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Epidural blood patch for the treatment of liquor hypotension after intrathecal chemotherapy in a 10-year-old: case report

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Abstract

Background and objectives: An epidural blood patch is used to treat postdural puncture and liquor hypotension headache. We report the use of an epidural blood patch in a critical pediatric patient.

Case report: A 10-year-old girl with acute leukemia developed venous cerebral thrombosis with hemorrhagic transformation one month after intrathecal chemotherapy. Given the unusual clinical and imagiological evolution even after decompressive craniectomy, we suspected cerebrospinal fluid hypotension. Spine imaging revealed signs of post-lumbar puncture fistula; we hence performed a blind blood patch.

Conclusions: Recognizing cerebrospinal fluid hypotension in critical pediatric patients is important. Less-conventional life-saving measures, such as a blind blood patch, may be considered in such patients.

KEYWORDS

Blood patch, epidural;

Chemotherapy;

Intracranial hypotension;

Cerebrospinal fluid leak

Introduction

Epidural Blood Patch (EBP) is a procedure wherein autologous blood is administered in the epidural space, which functions as a patch to the Cerebrospinal Fluid (CSF) leak, raising the epidural space pressure, and causing an inflammatory response that seals the leaking spot. It is used to treat postdural puncture headache and liquor hypotension headache.[1] Only few reports of the use of EBP in pediatric patients, including under general anesthesia, exist.[2]

The use of Intrathecal (IT) chemotherapy, which involves multiple lumbar punctures, is increasing for the treatment of some forms of cancer. The use of IT increases the risk of CSF hypotension.[3]

We report a case of CSF hypotension in a pediatric patient with clinical instability, who had acute lymphoblastic leukemia and was receiving IT chemotherapy, managed using EBP.

Case report

A 10-year-old girl (weight, 26 kg) received the diagnosis of acute lymphoblastic leukemia and was undergoing induction treatment with triple IT chemotherapy (methotrexate, cytarabine, and hydrocortisone) aimed at preventing central nervous system relapse along with intramuscular PEG-asparaginase and systemic corticotherapy. She had no relevant medical history. The patient was receiving 4 rounds of IT treatment through a serial lumbar puncture (under sedation) per month (one round a week). We observed no complications. However, we had experienced several unsuccessful lumbar punctures in the last round, without CSF leakage, and hence had to postpone the IT chemotherapy. The patient complained of increased orthostatic headaches, neck pain, and scotomas 4 hours after the last lumbar puncture. She developed a single complex focal seizure in the following 24 hours (conjugate deviation of the eyes to the right and clonic movements of the left arm). We administered intravenous diazepam (0.1 mg.kg^{-1}), which yielded resolution.

We observed superior sagittal sinus thrombosis and acute subcortical left parietal hemorrhage, with surrounding edematous areas and without signs of hydrocephaly on cranial computed tomography. We transferred the patient to the pediatric intensive care unit under sedation and analgesia and on mechanical ventilation. She was hemodynamically stable.

She presented coagulation abnormalities secondary to chemotherapy-induced myelosuppression; hence, significant bleeding risk was considered. Therefore, we postponed anticoagulant administration despite superior sagittal sinus thrombosis. Considering the possible expansion of intracerebral hemorrhage that could cause intracranial hypertension and clinical deterioration, we considered Intracranial Pressure (ICP) monitoring. Before introducing an ICP monitoring catheter, platelet transfusion was performed. The initial ICP was 8 mmHg (normal ICP for children, 3–12 mmHg).

We observed marked anisocoria (left pupil > right pupil) 1 hour after ICP catheter placement, without any variations in the ICP value. The patient underwent another cranial computed tomography, which revealed a volume increase of the left hemorrhage lesion with a marked shift of the midline structures to the right, although with the same basal cistern patency and a normal cerebellar tonsil position. Subsequently, we performed an urgent left partial decompressive craniectomy. Although a low ICP was maintained, her clinical status 24 hours after surgery did not improve. On the contrary, she experienced bradycardia, with a high arterial pressure and anisocoria persisting. Of interest, these signs improved with the patient supine at 0°. Another cranial computed tomography showed that the right deviation shift persisted; no cerebral herniation at the craniectomy side was observed, as expected.

Considering the initial clinical complaints of orthostatic headaches after several unsuccessful attempts of lumbar puncture and the unusual clinical and imagiological evolution after decompressive craniectomy (normal ICP values despite some neuroradiologic signs of augmented ICP), we suspected post-IT chemotherapy CSF hypotension syndrome.

Magnetic resonance imaging myelography revealed CSF collection in the epidural space, from T4 to the lumbar area, which increased in the lumbar cistern. These findings constituted an indirect evidence of a post-lumbar puncture fistula.

Conservative treatment including maintenance of the supine position at 0°, analgesia, and intravenous fluids was attempted, without improvements, however, by 72 hours.

A multidisciplinary team therefore decided to perform an EBP under sedation and analgesia. A trained anesthesiologist performed the procedure under proper aseptic conditions with the patient in the left lateral decubitus position. A 20G Tuohy needle was used to identify the epidural space at the L4–L5 level using the loss-of-resistance technique with saline. The epidural space was easily identified on the first attempt,

and 8 mL of autologous venous blood, collected from the saphenous vein of the left leg, was slowly injected. The anesthesiologist reported mild epidural resistance change after the administration of this volume, however, no complications were reported.

The patient had a favorable clinical and imagiological evolution in the first 24 hours after the procedure and was extubated 5-days later. Subsequently, an Ommaya reservoir was placed for IT chemotherapy to avoid further lumbar puncture.

Discussion

Acute lymphoblastic leukemia is the most common neoplasm among pediatric patients. Given the development of new treatment regimens, which often include IT chemotherapy, survival has increased among such pediatric patients. IT chemotherapy involves repeated lumbar puncture that may lead to a CSF fistula formation, which in turn causes liquor hypotension and related symptoms.[3]

With an incidence of 8%–25% among children after lumbar puncture with a 22G needle, postdural puncture headache is a relatively common complication. However, it is sometimes neglected by oncologists, particularly in young children with unspecific symptoms and with multiple medications that can lead to the same unexpected adverse effects.

In most extreme cases, like our case, the CSF leak can lead to liquor hypotension, with consequential sagging of the cerebellar tonsils, causing more intense neurologic symptoms.[1] Our patient complained of headache and neck pain after lumbar puncture; however, because of the simultaneous superior sagittal sinus thrombosis with a mass effect, we could not diagnose it earlier. Only when ICP did not increase and even the decompressive craniectomy did not improve the child's clinical status, a magnetic resonance imaging myelography was performed.

Magnetic resonance imaging has significantly improved the detection of CSF leaks, resulting in increased detection of cases and recognition of several clinical or imaging forms of CSF hypotension. We chose to perform a blind EBP at the lumbar region because of the large extension of CSF on magnetic resonance imaging; moreover, there are reports of successful treatment of postdural puncture headaches with EBP.[4] We performed this procedure at the L4–L5 level considering that IT chemotherapy was attempted in that region.

Traditionally, among adults, EBP is performed with the patient awake; this allows clinical monitoring of symptoms such as neck discomfort, according to which the volume

of autologous blood to be administered (typically 15–25 mL) can be adjust. However, performing this technique in children is more complex owing to the procedure being performed under sedation, thus making clinical monitoring and adjustment of volume to be administered impossible. Although few case reports of the EBP technique performed in children under sedation and analgesia do exist, the blood volume to be administered requires further studies. Ylonen and Kokki reported that a blood volume of 0.2–0.3 mL.kg⁻¹ should be administered.[5] Roy et al. reported a successful EBP in a 7-year-old with a volume of 10 mL[2] and Kandil et al. reported performing EBP in a 10-year-old boy with a volume of 14 mL; patients were sedated in both the reports. However, reports of successful EBP with a volume as low as 5 mL also exists. We decided to administer 8 mL because the senior anesthesiologist performing the technique noticed a difference in the syringe resistance after this volume was injected.

Central nervous system dissemination is another risk of performing EBP in patients with myeloproliferative disease. Regardless, most studies found that the risk of dissemination is very low. Hence, the benefits outweighed the risks in this case.

To our knowledge, no case reports of EBP use in children with CSF hypotension and similar clinical instability exist.

In conclusion, it is important to recognize CSF hypotension as a differential diagnosis, even in critical pediatric patients. Less-conventional life-saving measures, such as blind EBP, could be considered in similar cases. The optimal volume of blood to be administered in a blind EBP for the treatment of CSF hypotension in children under sedation needs further study.

Conflicts of interest

The authors declare no conflicts of interest.

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