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CASE REPORT

Subarachnoid hemorrhage in neurofibromatosis type 1: Case report of extracranial cerebral aneurysm rupture into a meningocele

Hémorragie sous-arachnoïdienne dans la neurofibromatose de type 1 : rupture d'un anévrysme extracrânien dans un méningocèle

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KEYWORDS
Subarachnoid hemorrhage;
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Spine malformation;
Cervical meningocele;
Neurofibromatosis type 1

Summary Described here is a case of subarachnoid hemorrhage due to rupture of an extracranial vertebral artery (V3 segment) aneurysm in a patient with neurofibromatosis type 1 (NF-1). The pathophysiology of this never-before reported complication of NF-1 is examined in the light of a focused literature review and with illustrations characteristic of this unique case, involving complex malformations of the spine and meningeal spaces, as well as of the vertebral artery wall itself. All these abnormalities are directly related to the underlying NF-1 disease.
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Introduction

von Recklinghausen's neurofibromatosis type 1 (NF-1) is characterized by cutaneous neurofibroma and so-called 'café-au-lait' spots. Vascular complications are not unusual, and most usually affect the kidney, gastrointestinal tract and heart vessels. However, cerebrovascular complications are rare, while many cases of stenosis, occlusion, aneurysm and arteriovenous malformations have been reported in the literature. The present case report is of a young woman with NF-1 and subarachnoid hemorrhage caused by rupture of an extracranial vertebral artery aneurysm into a giant cervical meningocele.

Case report

A 36-year-old woman with NF-1, but only cutaneous clinical symptoms such as multiple café-au-lait spots, freckles

Abbreviations: CT, computed tomography; LVA, left vertebral artery; MRA, magnetic resonance angiography; MRI, magnetic resonance imaging; NF-1, neurofibromatosis type 1.

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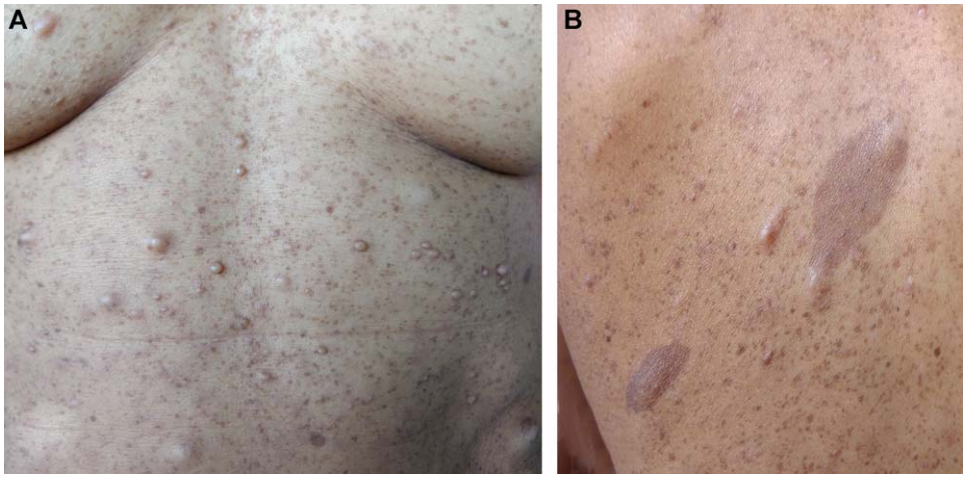

Figure 1 Thoracic lesions of the patient include (A) numerous cutaneous neurofibromas and freckles, and (B) so-called café-au-lait spots.

and numerous neurofibromas of the skin (Fig. 1), attended the emergency ward 12 h after the acute onset of violent and unusual headache, and left cervical neck pain and stiffness, followed by photophobia and vomiting without fever. Immediate brain computed tomography (CT) was normal, but the subsequent spinal tap disclosed subarachnoid hemorrhage. Brain magnetic resonance imaging (MRI) and magnetic resonance angiography (MRA) showed bilateral cortical post-central subarachnoid hemorrhage with neither proximal aneurysm nor cerebral venous thrombosis. Cervical MRA disclosed a branching left extracranial vertebral aneurysm of the V3 segment (Fig. 2). MRI and cervical CT disclosed dural ectasias and spinal malformations, with severe scalloping of the adjacent vertebrae (Figs. 3 and 4a). Cervical CT and CT angiography confirmed the presence of a voluminous extracranial aneurysm, measuring 17 × 13 mm, of the left vertebral artery. The cervical spinal cord was distorted by dural ectasias together with spinal subluxations associated with merging of vertebrae C2 to C5, abnormal vertebral bodies and aplasia of the left posterior arches of C3 to C5 (Fig. 4a, b). There was also an anterior meningocele extending from the left C3—C4 foramina close to the aneurysm. Although it was not possible to demonstrate blood in the meningocele, its close proximity to the aneurysm together with the dural ectasia and the negative findings in the rest of the workup led to the conclusion that rupture of the aneurysm into the meningocele was the cause of the subarachnoid hemorrhage (Fig. 4c, d).

Two days later, the patient experienced acute cervical pain that radiated into the left arm along the C5—C6 dermatome pattern, and was associated with left cervical swelling. Clinical examination revealed proximal motor deficit of the left arm, with abolition of the bicipital reflex. CT scan disclosed a twofold increase in the size of the aneurysm, with thickening of the surrounding soft tissue (Fig. 4 c). Conventional angiography (by Professor J. Moret, Neuroradiology Department, Adolphe

Figure 2 Cervical MRA shows a left vertebral artery (V3 segment) branching giant aneurysm (black flower).

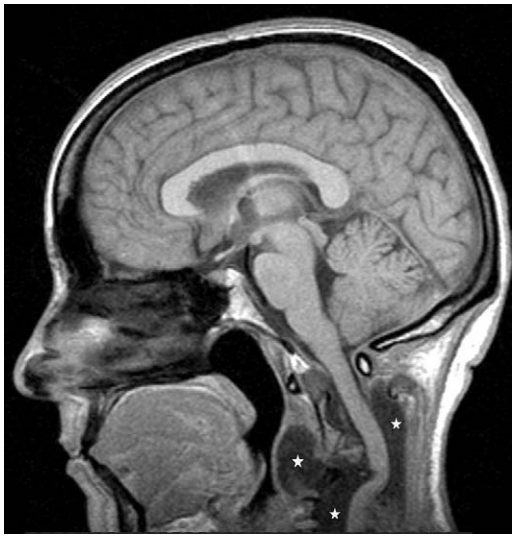

Figure 3 Sagittal T1-weighted MRI scan shows major cervical spinal cord deformation, vertebral abnormalities and meningeal ectasias (white stars = meningoceles).

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