CASE REPORT

Case Report: Late Sequela of a Muslinoma Involving the Optic Chiasm

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ABSTRACT

An 84-year-old woman presented with 3 months of vertical binocular diplopia and difficulty reading at near. She had a history of bilateral ophthalmic artery aneurysm repair involving use of muslin in the 1990s. The patient then developed bitemporal hemianopsia secondary to muslin-induced inflammation (“muslinoma”) extending to the optic chiasm, which required surgical decompression. She had a persistent bitemporal hemianopsia but was stable for two decades after surgery. In 2017, the patient re-presented with double vision. Exam showed a non-paretic diplopia due to a small angle comitant right hypertropia attributed to the retinal hemi-field slide. Repeat imaging showed no new aneurysm or recurrent muslinoma. This case had originally been reported two decades ago and represents the longest duration of recurrent symptoms from muslin-related optochiasmatic arachnoiditis in the English language ophthalmic literature. Clinicians should be aware of the potential of delayed and recurrent symptoms or signs years or even decades after muslin wrapping of aneurysms.

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Introduction

Muslin gauze has historically been used to wrap and repair intracranial aneurysms. Use of muslin in this context is to induce limited local inflammatory fibrosis to strengthen the walls, neck or base of aneurysms not amenable to definitive surgical clipping. In rare cases, patients have developed vigorous granulomatous inflammation extending into adjacent brain parenchyma with or without mass effect. Multiple reports in the literature have described the development of mass lesions affecting the optic chiasm, which have variously been described as “muslinoma”, “gauzoma”, or “optochiasmatic arachnoiditis”. We describe a patient who was originally reported two decades ago and returned with non-paretic diplopia due to the retinal hemi-field slide phenomenon. To our knowledge, this case is the longest duration between treatment of a muslinoma and recurrent visual symptoms in the English language ophthalmic literature.

Case report

A 58-year-old woman developed acute onset of severe headache, accompanied by nausea, vomiting, and nuchal rigidity in 1991. A computed tomography (CT) scan of the head revealed sub-arachnoid haemorrhage. Cerebral angiography showed a large right ophthalmic artery aneurysm. She underwent aneurysm clipping, and the neck and base of the aneurysm were wrapped with muslin, which was secured with gelfoam pledgets. In 1993, a new bitemporal field defect was noted. Imaging revealed a faintly enhancing suprasellar mass. Excisional biopsy of the mass revealed acute and chronic inflammatory cells, multinucleated foreign body giant cells, and birefringent filamentous material consistent with muslin gauze. The diagnosis of optochiasmatic arachnoiditis induced by muslin (“muslinoma”) was made.

In 1994, the patient reported progressive asymmetric bilateral loss of vision (left greater than right). A small but compressive left ophthalmic artery aneurysm was discovered and clipped

(without use of muslin). She was taken to surgery and a dense, adhesive scarring of the chiasm was noted at that time. In 1995, the patient reported worsening vision of her right eye. She was taken to surgery to remove as much muslin from the right ophthalmic artery and optochiasmatic region as possible. The surgeon noted bony overgrowth at the previous site of aneurysm clipping, along with deformation and compression of the right optic nerve/chiasm. This case was subsequently reported in this journal in 1997. The patient's vision stabilized and, over the next four years, she did not present with new visual complaints.

In 1999, on routine follow-up, the patient was noted to have a trace right relative afferent pupillary defect (RAPD). Visual field testing at this time revealed bitemporal hemianopsia superimposed on a bilateral superior altitudinal defect, with bilateral asymmetric constriction (right greater than left). Her vision remained stable for the next several years. In 2006, on routine follow-up, Humphrey visual field (HVF) testing showed little change from previously noted defects, with mean deviations (MD) of -24.31 OD and -22.85 OS (Figure 1). The patient did not complain of visual changes for the next 11 years.

In total, the patient was stable for 20 years, but in December 2017, the patient (now 84 years old) presented with a 3-month history of new onset vertical binocular diplopia and difficulty reading at near. On exam, her visual acuity was 20/50 OD and 20/30 OS. Colour vision was 9/14 OD and 7/14 OS. Pupils measured 4 mm bilaterally and constricted poorly to 3.5 mm bilaterally with light. There was light near dissociation in both eyes. No RAPD was observed in either eye. Both eyes demonstrated full ductions and versions. There was a comitant right hypertropia of 2 prism diopters, which was variable at near, stable on horizontal gaze, and mildly worse on right head tilt. Intraocular pressures were 13 mm Hg OD and 15 mm Hg OS. Fundoscopic exam showed optic atrophy OU. HVF testing demonstrated minimal progression from 2006, with MDs of -26.51 OD and -23.03 OS (Figure 2). A CT head with and without contrast (Figure 3) revealed only stable chronic post-operative changes compared to a scan from 2000. No acute or abnormal enhancing lesions were noted. CT angiography (CTA) of the head with and without contrast (Figure 4) showed only bilateral clipped and obliterated aneurysms at the internal carotid artery-ophthalmic artery junctions. Tests for myasthenia gravis and thyroid eye disease were

Figure 1. Humphrey visual field analysis on routine follow-up in 2006. These fields demonstrate a bitemporal hemianopsia superimposed on a bilateral superior altitudinal defect, with bilateral asymmetric constriction (right greater than left). Mean deviations: -22.85 OS and -24.31 OD.

Figure 2. Humphrey visual field analysis in 2017. These demonstrate minimal field changes from 2006. Mean deviations: -23.03 OS and -26.51 OD.

Figure 3. CT head scans from 2000 (left) and 2017 (right). The 2017 scan revealed bilateral aneurysm clips (arrows), stable chronic post-operative changes, and no acute developments, as compared to the baseline scan in 2000. No acute or abnormal enhancing lesions were noted in the most recent scan.

negative. The diagnosis of a non-paretic diplopia due to bitemporal visual field loss causing retinal hemi-field slide was made.

Discussion

The use of muslin gauze in the surgical repair of intracranial aneurysms was popularized through

the 1950s–1970s.^{1–3} By the 1980s, the practice was considered routine by many neurovascular surgeons, and the associated vessel wall fibrosis at the site of aneurysmal repair was well known.^{4,5} Thus, the rationale for use of muslin in repair of aneurysms deemed unsuitable for clipping was to strengthen aneurysm walls by inducing limited inflammation and local fibrosis.^{5–7} However, in

Figure 4. CT head angiography in 2017 revealed the previous bilateral aneurysms at the internal carotid artery–ophthalmic artery junctions status-post clipping (arrows).

some patients with muslin-wrapped aneurysms, this inflammatory reaction had been rarely reported to extend beyond the aneurysm into adjacent brain tissue,^{7,8} forming granulomas (i.e., muslinoma, gauzoma, gauze granuloma), with or without mass effect.⁷ Many reported cases have involved muslin-wrapped aneurysms at or near the ophthalmic artery or optic chiasm, producing optic neuropathy, chiasmopathy, or inflammation of the surrounding meninges (i.e., optochiasmatic arachnoiditis).^{4,7,9–14} Among these reports was the original publication describing our patient at the time of her diagnosis with muslinoma/optiochiasmatic arachnoiditis in 1997.¹⁰

The pathophysiological mechanisms underlying muslinoma-induced optic neuropathy or chiasmopathy are unclear, though a combination of infiltrative, ischemic, and/or compressive damage to the optic nerve has been proposed.^{6,8,9,13} Interestingly, symptomatic muslinomas have more commonly been reported in females^{12,13} and are largely sterile.^{6,13} Due to the rarity of muslinoma formation, the pathophysiology, natural history, and management approach are poorly defined.^{6,7,15} Thus, the long-term outcome for visual function of these patients is difficult to predict.

In a recent small-scale retrospective study, the incidence of muslinomas or optochiasmatic arachnoiditis in patients in whom intracranial aneurysms were repaired with use of muslin was estimated to be 8.8% over a 3-year period ($n = 57$ patients).¹⁶ This estimation may be artificially low as a result of historically suboptimal imaging modalities,^{4,6,13} subclinical or absent symptoms,⁸ and significantly delayed course of symptom onset.^{6,7,11–13} To date, 37 cases of muslin-induced optic neuropathy have been reported in the literature, only four of which were reported in the last decade.¹⁷ Such case reports are likely to continue to wane, since the use of muslin in aneurysm repair has steadily declined with expansion of both endovascular treatments^{14,15,18,19} and of wrapping fibre materials that have not been associated with uncontrolled inflammation.^{20,21} The use of material wrapping, however, remains an important means by which to repair of aneurysms with challenging morphology, for example, blister-like aneurysms,^{20–23} and thus the theoretical risk of muslinoma-like reactions may persist.

Of the reported cases of muslinoma-induced optic neuropathy and long-term outcome studies of patients with muslin-wrapped aneurysms,^{2,3,18,19,24} none has addressed the potential of delayed visual sequelae. Our patient (originally reported in 1997) presented to the clinic 22 years after her last surgical intervention complaining of new onset double vision, which was determined to be a long-term sequela of a muslinoma. Of note, due to the undetermined compatibility of the patient's aneurysm clips with magnetic resonance imaging, CT was the preferred imaging modality to work up the patient's diplopia. Thus, because localized inflammation can evade detection in CT imaging, it could not fully be excluded that some amount of inflammation persisted at the site of the muslinoma. At the time of the patient's original presentation with an ophthalmic artery aneurysm, the use of muslin in surgical repair was considered routine and the risk of developing uncontrolled inflammation considered outweighed by the benefits of expeditious intervention. Fortunately, the patient still has useful vision two decades after development and surgical resection of an optochiasmatic muslinoma.

To our knowledge, this case represents the longest known duration between treatment of a muslinoma and recurrent diplopia in the English language ophthalmic literature. Clinicians should be aware of the delayed effects of these lesions years or even decades following surgery.

Declaration of interest

The authors report no conflicts of interest.

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