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Intracranial hypotension syndrome following chiropractic manipulation of the cervical spine

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Abstract Cervical spine manipulation has been associated with several disorders such as cervical arteries dissection, but rarely has a relationship with intracranial hypotension been reported. We describe a patient showing intracranial hypotension syndrome following chiropractic cervical spine treatment. Magnetic resonance showed the presence of dural leakage at cervical level, suggesting the pathogenesis of the syndrome. We state that cervical spine manipulation should be considered a treatment with risk of neurological complications, including the occurrence of intracranial hypotension.

Key words Chiropractic •
Intracranial hypotension syndrome •
Headache

Introduction

Intracranial hypotension is an uncommon syndrome characterised by orthostatic headache, low cerebral spinal fluid (CSF) pressure and magnetic resonance imaging findings of diffuse pachymeningeal gadolinium enhancement. The aetiology of intracranial hypotension is not fully understood and often remains undetermined. When there is not a history of neck trauma or lumbar puncture it is defined “spontaneous”.

In some cases CSF leakage from spinal meningeal diverticula or dural tears may be involved. Recently, defects of the dura and abnormalities of connective tissue have been considered as predisposing factors to CSF leak, suggesting that an underlying connective disorder may lead to dural weakness [1]. Spinal manipulation has recently been associated with dissection of cervical arteries [2, 3], epidural haematoma [4], intracranial aneurysm [5], myelopathy and radiculopathy [6], but rarely has an association with intracranial hypotension syndrome been reported [7–9].

Case report

A 49-year-old man without a history of headache, other diseases or neck trauma consulted a chiropractor for neck pain and underwent sudden rotatory neck manipulation and hyperextensive head movements with poor benefit. The following day the patient presented with a severe and throbbing headache. The pain involved especially the frontal regions, increased in an orthostatic position, was associated with tinnitus and improved in a clinostatic position. The patient did not suffer other symptoms. General and neurological examination, ECG and blood exams were normal. Cervical arteries duplex scanner and transcranial Doppler were normal. Non-contrast cranial CT was negative. Magnetic resonance T1- and T2-weighted images obtained in the axial plane revealed a thin fluid extra-axial collection and a fronto-parietal pachymeningeal enhancement was appreciable after contrast medium administration (Fig. 1). These reports were indicative of intracranial hypotension. Magnetic resonance images of the total spine performed with myelographic sequences documented an arachnoid cyst of the third cervical root and suggested the presence of a dural leakage (Fig. 2). Magnetic resonance images of the dorsal and lumbar spine were normal. Skin biopsy did not show ultrastructural connective abnormalities. The patient was

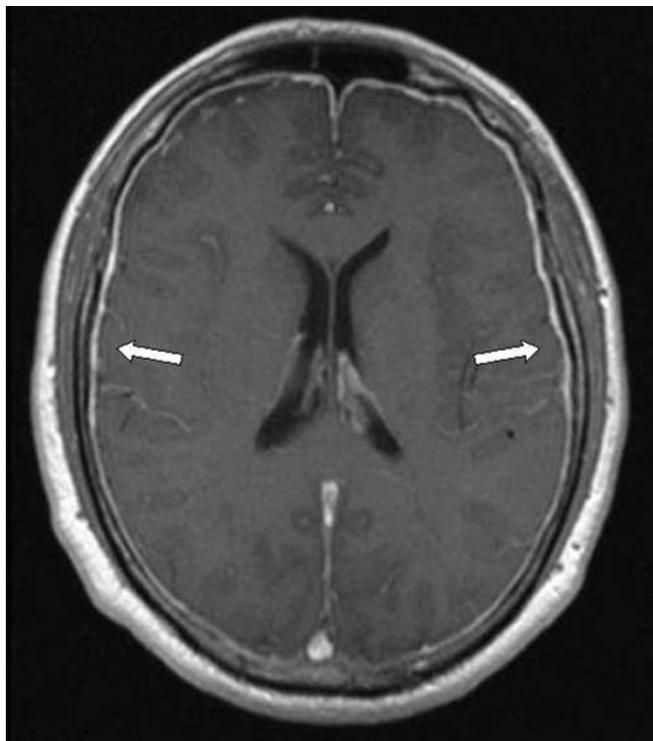


Fig. 1 Contrast-enhanced axial MR images of the brain showing diffuse pachymeningeal enhancement (*arrows*)

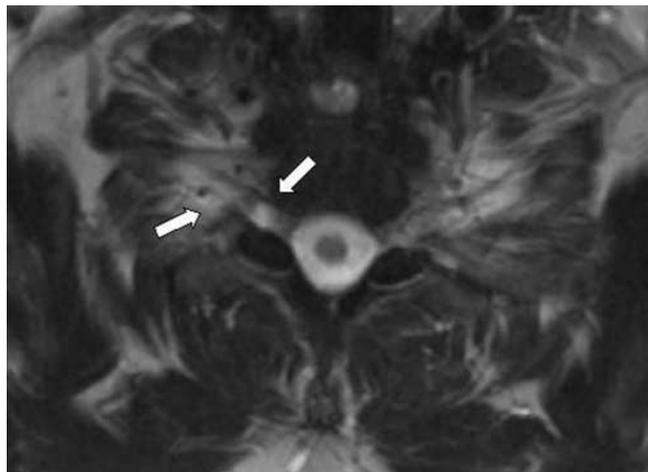


Fig. 2 Contrast-enhanced axial MR images of the cervical spine show an arachnoid cyst of the third cervical root suggesting the presence of a dural leakage (*arrows*)

treated with intravenous rehydration and headache improved gradually over the following days. Three months later cranial and cervical spine magnetic resonance images showed the disappearance of the pachymeningeal enhancement and of the dural CSF leak at the level of the third cervical root. History, clinical picture and neuroimaging findings allowed diagnosis of CSF fistula headache (code 7.2.2) according to International Headache Society criteria [10].

Comment

This unusual case documents that intracranial hypotension syndrome may follow cervical spine manipulation. Mechanical disruption of the spinal dural thecal sac with subsequent loss of CSF seems to be the main pathophysiological mechanism. One hypothesis is that fragility of the spinal meninges at the level of the radicular nerve root sleeves predisposes to the formation of meningeal diverticula or to dural tears following a traumatic event, often misdiagnosed. Spine manipulation may be considered a trigger factor, even if it is debated how a mild external force generated by hands could tear a healthy dura. Nevertheless it is known that “spontaneous” intracranial hypotension, like cervical artery dissection, may be associated with an underlying hereditary disorder [11]. Recently, in fact, abnormal or deficient fibrillin, elastin or both in dermal fibroblast cultures of patients diagnosed with spontaneous CSF leaks and connective disorders have been described [1], but our case did not document connective tissue abnormalities. In this case spinal chiropractic manipulation preceded symptom onset by a few

hours, in the absence of a history of head or neck trauma. Cervical magnetic resonance imaging suggested the presence of a dural leakage and did not show meningeal diverticula or other anatomical abnormalities. Moreover, magnetic resonance imaging of the spinal axis excluded the presence of other sites of CSF loss, suggesting that the

dural tear in the cervical spine was related to intracranial hypotension. We state that cervical spine manipulation should be considered a treatment at risk of neurological complications including intracranial hypotension occurrence. Intracranial hypotension syndrome must be added to the list of the differential diagnosis in case of headache following manipulation of the spine [7].

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