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Case Report

High cervical spinal subdural hemorrhage as a harbinger of craniocervical arteriovenous fistula: an unusual clinical presentation

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Abstract

BACKGROUND CONTEXT: Craniocervical dural arteriovenous fistula (dAVF) is rare as compared with the typical thoracolumbar dAVFs of the spine and usually presents with hemorrhagic manifestation, predominantly intracranial subarachnoid hemorrhage.

PURPOSE: We describe the first case of craniocervical dAVF with initial presentation as neck pain and spinal subdural hemorrhage.

STUDY DESIGN: Case report.

METHODS: We present the case of a 59-year-old woman who presented with sudden onset of neck pain at an outside institution emergency department (ED) and was discharged after negative cervical spine radiographs. Magnetic resonance imaging of the cervical spine performed because of persistent pain demonstrated presence of high cervical spinal subdural hematoma and she was managed conservatively. She subsequently presented to our ED a week later with headache and was found to have an intraventricular hemorrhage on computed tomography scan of the head, which on subsequent workup with an angiography revealed the presence of a craniocervical dAVF.

RESULTS: Surgical obliteration of the fistula was performed with use of intraoperative angiography as an adjunct to confirm complete fistula obliteration. She had an excellent clinical outcome with no deficits at her last follow-up at 9 months.

CONCLUSIONS: Even though hemorrhagic presentation is fairly common in craniocervical dAVFs, there is no report of a craniocervical dAVF presenting with spinal subdural hemorrhage. The present case further highlights the propensity of these vascular lesions to bleed and emphasizes the clinical importance of including these lesions in the differential diagnosis of hemorrhage in the vicinity of foramen magnum region, whether subarachnoid or subdural in location. Physicians treating spinal pathologies should be aware of this entity and clinical presentation, as an angiography needs to be considered in these cases to direct appropriate referral and treatment.

Keywords: Craniocervical junction; Spine; Dural arteriovenous fistula; Perimedullary arteriovenous fistula; Subarachnoid hemorrhage; Subdural hemorrhage; Vascular disorders

Introduction

Dural arteriovenous fistula (dAVF) accounts for most cases of spinal arteriovenous malformation and most commonly occurs in the thoracolumbar region [1,2]. Hemorrhagic presentation is uncommon with dAVFs, which classically manifest as progressive congestive myelopathy secondary to stagnation of the venous outflow from the spinal cord, resulting in intramedullary venous hypertension and ischemic insult to the spinal cord [1–4]. In contrast, dAVFs located in the craniocervical region are rare, accounting for only up to 2% of such cases; albeit they fairly

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commonly present with hemorrhage [4–8]. Subarachnoid hemorrhage (SAH) remains the most common mode of presentation of craniocervical/high cervical dural and perimedullary AVFs and is reported to occur in up to 45% of cases, which forms the basis of obtaining vascular imaging of the neck to rule out high cervical vascular pathologies such as craniocervical dAVF in case of negative cerebral angiogram in a patient with SAH [4,6,9–12]. Spinal subdural hemorrhage (SDH) is less common as compared with cranial SDH, and usually occurs as a result of trauma, iatrogenic secondary to lumber puncture, or rarely spontaneously in cases of hematological disorders, tumors, or anticoagulant use [13]. No case of cervical spinal SDH secondary to a craniocervical dAVF has been reported earlier as per our review of the literature. Considering the common hemorrhagic presentation of dAVF involving the craniocervical region, the presence of high cervical spinal SDH should lead to a suspicion and appropriate workup to unravel the presence of craniocervical dAVF, as it can be a sentinel event before the usual clinical presentation as an SAH or an intraventricular hemorrhage (IVH), as happened in the present case. We report a case of craniocervical dAVF that presented with neck pain. The patient rebled and sustained an IVH. Further workup revealed presence of ventral spinal SDH, which was subsequently diagnosed to be secondary to a craniocervical dAVF.

Case report

A 59-year-old woman with significant past medical history of breast carcinoma presented with sudden onset of neck pain. She was evaluated at an outside institution emergency department and was sent home after negative cervical spine radiographs. Because of persistent neck pain over the next few days, she underwent magnetic resonance imaging (MRI) of the cervical spine, which revealed presence of a ventral high cervical spinal subdural collection with signal intensity suggestive of a small subdural hematoma (Fig. 1). In view of previous history of breast carcinoma in the past, meningeal metastasis was suspected and she underwent contrast MRI of the spinal column and brain, along with magnetic resonance angiography of the brain to rule out any vascular anomaly. All were negative apart from the presence of ventral spinal SDH. She subsequently presented to our emergency department a couple of days later with worsening headaches and a computed tomography scan showed the presence of an isolated IVH involving the left occipital horn with no evidence of SAH (Fig. 2). A digital subtraction angiography was performed, which showed the presence of an early draining vein following left vertebral artery injection, suggestive of a craniocervical dAVF supplied by meningeal branches of radicular artery originating from the left vertebral artery with no evidence of an aneurysm or intracranial arteriovenous malformation (Fig. 3). A transcondylar far lateral approach was performed with clipping of the arteriovenous fistula. Intraoperative angiogram was performed during surgery to confirm the obliteration of AVF. The patient had an uneventful postoperative course and was discharged home with no neurologic deficits. She was doing well at 9 months follow-up with an intact neurologic examination and a modified Rankin scale score of 0.

Discussion

Spinal arteriovenous malformations are rare vascular lesions with spinal dAVFs being the most common type [2]. Classically, a spinal dAVF is located in the thoracolumbar region, has an arteriovenous shunt within the dural root sleeve fed by the radiculo-meningeal arteries, drains through a single radicular vein, and generally presents in

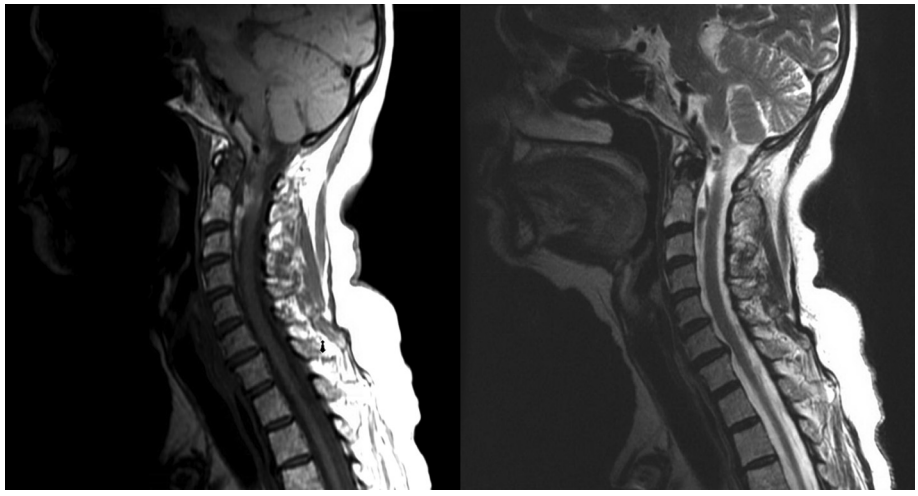

Fig. 1. Sagittal T1-weighted (Left) and T2-weighted (Right) magnetic resonance image of the cervical spine showing presence of a ventral high cervical spinal subdural collection with signal intensity suggestive of a subacute subdural hematoma.