Intramedullary tuberculoma of the spinal cord

Case report and review of the literature

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Intramedullary spinal tuberculosis infection remains an extremely rare disease entity. In the most recent reviews only 148 cases have been reported in the world literature, although numerous recent reports from developing countries and on human immunodeficiency virus (HIV)–positive patients have increased this number. The authors present an unusual case of intramedullary tuberculoma in an HIV–negative patient from the southern United States who demonstrated no other signs or symptoms of tuberculosis infection. The authors believe that this is the first case of its kind to be presented in recent literature. The presentation of miliary disease via an isolated intramedullary spinal mass in a patient with no evident risk factors for tuberculosis infection emphasizes the importance of including tuberculosis in the differential diagnosis of spinal cord masses.

KEY WORDS • spinal cord • tuberculosis • intramedullary infection

TUBERCULOSIS remains an important pathological entity in developing countries. It is also a common opportunistic infection in individuals who test positive for human immunodeficiency virus (HIV), and it remains an important public health problem. In the industrialized world, tuberculosis infection that involves the central nervous system is rare, and intramedullary spinal tuberculomas are even more unusual. In the most recent reviews by Lin, et al.,13 and MacDonnell, et al.,15 148 cases of intramedullary spinal tuberculosis infection are recorded in the world literature. Recent case reports have presented intramedullary infection found in HIV-positive patients,5,16,26 patients with other evidence of tuberculosis infection,19 and individuals from developing countries.3,8–10,13,21–24

We present a case of pathologically confirmed intramedullary spinal tuberculosis in an otherwise healthy, HIV-negative North American patient who showed no other evidence of systemic disease. To our knowledge, this is the only case of its kind to be reported in recent literature.

Case Report

Presentation. This 46-year-old woman from Mississippi was referred to our institution for evaluation of bilateral lower-extremity weakness and numbness, low-back pain, and bowel and bladder incontinence. The patient noted the onset of symptoms 18 months prior to presentation, with initial numbness and weakness, followed by onset of back pain in the upper lumbar region. No radicular pain was noted. After the onset of bladder incontinence, medical evaluation was performed.

The patient was employed as a physical therapist and had never traveled outside the United States. Her medical history was significant for gastric ulcers, diagnosed in the 1980s, but was otherwise negative. She reported sporadic tobacco use (one pack of cigarettes per month), and occasional alcohol consumption. Review of systems was otherwise negative. No significant family history was offered. She was not at high risk for contracting HIV infection, and she tested negative for HIV postoperatively. Purified protein derivative skin testing, which was also performed postoperatively and while the patient was undergoing a tapering regimen of steroid therapy, was negative.

Examination. On examination, the patient appeared to be healthy. Bilateral lower-extremity weakness was evident, and the left leg was more profoundly affected than the right. Lower-extremity reflexes were diffusely hyperactive, with a positive left Babinski’s sign and no clonus. Sensory deficit to pinprick and light touch extended to the T-6 level. Her gait was unsteady and faltering, and she favored the left lower extremity. No upper-extremity motor or sensory deficits were evident. Preoperative laboratory results and complete blood count were all within normal limits. Chest x-ray films were obtained and revealed no abnormalities; bilateral apexes were clear. Magnetic resonance imaging (MR) of the thoracic spine revealed an enhancing intramedullary mass with an inferior area of syringomyelia (Fig. 1).
Operation. A T7–12 laminectomy was performed. Intraoperative examination via the initial myelotomy at T10–11 revealed very poorly demarcated grayish tissue. A frozen section showed chronic inflammation. A biopsy sample of the permanent section was obtained.

Pathological Findings. Pathological examination of the tumor biopsy specimen revealed granuloma formation and perivascular lymphocytic infiltration. Special staining demonstrated acid-fast bacilli (Fig. 2). Further polymerase chain reaction examination confirmed the presence of Mycobacterium tuberculosis.

Postoperative Course. The patient was started on a course of four-drug antituberculous antibiotic therapy (ethambutol 1200 mg/day, rifampin 600 mg/day, isoniazid 300 mg/day, pyrazinamide 2000 mg/day). She has done well postoperatively, with no new neurological worsening. At time of this report, she continued to undergo antituberculous antibiotic therapy, was able to walk using a walker, and was actively enrolled in a rehabilitation program. She anticipates returning to work after her antibiotic course is completed.

Discussion
Intramedullary spinal tuberculosis is a rare disease entity, which was first reported by Abercrombie in 1828.1 Spinal tuberculosis infection more commonly presents with tuberculous spondylitis2 or arachnoiditis.2,12 An extensive review of the world literature conducted by Lin14 in 1960 revealed 105 cases of intramedullary tuberculosis infection, 88 of which were diagnosed at postmortem examination. More recently MacDonnell, et al.,15 have described cases from developing nations: three reports from India (12 cases),7,8,10 two each from Taiwan13,22 and Turkey,9,23 and one each from Lebanon,7 Nigeria,24 and Argentina.21 Additional reports have come from Spain16 and the Middle East.11 In six of these cases, the pathological confirmation of tuberculosis myelitis was obtained after examination of cerebrospinal fluid (CSF) culture or by direct pathological examination and/or evaluation of the culture of the surgical specimen.3,8,9,11,13 In the remaining cases, diagnosis was made using MR imaging and CSF analysis without culture,10 response to antituberculous chemotherapy after surgical decompression and abscess aspiration,25 and imaging characteristics in conjunction with miliary tuberculosis infection.20–23 Of the cases reported on international patients in whom clinical information was available, nine featured miliary disease at the time at which intramedullary spinal infection was diagnosed.3,9–11,13,20,21,24 In reviewing the Sanjay Gandhi Postgraduate Institute’s experience with MR imaging in intraspinal tuberculosis, Gupta, et al.,7 presented eight cases of intramedullary tuberculoma. Only one of these cases, however, was confirmed by open biopsy sampling and pathological examination. The specific mechanism of diagnosis in the remaining tuberculoma cases was not addressed. Clinical information on intramedullary tuberculoma cases was not provided. A separate review of tuberculous spondylitis by Nussbaum, et al.,18 presented two additional cases of intramedullary tuberculoma but offered no details. Diagnosis in these cases was determined by surgical exploration and pathological examination of biopsy specimen.

Magnetic resonance imaging appearance of intramedullary spinal tuberculosis was first described by Rhoton, et al.,19 and additional descriptions of its radiographic appearance have been offered by numerous authors.3,6–11,13,16,20,22 No pathognomonic radiographic appearance has been described, although thorough description of the lesion’s imaging characteristics have been offered.4–8,10,13,16,22 The absence of a clearly demarcated lesion on MR imaging in our case differs from previously described reports.
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Fig. 2. Photomicrographs. Upper: Photomicrograph demonstrating diffuse lymphocytic infiltrate with perivascular concentrations and granuloma formation. H & E, original magnification × 400. Lower: Photomicrographs revealing a centrally located acid-fast bacillus. Ziehl-Neelsen, original magnification × 1000 (oil).
This finding is echoed by the intraoperative appearance of the mass: no clear boundary or plane was observed. This is in contrast to the well-circumscribed, firm masses described by Rhoton, et al., Lin,14 and Dastur.4 The age of the lesion and its degree of caseation necrosis are possible explanations for this finding. Radiographic findings in our case are similar to those described by Lin, et al.,13 in which diffuse contrast enhancement was compatible with myelitis or early tuberculoma formation. The alteration in imaging characteristics throughout the course of antituberculous chemotherapy is well reviewed by Lin, et al. In a review of intradural inflammatory processes, Gero, et al.,6 presented three additional cases of intramedullary tuberculosis, which were diagnosed by analyzing CSF and by response to antituberculous therapy. Two of these cases were noted to originate from “endemic areas.” No other clinical analyses were presented.

*Mycobacterium tuberculosis* opportunistic infection in HIV-positive individuals has been extensively reviewed in the literature. In 1993, pulmonary and extrapulmonary tuberculosis infection were classified as indicators of the presence of acquired immune deficiency syndrome by the Centers for Disease Control and Prevention.25 Intramedullary tuberculosis infection was first bacteriologically confirmed by Gallant, et al.,4 and two similar cases have been recently described.16,26 In each of these cases, systemic tuberculosis infection was evident. As noted by Woolsey, et al.,26 the treatable nature of intramedullary spinal tuberculosis mandates its inclusion in the differential diagnosis of spinal cord masses in HIV-positive patients. Tuberculosis infection may also manifest in individuals whose immune systems are compromised by sources other than HIV infection. In a review of spinal cord myelitis, Murphy, et al.,17 reported a single case of intramedullary tuberculoma in a patient with systemic lupus erythematosus who underwent treatment with prednisone. Diagnosis in this case was confirmed by analysis of CSF isolate.

**Conclusions**

This search of the international literature presents a total of 31 cases of intramedullary tuberculosis infection that were published after the review by MacDonnell, et al.5 The majority of these patients were from less developed regions and in areas where tuberculosis infection is endemic. A significant number of the patients also had HIV and otherwise compromised immune systems. To our knowledge, ours is the only case described in the recent literature of miliary tuberculosis infection presenting with an intramedullary spinal cord mass in an HIV-negative patient from a region with presumably low incidence of tuberculosis. This unusual de novo presentation emphasizes that tuberculosis infection should be considered in the differential diagnosis of intramedullary spinal cord masses.

**References**


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