Recurrent Post-operative Aseptic Meningitis After Cranioopharyngioma Resection

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ABSTRACT

INTRODUCTION: Aseptic meningitis is an uncommon phenomenon related to chemical irritation of the meninges. Among other cystic lesions, intracranial rupture of cranioopharyngiomas has been reported to cause an aseptic meningitis. Cholesterol crystals released from the cyst cavity are implicated as chemo-irritant. Surgical manipulation and blood have also been implicated triggers for a chemical-mediated inflammatory response. Aseptic meningitis may present similar to infectious meningitis inclusive of fever, headache, neck stiffness, and photophobia. Work-up can be extensive and treatment decisions can be a conundrum. Herein we present a case of a female who underwent cranioopharyngioma resection and returned with multiple episodes of aseptic meningitis.

METHODS: This is a case report of a 65-year-old female with a compressive cranioopharyngioma who underwent endoscopic trans-sphenoidal, trans-sellar resection followed by aseptic meningitis, regrowth, re-resection and recurrence of meningitis.

RESULTS: The patient was found to have a cranioopharyngioma on Magnetic Resonance Imaging (MRI) after presentation with 2-months of dizziness. She underwent trans-sphenoidal resection with improved vision post-operatively. However, she presented to the emergency department (ED) with fatigue and fever concerning for meningitis. Cerebrospinal fluid (CSF) analysis revealed elevated leukocytes and total protein, and normal glucose levels consistent with meningitis. Cultures returned sterile. She was discharged on long-term antibiotics. Unfortunately, her cranioopharyngioma regrew requiring open craniotomy re-resection. She did well post-operatively but presented to the ED with altered mental status, emesis, and fever. Again, CSF analysis was sterile with elevated leukocytes and total protein. She was treated with another course of long-term antibiotics. After radiation therapy, repeat MRIs found no regrowth of her cranioopharyngioma.

CONCLUSION: Aseptic meningitis after cranioopharyngioma resection is a rare phenomenon but can introduce a diagnostic and management conundrum. This case highlights the importance of awareness for chemical-induced meningitis following cranioopharyngioma resection. While symptomatic relief is important, the course of antibiotic coverage is controversial. Coordinated multi-team efforts are necessary to provide optimal care for these patients.

CASE: A 65-year-old female presented with a 2-month history of progressive dizziness along with dry skin and fatigue. She was found to have a bitemporal hemianopia. Magnetic resonance imaging (MRI) demonstrated a 2.4 x 2.1 cm mixed cystic and solid mass suggestive of a cranioopharyngioma with mass effect on the optic tracts and chiasm [Figure 1]. Given the progression of her symptoms, the patient underwent an image-guided endoscopic trans-sphenoidal, trans-sellar decompression of the suprasellar mass with an ablational fat graft and pedicled septal mucosal graft repair. Pathology returned as papillary-type cranioopharyngioma. She did well post-operatively in the hospital with improvement of her vision and was discharged to a rehabilitation facility in good condition.

However, she presented to the Emergency Department (ED) with fever and fatigue two days after discharge. She denied any meningitic symptoms or any nasal drainage. Neurologic examination was non-focal and her white blood cell count was mildly elevated. MRI demonstrated a focus of restricted diffusion around the operative site but no leptomeningeal enhancement. A lumbar puncture (LP) found elevated cerebrospinal fluid (CSF) white blood cells with a neutrophil predominance, normal glucose and elevated total protein levels. Infectious Disease was consulted the patient was started on vancomycin and ceftriaxone. She improved clinically during her hospitalization and deinfected. Her cerebrospinal fluid cultures returned sterile. She was discharged to complete a 3-week course of intravenous antibiotics followed by an additional week of oral antibiotics.

CASE (continued): The patient again re-presented to the ED with right-sided weakness and headache two days after discharge. MRI demonstrated re-growth of the suprasellar cyst along with multifocal stenosis in the distal branches of the left anterior communicating artery and the left middle cerebral artery concerning for septic emboli. She also had a rim-enhancing focus in the left temporal lobe concerning for an abscess for which she was taken to the operating room for a left frontotemporal craniotomy for resection of her cranioopharyngioma. She recovered and went to a rehabilitation facility in good condition.

A few weeks later, the patient developed worsening of her vision once again. MRI demonstrated regrowth of her cranioopharyngioma with compression on the optic chiasm [Figure 2]. She underwent a right prefrontal craniotomy for decompression and subtotal resection of the mass which subsequently regrew. She was again taken to the operating room for resection of the cranioopharyngioma. She recovered. She was then discharged back to a rehabilitation facility, but eventually improved and underwent radiation treatment to inactivate the tumor and prevent cystic re-accumulation. Her vision improved with only a residual right upper quadrantopia.

A follow-up MRI found no new re-growth of her cranioopharyngioma.

Figure 1: MRI imaging, pituitary protocol. T2-weighted, (a) axial and (b) coronal sections. T1-weighted, post-gadolinium, (c) sagittal section. There is a mixed cystic and solid mass situated superior and posterior to the sella measuring 2.5 x 2.4 x 2.1 cm. The mass displaces the optic chiasm and tracts.

DISCUSSION: Aseptic meningitis following cranioopharyngioma resection is an uncommon entity that can pose a difficult clinical challenge. Post-operative infection, cyst fluid and the surgery itself are potential culprits for meningitis in the operative setting. With rupture of a cranioopharyngioma, chemo-irritation from cystic release of cholesterol crystals has been implicated as a cause [3]. With respect to surgery, manipulation of cerebral vessels and exposure of blood leading to an inflammatory response has been discussed [2]. Surgical material and debris have also been implicated as initiators of the inflammatory response [3]. A review of 64 patients undergoing endoscopic endonasal surgery for cranioopharyngioma found 7.8% rate of post-operative meningitis but did not delineate between infectious and non-infectious etiologies [4].

Patients may present similarly to those with infectious meningitis with signs and symptoms inclusive of fever, headache, vomiting, neck stiffness, and mental status changes. In one review of post-neurosurgical patients with post-operative meningitis, the only statistically significant differentiating signs denoting infectious meningitis were fever over 39.4°C, fever lasting longer than 7 days and a reported period of unconsciousness [5]. Given aseptic meningitis is largely a case reportable phenomenon and its entity may be underreported, the sample size is too small to garner meaningful conclusion.

Figure 2. MR imaging. T2-weighted, (a) axial and (b) coronal sections. T1-weighted, post-gadolinium, (c) sagittal section. There is a rim-enhancing focus in the left temporal lobe tip measuring 2.3 x 2.4 x 1.9 cm which exerts mass effect on the optic chiasm and tracts.

DISCUSSION (continued): Work-up of meningitis includes lumbar puncture with CSF analysis and culture along with serum white blood cell count and MRI to rule out abscess in the post-operative setting. CSF culture growth of an infectious agent clearly differentiates an infectious etiology from aseptic meningitis. However, infectious meningitis may yield sterile cultures, particularly in the setting when empiric antibiotics are initiated prior to obtaining the culture. Gram stain sensitivities have been reported at 56-80% [6]. CSF lactate levels have been shown to be a positive biomarker in the early diagnosis of bacterial meningitis with one meta-analysis yielding a sensitivity and specificity of 93% and 96%, respectively [6]. A clinical value above 39 mg/dl was the recommended CSF lactate level below which bacterial meningitis could be ruled out.

Given the severity of sequela from an infectious meningitis, empiric broad-spectrum antibiotics is usually commenced after cultures obtained. However, as aseptic meningitis is a self-limiting process improved with steroid administration, empiric antibiotic therapy for culture negative meningitis increases patient cost and risk of medication toxicity as well as promotes bacterial resistance. It has been proposed to commence empiric antibiotics until cultures return with cessation of the antibiotics should the cultures prove sterile in the post-operative setting [5].

Aseptic meningitis should be considered in the post-operative setting after cranioopharyngioma resection. Work-up for an infectious etiology inclusive of MRI and lumbar puncture are imperative. CSF lactate may be useful in assessing for aseptic meningitis, especially in presentations of recurrent post-operative meningitis when cultures are sterile. Initially management with empiric, broad-spectrum antibiotics is prudent given the potentially devastating sequela of infectious meningitis.

REFERENCES