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Muslin-induced intracranial vasculopathic stenosis: A report of two cases

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Abstract

Muslin wrapping is a commonly utilized alternative technique in the treatment of aneurysms that are not amenable to direct clipping. In this case report, we describe two patients from different institutions who both required aneurysm wrapping with gauze/muslin for aneurysm reinforcement. Both patients developed an inflammatory foreign body response to muslin visible on MRI that resulted in a vasculitic stenosis. The onset of TIAs was at 6 months and 1 month postoperatively, respectively. The stenoses rapidly progressed to near occlusion despite antiplatelet therapy, and in one case, an aggressive corticosteroid regimen. One patient eventually developed leptomeningeal collateral flow that allowed tolerance of the stenosis, while the other patient required microsurgical bypass. These cases reports are the first to our knowledge that describe the adverse effects of muslin wrapping without adhesive reinforcement, as well as one of few reports to include follow-up angiographic imaging.

Keywords

Muslin; Gauzoma; Intracranial stenosis; Vasculitis; MRI; Aneurysm

1. Introduction

Muslin wrapping is commonly utilized as an alternative technique in the treatment of aneurysms that are not amenable to direct clipping, particularly in either very small, “blister-like” aneurysms or large fusiform aneurysms. Interestingly, foreign-body inflammatory responses related to muslin have been reported, with the majority of cases resulting in mass-like granulomas and compression of the optic chiasm with loss of visual acuity [1–3]. At least three case studies have also described vascular stenosis and cerebral ischemia as an unfortunate variant manifestation [4–6]. Complicating the analysis of this pathophysiology is the common, but not universal technique of adjunctive gauze reinforcement with bioadhesive agents, such as Biobond. Some of these adhesives have been shown to cause

direct neurotoxic and inflammatory effects [7–9]. All of the three aforementioned case reports included gauze reinforcement with adhesive, and only Kawamura included radiographic follow-up. We describe two cases of delayed progressive arterial stenosis after gauze wrapping without adhesive reinforcement.

2. Case 1

A 44-year-old African-American woman presented in May 2008 with sudden onset of headache and Hunt and Hess grade 1 subarachnoid hemorrhage. Cerebral angiography demonstrated a 2-mm left internal carotid artery (ICA) aneurysm in the superior hypophyseal region (Fig. 1). During craniotomy, a small 2-mm blister aneurysm was exposed that was deemed too small for microsurgical clipping. The aneurysm was therefore wrapped with muslin gauze. The patient was discharged to home on postoperative day 6 without neurologic deficit.

Six months later, the patient began experiencing recurring headaches and transient ischemic attacks (TIAs), manifesting as right arm and hand paresthesias. A full imaging workup ensued, including CT angiography (CTA), MRI of the brain, and ultimately cerebral angiography. An MRI imaging protocol tailored for cerebral vasculitis was formulated, consisting of thinly acquired, magnified views of the distal ICA and Circle of Willis region in multiple planes after administration of intravenous contrast. This scan demonstrated vessel wall thickening and enhancement of the ICA extending from the posterior communicating artery (PCOM) region to the anatomic terminus, including the proximal A1 segment of the anterior cerebral artery (ACA) and proximal M1 segment of the middle cerebral artery (MCA) (Fig. 2). Cerebral angiography confirmed corresponding moderate luminal stenoses in these areas, with resultant partial flow obstruction in the left MCA and no antegrade flow in the left ACA (Fig. 3). The imaging findings corresponded to the area including, and distal to, the area of muslin wrapping.

Rheumatologic laboratory testing was notable for an elevated C-reactive protein (CRP) level of 4.9 mg/dL and elevated erythrocyte sedimentation rate (ESR) of 55 mm/h. The patient had a positive ANA antibody at a titer of 1:1280 with strongly positive SS-A and SS-B antibodies. CSF analysis ruled out meningitis. Therefore, a diagnosis of local muslin-induced foreign body vasculitic reaction was made. The patient was treated with an inpatient course of methylprednisolone, and later discharged on oral prednisone, methotrexate and clopidogrel.

Three weeks later, the patient again returned with multiple episodes of slurred speech over 3 days. Cerebral angiography showed progression of the left terminal ICA stenosis (not shown), with interval enlargement of leptomeningeal collateral vessels. A follow up MRI showed no change in ICA vessel wall thickening and enhancement. Laboratory tests demonstrated normalization of the previously elevated inflammatory markers. A second short inpatient course of intravenous SoluMedrol was begun, and the clopidogrel dosage was increased to 150 mg daily. Medication regimens were altered to allow permissive hypertension. Angioplasty and stenting was deferred due to concerns for rupture with stressing of an inflammatory vessel. Revascularization surgery was declined by the patient.

Routine follow-up imaging was performed 5 months after the second presentation (approximately 1 year after the original surgery). Cerebral angiography (Fig. 4) demonstrated severe, near-occlusive stenosis of the left terminal ICA with further leptomeningeal collateral vessel enlargement to the suprasylvian MCA territory. The MRI brain appearance was again unchanged. A slow taper of oral prednisone and methotrexate was continued commenced. At most recent follow up, now 2 years after clinical onset and

2.5 years post-operatively, the patient continues to be without neurologic deficit or TIA. A follow up CTA of the brain demonstrated slight improvement in the left ICA stenosis.

3. Case 2

A 31-year-old woman presented with Hunt and Hess grade 2 subarachnoid hemorrhage. Cerebral angiography demonstrated a blister aneurysm arising medially from the left ICA, near the region of the left superior hypophyseal artery (Fig. 5). During craniotomy, the aneurysm was not amenable to clipping, and was instead wrapped with muslin gauze. Intraoperative microvascular doppler confirmed preservation of normal flow in the ICA. She was discharged 14 days later, neurologically intact.

The patient returned 1 month later with transient episodes of word-finding difficulty. Cerebral angiography demonstrated an 85% stenosis of the left ICA, with occlusion of the A1 segment of the left ACA and mild narrowing of the proximal M1 segment of the MCA (Fig. 6). MRI of the brain demonstrated an enhancing mass around the muslin-wrapped artery, extending to the distal ICA bifurcation, consistent with granuloma. Serum CRP and CSF laboratory values were normal and without evidence of infection.

Angioplasty with or without stenting was deferred because of concerns for vessel rupture. The patient underwent occipital artery to MCA bypass with saphenous interposition grafting. Treatment with aspirin and heparin was started postoperatively and continued upon discharge. At the most recent follow-up examination, the patient had full bilateral motor strength and mild dysphasia. A follow up CT angiogram demonstrated stable carotid artery stenosis with patency of the saphenous bypass graft (not shown).

4. Discussion

In this communication, we report two cases of a delayed, post-operative stenosis after muslin wrapping of intracranial aneurysms. As in similarly reported cases, history and workup strongly suggest a foreign-body response to muslin. In both cases, the stenotic segments corresponded directly to the area of muslin wrapping. MRI studies clearly demonstrated perivascular enhancement and inflammation. In contrast, a generalized cerebral vasculitis usually involves multiple lesions in varying distribution throughout the cerebral vasculature. The timing of onset of both cases was within months, not days, of the muslin application; a timeline suggestive of a chronic inflammatory response. The onset was slower than would be expected of an infection, and yet faster than would be expected for an atherosclerotic process. Finally, most vasculitides would be expected to show angiographic improvement over time with steroid treatment. This lack of resolution supports the concept of the muslin mesh as a persistent inflammatory irritant.

In addition to providing physical tamponade in the event of aneurysm rupture, muslin and other cotton-based gauze wrap substances have also been found to promote a mild chronic inflammatory response. It is this resultant layer of fibrotic perivascular tissue which, in theory, thickens and strengthens the aneurysm sac [10]. Presumably, it is a rare, excessive variant response that results in the phenomena described in these case reports.

Complicating the understanding of this pathology is the common technique of gauze saturation with reinforcing agents such as cyanoacrylate or fibrin glue. These adhesives have been shown in experimental and clinical studies to have a direct vascular and neurologic toxicity [6,8,9,11]. Herrera showed in rats that cyanoacrylate (Biobond) glues, fibrin glues, and untreated pure cotton gauze (Bemsheets) can each have direct inflammatory effects, with cyanoacrylates causing the most vigorous response. Cotton-gauze elicited a chronic granulomatous inflammatory response, eventually resulting in adventitial replacement with

fibrotic tissue. It was also suggested that wrapping restricts the natural dispersion of the glue agent in the subarachnoid space and regional tissues, resulting in a retained high local concentration that causes a greater cumulative effect than would be expected for the agents individually. These investigators postulated that the majority of the biological response observed likely resulted from the adjunctive agent. However, our case study is the first to our knowledge to demonstrate that muslin alone can result in a clinically significant response in humans.

The natural history of this granulomatous reaction to muslin is poorly understood. Perianeurysmal granuloma formation with optic neuropathy is the best described, and likely, most common response. Bhatti et al. reviewed 23 reported cases of muslin-induced granulomatous disease, including two personally reported [1]. These all manifested as optic neuropathy without clinically significant vascular stenosis. The onset of clinical presentation was broad and ranged between 1 and 54 months postoperatively. Although the patients in their series were treated with corticosteroids, there was no conclusive evidence that pharmacological therapy provided any benefit, as some patients were found to improve spontaneously.

Including this report, there are now four case series describing vascular stenosis as a variant presentation of this phenomenon [4–6]. Yoon described three patients presenting with artery narrowing between 2 and 16 months post-operatively. Of these, one presented with headaches, one experienced sudden decreased vision, and another experienced sudden right-side weakness. Angiography showed severe stenosis of the wrapped aneurysm in two patients, and in the third patient, delayed segmental MCA narrowing resulting in infarction in the upper basal ganglia. The first two patients underwent conservative treatment with steroid therapy and fully recovered within a few months. The third patient suffered from recurrence and cerebral infarction despite treatment with dexamethasone, clopidogrel, and heparin.

Kurita described two patients presenting at 3 and 6 months post-operatively. The first patient presented with left hemi-paresis, dysarthria, and dysphagia. Angiography showed severe stenosis of the inferior trunk of the right MCA, precisely where wrapping had occurred. The second patient presented with infarction in the right basal ganglia. Angiography showed stenosis of the superior trunk of the right MCA. Details regarding treatment and follow-up were not included.

Of these case reports, only Kawamura included follow-up imaging beyond the initial presentation of complication. Kawamura described seven patients with post-operative stenosis, with five patients presenting with TIAs or stroke. Of these seven patients, three presented with arterial occlusion, with one of these patients having follow up angiography showing no change. The other four patients presented with non-occlusive stenosis, only two of which presented with ischemic symptoms. Of these four patients, three patients demonstrated improvement in their stenosis at between 6 and 12 months, while the last patient showed an unchanged stenosis at 2 years. Medical management in this series included antiplatelets, anticoagulants, low-molecular dextran, dobutamine, steroid, and thromboxane synthetase inhibitors.

In our patients treated without adhesive reinforcement, patient #1 presented with TIAs and a progressive distal ICA stenosis. This stenosis progressed to near occlusion after 5 months, despite medical management consisting of antiplatelets, prednisone, and methotrexate. However, she had no further neurologic sequelae. Patient #2 presented with TIAs and distal ICA occlusion after 1 month, which then prompted bypass surgery. CTA follow up at 6 months showed no change in the ICA stenosis and patency of the graft.

Because these case series are small, it is difficult to identify definitive trends regarding the natural history of the pathological process. However, the initial theory that the majority of the inflammatory effect emanates from the adhesive could be inaccurate. We believe that the direct foreign-body response to the cotton fibers is underestimated. Both of our patients treated by gauze wrapping without stenosis presented with a progressive severe arterial stenosis. Whereas three of seven patients in Kawamuras series treated with adhesive actually showed some angiographic improvement on follow-up.

In regards to clinical course and management, it seems that a fraction of the patients will present very early in follow-up, within 1–2 months, with occlusion or rapidly progressive stenosis that requires immediate treatment. Others will have a longer, more protracted course, allowing for medical intervention. Clinical cerebrovascular insufficiency is dependent on two competing factors: the rate of progression of stenosis versus the rate of leptomeningeal collateral formation. In our first case, although the stenosis was progressive, medical management may have delayed the process and prevented thromboembolic complications long enough for leptomeningeal collaterals to form and prevent cerebrovascular insufficiency.

However, in those patients who do not adequately compensate, intervention will be required. Whether stenotic lesions in this setting are amenable to angioplasty and stenting is of some debate. In our first case, after normalization of the inflammatory markers with corticosteroids, we considered whether the inflammatory process had subsided. We initially hoped that a scar-like process was causing a progressive contractile-type vessel stenosis. Such a firmer fibrous tissue might be better able to withstand the transluminal pressure inherent in angioplasty. This hypothesis was at least partially supported by descriptions of operative re-explorations in both the literature [12] and in personal communications with colleagues. However, continuing MRI follow-up demonstrated persistent vessel wall thickening and enhancement. Therefore, fragile, acute inflammatory adventitial tissue could still be present. Furthermore, the stimulus of angioplasty and a persistent foreign body, such as a stent, could theoretically prolong and/or accelerate the inflammatory reaction. Microvascular bypass, as performed in our second case, has the advantage of vascular manipulation distant from the inflammatory site and may be safer.

These cases raise the question as to whether better, more inert materials are available for aneurysm wrapping. Some investigators have utilized woven Dacron-based materials as constructs for thoracoabdominal aneurysms wrapping, vascular conduits, patch grafts, and components for covered stents with good efficacy [13–17]. Accrual experiments and clinical trials are needed to find a suitable replacement for cotton-based materials, which have a well-recognized tendency to cause intracranial granulomatous reactions [18–21].

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Fig. 1. Case 1. Preoperative cerebral angiogram showing the aneurysm in the left ICA. (a) Oblique view, showing aneurysm. (b) Conventional anteroposterior (AP) view, showing original caliber of the ICA terminus. Note the accessory left MCA off the ICA, supplying the posterior left temporal lobe, an anatomic variant.

Fig. 2.
Case 1. Coronal T1-weighted and MR imaging after contrast enhancement showing marked left supraclinoid ICA thickening and enhancement (arrows).

Fig. 3.

Case 1. Left ICA angiograms obtained at 5-month follow-up examination showing left ICA terminus stenosis. AP view. Compare to original preoperative angiogram (Fig. 1b). Note the lack of antegrade flow in the left ACA, as well as the stenosis at the ICA terminus/proximal MCA region (arrow).

Fig. 4.

Case 1. Cerebral angiogram obtained at 9-month follow-up. (a) AP view of the ipsilateral left ICA. Note progression of the terminal ICA stenosis to occlusion (arrow). (b) Anteroposterior view of the contralateral right ICA. Note retrograde opacification of the left A1 stenosis (arrow) and extensive leptomeningeal collateral formation.

Fig. 5.
Case 2. Preoperative anteroposterior cerebral angiogram showing the left ICA aneurysm before wrapping (arrow). Note the baseline vessel luminal caliber.

Fig. 6. Case 2. One-month follow-up: (a) lateral left ICA cerebral angiogram. Note the significant ICA stenosis (arrow) and lack of left ACA filling. (b) Angiogram of the contralateral right ICA. Note retrograde opacification of the left carotid terminus stenosis (arrow).