appearance in internal carotid artery is consistent with fibromuscular dysplasia (*bracket*).

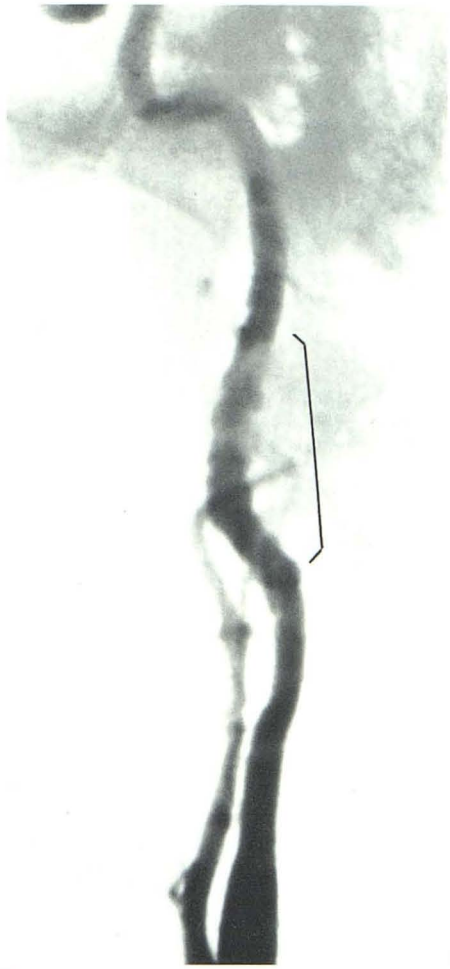

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Fig. 2.—Left common carotid angiogram, lateral projection. Fibromuscular dysplasia is also noted in proximal internal carotid artery (*bracket*). There is a direct carotid-cavernous fistula, and ophthalmic veins are filled (*long arrow*). Exact fistula site is undetermined, although carotid artery is slightly tented at point of fistula (*short arrow*).

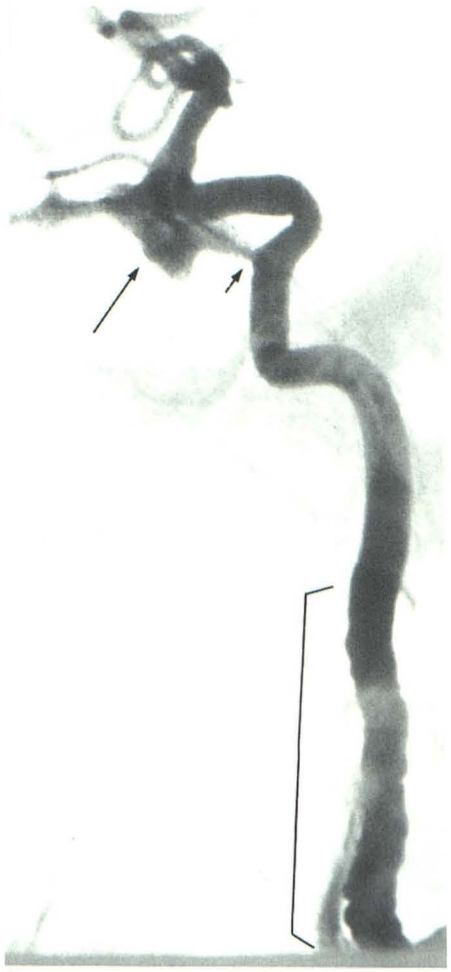

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Fig. 3.—Left lateral vertebral angiogram with carotid compression clearly demonstrates tenting of carotid artery indicating orifice of fistula (*arrow*). Note good collaterals to left intracranial circulation.

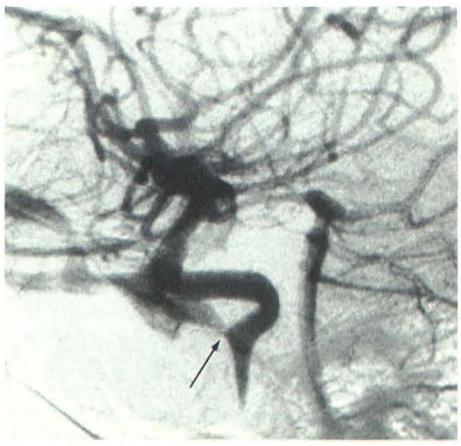

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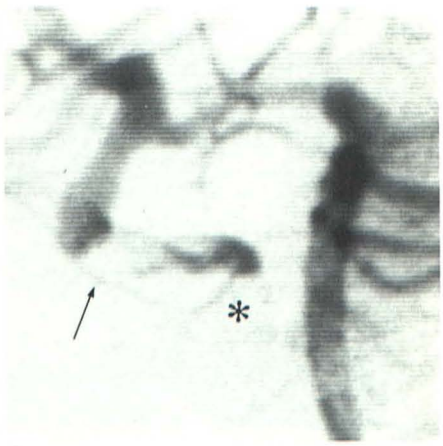
Fig. 4.—Immediate postembolization left vertebral digital arteriogram, lateral projection, shows that fistula is no longer seen. *Arrow* indicates filling defect caused by balloon placed in carotid siphon. *Asterisk* indicates site of second balloon in carotid artery.



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MR in Diffuse Angiomatosis

Angiomatous malformations of the CNS have been classified as capillary telangiectasis and as venous, arterial, and cavernous angiomas [1]. In this instance, a 37-year-old man presented with leg weakness and slight mental deterioration over a 3-year-period. Angiography showed large vessels and diffuse angiomatosis that seemed to involve every part of the brain except the cerebellum (Fig. 1). MR revealed large arteries, veins, and sinuses, suggesting an underlying increase in the capillary bed (Fig. 2). The cerebellum was clearly not involved. Our working diagnosis is that of capillary telangiectasis on the basis of the angiographic and MR findings. A direct biopsy was not performed since it entailed serious risk to the patient. Biopsy of a branch of the external carotid artery proved inconclusive.

Although our diagnosis is radiologic, considering the unusual radiologic features, we felt there was a need to document this case. We would welcome comments from the readers of *AJNR* on the frequency of such diffuse vascular lesions [2–4].

V. Wagle
I. McCutcheon
D. Melanson
R. Ethier
L. E. Roy
Montreal Neurological Hospital and Institute
Montreal, QC, Canada HIT 2H1

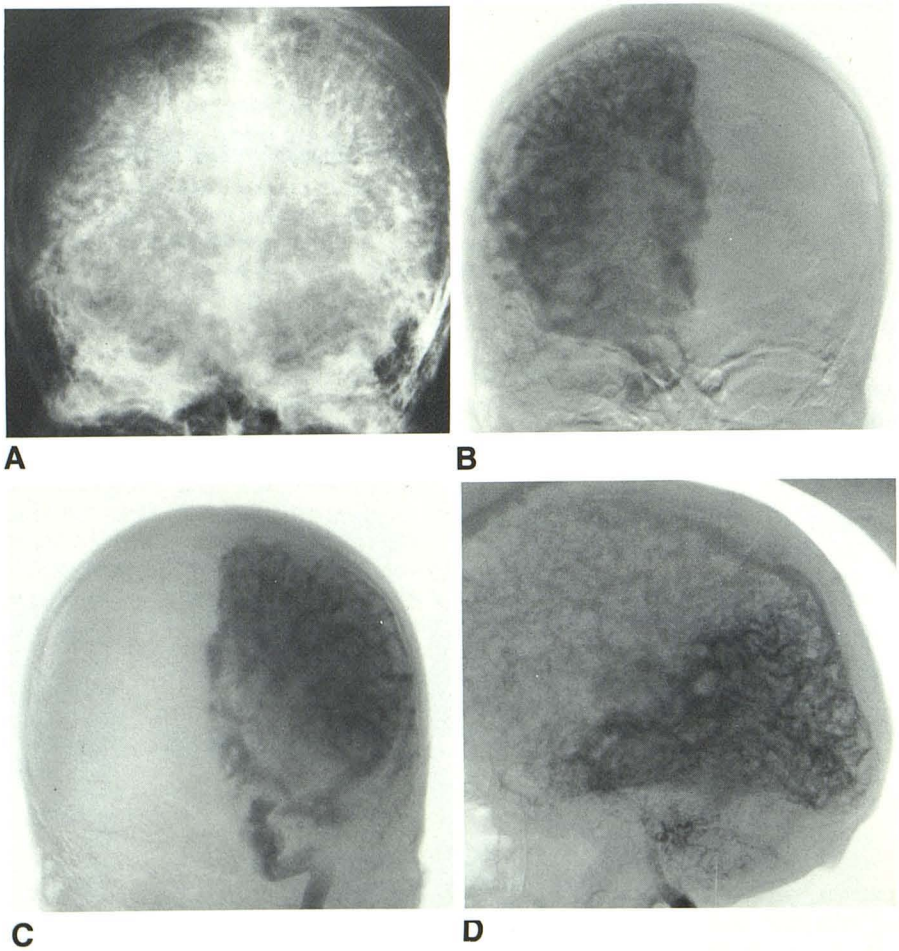

Fig. 1.—Arch and selective angiography showing extensive angiomatosis. Arch study (*A*), right carotid (*B*), left carotid (*C*), vertebral (*D*). Cerebellum is the only spared portion.