

CASE REPORT

A rare life-threatening complication of acute mastoiditis: Case report and literature review

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Abstract

Severe complications may not always present with “classic” signs and symptoms. In the setting of recent mastoiditis, complications including cerebral venous sinus thrombosis, skull base osteomyelitis, and retropharyngeal abscess should be considered, particularly with persistent or worsening symptoms. A broad differential can lead to prompt diagnosis and treatment, thereby reducing the likelihood of morbidity and mortality.

KEYWORDS

cerebral venous sinus thrombosis, mastoiditis, retropharyngeal abscess, skull base osteomyelitis

1 | INTRODUCTION

An 88-year-old man with recent history of mastoiditis status after completing outpatient antibiotic regimen presented with worsening neck pain. Found to have complications of cerebral venous sinus thrombosis, skull base osteomyelitis, and a retropharyngeal mass. This is the eleventh case in medical literature reporting on this phenomenon.

Mastoiditis is a bacterial infection of the mastoid air cells, most commonly secondary to acute otitis media (AOM). Mastoiditis can be effectively managed with antibiotics; however, incomplete treatment can potentially result in worsening infection with contiguous and hematogenous spread leading to fatal complications. Some of these complications include cerebral venous sinus thrombosis (CVST), skull base osteomyelitis (SBO), and retropharyngeal abscess (RPA). Although these complications are rare, they often have a subtle presentation and can result in significant morbidity.¹

Cerebral venous sinus thrombosis is a stroke that involves a thrombus forming in the cerebral veins or dural sinuses.

The exact symptomology and pathogenesis is complex and not entirely understood, however, two major accepted mechanisms include the following: (a) Thrombosis to the cerebral veins or dural sinuses impairing blood drainage from the brain tissue leading to cerebral parenchymal destruction; (b) obstruction in dural sinus decreasing cerebrospinal fluid absorption leading to increased intracranial pressure.²

Skull base osteomyelitis is an inflammation of the skull bones. Clinical presentations of SBO vary, depending on the location of osteomyelitis. The most often affected regions are the sphenoid and/or temporal bones, due to contiguous spread and resultantly present with severe otalgia, fevers, aural fullness, and purulent otorrhea.³

Retropharyngeal abscess is a purulent fluid collection in the retropharyngeal space, located between the buccopharyngeal and alar fascias. In adults, infection of this space and subsequent RPA formation is generally due to trauma of the posterior pharynx, while in children, is often due to compartmental spread of neighboring infection.⁴

This is a case report that presents a patient who concomitantly developed all three of these complications.

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2 | CASE PRESENTATION

An 88-year-old man presented to the emergency department (ED) with progressively worsening neck pain. He had a past medical history of hypertension, right eye prosthesis status after remote injury, and recent mastoiditis status after full course of intravenous and oral antibiotics. His neck pain initially started 1 week ago although significantly worsened over the past 3 days after receiving a magnetic resonance imaging (MRI) brain to assess for resolution of infection. The neck pain was significantly worsened with minimal movement and associated with bilateral upper extremity weakness, which limited his ability to hold himself up with his walker. He denied any recent trauma or having any headaches, dizziness, fever, odynophagia, difficulty breathing, numbness, tingling, saddle anesthesia, bladder, or bowel incontinence. No history of illicit drug use, spine surgery, or malignancy. The MRI of the brain with contrast was read as negative and reported resolution of previously identified mastoiditis.

Upon arrival at the emergency department, physical exam showed vital signs: heart rate 107, blood pressure 176/110, temperature 36.9 C, and respirations 16. The patient was overall well appearing and nontoxic. He did have a stiff neck with significant limitation in mobility. On evaluation of the right ear, granulation tissue and mild erythema were noted in the right external auditory meatus, the tympanic membrane could not be visualized, there were no necrotic or vesicular lesions, and there was no active drainage. He had bilateral trapezius and cervical paraspinal muscle tenderness and 4/5 bilateral upper extremity strength.

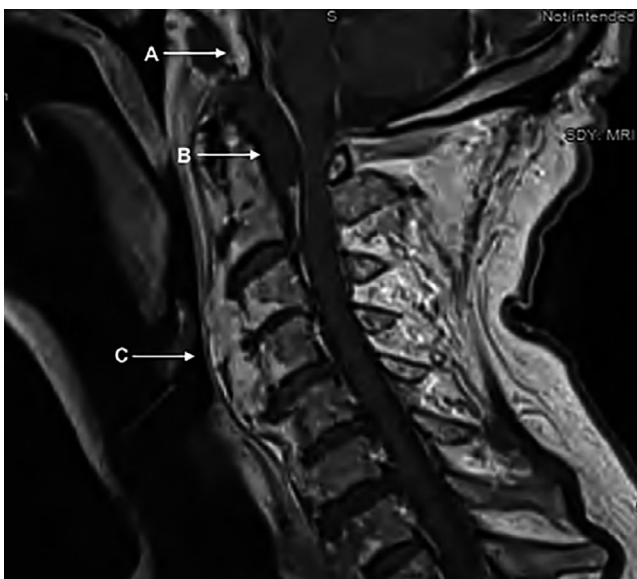


FIGURE 1 MRI T1, sagittal. Clivus osteomyelitis (Arrow A). Pannus causing spinal cord compression (Arrow B). Retropharyngeal inflammatory changes (Arrow C)

On laboratory blood analysis, complete blood count with differential revealed leukocytosis of 12.4, and coagulation studies were elevated as follows: PT 16.6, INR 1.5, and PTT 44.1. At this point, a lumbar puncture was deferred due to low clinical suspicion for meningitis or intracranial hemorrhage in the setting of a mildly elevated INR. Though the patient's MRI report from 3 days prior noted resolution of mastoiditis and no other abnormalities, a repeat MRI brain including cervical spine was ordered as the patient had leukocytosis, persistent neck stiffness, and objective bilateral upper extremity weakness. The MRI of the cervical spine demonstrated findings suggestive of osteomyelitis of the clivus and upper cervical spine, a prevertebral abscess possibly retropharyngeal, spinal canal narrowing, and spinal cord edema (Figure 1). MRI brain revealed necrosis and abscess in the right nasopharynx, patchy sclerosis in the adjacent central skull base suspicious for osteomyelitis, and venous thrombosis involving the distal right sigmoid sinus, jugular bulb, and proximal aspect of the right internal jugular vein (Figure 2).

3 | MANAGEMENT AND OUTCOMES

Given these critical findings, Vancomycin 20 mg/kg, Piperacillin/Tazobactam 4.5 g, and Dexamethasone 10 mg

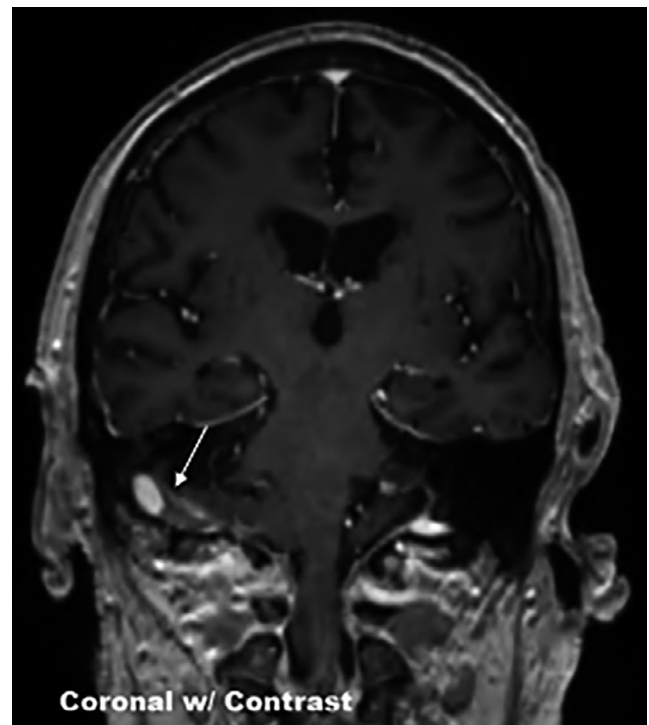


FIGURE 2 MRI T1 with contrast, coronal. Arrow shows filling defect from intraluminal thrombus, identifying cerebral venous sinus thrombosis

were immediately given. A heparin drip 18 u/kg/h was initiated, and the patient was transferred to a tertiary care facility for a higher level of care. Over the patient's hospitalization, he was seen by a team of consultants, including interventional radiology, infectious disease, otolaryngology, hematology, neurotology, and neurosurgery. The patient underwent exploration by otolaryngology for possible incision and drainage of suspected RPA that revealed a soft tissue mass with no evidence of abscess or malignancy after biopsy. A biopsy of the vertebrae did reveal osteomyelitis. Ultimately, the patient was continued on 1 month of intravenous antibiotics and 3 months of anticoagulation therapy. Cervical spine surgery was deferred until resolution of infection, to avoid placing hardware in infected bone. The patient was discharged from the hospital 8 days after admission in stable condition.

Over the course of the next 2 months, the patient returned to the hospital with anemia secondary to gastrointestinal bleeding from duodenal ulcers that required microembolization and nephropathy secondary to medication effects that required a change in antibiotic regimen. The patient was otherwise improving and ultimately underwent neurosurgery for a C1-C2 laminectomy and decompression of spinal cord with occipital fusion (Figure 3).

4 | DISCUSSION

This case illustrates the rare and life-threatening presentation of CVST, SBO, and retropharyngeal mass in the setting of recent mastoiditis. There have been only 10 other cases of CVST with SBO reported in literature to date (Table 1).⁵⁻¹⁴ Patient age ranged from 17 to 94 years old with eight out of 10 cases occurring above the age of 50 years old. The initial



FIGURE 3 CT without contrast, sagittal. Occiput to C5 fusion status post decortication and laminectomy

chief complaint in each case varied, however, eight out of the 10 cases presented with neurologic symptoms, the most common being a headache.^{5-7,9,11,13,14} In all cases, CVST and SBO developed as secondary complications from a primary infectious etiology. Of note, eight of the 10 cases were male patients. The etiology for this gender trend is unclear at this time given the limited number of cases, as it could also be due to an artifact of small sample size.

Cerebral venous sinus thrombosis itself is a rare pathology. The annual incidence ranges from 0.22 to 1.57 per 100 000 patients.¹⁵ It is most commonly found in young females. Presentation is highly variable, and diagnosis requires imaging via a head computed tomography (CT), preferably with venography, or a brain MRI with contrast. Some previously described symptoms of this pathology include headaches, blurred vision, seizures, ophthalmoplegia and/or coma.¹⁶ Early discovery and treatment with anticoagulation carries a favorable outcome. One multinational study consisting of 624 patients reported 79% of patients having complete recovery or minor residual symptoms after treatment, with 4.3% mortality during hospitalization and 3.4% mortality within 30 days of symptom onset.¹⁷ Recurrence after anticoagulation therapy was found to be 2%-4%.¹⁸

Skull base osteomyelitis is also a rare pathology, as only 84 cases of head or neck osteomyelitis have been reported to date.³ Diagnosis is typically established with an MRI with contrast or bone single photon emission computed tomography (SPECT) imaging. Gold standard diagnosis modality is via tissue biopsy which is frequently done in these instances in order to rule out malignancy.¹⁹ Early treatment with culture guided long-term antibiotic therapy carries a favorable prognosis. Although, patients might require surgical debridement with hyperbaric oxygen therapy.²⁰ In one study, prognosis for SBO at 18-month follow-up was reported to have a mortality rate of 9.5%,²¹ while another study quoted a mortality rate as high as 21%-70%.³

Retropharyngeal abscess clinical presentation varies according to the stage of illness; common signs and symptoms include odynophagia, neck stiffness, hoarseness, neck mass/swelling and respiratory distress.²² Diagnosis can be made via plain radiography, CT neck with contrast or MRI neck with contrast. Gold standard method of diagnosis is a tissue biopsy. Treatment consists of antibiotics with possible incision and drainage, depending on the severity of pathology.

The final diagnosis for our patient necessitated utilization of almost all of these modalities. More importantly, this case highlights the value of reviewing one's own ordered imaging and reassessment of patients. In this case, the patient's MRI brain with contrast report from 3 days prior indicated resolution of mastoiditis with no acute abnormalities; however, the patient only had the radiology interpretation available with them when they presented to our ED. With ongoing symptoms and the inability to review previous outpatient imaging,

TABLE 1 Summary of case reports with skull base osteomyelitis (SBO) and cerebral venous sinus thrombosis (CVST)

Case reference	Age/Sex	Chief complaint	Complications
5	65/M	6 mo of pain and discharge in right ear	Complete right facial paralysis, occlusion of right lateral venous sinus, and SBO
6	51/M	7 d of left temporooccipital and retro-orbital facial pain, hoarseness, and dysphagia	Mastoiditis, SBO, cranial nerve palsies, sigmoid sinus thrombosis, and septic pulmonary embolus
7	17/M	5 d of sore throat, myalgias, and vomiting	Internal jugular vein, cavernous sinus, internal carotid artery, and sigmoid sinus thrombosis, vertebral artery dissection, SBO, and epidural abscess
8	50/M	Right eye pain, swelling, and progressive vision loss after a fall	Superior ophthalmic vein thrombosis, bilateral cavernous sinus thrombosis, internal jugular vein thrombosis, orbital cellulitis, and clival osteomyelitis
9	58/M	Headache, hoarseness of voice, dysphagia, cough, and tiredness	SBO, left jugular vein, left sigmoid sinus, and lateral third of transverse sinus thrombosis
10	70/F	Left temporal headache	Aseptic meningitis, septic pulmonary embolism, cryptococcal pneumonia, SBO, and cavernous sinus thrombosis
11	94/M	8 d of fever, unilateral hearing loss	Subdural empyema, SBO, and venous sinus thrombosis
12	66/M	Sudden epistaxis	Facial nerve palsy, SBO, sigmoid sinus thrombosis, maxillary artery pseudoaneurysm
13	71/M	Worsening headaches	SBO and sigmoid sinus thrombosis
14	45/F	Left hemiplegia, dysarthria, and dysphagia	Transverse sigmoid thrombosis, retropharyngeal abscess, and SBO

Abbreviations: F, female; M, Male.

the decision was made to obtain repeat MRI testing; which ultimately lead us to identify the previously missed diagnosis.

Additionally, it is worth noting that none of the patients in previously reported similar cases presented with “classic” signs and symptoms befitting a common pathological process. In situations of atypical patient presentations, a broad differential, a high index of suspicion, and a robust and complete physical examination are essential to diagnosing life-threatening illnesses and complications.

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CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

JO, HV, HR, and PS drafted the article or revised it critically for important intellectual content; and gave final approval of the version of the article to be published.

ETHICAL APPROVAL

Informed consent was obtained for publication of this case report and accompanying images.

DATA AVAILABILITY STATEMENT

No data were generated or analyzed for this publication.

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