Clinical note

Spontaneous intracranial hypotension syndrome: Contribution of radioisotope cisternography $\stackrel{\mbox{\tiny Ξ}}{\sim}$



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ABSTRACT

Spontaneous intracranial hypotension is a clinical syndrome caused by a loss of cerebrospinal fluid volume, usually secondary to leaking through structural defects of the spinal dura mater. Radioisotope cisternography (RC) can confirm the diagnosis of spontaneous intracranial hypotension, especially in doubtful or atypical case presentations. A retrospective study was conducted on 8 patients who underwent RC because spontaneous intracranial hypotension was suspected, and they presented with atypical clinical manifestations and/or inconclusive findings in other imaging techniques. RC detected paradural extravasation of cerebrospinal fluid in 7 patients. Moreover, there was indirect evidence of cerebrospinal fluid leaks in all 8 patients (early appearance of radioactivity in the bladder, soft tissue uptake of radioisotope and/or reduction in the amount of radiotracer in the brain at 24 h). RC had a significant impact on the diagnosis of 6 patients, and on the therapeutic management of 4 patients.

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Síndrome de hipotensión intracraneal espontánea: aportación de la cisternografía radioisotópica

RESUMEN

La hipotensión intracraneal espontánea es un síndrome clínico debido a la pérdida de volumen de líquido cefalorraquídeo, generalmente secundario a su fuga por defectos estructurales de la duramadre espinal. La cisternografía radioisotópica (CR) es una técnica que puede aportar un diagnóstico confirmatorio en este síndrome, principalmente en casos dudosos o de presentación atípica. Estudiamos retrospectivamente 8 pacientes con CR realizada por sospecha de hipotensión intracraneal espontánea con clínica atípica y/o hallazgos no concluyentes en las técnicas de imagen. La CR detectó extravasación paraespinal de líquido cefalorraquídeo en 7 de ellos. Además, se apreciaron signos indirectos de fuga de líquido cefalorraquídeo en los 8 pacientes, consistentes en la presencia precoz del radiotrazador en orina, presencia de actividad de fondo corporal y/o reducción de la cantidad de radiotrazador en cráneo a las 24 h. La CR tuvo un impacto significativo en el diagnóstico de 6 pacientes y en el manejo terapéutico de 4 pacientes.

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Introduction

Spontaneous intracranial hypotension (SIH) is an unusual syndrome (5 per 100,000) secondary to a reduction of volume of cerebrospinal fluid (CSF), mainly characterized by orthostatic headache.¹ Most cases are due to structural defects of the spinal dura mater, CSF extends into the epidural space and the paraspinal soft tissues, so there is a decrease in intracranial pressure (ICP).¹ Although diagnosis of CSF hypotension is established by ICP measurement, the detection of parathecal activity pointing to the level of the leak has a higher diagnostic value in SIH syndrome, especially if clinical and imaging findings are inconclusive. Radioisotope cis-

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ternography (RC) may be very useful to determine etiological diagnosis of the disease.

Clinical cases

We studied retrospectively 8 patients who underwent a RC from January 2011 to January 2016 because SIH had been suspected. RC was performed by the intrathecal lumbar injection of 37MBq (1 mCi) of ¹¹¹In-DTPA. Planar scintigraphic images of spine (anterior and posterior projections) and head (anterior, posterior and lateral projections) were obtained with a Millenium VG gammacamera (GE) 3 and 24 h after injection. Paradural extravasation of the radioisotope was the direct evidence of spinal CSF leak. Three signs were considered as indirect evidence of CSF: early appearance of radioactivity in the kidneys and urinary bladder, radioactivity in the patient's circulation outlining the whole patient in the images ("silhouette sign"), and absence or paucity of radioactivity over the cerebral convexities at 24 h (PAC). CSF leaks were classified

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Table 1		
Clinical	features	of patients.

Patient	Age	Sex	SIH signs on cranial MRI	Spine MRI	CSF pressure	Radioisotopic Cisternography		
						Direct signs (location)	Indirect signs	Flow grade
1	56	М	NO	Normal	NP	Thoracic and lumbar	EBF, SS, PAC	High
2	48	Μ	NO	Normal	Hypotension	Thoracic, lumbar and cauda equine	EBF, SS, PAC	High
3	68	Μ	NO	Normal	NP	Lumbar (24 hours)	NO	Very low
4	53	F	NO	Normal	NP	Lumbar and cauda equine	EBF, SS, PAC	High
5	26	F	NO	Normal	NP	Thoracic	EBF, SS, PAC	Low
6	33	Μ	NO	Normal	NP	Thoracic	EBF, SS, PAC	Low
7	53	F	YES	NP	Hypotension	Cervical	EBF, SS, PAC	Low
8	47	F	YES	Normal	Hypotension	NO	EBF, SS	Very low

NP, not performed; EBF, early bladder filling; SS, "Silhouette sign"; PAC, paucity of activity over cerebral convexities at 24 h; Conservative treatment, postural and diet measures and/or pharmacologic treatment.

according to visual assessment at 24-h cranial images as: high-flow (absence or paucity of radioactivity), slow-flow (slight decreased activity) or very low-flow leaks (normal activity). Definitive diagnosis was established according to the measurement of CSF pressure and/or direct evidence for spine CSF leak. Table 1 summarizes clinical features of patients.

Patient 1

A 56-year-old male had been diagnosed of recurrent aseptic meningitis caused by HIV infection for 5 years. He had been hospitalized three times, and during last one SIH was suspected because of an atypical neurologic syndrome. There were no signs of SIH at head magnetic resonance imaging (MRI), and spine MRI was normal too. RC showed thoracic and lumbar paraspinal radioactivity and indirect evidences of CSF leaks: early bladder filling (EBF), "silhouette sign" and PAC suggesting high-flow leak (Fig. 1). Although computed tomography (CT) myelography was normal, conservative treatment was begun with good results.

Patient 2

A 48-year-old male suffered from headaches at frontotemporal area and they improved at supine position. The CSF opening pressure was low. RC showed direct evidences of CSF leak at thoracic and lumbar spine, and at cauda equine, and all three indirect signs (Fig. 2A). Bed rest, increased fluid intake and caffeine improved the symptoms, and a RC performed 3 months later (Fig. 2B) showed a decrease in the paraspinal activity and in the flow of the CSF leak. A new measurement of pressure still showed hypotension, although spinal MRI was normal and there were no signs of SIH at head MRI.

Patient 3

A 68-year-old man had bilateral frontal hygromas that presented slight morphological changes on CT, so SIH was suspected. 24-h imaging of RC only showed 3 paraspinal lumbar foci with mild activity, and there were no indirect evidences of CSF leak, mimicking meningeal diverticula. Lumbar MRI and early images of RC were normal, and together with his clinical evolution, truly very low-flow



Fig. 1. (A) Anterior projection of RC performed 2 h after injection of patient 1. Focal paraspinal radioactivity (arrows), early bladder filling and "silhouette sign" are shown; (B) 24-h anterior projection shows paucity of radioactivity over the cerebral convexities, suggesting high-flow CSF leak.

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