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Malignant PRES and RCVS after brain surgery in the early postpartum period

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ABSTRACT

The management of women with brain tumors in the early post-partum period may be demanding as the patho-physiological changes that occur during pregnancy may also manifest in the early post-partum period. The aim of our paper is to report a case of late-onset post-partum pre-eclampsia after brain tumor surgery, complicated by posterior reversible encephalopathy syndrome (PRES) and reversible cerebral vasoconstriction syndrome (RCVS). Hemicraniectomy and intensive care management were necessary to obtain a favorable neurological outcome.

The inherent literature on the subject is also analyzed through a systematic research. This is the first case of supratentorial decompressive hemicraniectomy in post-partum PRES, while there has been only one other case of posterior fossa decompression described in this cohort of patients.

PRES and RCVS can complicate the neurosurgical management of women in the postpartum period. A careful evaluation of the clinical presentation is necessary as in some particular cases an aggressive medical and surgical treatment is required to obtain a favorable outcome.

1. Introduction

Pregnant women with concurrent neurosurgical pathology represent a therapeutic challenge. The patho-physiological changes occurring during pregnancy may extend to the early postpartum period and may complicate the management of these patients. Posterior reversible encephalopathy syndrome (PRES) and reversible cerebral vasoconstriction syndrome (RCVS) may accompany late-onset post-partum pre-eclampsia, a rare condition secondary to a systemic endotheliopathy, that may occur in the first weeks after delivery, even after uncomplicated pregnancies.

PRES manifests as a transient subcortical vasogenic edema typically in the posterior lobes and its clinical manifestations should not be underestimated, as an aggressive management may be required in some cases. A recent paper dealing with malignant forms of PRES (defined as patients presenting with GCS < 8 and clinical deterioration despite optimal medical management for intracranial hypertension), reported favorable outcomes after decompressive craniectomy [1].

We would like to illustrate a case of malignant PRES associated with RCVS and secondary to a late-onset post-partum pre-eclampsia. The aim of this paper is to discuss the related literature on this subject.

2. Material and methods

A systematic literature search was performed through PubMed database using the terms “postpartum” AND “posterior reversible encephalopathy syndrome” coupled or not with “surgery” OR “decompression”. Articles included were published till March 2019.

A P.I.C.O. question was formulated to select articles reporting cases of post-partum PRES requiring a surgical treatment. The outcome was intended as functional neurological recovery. Reviews and articles not in English were excluded.

3. Case presentation

A 39-year old woman presented to the emergency department with severe headaches and acute weakness on the right side. She had an uncomplicated pregnancy with spontaneous delivery 1 week prior to presentation at hospital. She had no other relevant medical history and she denied substance abuse. She was found to have mild hypertension (BP 150/90 mmHg) and was normocardic. Neurologic examination confirmed the mild right proportional hemiparesis and optic fundoscopy was normal. Head CT and MRI showed the presence of a hemorrhagic lesion of the left thalamus with a mass effect on the internal

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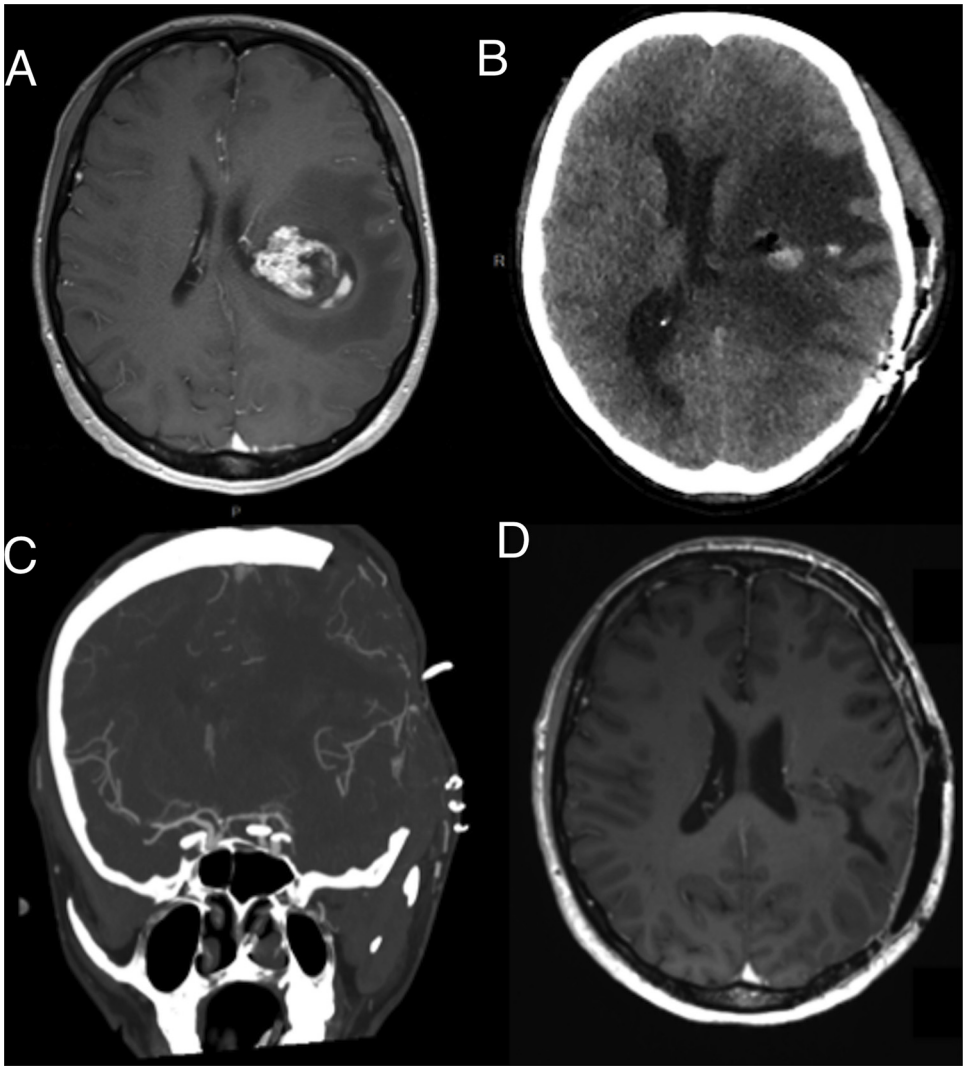

Fig. 1. Axial T1-weighted MRI with gadolinium administration showing an intra-axial lesion centered in the left thalamus with an antero-medial contrast enhancement, a hemorrhagic portion and a perilesional edema (Figure A).
The early postoperative CT-scan shows no major surgical complication with a massive perilesional edema extending in the left fronto-parietal lobe and to the splenium (Figure B).
A coronal cerebral angio-CT demonstrates the presence of vasospasm in the distal internal carotid artery and proximal middle cerebral artery (Figure C). A large decompressive craniectomy was performed.
The bone flap was repositioned 3 weeks after the decompression and the cerebral MRI confirmed a resolution of the postoperative edema (Figure D).

capsule, with a small anterior contrast enhancement following gadolinium administration (Fig. 1A).

Steroids were started and surgery was planned 1 week after admission with intraoperative electrophysiological neuromonitoring. A gross total resection was possible through a transparietal approach (inferior parietal lobe) and pathology confirmed a pilocytic astrocytoma. The patient started becoming hypertensive during the second half of the surgical procedure and a treatment with intravenous labetalol was started. In the immediate postoperative period she presented a worsened hemiparesis associated with a mild Wernicke aphasia. During the first postoperative night she presented an altered mental status with agitation and increased hypertension and 24 -h postoperatively, she further deteriorated with a left non-reactive mydriasis. A cerebral angio-CT scan showed massive perilesional edema extending also to the splenium and a vasospasm of the left anterior circulation in the absence of subarachnoid hemorrhage (Fig. 1B and 1C). No postoperative stroke or venous thrombosis was visible. She was immediately transferred to the OR for a decompressive craniectomy and ICP monitoring. The ICP monitoring showed normal pressures in the postoperative period and she could be extubated 4 days after the craniectomy. She had completely recovered from her hemiparesis and aphasia at 3 months follow-up. The radiological follow-up was also favorable with a complete spontaneous resolution of cerebral edema and vasospasm and no ischemic or hemorrhagic complication (Fig. 1D). The bone flap was repositioned 3 weeks after the craniectomy.

4. Results of literature review

With our search, 124 articles were identified and 60 papers reported cases of post-partum PRES. Only one article described a surgical decompression of the posterior fossa in the early postpartum period [2], while in the other papers patients were treated with conservative measures (Fig. 2). This is the first report of supratentorial craniectomy for a postpartum PRES.

Eclampsia and pre-eclampsia are well-known conditions associated with a high risk for cardio- and cerebro-vascular accidents [3]: inflammation and placental ischemia, hypertension with failed autoregulation and injury to the capillary bed may determine the development of cerebral edema during or after pregnancy, accompanied by an increased permeability of the blood-brain barrier [4]. The interesting fact, though less well known, is that the pro-inflammatory environment may also extend to the postpartum period [5]. Eclampsia and pre-eclampsia represent a multi-organ endotheliopathy and cerebral complications such as PRES [6–8] and RCVS may be associated [9]. The incidence of PRES was higher in cases of postpartum eclampsia when compared to antepartum eclampsia. The other risk factors identified have been no previous pregnancies and a younger age [10]. Late-onset post-partum pre-eclampsia is a rare manifestation and constitutes only 6% of all pre-eclampsia cases [11]. It may occur in the first weeks after delivery even after uncomplicated pregnancies and previously non-hypertensive women and it may associate with the same complications.

PRES classically presents with a full recovery in 75–90% of patients

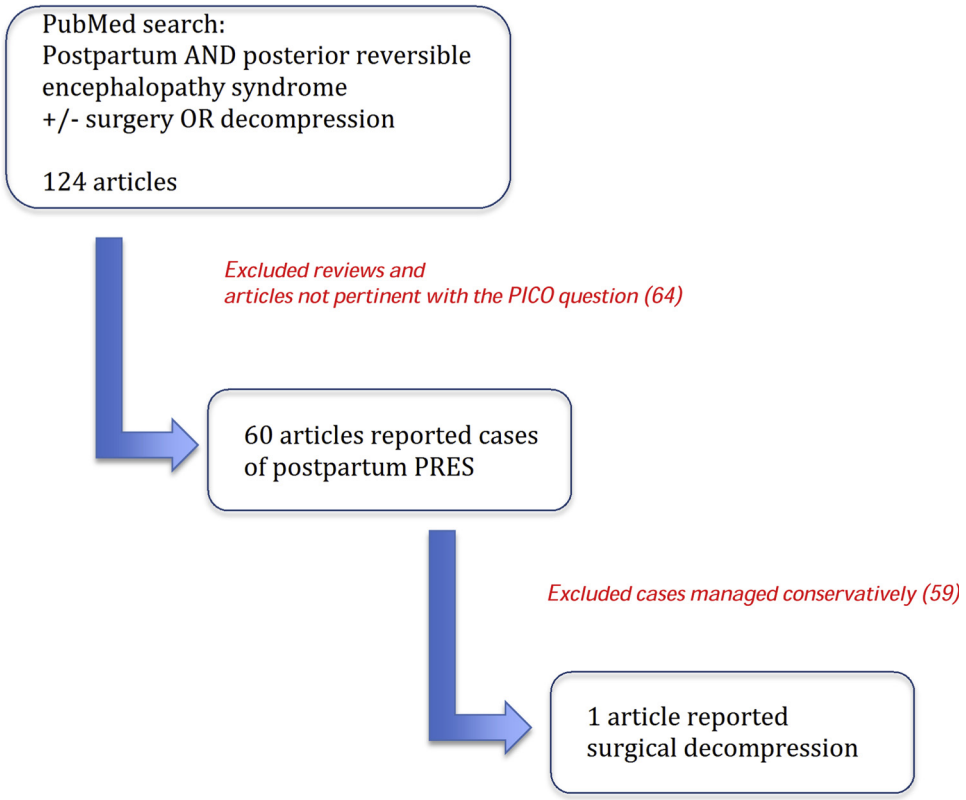

Fig. 2. This figure shows the literature search and the process of articles' selection.

after a symptomatic treatment of the underlying cause [6]. A recent review on the malignant form of PRES needing surgery was performed [2] and if we conjugate these results with those of Akins et al. [1], 9 cases treated with surgical decompression are reported in literature [1,2,12–14] of which 6 were supratentorial [1,12]. Four of these cases were associated with hemorrhagic transformation [1] while two presented a malignant edema with midline shift and brainstem compression [1,12]. The factors determining the localization of the edema and the hemorrhagic transformation were not characterized.

Both cases of postpartum PRES treated with surgical decompression (including ours and those reported by Katsevman et al. [2]) presented a modified Rankin Scale at 0 at last follow-up. In our case the patient presented an hemorrhagic transformation of a benign tumor with a malignant edema in the early postoperative period, associated with clinical manifestations of late-onset post-partum pre-eclampsia and RCVS. Hemicraniectomy was necessary to obtain neurological recovery while RCVS was treated conservatively with intravenous Nimodipine. The surgery for the astrocytoma might have had a role in our case in exacerbating these complications. Traditionally PRES and RCVS are associated with a favorable prognosis after a conservative treatment but it should be kept in mind that in some particular cases they may require an aggressive medical and surgical management to obtain a favorable outcome [1].

5. Conclusion

Neurosurgical procedures should be considered with caution in the early postpartum period. Complications associated with pre-eclampsia/eclampsia, such as PRES and RCVS should be kept in mind when faced with an unusual neurological worsening following elective brain surgery. A high index for suspicion will help diagnose this rare problem and allow early medical and/or surgical therapy to enable a good outcome in these patients.

Patient consent statement

The patient consented to the submission of this paper for publication.

Declaration of Competing Interest

None.

References

[1] P.T. Akins, Y. Axelrod, J.W. Silverthorn, et al., Management and outcomes of malignant posterior reversible encephalopathy syndrome, Clin. Neurol. Neurosurg. 125 (2014) 52–57.
[2] G.A. Katsevman, R.C. Turner, C. Cheyuo, et al., Post-partum posterior reversible encephalopathy syndrome requiring decompressive craniectomy: case report and review of the literature, Acta Neurochir. (Wien) 161 (2019) 217–224.
[3] L.H. Theilen, A. Fraser, M.S. Hollingshaus, et al., All-Cause and Cause-Specific Mortality After Hypertensive Disease of Pregnancy, Obstet. Gynecol. 128 (2016) 238–244.
[4] W.S. Bartynski, Posterior reversible encephalopathy syndrome, part 2: controversies surrounding pathophysiology of vasogenic edema, AJNR Am. J. Neuroradiol. 29 (2008) 1043–1049.
[5] A.M. Clayton, Q. Shao, N.D. Paauw, et al., Postpartum increases in cerebral edema and inflammation in response to placental ischemia during pregnancy, Brain Behav. Immun. 70 (2018) 376–389.
[6] J.E. Fugate, A.A. Rabinstein, Posterior reversible encephalopathy syndrome: clinical and radiological manifestations, pathophysiology, and outstanding questions, Lancet Neurol. 14 (2015) 914–925.
[7] Q.Y. Bo, X.H. Zhao, X. Yang, et al., Reversible posterior encephalopathy syndrome associated with late onset postpartum eclampsia: a case report, Exp. Ther. Med. 12 (2016) 1885–1888.
[8] J.P. Rijal, S. Giri, S. Dawadi, et al., Posterior reversible encephalopathy syndrome (PRES) in a patient with late postpartum eclampsia, BMJ Case Rep. 2014 (2014).
[9] M. McDermott, E.C. Miller, T. Rundek, et al., Preeclampsia: association with posterior reversible encephalopathy syndrome and stroke, Stroke 49 (2018) 524–530.
[10] S. Bembalgi, V. Kamate, K.R. Shruthi, A study of eclampsia cases associated with posterior reversible encephalopathy syndrome, J. Clin. Diagn. Res. 9 (2015) QC05-7.
[11] Z. Al-Safi, A.N. Imudia, L.C. Filetti, et al., Delayed postpartum preeclampsia and eclampsia: demographics, clinical course, and complications, Obstet. Gynecol. 118

(2011) 1102–1107.
[12] S. Hernandez-Duran, A. Barrantes-Freer, V. Rohde, et al., Posterior reversible encephalopathy syndrome presenting in the anterior circulation with malignant intracranial hypertension requiring surgical decompression: a case report and literature review, Acta Neurochir. (Wien) 159 (2017) 1321–1324.
[13] A.J. Grossbach, T.J. Abel, B. Hodis, et al., Hypertensive posterior reversible encephalopathy syndrome causing posterior fossa edema and hydrocephalus, J. Clin. Neurosci. 21 (2014) 207–211.
[14] Y. Abusabha, A.K. Petridis, B. Kraus, et al., Life-threatening posterior reversible encephalopathy syndrome in the cerebellum treated by posterior fossa decompression, Acta Neurochir. (Wien) 159 (2017) 1325–1328.