Postpartum workup of postdural puncture headache leads to diagnosis and surgical treatment of thoracic pseudomeningocele: a case report

Une investigation postpartum de céphalées post-ponction durale débouche sur le diagnostic et le traitement chirurgical d'un pseudoméningocèle thoracique: une présentation de cas

Lindsay Hurlburt, MD · Christine Lay, MD ·
Michael G. Fehlings, MD, PhD · Jose C. A. Carvalho, MD, PhD

Received: 24 August 2012 / Accepted: 19 November 2012 / Published online: 7 December 2012
© Canadian Anesthesiologists' Society 2012

Abstract

Purpose Postdural puncture headache is a well-known complication of neuraxial anesthesia in childbirth. There are, however, many other causes of postpartum headache, some of which may present like postdural puncture headache and confuse the diagnosis. We report a case of postpartum headache due to pseudomeningocele.

Clinical features A 31-yr-old primigravida presented at 35 weeks gestation for induction of labour. A pre-pregnancy history of migraines was suspicious for low cerebrospinal fluid headache, but this had been ruled out by normal brain

magnetic resonance imaging (MRI), and the headaches had resolved with pregnancy. A labour epidural was easily placed at L3/4. On postpartum day one the patient complained of mild headache, and the severe pre-pregnancy headache returned within weeks. The pounding occipital/hemispheric headache was associated with nausea and vomiting; it worsened over the day, and improved when the patient was supine. A repeat brain MRI showed evidence of intracranial hypotension, and an epidural blood patch was performed at L3/L4 with no improvement. A second blood patch was performed ten days later, still with no improvement. A subsequent brain MRI showed unchanged intracranial hypotension, and a spine MRI revealed a pseudomeningocele at T7. A computed tomography myelogram confirmed a T7/8 pseudomeningocele. The patient underwent direct microsurgical exploration and repair four months later, which was followed by re-operation ten months later. The patient's condition has since remained significantly improved.

Discussion This case highlights the importance of maintaining a wide differential in the workup of postpartum headache after regional anesthesia, particularly in cases refractory to blood patch. This case also reveals a T7 pseudomeningocele causing spontaneous intracranial hypotension that otherwise may not have been detected and corrected.

Résumé

Objectif Les céphalées post-ponction durale sont une complication bien connue de l'anesthésie neuraxiale pour l'accouchement. Il existe toutefois de nombreuses autres causes de céphalées postpartum, dont certaines pourraient

Author contributions

All authors cared for the patient. Lindsay Hurlburt and Jose C.A. Carvalho reviewed the literature and wrote the manuscript. Michael G. Fehlings provided the surgical pictures. Michael G. Fehlings, Christine Lay, and Jose C.A. Carvalho revised the manuscript, and Jose C.A. Carvalho submitted the final version of the manuscript.

L. Hurlburt, MD
Department of Anesthesia, University of Toronto, Toronto, ON, Canada

C. Lay, MD
Department of Neurology, Women's College Hospital, University of Toronto, Toronto, ON, Canada

M. G. Fehlings, MD, PhD
Department of Neurosurgery, Toronto Western Hospital, University of Toronto, Toronto, ON, Canada

J. C. A. Carvalho, MD, PhD (✉)
Department of Anesthesia and Pain Management, Mount Sinai Hospital, University of Toronto, 600 University Avenue Room 781, Toronto, ON M5G 1X5, Canada
e-mail: jose.carvalho@uhn.on.ca

se présenter comme des céphalées post-ponction durale et fausser le diagnostic. Nous rapportons le cas d'une céphalée postpartum due à un pseudoméningocèle.

Éléments cliniques Une primigeste de 31 ans s'est présentée à 35 semaines de grossesse pour l'induction du travail obstétrical. Des antécédents pré-grossesse de migraine ont laissé soupçonner une céphalée due à un liquide céphalorachidien peu abondant, mais cette hypothèse a été écartée à la suite d'une imagerie par résonance magnétique (IRM) normale du cerveau; en outre, les maux de tête s'étaient résolus pendant la grossesse. Une péridurale pour le travail obstétrical a été facilement positionnée au niveau L3/4. Le premier jour postpartum, la patiente s'est plainte de légers maux de tête, et les graves céphalées pré-grossesse se sont manifestées à nouveau dans les semaines suivantes. Les céphalées occipitales / hémisphériques martelantes étaient associées à des nausées et vomissements; elles empiraient au cours de la journée mais s'amélioraient lorsque la patiente était couchée. Une seconde IRM du cerveau a révélé des signes d'hypotension intracrânienne, et un colmatage sanguin péridural a été réalisé au niveau L3/4, sans entraîner d'amélioration. Un deuxième colmatage sanguin a été réalisé dix jours plus tard, toujours sans amélioration. Une IRM du cerveau subséquente a révélé une hypotension intracrânienne inchangée, et une IRM rachidienne a révélé un pseudoméningocèle au niveau T7. Une myélographie par tomodensitométrie a confirmé la présence d'un pseudoméningocèle au niveau T7/8. La patiente a subi une exploration microchirurgicale directe et une réparation quatre mois plus tard, suivies par une réopération dix mois plus tard. L'état de la patiente est depuis demeuré significativement amélioré.

Discussion Ce cas souligne l'importance de faire un diagnostic différentiel élargi lorsqu'on cherche la cause de céphalées postpartum après une anesthésie régionale, particulièrement dans les cas réfractaires aux colmatages sanguins. Ce cas révèle également un pseudoméningocèle au niveau T7 provoquant une hypotension intracrânienne spontanée qui n'aurait autrement pas été dépistée ou corrigée.

Postdural puncture headache (PDPH) is a well-known complication of neuraxial anesthesia in childbirth and may occur even when dural puncture goes unrecognized.^{1,2} There are, however, many other causes of postpartum headache, some of which may present like a PDPH and confuse or delay the correct diagnosis and treatment.^{1,2}

In one prospective cohort study of 985 women in the postpartum period, the incidence of headache was 39%; however, PDPH accounted for only 4.7% of all postpartum

headaches. A large proportion of women (8.1%) exhibited headache of undetermined cause.¹

The differential diagnosis of postpartum headache is wide and includes primary headache (migraine, tension-type, cervicogenic, and cluster headaches) as well as secondary headache. Secondary headaches are those resulting from obstetric disease (hypertensive disorders, pituitary apoplexy), intracranial pathology (central venous thrombosis, tumour, meningitis, stroke), and intracranial hypotension (spontaneous or secondary to regional anesthesia).

We report a case of postpartum headache due to pseudomeningocele, a cause of postpartum headache not traditionally included in published lists of the differential diagnosis. The patient's written consent was obtained for publication of this report.

Case report

A 31-yr-old primigravida presented to hospital at 35 weeks gestation for induction of labour due to a low biophysical profile score.

Her medical history revealed intermittent migraines, with and without aura, complicated by two years of severe debilitating, chronic, daily, right-sided headache with a postural component. Low cerebrospinal fluid (CSF) pressure headache was suspected, but brain magnetic resonance imaging (MRI) ordered by her neurologist was reported as negative. She was treated with moderate success with indomethacin until pregnancy, and the headache resolved within the first trimester.

A labour epidural was placed without complication at the L3/L4 interspace. On postpartum day one, she complained of mild headache that responded to ibuprofen, and she was discharged to home on postpartum day two. The severe debilitating pre-pregnancy headache gradually returned within weeks. This pounding headache radiated from the neck into the occipital and right hemispheric regions and was associated with nausea and vomiting. The headache worsened over the day but improved when the patient was supine. Neurology follow-up prompted a repeat brain MRI that showed downward displacement of the brainstem and posterior fossa with tonsillar herniation, in keeping with intracranial hypotension.

The patient was referred to the Department of Anesthesia, and an epidural blood patch was performed at the L3/L4 interspace. No clinical improvement resulted, and a second lumbar epidural blood patch was performed ten days later, still with no improvement. The anesthesiologist then requested an MRI of both the brain and spine, which showed unchanged intracranial hypotension (Fig. 1) and a posterior epidural fluid collection at T7 with a small lateral

Fig. 1 Preoperative C-spine magnetic resonance imaging shows an acquired Chiari malformation with herniation of the cerebellar tonsils down below C1 and marked crowding of the foramen magnum

Fig. 2 Preoperative axial and sagittal computed tomography myelograms show leak of contrast outside the thecal sac at the right T7 nerve root region

pseudomeningocele from the right sac at T7. Upon discussion with the neurologist and the neuroradiologist, a computed tomography myelogram was performed and confirmed a pseudomeningocele with CSF collection on the right of T7/T8 (Fig. 2). While discussing treatment options, a lesion-specific epidural blood patch was considered given that two lumbar epidural blood patches had failed. Nevertheless, given the size of pseudomeningocele in this case, it was judged unlikely to respond to such a procedure.

The patient was then referred to neurosurgery and underwent repair of the thoracic pseudomeningocele four months later. A dural repair and a duraplasty were fashioned (Figs. 3 and 4), resulting in radiographic and clinical

Fig. 3 Intraoperative microphotograph pre-repair: right T7 nerve root sleeve with spontaneous tear in the dura mater

improvement. Occasional suboccipital headache that resolved with caffeine still persisted, and further workup was planned. The patient eventually developed progressive recurrence of her preoperative symptoms. Follow-up imaging showed a partial recurrence of the acquired Chiari I malformation, although the changes in the posterior fossa were not as severe as prior to surgery (Fig. 5). In addition, the findings on MRI in the thoracic spine were somewhat equivocal but suggestive of a possible recurrence of the pseudomeningocele. In light of this clinical and radiographic picture, the neurosurgeon elected to re-operate on the patient ten months after the first operation. At surgery, the previous T6-7 laminectomy was re-opened and re-explored. Upon removal of scar tissue, a small CSF space adjacent to the T6-7 level was entered. Although a definite leakage of CSF could not be identified, the neurosurgeon elected to clip the right T6 nerve route. This was then combined with a repeat duraplasty using a combination of autogenous fat, collagen matrix, and fibrin sealant.

To date (i.e., 11 months later), there has been no clinical or radiographic evidence that the pseudomeningocele has recurred, and the patient's headaches have substantially improved.

Discussion

This case report shows a T7 pseudomeningocele causing spontaneous intracranial hypotension in the postpartum period. This condition may not have been detected and corrected without diligent consideration of the differential diagnosis of PDPH and without investigations that included an MRI of the spine and a computed tomography myelogram. Furthermore, this case emphasizes the role of the

Fig. 4 Intraoperative microphotograph post-repair: collagen matrix and fibrin sealant in place over the exposed dura mater

Fig. 5 Postoperative C-spine magnetic resonance imaging shows marked improvement in the extent of the acquired Chiari malformation with cerebrospinal fluid spaces present around the cervicomedullary junction in the foramen magnum

anesthesiologist as part of the care team and the need for clear and continued communication among all professionals involved.

Spontaneous intracranial hypotension is uncommon but not a rare cause of persistent headache. Similar to PDPH, it is orthostatic in nature. The etiology and pathogenesis of spontaneous intracranial hypotension is multifactorial. In cases where surgery has been indicated, a variety of dural abnormalities have been reported.³

True meningoceles are extradural collections of CSF in an arachnoid-lined capsule, while pseudomeningoceles are extradural collections of CSF that occur when the dura is breached and may be contained in an arachnoid-lined membrane or in a fibrous capsule.⁴ Pseudomeningoceles are primarily iatrogenic, caused by incidental durotomies during spinal or intradural surgery, but they may also occur

traumatically or congenitally. In many cases, they are asymptomatic, but they may cause various signs and symptoms, including headache, infection, back pain, radiculopathy, and myelopathy. When headache occurs, it is thought to result from a reduction in CSF volume and intracranial hypotension resulting in traction on the meninges and vasculature. Magnetic resonance imaging is the diagnostic imaging of choice, but computerized tomography myelography is sometimes better for visualizing its location. Treatment options range from conservative treatment to epidural blood patch. Surgical dural repair is the definitive treatment.⁵

This case shows the occurrence of spontaneous intracranial hypotension secondary to a spontaneous dural tear causing CSF leak and a pseudomeningocele. Despite the patient's medical history of chronic headache suggesting a low CSF pressure etiology, a previous MRI had been reported as normal and the diagnosis was never made. The improvement of the chronic headache during pregnancy is likely explained by an increase in extradural pressure⁶⁻⁹ and a reduction in CSF leakage. The fact that the patient remained asymptomatic during pregnancy certainly influenced the diagnosis of an epidural-related PDPH, prompting the treatment with two blood patches. Eventually, when the MRI showed a posterior epidural fluid collection at T7, the question was raised as to whether this could be remnants of blood injected with the epidural blood patch. Nevertheless, this possibility was quickly ruled out because the imaging was performed seven days after the most recent blood patch, and as a rule, there is evidence of clot resolution within seven to 18 hr following an epidural blood patch.¹⁰ Similarly, the possibility that an epidural could have caused the pseudomeningocele was dismissed because there was a long distance between the T7 dural breach and the lumbar region where all epidurals had been performed.

While discussing treatment options, a lesion-specific epidural blood patch was considered given that two lumbar epidural blood patches had failed. Although epidural blood patch has been used successfully to treat cases of spontaneous intracranial hypotension, it was judged that given the size of pseudomeningocele in this case, the response to such procedure, even if lesion-specific, was unlikely. A surgical correction was therefore deemed necessary. Yoon *et al.*⁵ reported the use of both blind (15 out of 24) or lesion-specific (4 out of 24) blood patches in a cohort of 30 patients with spontaneous intracranial hypotension. Four patients received two blood patches and one patient required three blood patches. Symptoms improved in 22 out of these 24 patients; however, the improvement was not related to the specific site of the epidural blood patch.

Despite what seemed to be a successful first surgical correction in our patient, the fistula eventually

re-established, and a second surgical correction was required. This outcome is in keeping with the literature. Data on long-term outcomes of patients treated for spontaneous intracranial hypotension are not conclusive, but recurrence of a spinal CSF leak is seen in approximately 10% of patients regardless of the treatment modality.¹¹

This case report highlights the importance of maintaining a wide differential diagnosis in the workup of postpartum headaches after regional anesthesia, particularly in cases that are refractory to epidural blood patches. Furthermore, this case report emphasizes the role of the anesthesiologist as part of the care team and highlights the need for clear and continued communication among all professionals involved.

Acknowledgement The authors gratefully acknowledge Dr. Robert Willinsky, neuroradiologist at the Toronto Western Hospital, Toronto, for his assistance with the interpretation of the MRI images.

Conflicts of interest None declared.

References

1. Goldszmidt E, Kern R, Chaput A, Macarthur A. The incidence and etiology of postpartum headaches: a prospective cohort study. *Can J Anesth* 2005; 52: 971-7.
2. Klein AM, Loder E. Postpartum headache. *Int J Obstet Anesth* 2010; 19: 422-30.
3. Schievink WI. Spontaneous spinal cerebrospinal fluid leaks. *Cephalalgia* 2008; 28: 1345-56.
4. Couture D, Branch CL Jr. Spinal pseudomeningoceles and cerebrospinal fluid fistulas. *Neurosurg Focus* 2003; 15: 1-5.
5. Yoon SH, Chung YS, Yoon BW, Kim JE, Paek SH, Kim DG. Clinical experiences with spontaneous intracranial hypotension: a proposal of a diagnostic approach and treatment. *Clin Neurol Neurosurg* 2011; 113: 373-9.
6. Bromage PR. Continuous lumbar epidural analgesia for obstetrics. *Can Med Assoc J* 1961; 85: 1136-40.
7. Usubiaga JE, Wikinski JA, Usubiaga LE. Epidural pressure and its relation to spread of anesthetic solutions in epidural space. *Anesth Analg* 1967; 46: 440-6.
8. Galbert MW, Marx GF. Extradural pressures in the parturient patient. *Anesthesiology* 1974; 40: 499-502.
9. Messih MN. Epidural space pressures during pregnancy. *Anaesthesia* 1981; 36: 775-82.
10. Beards SC, Jackson A, Griffiths AG, Horsman EL. Magnetic resonance imaging of extradural blood patches: appearances from 30 min to 18 h. *Br J Anaesth* 1993; 71: 182-8.
11. Schievink WI. Spontaneous spinal cerebrospinal fluid leaks and intracranial hypotension. *JAMA* 2006; 295: 2286-96.