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Case Report

Intradural cavernous lymphangioma of the thoracic spine: case report, technical considerations, and review of the literature

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Abstract

BACKGROUND CONTEXT: Cavernous lymphangioma is a rare slow-growing tumor that can cause neurologic compromise when it involves the central nervous system. Involvement of the spinal column is rare but may involve the osseous structures or the epidural space of the spinal column.
PURPOSE: We report the first case of an intradural, extramedullary cavernous lymphangioma involving the thoracic spinal cord.
METHODS: An 83-year-old woman presented with progressive gait ataxia, bilateral lower extremity weakness, and a band-like sensation in the middle and lower thoracic dermatomes. Magnetic resonance imaging of the thoracic spinal cord revealed hyperintensity on T2 and enhancement of an intradural cystic mass along the dorsal aspect of the T5–T8 levels with significant compression of the spinal cord.
RESULTS: Complete surgical resection was difficult owing to the adherence of the tumor to the pial surface and microvasculature of the thoracic spinal cord. Recurrence of the mass was ultimately treated with cystic fluid diversion into the peritoneum. At her follow-up visit after 28 months, the patient was able to ambulate with minimal assistance. A comparative literature review is presented. There are no reports of intradural thoracic spinal cord involvement in the literature.
CONCLUSIONS: Intradural cavernous lymphangioma of the spine poses a unique surgical challenge for complete resection. Cystic fluid diversion appears to be a viable treatment option with lasting benefit if complete resection is not achieved.

Introduction

Lymphangiomas are benign, slow-growing soft-tissue tumors with predilection for the head and neck but may involve any organ. Lymphangiomas are classified into congenital or acquired forms and histologically divided into cavernous, capillary, or cystic type [1–9]. Tumor involvement of the spinal column is exceedingly rare and presents either as a primary osseous tumor or as a soft-tissue tumor in the epidural space [3,5,8,10–13]. When lymphangiomas involve the epidural space, they usually occur as an isolated mass or an extension from a primary mediastinal lesion. Treatment for symptomatic lymphangiomas of the spine requires complete surgical resection. Recurrence is common when residual disease is left behind [2,3,6,7,10]. We report the first case of an intradural, extramedullary cavernous lymphangioma (CL) involving the thoracic spinal cord.

Clinical Case Report

An 83-year-old right-handed woman with a history of shingles presented to the clinic with an 8-month history of an abnormal band-like sensation in the middle and lower thoracic dermatomes and progressive gait ataxia requiring a walker for ambulation. Physical examination was significant for 5 of 5 bilateral lower extremity strength, decreased proprioception in the left lower extremity, positive Romberg sign, decreased sensation over the mid-thoracic dermatomes, and 3+ bilateral patellar and Achilles tendon reflexes.

Initial magnetic resonance imaging (MRI) of the thoracic spine revealed an intradural, extramedullary cystic lesion along the dorsal aspect of the T5–T8 levels with significant compression of the thoracic cord (Fig. 1A and B). Systemic imaging included a positron emission tomography scan and

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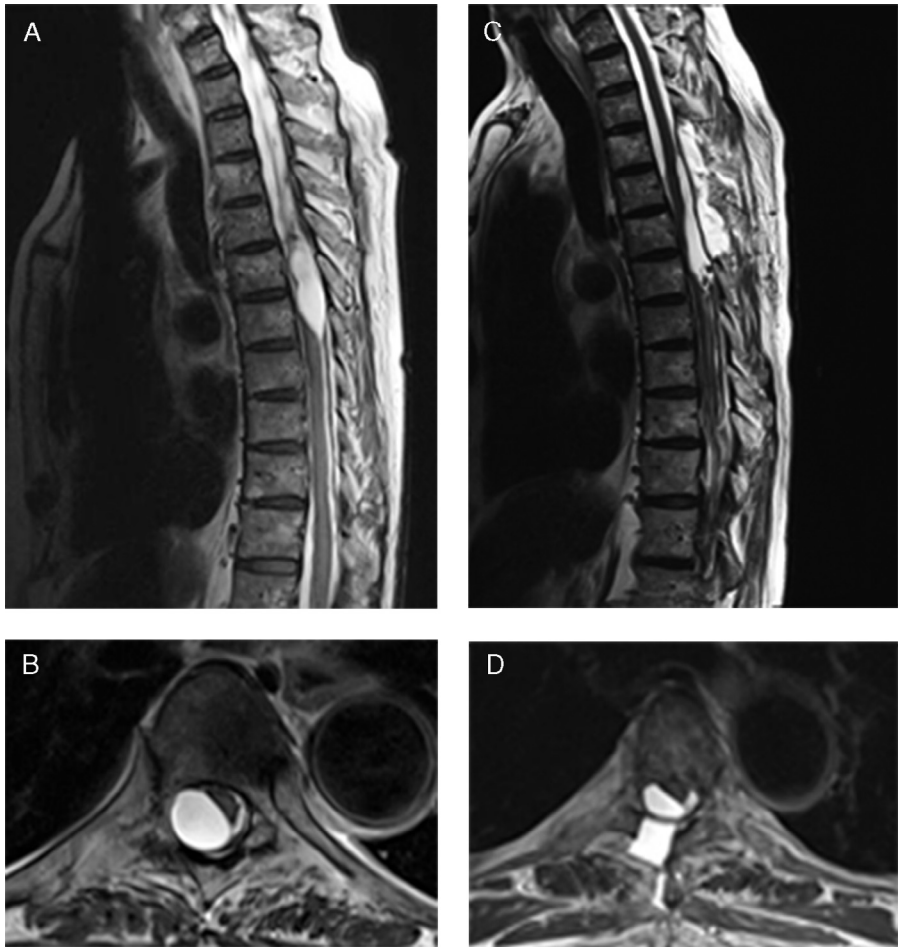

Fig. 1. Pre- and postoperative magnetic resonance imaging (MRI) of the thoracic spine. T2-weighted preoperative (A) sagittal and (B) axial images demonstrating a cystic intradural mass at the T5–T8 levels with cord compression. Postoperative (C) sagittal and (D) axial images demonstrating a right-side laminectomy and durotomy with partial decompression of the spinal cord.

computed tomography of the chest, abdomen, and pelvis, which were unrevealing for a contributing disease process.

The patient underwent a T5–T8 laminectomy and dural opening for lesion resection. Intraoperative neuromonitoring included somatosensory evoked potentials, motor evoked potentials, and electromyographic modalities. During surgical exploration, the cyst was found to be dark in color with a thick and uniform fibrous capsule. The capsule was incised, revealing opaque fluid with no septations. Under the microscope, sharp dissection was used for successful release of the dural surface of the tumor. However, attempts at dissecting the capsule from the pial surface proved problematic as the microvasculature of the pia and the capsule were intimately involved. As such, residual capsule was left attached to the pial surface (Fig. 1C and D). There were no electrophysiological changes noted intraoperatively. Final pathologic review was consistent with CL (Fig. 2A and B).

At her fourth week follow-up, the patient was able to walk independently but continued to experience decreased proprioception in the left lower extremity and hyperactive (3+) reflexes in both her patellar and Achilles tendons. However, at 4 months, she presented with new onset weakness (3 of 5) in dorsiflexion of the left lower extremity, a positive Romberg sign, and progressive gait ataxia requiring a walker. Repeat MRI done at this time revealed accumulation of fluid within the cystic mass, now extending between T5 and T9 with compression of the thecal sac (Fig. 3, Left). The patient returned to surgery for another incomplete resection (Fig. 3, Middle). Immediately after surgery, her dorsiflexion strength (5 of 5) and gait ataxia improved, although she continued to require a walker for ambulation.

Three months later, she presented again with progressive neurologic decline. Examination showed bilateral lower extremity strength of 4 of 5 with severe ataxia requiring a wheelchair for mobility. Repeat MRI revealed tumor recurrence with cord compression (Fig. 3, Right). Given the large cystic component of the mass, the patient was taken back to the operating room for placement of a cystoperitoneal shunt. Her postoperative course was consistent with slow neurologic improvement. At her follow-up visit 9 months later, lower extremity strength was 5 of 5 and she was ambulating with a walker. During her latest follow-up visit 28 months after