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Case Report

Exclusively epidural spinal metameric arteriovenous shunts: case report and literature review

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Abstract

BACKGROUND CONTEXT: Spinal arteriovenous metameric syndrome (SAMS) is a subgroup of spinal arteriovenous malformations (AVMs). Most SAMS cases have intra- and extradural AVMs and suffer from hematomyelia, subarachnoid hemorrhage, or venous congestive myelopathy.
PURPOSE: To present a rare case of SAMS in which spinal AVMs were exclusively epidural. We reviewed previous literature and evaluated the feasibility of a treatment strategy using endovascular interventions, followed by surgical obliteration.
STUDY DESIGN: A case report and literature review of SAMS.
METHODS: We report a case of a 15-year-old boy suffering from SAMS in which epidural venous ectasia because of extradural AVMs caused spinal cord compression.
RESULTS: The patient was successfully treated with multiple sessions of transarterial embolization followed by open surgery. After the treatment, his neurologic deficits resolved. Postoperative angiography confirmed complete obliteration of extradural AVMs.
CONCLUSIONS: Although exclusively epidural spinal AVM is an uncommon type of SAMS, combined endovascular and surgical interventions can be an effective treatment for AVMs to achieve better radiologic outcomes and complete resolution of patient symptoms.

Keywords: Arteriovenous malformation; Cobb syndrome; Epidural arteriovenous shunt; Endovascular treatment; Metameric; Myelopathy; Spinal cord

Introduction

Spinal arteriovenous metameric syndrome (SAMS) is a rare clinical form of spinal arteriovenous malformation (AVM) whose hallmarks include spinal vascular malformations and skin nevi in the same dermatome [1–3]. Spinal arteriovenous metameric syndrome is also termed Cobb syndrome based on a case described by Stanley Cobb in 1915 [4]. Most patients with SAMS suffer from intradural spinal vascular pathology [5], which is consistent with the fact that it was classified as "*extradural-intradural arteriovenous malformations*" in one of the most widely used classification schemes [6].

Here we report an unusual case of SAMS exclusively involving epidural spinal AVM. The patient suffered myelopathy because of midthoracic spinal cord compression. Multiple sessions of endovascular interventions followed by surgical obliteration of extradural arteriovenous shunts (AVSs) led to complete regression of the epidural venous engorgement and a good clinical outcome.

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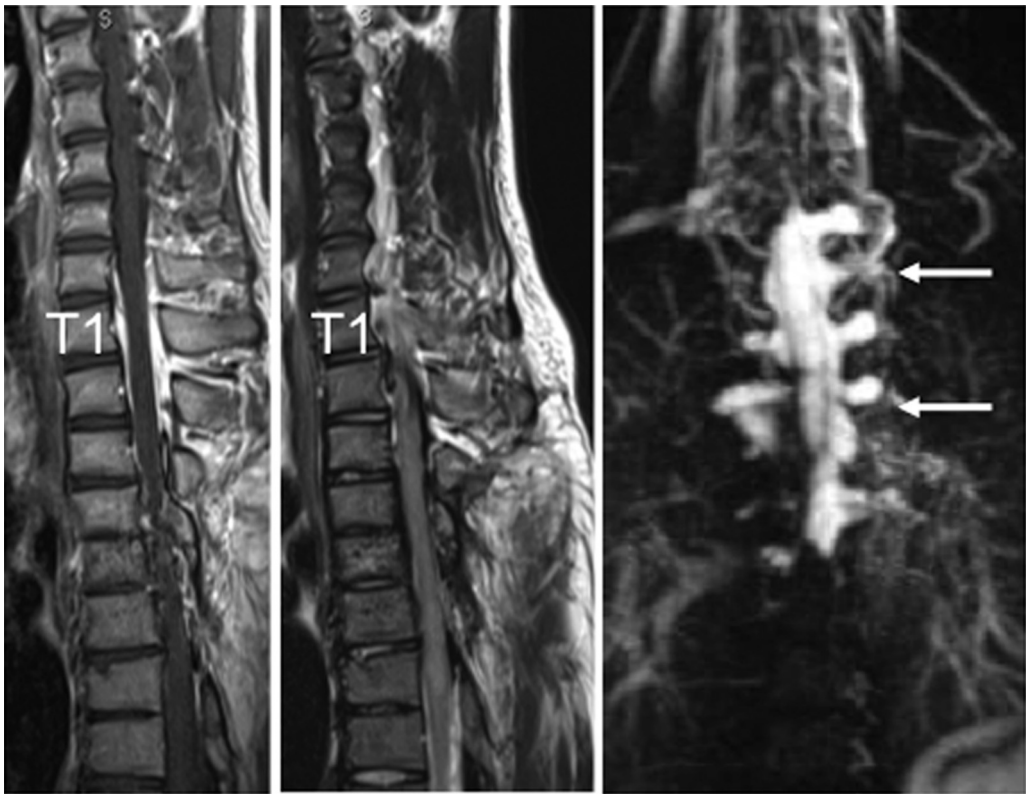

Fig. 1. Magnetic resonance image of the patient's cervical and thoracic spine on admission. (Left) T1-weighted gadolinium enhanced sagittal image showing enlargement of the epidural venous plexus from C7 to T3. (Middle) T2-weighted sagittal image showing no abnormal flow void along the spinal cord surface. There was no T2 high-intensity area in the spinal cord. (Right) Magnetic resonance angiogram demonstrating ectasia of the epidural venous plexus (arrows). ''T1'' indicates (Left and Middle) the vertebral level.

Case report

Presentation

A 15-year-old boy with a history of acute thoracic (T4–T7) epidural hematoma when he was 1 year old was admitted to and treated at our department. At 1 year of age, he had developed acute flaccid paraparesis and the hematoma was surgically resected. According to his record, no vascular malformation was confirmed during the surgery. Although the patient was followed for 5 years postoperatively, angiography was not performed. No spinal or paraspinal vascular anomaly was detected in magnetic

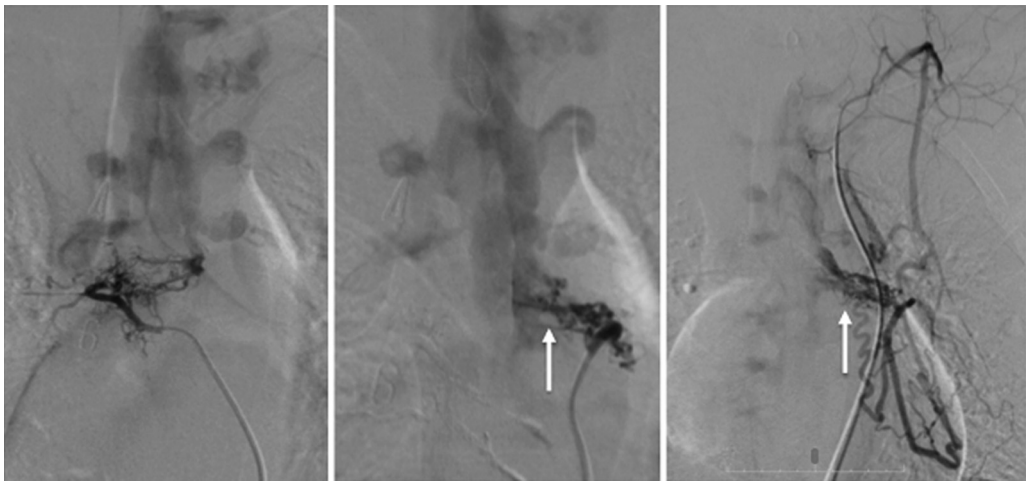

Fig. 2. Preoperative digital subtraction spinal angiograms. Selective angiograms through (Left) right T5 intercostal, (Middle) left T5 intercostal, and (Right) left descending scapular arteries. Epidural arteriovenous shunts were along the left T5 root sleeve (arrows). (Left and Middle) An enlarged epidural venous plexus was also apparent.