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Case Report

Intramedullary histoplasmosis of the thoracic cord as an isolated lesion: A rare case report and literature review

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ABSTRACT

Background: Disseminated histoplasmosis involving the central nervous system occurs in 5–10% of cases. However, intramedullary spinal cord lesions are extremely rare. Here, 45-year-old female with a T8–9 intramedullary lesion did well following surgical extirpation.

Case Description: For 2 weeks, a 45-year-old female experienced progressive lower back pain, paresthesias, and paraparesis. The magnetic resonance imaging showed an intramedullary expansive lesion at the T8–T9 level that markedly enhanced with contrast. Surgery, consisting of T8–T10 laminectomies performed using neuronavigation, an operating microscope, and intraoperative monitoring, revealed a well-demarcated lesion that proved to be a focus of histoplasmosis; it was readily completely excised.

Conclusion: Surgery is the gold standard for treating spinal cord compression attributed to intramedullary histoplasmosis unresponsive to medical management.

Keywords: Histoplasmosis, Neurosurgery, Spine infection, Spine surgery, Spine

INTRODUCTION

Disseminated histoplasmosis involves the central nervous system (CNS) in 5–10% of cases, but only rarely is found within the spinal cord itself (i.e., intramedullary/intradural). The most common neurological symptoms/signs correlate with the various brain and spinal lesion locations, while other findings may include chronic meningitis, hydrocephalus, and/or encephalitis. Approximately 1/3 of cases occur in immunocompetent versus 2/3 in immunosuppressed individuals.[4,6,7] Here, a 45-year-old female presented with a T8–T9 intramedullary focus of histoplasmosis resulting in spinal cord dilatation/compromise responsible for a severe paraparetic deficit that largely resolved following lesion resection.

CASE REPORT

Clinical presentation

An immunocompetent 45-year-old female presented with a 2-week history of progressive lower back pain, paresthesias, and paraparesis. Within a few days, the patient progressed from 3/5 to 1/5 lower extremity weakness; her neurological findings also included diffuse hyperreflexia,

bilateral Babinski signs, and urinary retention. The magnetic ressonance imaging (MRI) of the spine demonstrated an intramedullary expansive lesion at the T8–T9 level that markedly enhanced with contrast [Figures 1 and 2]. Moreover, an MRI of the brain revealed no intraparenchymal lesions, and the computed tomography of the chest/abdomen failed to reveal any other focus of histoplasmosis (i.e., specifically no pulmonary involvement). A lumbar puncture for cerebrospinal fluid (CSF) analysis revealed; 94 cells; 84% neutrophils; 1% glucose; proteins 1779; and lactic acid 13.4 mmol/L. The serology was negative for: toxoplasmosis immunoglobulin M (IgM), human immunodeficiency virus (HIV), hepatitis C virus (HCV), hepatitis B surface antigen (HBsAg), venereal disease research laboratory (VDRL), and the patient denied a history of tuberculosis. Notably, the patient had a long-standing history of requiring a ventriculoperitoneal shunt (with multiple revisions), and later, a ventriculoatrial shunt, despite an undiagnosed or apparent cause.

Surgery

Utilizing neuronavigation, an operative microscope, and intraoperative monitoring, the patient underwent a T8–10 laminectomy. As soon as the dura and arachnoid were opened, an expansile intramedullary cord lesion was visualized, and xanthochromic/purulent fluid immediately drained under increased pressure. A well-demarcated cleavage plane between the lesion and cord facilitated gross-total lesion resection. The pathological analysis was diagnostic for histoplasmosis [Figure 3]. Postoperatively, the patient's paraparesis markedly improved, with full recovery of the motor/sensory deficits, and sphincter function.

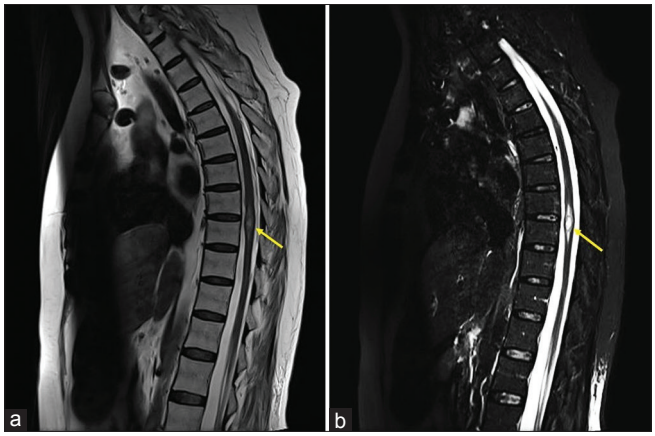

Figure 1: Magnetic resonance imaging (MRI) of the thoracic spine. (a) Sagittal T2-weighted revealed an intramedullary expansive lesion (yellow arrow). (b) Sagittal T2-weighted short-tau inversion recovery (STIR) image revealing an intramedullary intradural hyperintense mass (yellow arrow) at the T8–T9 level.

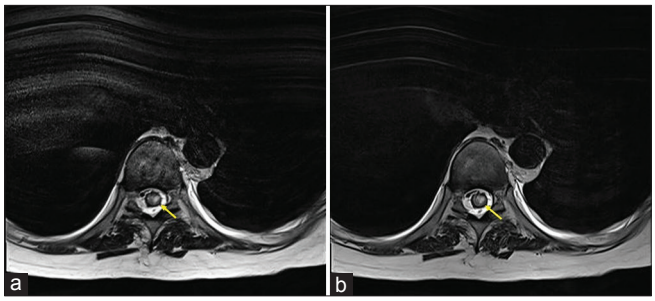

Figure 2: MRI of the thoracic spine. (a) Axial T2-weighted and (b) axial T2-weighted Gd-enhanced image demonstrating a hyperintense lesion infiltrating the spinal cord located at T8–T9 (yellow arrow).

DISCUSSION

Definition and frequency of isolated versus disseminated histoplasmosis

Histoplasmosis is defined as an intracellular infection caused by the fungus histoplasma capsulatum. The first case involving the CNS was described in an infant in 1934.[4,8] CNS histoplasmosis can present with/without pulmonary involvement, and either as an isolated (i.e., less common) or disseminated process (i.e., more severe/more typical).[2,9,12] CNS involvement occurs in 5–20% of patients with disseminated disease (i.e., infects extrapulmonary sites through hematogenous spread).[1,9,10] Immunosuppression increases the risk 10 fold for progression of histoplasmosis to disseminated disease versus immunocompetent individuals.[1]

Pathogenesis of spinal intramedullary histoplasmosis

Intramedullary spinal histoplasmosis is extremely rare, and the literature suggests that 50–90% of immunocompetent individuals with histoplasmosis remain asymptomatic. Symptomatic cases are often observed in immunocompromised patients, and the signs/symptoms correlate with the location of brain or spine lesions (i.e., myelopathy). Other findings include; chronic meningitis, cerebral vasculitis/stroke syndromes, hydrocephalus (chronic recurrent), and encephalitis [Table 1].[1,2,5]

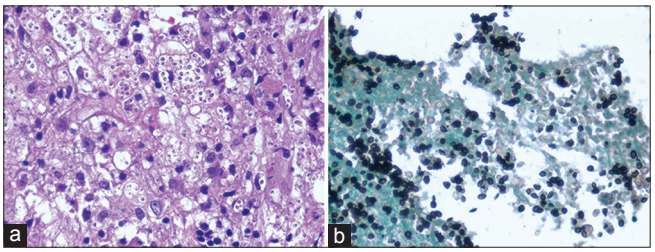

Figure 3: Histology of intramedullary histoplasmosis lesion. (a) Hematoxylin and eosin (H&E) staining revealed inflammatory process, where epithelioid macrophages, lymphocytes, and multinucleated giant cell are observed. (b) Grocott methenamine silves (GMS) staining identified fungal microorganisms that are morphologically consistent with histoplasma.

Table 1 : Summary of the cited cases of CNS histoplasmosis, including the present study.

Article	Patient's age	Sex	Level/ Presentation	HIV/ Immunosuppression	Symptoms	Surgery/Treatment
Gonzalez *et al.*[1]	62	Male	Brain; C2, T2 and T11	Negative	Altered mental status, fevers, weakness, and headache	Meropenem, amphotericin and itraconazole
Hariri *et al.*[2]	32	Male	Brain	Positive	Altered mental status and confusion; Weight loss, cough and fever	Amphotericin and itraconazole
Hott *et al.*[3]	47	Female	T2. Intramedullary	Negative	Paraparesis and bowel and bladder incontinence	T2–T3 laminectomy. Intrathecal amphotericin and voriconazole
Manning *et al.*[4]	27	Male	C7–T1. Intramedullary	Positive	Progressive paraparesis	C7–T2 laminectomy and liposomal amphotericin B
N'dri-Oka *et al.*[5]	42	Female	T2. Intramedullary	Negative	Progressive paraparesis	T12–L1 laminectomy and itraconazole
Recker *et al.*[6]	44	Male	T3. Intramedullary	Positive	Clumsiness and numbness of his left-lower extremity	Amphotericin B and methylprednisolone; followed by itraconazole and prednisone
Renan *et al.*[7]	4	Female	C4–C6. Intramedullary	Positive	Fever and symmetrical tetraparesis with alterations of the upper motor neuron syndrome	C4–C6 posterior laminectomy. Liposomal amphotericin followed by fluconazole
Simms *et al.*[9]	84	Male	T7 inferiorly into the conus	Positive	Fever, confusion, and acutely bilateral lower extremity weakness and loss of sensation	Liposomal amphotericin B and itraconazole
Voelker *et al.*[10]	28	Male	C4. Intramedullary	Negative	Neck pain and left arm weakness	C3–5 laminectomy and amphotericin B
Wheat *et al.*[12]	29	Female	Meningitis	Positive	NA	Amphotericin B and fluconazole
	73	Male	Brain	Positive	Confusion, lethargy, weakness, fever and aphasia	Liposomal amphotericin B followed by fluconazole
Piovesan *et al.* (Present Study)	45	Female	T8–T9. Intramedullary	Negative	Lower back pain; paresthesias and paraparesis, with urinary retention	T8–T10 laminectomy

NA: not available. CNS: Central nervous system.

MRI of intramedullary histoplasmosis

MRI findings for intramedullary histoplasmosis include focal nodular low-intensity T1 lesions, and high-intensity T2 lesions that heterogeneously enhance. The lesion is nodular or like a ring one and, if a lesion is found, the entire neuraxis should be assessed.[5] Differential diagnoses for these intramedullary lesions include: infections (i.e., syphilis, fungal infections, tuberculosis, and other mycobacteria), demyelinating diseases (acute demyelinating encephalomyelitis), or neoplastic causes.[7]

Delayed diagnosis of CNS histoplasmosis utilizing antibody/antigen testing, cultures, and biopsy

The diagnosis of CNS histoplasmosis is challenging and typically delayed.[7] The most sensitive method to document histoplasmosis is the antibody and antigen tests of the CSF.[11] Although a CSF positive culture is the gold standard for confirming the diagnosis of CNS histoplasmosis, it has a low sensitivity, and culture growth may take several weeks potentially delaying diagnostic confirmation.[1] Alternatively,

biopsy for accessible lesions may be the most rapid and definitive diagnostic technique.[7]

Morbidity and mortality for CNS histoplasmosis

Patients with CNS histoplasmosis exhibit high morbidity, mortality, and relapse rates.[11] Immunocompromised individuals have a 20–40% mortality and a 50% relapse rate. The most commonly reason for these failures includes inadequate treatment.[1,3]

Medical management of CNS histoplasmosis

For those without CNS involvement not requiring hospitalization, itraconazole is the treatment of choice. For those hospitalized or with CNS involvement, initial therapy should include amphotericin B with transitioning to itraconazole.[9]

Medical and/or surgical management of CNS histoplasmosis

Fewer than 50% of cases of CNS histoplasmosis are cured by antifungal treatment alone. Therefore, the treatment of choice for those patients, particularly with progressive neurologic deficit/deterioration despite medical management, includes amphotericin B and surgical lesion resection.[3]

CONCLUSION

CNS histoplasmosis can be difficult to diagnose due to its atypical features and because it can mimic other systemic diseases. CNS histoplasmosis should be considered even in non-endemic areas, with or without a history of immunosuppression, and even with isolated spinal cord injury. In the present study, a 45-year-old female presented with myelopathy/paraparesis attributed to a T8–T9 intramedullary histoplasmosis that presented as an isolated lesion. Following complete lesion resection, the patient recovered the neurological function.

Declaration of patient consent

Patient's consent not required as patient's identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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