CASE REPORT

Spontaneous intracranial hypotension treated with epidural blood patch. Case report*

Hipotensão intracraniana espontânea tratada com tamponamento sanguíneo peridural. Relato de caso

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SUMMARY

BACKGROUND AND OBJECTIVES: Spontaneous intracranial hypotension (SIH) is a syndrome characterized by postural headache associated to low CSF pressure, which is rapidly resolved with decubitus. Therapy varies from conservative approaches to invasive procedures, such as epidural blood patch (EBP). This study aimed at presenting a case of postural headache secondary to SIH and treated with EBP.

CASE REPORT: Female patient, 33 years old, Caucasian, for seven months suffering from daily orthostatic holocrainal headache, followed by nausea and vomiting, triggered by orthostatism and relieved by decubitus. No history of dural puncture or other reason for fistula. Previous history of migraine for more than ten years. Neurological evaluation has shown no deficits, however she had difficulties to walk due to dizziness and headache. Lumbar puncture has shown CSF hypotension. An EBP was performed in L_3-L_4 with autologous blood, without intercurrences and resolving her headache.

CONCLUSION: EBP was effective to treat SIH not resolved with conservative approaches.

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Keywords: CSF pressure, Epidural blood patch, Intracranial hypotension, Lumbar puncture, Postural headache, Treatment.

RESUMO

JUSTIFICATIVA E OBJETIVOS: Hipotensão intracraniana espontânea (HIE) é uma síndrome caracterizada por cefaleia postural associada à baixa pressão liquórica e que desaparece rapidamente ao decúbito. A terapia varia de tratamento conservador a procedimentos invasivos, como a realização de tampão sanguíneo peridural (TSP). O objetivo deste estudo foi apresentar o caso de uma paciente com cefaleia postural secundária à HIE tratada com TSP.

RELATO DO CASO: Paciente do sexo feminino, 33 anos, branca, há 7 meses com quadro de cefaleia ortostática diária, holocraniana, acompanhada de náuseas e vômitos, desencadeada pelo ortostatismo e aliviada pelo decúbito. Sem história de punção dural ou outra causa de fístula. História prévia de enxaqueca há mais de 10 anos. Ao exame neurológico sem déflcits, porém com diflculdade para deambular devido a tonturas e cefaleia. Punção lombar evidenciou hipotensão liquórica. Foi realizado TSP em nível de L_3-L_4 com 20 mL de sangue autólogo, sem intercorrências e com resolução da cefaleia.

CONCLUSÃO: O TSP foi uma opção efetiva no tratamento da HIE não solucionada com o tratamento conservador.

Descritores: Cefaleia postural, Hipotensão intracraniana, Pressão do líquido cefalorraquidiano, Punção lombar, Tampão sanguíneo peridural, Tratamento.

INTRODUCTION

Spontaneous intracranial hypotension (SIH) is a syndrome characterized by postural headache, associated to low CSF pressure, which starts soon after standing by and is rapidly relieved by decubitus[1].

Although being uncommon and with well characterized clinical presentation[2], SIH is still unknown by several professionals who end up erroneously diagnosing it as migraine, tension headache and viral meningitis, among others, which delays the correct treatment and contributes to worsen clinical presentation[3].

Therapy varies from conservative approaches, such as rest, hydration and analgesics, to invasive procedures, such as epidural blood patch (EBP)[4].

This study aimed at describing a typical case of patient with postural headache secondary to SIH treated with lumbar EBP, to highlight diagnostic criteria of the disease and its effective treatment.

CASE REPORT

Female patient, 33 years old, Caucasian with almost daily severe holocranial orthostatic headache, followed by nausea and vomiting with 7 months of evolution. She denied phono and photophobia. Pain would appear few seconds after adopting the orthostatic position and would rapidly resolve with horizontal decubitus. There was no report of fever and patient denied previous dural puncture or any other possible cause of CSF flstula. History of migraine for more than 10 years, characterized by hemicranial, pulsatile and severe headache with nausea, phono and photophobia.

Patient was under sibutramine for three months previous to symptoms onset and had lost approximately 10 kg. At admission she was under no medication. She denied smoking, use of alcohol and illegal drugs.

Patient was submitted to cranial MRI and the report described hyperintensity in T_1, in posterior pituitary gland. At physical evaluation she was in good general status, lucid, oriented and conscious. Neurological evaluation has shown no deflcits, however with diffIculty to walk due to dizziness and headache.

According to history and physical evaluation, diagnostic impression was that headache was caused by low CSF pressure. Lumbar puncture has shown initial ‡uid pressure of 25 mmH_2O, which has conflrmed the diagnosis.

No cisternogram by CT was performed because patient had history of allergy to contrast. She was treated with bed rest and hydration for two days without improvement. After written informed consent, EBP was performed in the operating center, at the level of L_3-L_4, with 15 Tuohy needle, with the patient in lateral and head down position. After locating the epidural space by the loss of resistance technique, 20 mL of autologous blood collected with aseptic technique from the left antecubital fossa were injected in approximately 90 seconds. There were no technical intercurrences and patient was referred to the post-anesthetic recovery unit. Postural headache resolved and patient was discharged three days after the procedure with the recommendation to use paracetamol, if necessary.

DISCUSSION

SIH is uncommon among headache patients with estimated incidence of 1:50000, but it is an important differential diagnosis for patients with new, persistent and daily headache[1,2].

SIH patients are often poorly diagnosed and have their effective treatment delayed, probably due to the lack of understanding of the disease by physicians. In a study where 94% of patients had diagnosis of SIH, it took from 4 months to 13 years to reach the correct diagnosis, which has exposed many patients to risks of unnecessary diagnostic and therapeutic procedures, such as arteriography and craniotomy[3].

Spontaneous CSF leak is the typical cause of SIH[1]. For most patients, neuroimaging studies reveal the leak site[3,4]. The precise etiology of such leak remains unknown, but it is suspected that there is underlying structural weakness of spinal meninges, with the presence of dural slits or arachnoid cysts[5,6], similar to those seen in generalized connective tissue disease, such as Marfan syndrome[7-9]. In our case, the diagnosis of headache caused by spontaneous low CSF pressure was possible by history and complementary tests, which have met the diagnostic criteria proposed by the International Headache Society[10]. In some patients, however, in spite of the presence of typical orthostatic headache, diagnostic studies fail to show any evidence of leak or low CSF pressure[11].

It is possible that there is a different mechanism responsible for low CFS pressure and for the headache, probably related to higher dural sac compliance in the lower medullar segment. When the patient stands up, the abnormal distribution of the craniospinal elasticity determines abnormal hydrostatic pressure distribution, thus decreasing intracranial CSF pressure[12].

Inside the skull, according to Monroe-Kellie hypothesis, the sum of intracranial CSF and blood volumes is constant. So, the decrease in one should cause an equivalent increase in the other, or in both. Thus, decreased intracranial CSF pressure leads to compensatory dilatation of intracranial pain-sensitive venous structures, causing orthostatic headache[13].

The treatment proposed by the literature is bed rest with ‡uid supplementation, analgesic agents and caffeine. If conservative measures do not totally relief symptoms and headache persists, EBP is indicated[2,6]. EBP is the most effective treatment for SIH[3]. The mechanism responsible for its effect remains undetermined, but it may involve the temporary increase in the epidural space pressure, leak sealing by injected blood coagulation and dura mater slit healing by the in‡ammatory response[14].

CSF flstulas are more often located in cervical-thoracic and lumbar spaces, which are more mobile spinal segments. A study has shown that image-guided EBP has a better result as compared to blind puncture[4].

Image-guided EBP may be associated to higher risk for medullar and nervous root compression, chemical meningitis, subarachnoid blood injection and cervical stiffness. In addition, just a small volume of blood may be injected at thoracic or cervical level, as compared to the volume injected at lumbar level. For safety reasons, current recommendation for EBP is the injection of 20 mL of autologous blood in the epidural space, in lumbar spine[15], since there, 90% of EBP are effective to treat SIH[16] even when EBP is performed distant from the CSF flstula, or when the leak site is not determined[17]. It is important to apply in the epidural space because injection in the paraspinal muscle is not enough to relief headache and requires additional intervention[18].

EBP is considered an effective treatment for post-dural puncture headache. There is no precise indication of the puncture site for SIH. The presence of flstulas or dura mater hernias is more common in more mobile spinal segments. In the absence of objective data, the lumbar region should be chosen for the procedure. Initially, 10 to 20 mL of blood are used, being effective to relieve symptoms in approximately one third of patients, probably due to dural buffering, stopping CSF leak.

CONCLUSION

EBP was effective to treat spontaneous intracranial hypotension not controlled with conservative approaches.

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