

SPONTANEOUS INTRACRANIAL HYPOTENSION AND RECURRENT BRAINSTEM ISCHEMIC STROKE

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ABSTRACT

Objective: To present a rare case of spontaneous intracranial hypotension and recurrent brainstem ischemic stroke. **Case description:** A case is reported of a 74-years-old man with permanent atrial fibrillation on vitamin K antagonist anticoagulation presented with confusion, left internuclear ophthalmoplegia and worsening of previous orthostatic headache. Brain and spine images showed left paramedian mesencephalic acute infarction and signs compatible with CSF hypotension. The patient evolved with subdural hematoma rebleeding requiring surgical evacuation. A new incidental pontine infarction on the right paramedian pons was revealed in a follow-up MRI seven days after surgery. **Conclusions:** To our knowledge, this is the first case reported of recurrent brainstem infarctions prior and after subdural hematoma evacuation with excellent clinical outcome, which illustrates a rare but potentially fatal complication of intracranial hypotension.

Keywords: Stroke; Subdural hematoma; Intracranial hypotension; Hidden dural fistula; Blood patch.

HIPOTENSÃO INTRACRANIANA ESPONTÂNEA E ACIDENTE VASCULAR CEREBRAL ISQUÊMICO RECORRENTE DO TRONCO CEREBRAL

RESUMO

Objetivo: Apresentar um caso raro de hipotensão intracraniana espontânea e acidente vascular cerebral isquêmico recorrente do tronco cerebral. **Descrição do caso:** Relata-se um caso de um homem de 74 anos com fibrilação atrial permanente em anticoagulação com antagonista da vitamina K, que apresentava confusão, oftalmoplegia internuclear esquerda e pioria da cefaléia ortostática prévia. As imagens do cérebro e da coluna vertebral mostraram infarto agudo do mesencéfalo paramediano esquerdo e sinais compatíveis com hipotensão no LCR. O paciente evoluiu com ressangramento subdural de hematoma, necessitando de evacuação cirúrgica. Um novo infarto pontino incidental na ponte paramediana direita foi revelado em uma ressonância magnética de acompanhamento sete dias após a cirurgia. **Conclusão:** Até onde sabemos, este é o primeiro caso relatado de infartos de tronco encefálico recorrentes antes e após a evacuação do hematoma subdural com excelente resultado clínico, que ilustra uma complicação rara, mas potencialmente fatal, da hipotensão intracraniana.

Palavras-chave: Acidente vascular cerebral; Hematoma subdural; Hipotensão intracraniana; Fístula dural oculta; Tamponamento sanguíneo peridural.

INTRODUCTION

Ischemic strokes are a rare complication in spontaneous intracranial hypotension (SIH), even more exceptional is recurrence.

In this report we present a patient with recurrent brainstem infarctions secondary to SIH, prior and after subdural hematoma evacuation.

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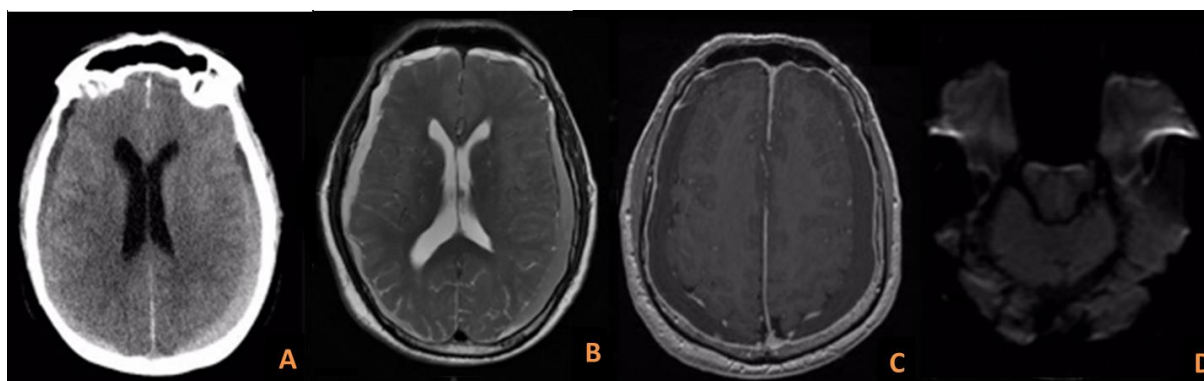
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CASE REPORT

A 74-years-old man with permanent atrial fibrillation on vitamin K antagonist anticoagulation presented to the emergency department 2 days after starting with confusion, diplopia and worsening of previous occipital headache. His family referred headache with orthostatic features and bilateral tinnitus for the past 3 months. They denied prior trauma or recent lumbar puncture.

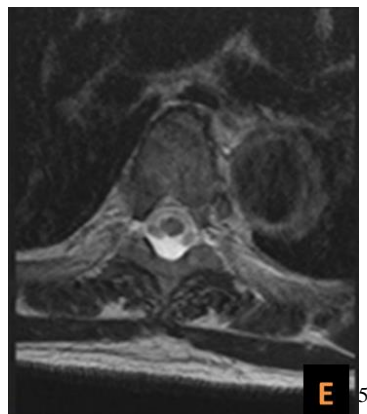
Physical examination upon admission revealed disorientation and left internuclear ophthalmoplegia (INO). A brain computed tomography (CT) demonstrated bilateral frontoparietal subdural hematomas (SDH), brain magnetic resonance imaging (MRI) confirmed the presence of SDH, cerebellar tonsillar ectopia, diffuse pachymeningeal enhancement and also revealed left paramedian mesencephalic acute infarction. [Fig A,B,C,D]. Angiography MRI, carotid duplex, transthoracic echocardiogram and 48 hour heart rhythm monitory were normal. International normalised ratio (INR) was 3.02.



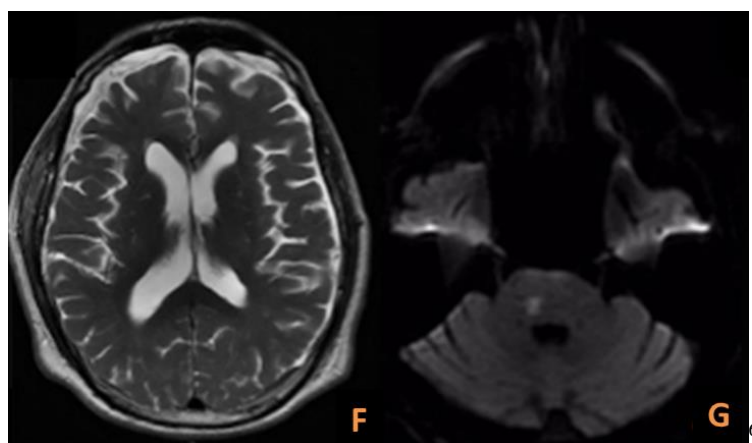
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The case was interpreted as SIH complicated with bilateral SDH and left mesencephalic ischemic stroke. Anticoagulation was stopped without reversal, and conservative treatment including strict supine positioning with ample hydration was started. Contrast-enhanced spinal MRI with myelographic technique showed multiple bilateral periradicular cysts at the cervico dorsal region but a CSF leak was not identified.[Fig E]

⁴A: Initial brain non-enhanced CT showing bilateral frontoparietal SDH hypodense with the adjacent cerebral cortex on the right side and isodense on the left side. B: Initial brain MRI (T2-WI) showing iso-hyperintense bilateral frontoparietal subdural hematomas. C: Initial brain MRI (contrast-enhanced T1-WI) showing bilateral hypointense frontoparietal subdural hematomas and diffuse pachymeningeal enhancement. D: Initial brain MRI (DWI) showing left paramedian midbrain acute infarction.



Six hours after admission the patient became stuporous with right mydriasis. A brain CT revealed SDH progression with signs of bilateral rebleeding. A new MRI confirmed these findings without changes in the ischemic brainstem lesion. A blind blood patch was performed as empirical treatment for a presumed hidden CSF fistula. After this, the patient underwent surgical evacuation of bilateral SDH. He evolved with complete recovery of sensorium and normal pupils. A follow-up MRI seven days after surgery revealed a new incidental DWI pontine lesion consistent with infarction on the right paramedian pons, which was asymptomatic, a marked reduction of bilateral subdural collections, resolution of cerebellar tonsils ectopia and persistence of pachymeningeal enhancement [Fig F,G]



Twelve days after admission he was discharged with left INO as his only deficit and with good brain expansion by CT [Fig. H].

⁵ **E:** Dorsal spine MRI axial T2-WI showing bilateral meningeal cysts.

⁶ **F:** Brain MRI (T2-WI, 7 days after SDH evacuation) showing absence of both hematomas. **G:** Brain MRI (DWI, 7 days after SDH evacuation) showing a new brainstem acute infarction during the postoperative period on the pons.



DISCUSSION

We report a case with SIH, bilateral subdural hematomas and recurrent brainstem infarcts. SIH is a relatively common condition (1-2,5 per 50.000 persons/year) [1] typically caused by cerebrospinal leakage, leading to a decrease in CSF volume. It is a clinically heterogeneous condition and its most common manifestation is orthostatic headache [2]. Some authors suggest that the sagging of the brain from the reduced CSF volume may result in traction of pain-sensitive structures leading to orthostatic headaches [2]. Asymptomatic cases diagnosed as incidental findings on MRI have been reported [3]. Less common manifestations include neck stiffness, nausea and vomiting, tinnitus, cranial neuropathies and altered mental status [1]

Underdiagnosed, this condition may lead to more severe complications as the development of SDH, with reported incidence ranging from 20% to 45%. [4,5] Conservative management, including bed rest, hydration and blood patch may be an effective first line treatment. Early surgical evacuation may be necessary in cases of symptomatic SDH to avoid irreversible herniation and death. SDH evacuation prior to control of the underlying CSF leak may result in clinical deterioration, SDH recurrence and stroke. [6]

Ischemic strokes have been reported previously in SIH, but they are rare. [3] Even more exceptional is the recurrence of stroke in this setting. This condition was previously described in a 54-year-old man who had two brainstem infarcts prior to SDH evacuation and one after the intervention. [7]

⁷ **H:** Brain non-enhanced CT (12 days postoperative) showing excellent cerebral expansion.

The exact mechanism of arterial cerebral infarcts in ISH remains unknown. It may be related to mechanical stretching of arteries due to downward displacement of the brain. SDH evacuation can also precipitate the event [8]. Prompt treatment of the underlying CSF leak and placement of the patient in Trendelenburg position may help to prevent cerebrovascular complications.

CONCLUSION

This case of recurrent cerebral infarctions prior and after SDH evacuation, illustrates a rare but potentially fatal complication of SIH.

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