

Cervical intramedullary tuberculoma and tuberculous kyphosis in a 23-month-old child: case report

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Mycobacterium tuberculosis is a serious pathogen in many parts of the world being increasingly responsible for disease.^{1,2} Worldwide 7–10 million people have tuberculosis; in 1995 in the United States 22 201 new cases were reported.² Neurotuberculosis accounts for about 5% of extrapulmonary cases of tuberculosis.² It is a result of the bacteremic stage of the disease, during which tuberculous lesions (Rich's foci) may form in the meninges or within the substance of the brain or spinal cord, or both.³ Months or years later, these foci may rupture into the cerebrospinal fluid, causing meningitis, or expand into a tuberculoma within the parenchyma of the brain or spinal cord, causing focal neurologic signs. The most common manifestations of tuberculosis in the central nervous system are tuberculous meningitis and intracranial tuberculoma. Tuberculous meningitis is more common than intracranial tuberculoma,⁴ more frequent in children than adults⁵ and is often found in patients of low socioeconomic status.⁶

The majority of spinal cord disease, however, is secondary to tuberculous spondylitis (Pott's disease), causing vertebral body destruction and spinal cord compression,⁷ and accounting for 50%–60% of osseous tuberculosis.⁸ The usual sites of involvement are the thoracolumbar and lower thoracic spine, with infection seeding the metaphysis of a single vertebral body, spreading under

the anterior longitudinal ligament and causing osseous destruction or abscess formation.⁹ Ensuing osseous destruction may lead to anterior vertebral collapse and kyphosis.

Intramedullary tuberculoma of the spine is a rare manifestation of disseminated hematogenous tuberculosis. Approximately 180 cases of intramedullary tuberculoma have been reported in the world literature, with most recent case reports from developing countries and HIV-positive populations.^{1,10–23}

We report a case of intramedullary tuberculoma in a young child. Signed informed consent was obtained. The child's chart and imaging studies were reviewed.

Case report

A 23-month-old boy from a small town in the interior of British Columbia had a seizure at 3 months of age, requiring admission to hospital followed by anti-seizure medication until he was 15 months old.

Three months before presentation he had a tender left-sided neck mass with associated fever and anorexia but no rigors, night sweats or weight loss. Broad-spectrum antibiotic treatment (14-day course) alleviated the fever and reduced the size of the neck mass.

Two months later, the left-sided neck mass again became tender and increased

in size. The mass did not regress despite another course of broad-spectrum antibiotics. Although the boy had anorexia initially he again had no fever, rigors, night sweats or weight loss. His mother reported an asymmetrical gait and apparent pain in his left arm. Her pregnancy had been complicated by pyelonephritis treated with antibiotics intravenously but was otherwise normal. The boy's antenatal, perinatal and neonatal courses had been unremarkable. His immunizations were up-to-date, and development was appropriate for his age.

On examination, he was pale but alert, active and afebrile. Findings on neurologic examination were normal. Head and neck examination revealed a 4 × 3-cm submandibular mass that was nontender, fixed and firm. No other lymphadenopathies were identified. Musculoskeletal examination showed marked lower thoracic kyphosis with no other abnormalities. Results of respiratory, abdominal and dermatologic examinations were normal.

Initial laboratory investigations demonstrated only iron-deficiency anemia. Mantoux skin testing for tuberculosis exposure was positive within 48 hours. Culture of a gastric aspirate was positive for *Mycobacterium tuberculosis*, and cervical mass biopsy showed necrotizing granuloma with no organisms seen on Gram's staining.

Plain radiography showed anterior

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collapse of vertebral bodies T10 and T11 with focal gibbous formation and a large paravertebral mass (Fig. 1). Chest radiography demonstrated a density in the left upper lobe with ipsilateral hilar adenopathy.

CT of the thoracic spine showed classic tuberculous spondylitis involving the T10 and T11 vertebrae with a large paravertebral mass measuring 5 cm in diameter (Fig. 2). It displaced the aorta anteriorly and narrowed the spinal canal by 50%. Chest CT showed a dense parenchymal mass in the left lower lobe with evidence of calcification and nodal mass lesions in the left hilar region.

MR images of the spine, spinal cord and head were obtained. The vertebral bodies were destroyed at T10 and T11, with a large associated rim-enhancing paravertebral lesion (Fig. 3). Although the T9–10 disc showed signal change suggesting disease involvement, the T11–12 disc was intact. In addition, there was diffuse cord swelling between C5 and C7. The transverse T_1 -weighted images following gadolinium contrast enhancement showed a 1-cm rim-enhancing lesion with central nonenhancing necrosis occupying the right side of the cord at C5–C7 (Fig. 3).

Gadolinium-enhanced images of the head showed multiple echogenic enhanc-

ing intracerebral lesions. Most of these were at the grey–white interface. The scrofulous cervical node was also identified with multiple low-density centres suggesting necrosis.

Triple antituberculous therapy was initiated with daily oral administration of isoniazid 51 mg, rifampicin 200 mg and pyrazinamide 400 mg. The ongoing concern at this point was the status of the boy's tuberculous spondylitis and subsequent risk of progressive kyphosis. He was fitted with a spinal brace. He was admitted to a step-down unit because of concerns regarding brace compliance and completion of medical therapy. Surgical stabilization was recommended 6 weeks later when clinical examination and repeat radiographs demonstrated progression of the kyphosis. He underwent anterior spinal débridement and vascularized rib placement with posterior spinal instrumentation and fusion. A fibrocartilaginous mass had destroyed and replaced the vertebral bodies of T10 and T11. This was resected. The spine was grossly unstable both before and after resection and was acceptably reduced with vascularized vertical rib struts. Posterior fusion was completed with the use of hook stabilization and the pediatric Moss–Miami system. A stable configuration was achieved. The boy was discharged home 10 days later in a hip spica brace.

Three months postoperatively, the boy showed acceptable spinal alignment, and his brace was converted to a thoracolumbosacral orthosis. Findings on neurologic examination were normal. The combination antituberculous therapy was continued. Follow-up clinical examination at 6 months demonstrated normal spinal alignment and radiographic evidence of fusion. The boy was healthy and

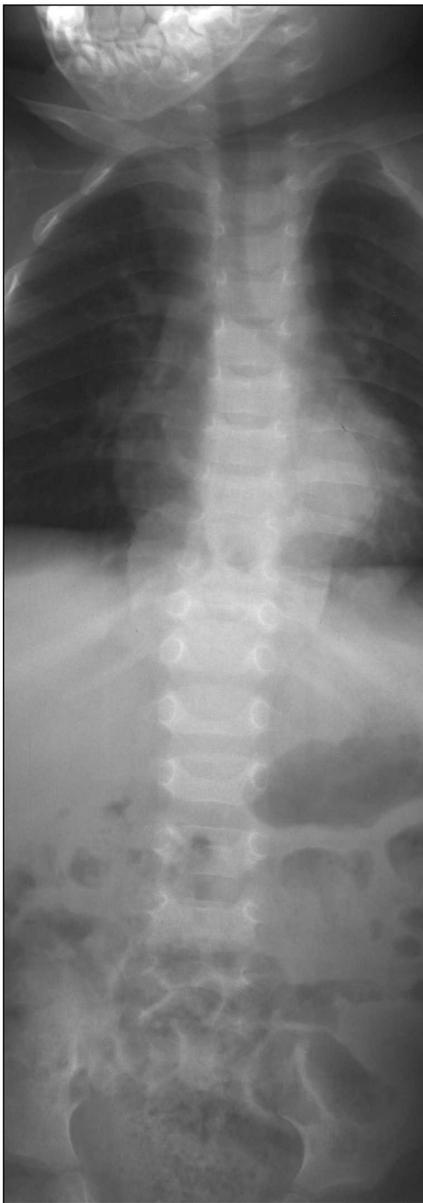


FIG. 1. Anteroposterior (left) and lateral (right) radiographs demonstrate focal gibbous formation and a large paravertebral mass.



FIG. 2. A CT scan demonstrates a significant mass lesion with notable vertebral destruction.

active. All bracing and restrictions were discontinued. Follow-up MRI demonstrated complete resolution of the cervical cord and brain lesions (Fig. 3).

Discussion

Tuberculosis is still a serious pathogen in many parts of the world and is increasingly responsible for disease as its incidence rises with the HIV population.^{1,2} The 22 201 new cases reported in the United States in 1995 exceeded the previous years' incidence.² Disseminated tuberculosis in children is rare in Canada, and potential sources must be considered. Further investigation of this child's history revealed that his neonatal caregiver was a grandparent with a history of tuberculosis.

This is a rare case of intramedullary tuberculoma. The boy is the youngest patient with such a lesion to be documented in the literature. In the absence of neurologic deficits, the cervical intramedullary tuberculoma was managed medically, in

agreement with earlier literature.^{10,21,23} In patients manifesting neurologic deficits, a combination of surgical decompression and medical therapy is indicated.^{12,14} At 6 months' follow-up from the initiation of maximal medical therapy, our patient remained neurologically intact.

MRI findings characteristic of intramedullary tuberculoma have been described.^{13,24,25} The presumed cervical intramedullary tuberculoma in this case demonstrated a 1-cm rim-enhancing lesion with a central nonenhancing area of necrosis on transverse T₁-weighted images. Based on the clinical presentation and the classical appearance of this lesion, biopsy was deemed unnecessary. The gadolinium-enhanced images also showed intracerebral lesions, presumed to be tuberculomas.

This case demonstrates that tuberculosis should be considered in the differential diagnosis of kyphosis in infants. The clinical and radiologic findings were advanced, with severe vertebral destruction at 2 levels and a large intramedullary

lesion. This suggests our patient had had the disease for a long time, and we felt his initial seizure may have been a presentation of cerebral involvement. The use of anterior vertebrectomy and fusion with vascularized rib placement plus posterior fusion and instrumentation was effective in this patient based on outcome measures of spinal alignment and a normal neurologic findings 5 months postoperatively. Circumferential fusion with anterior vascularized struts and posterior instrumentation was undertaken to provide rapid stability and prevent recurrent kyphosis. The risk of late kyphosis was increased in this child because of the marked anterior vertebral destruction, noncompliance with brace wear and the potential for future posterior growth. Progressive kyphosis was noted in 46.4% of patients treated with anterior decompression and autologous rib grafts over a 10-year period in one series.²⁶ In another series structural support after anterior arthodesis with rib grafting was attained in 41%.²⁷ Alone, therefore, anterior arthodesis with rib grafting did not effectively stabilize the spine in the majority of cases. Posterior fusion in addition to anterior rib grafting has been effectively adopted,^{17,28} and this case has shown that circumferential fusion can be used successfully in infants.

Competing interests: None declared.

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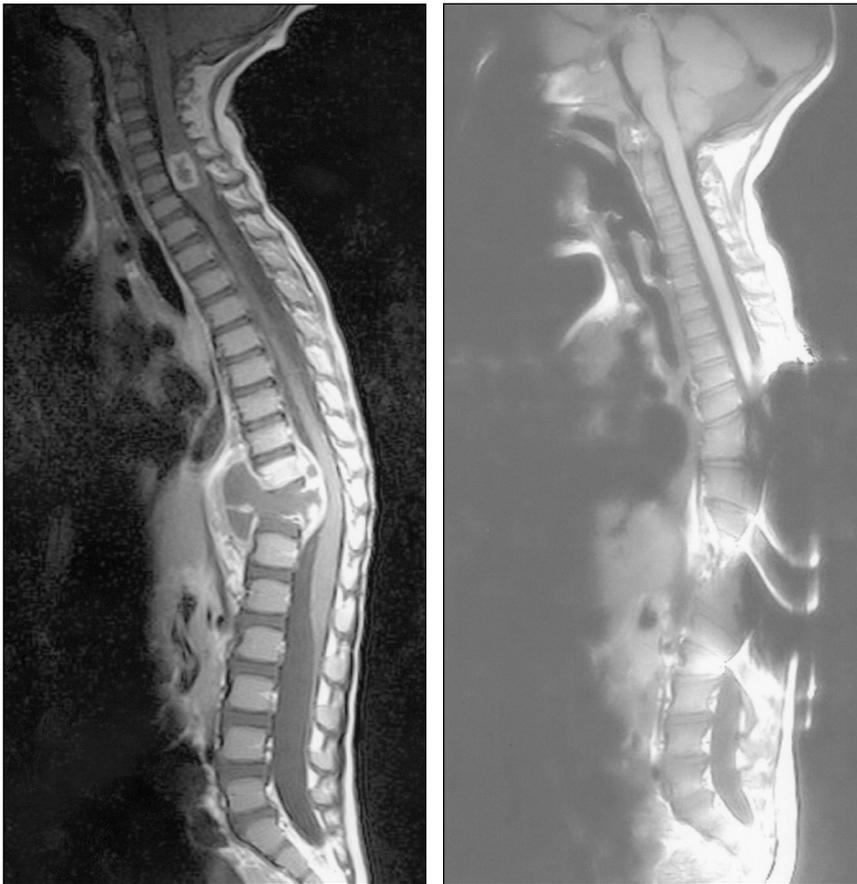


FIG. 3. Sagittal MRI studies with gadolinium enhancement. Pretreatment view (left) shows the tuberculoma; on the posttreatment view (right) there is almost complete resolution of the lesion.

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