

## CASE REPORT

### SPINAL GNATHOSTOMIASIS RESEMBLING AN INTRINSIC CORD TUMOR/MYELITIS IN A 4-YEAR-OLD BOY

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**Abstract.** A 4-year-old boy presented with fever, myalgia followed by progressive quadriparesis and urinary retention. Spinal fluid from a lumbar puncture showed 42 WBC/ $\mu$ l with 100% lymphocytes, no RBC, a glucose of 54 mg/dl (blood glucose 107 mg/dl), and a protein of 39 mg/dl. The cerebrospinal fluid culture was negative. His white blood cell count was 10,860 cells/ $\mu$ l with a normal differential count. An MRI of the brain was negative. An MRI of the whole spine showed fusiform dilatation of the cervical cord from the cervicomedullary junction to the T4 level. The tentative diagnosis was acute hemorrhage of an intrinsic cord tumor versus acute myelitis. Intravenous dexamethasone was administered which resulted in a slight improvement in strength. One week later, he deteriorated precipitously and became flaccidly quadriplegic. Since the patient deteriorated rapidly and no definitive diagnosis was made, the patient underwent cervical cord biopsy. Intraoperatively, after the cervical cord had been opened, a living *Gnathostoma spinigerum* was found in the spinal cord parenchyma. The nematode was removed. Following the operation the patient was placed on albendazole 400 mg/d and metronidazole 250 mg three times per day for 3 weeks. He gradually improved over the next several weeks.

#### INTRODUCTION

Gnathostomiasis is caused by ingestion of undercooked fish or meat harboring the third stage larva of *Gnathostoma spinigerum*. The diagnosis is usually made by a history of raw food ingestion, localized migratory swellings of the skin, peripheral eosinophilia combining with an immunological study. In rare cases, the diagnosis is made by retrieval of the larva from the eye or subcutaneous tissue. We present a first case report in the literature of retrieval of *Gnathostoma spinigerum* from the

spinal cord during the operation.

#### CASE REPORT

A previously healthy 4-year-old boy presented to a local medical office with several days of fever and myalgia. These symptoms then subsided. The following week, he again developed high fever and severe myalgia without other associated symptoms. Subsequently, his arms and legs became increasingly weaker. At the local hospital, he was afebrile and hemodynamically stable. He was awake and coherent. His strength was 4/5 on the right side and 3/5 on the left side. The patient responded to pinprick but could not reliably indicate the location. The reflexes in all extremities were decreased. He also had urinary retention. The rest of physical and neu-

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rological examinations were unremarkable. Spinal fluid from lumbar puncture showed a WBC of 42 cells/ $\mu$ l, (lymphocytes 100%), no RBC, a glucose of 54 mg/dl (blood glucose of 107 mg/dl) and a protein of 39 mg/dl. The cerebrospinal fluid culture was negative. The urine culture was also negative. A CT of the brain was normal. Serum electrolytes were normal. On hospital Day 5 his motor power decreased to 3/5 on the right and 2/5 on the left; he was then transferred to our hospital for further management. At our hospital, he was afebrile, awake and alert. His blood WBC count was 10,860 cells/ $\mu$ l with a normal differential count. An MRI of the brain was negative. An MRI of the whole spine showed fusiform dilatation of cervical cord from the cervicomedullary junction to the T4 level (Fig 1, 2). The tentative diagnosis was acute hemorrhage of an intrinsic cord tumor versus acute myelitis. Intravenous dexamethasone was administered which resulted in slight improvement in motor strength. One week later he deteriorated precipitously and became flaccidly quadriplegic. The rest of the physical examination was unremarkable. As the patient rapidly deteriorated and no definitive diagnosis could be reached, the patient underwent cervical cord biopsy. Intraoperatively, after the cervical canal and cord were opened, a living nematode was found in the spinal cord parenchyma (Fig 3). Morphological examination revealed a male *Gnathostoma spinigerum*. Pathological examination of the surrounding spinal cord tissue revealed acute and chronic inflammation with abscess formation. Post-operatively the patient was placed on albendazole 400 mg/d and metronidazole 250 mg three times per day for 3 weeks. He gradually improved over the next several weeks. His motor strength improved to grade 4/5.

## DISCUSSION

Gnathostomiasis, an endemic zoonosis found in Southeast Asia, Japan, Mexico, and



Fig 1—Sagittal T1W (upper), T1W with contrast (middle), T2W (lower) MRI of spine showing fusiform dilatation of the cervical cord from the cervicomedullary junction to the T4 level.



Fig 2—Axial T1W (upper), T2W (lower) MRI showed enlargement of the spinal cord.

South America, is transmitted to humans by ingestion of undercooked fish. Once ingested, the third stage larva can migrate to various organs, including most devastatingly, the central nervous system. Typical presentations include a history of raw food ingestion, localized migratory swellings of the skin, peripheral eosinophilia combining with a positive immunological study (Daengsvang, 1981; Schmutzhard *et al*, 1988; Ogata *et al*, 1998; Chandenier *et al*, 2001; Hughes and Biggs, 2002; Gorgolas *et al*, 2003; Hale *et al*, 2003;



Fig 3—After the spinal cord was opened, a living nematode was found (arrow). Marked inflammation of the surrounding spinal cord parenchyma was also noted (double arrows).

Moore *et al*, 2003; Ligon, 2005; Hennies *et al*, 2006). On rare occasions the diagnosis is made by recovering the nematode from the eye or skin (Gorgolas *et al*, 2003; Basak *et al*, 2004; Magana *et al*, 2004; Barua *et al*, 2007).

This patient presented with acute spinal cord syndrome without other typical features suggestive of gnathostomiasis. Although MRI findings of spinal cord gnathostomiasis are not specific, those described in the literature include enlargement of the spinal cord, hyperintensity on a T2 weighted image, and an iso-hypersignal intensity on a T1 weighted image. There may be a hemorrhagic tract or some enhancement of the spinal cord following gadolinium injection (Chandenier *et al*, 2001; Sawanyawisuth *et al*, 2004, 2005).

Spinal gnathostomiasis may clinically/radiographically mimic intrinsic cord tumor and acute myelitis. Thus, it should be in the differential diagnosis of patients with acute spinal cord syndrome, particularly in the endemic areas.

To the best of our knowledge, this is the first reported case of recovering a living *Gnathostoma spinigerum* from the spinal cord.

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