MANAGEMENT OF A CEREBRAL CAVENOMA DURING PREGNANCY: CASE REPORT

T. Mesbahi, Y. Tahrir, K. Guettabi, K. Baayoud, K. Ibahiouin and A. Lakhdar

ABSTRACT

Intracranial cavernoma is a vascular malformation composed of thin-walled vascular vessels. Blood flow in these lesions is much lower than in AVMs and their hemorrhages are usually small. The presentation is often subacute with seizures or focal neurological deficit, which can be confused in pregnant women with pregnancy toxemia especially if the malformation is bleeding. We report the case of a 35-year-old patient, 30 weeks pregnant, admitted for delivery who presented with acute intracranial hypertension syndrome with obtundation of consciousness. Emergency brain CT showed a right fronto-parietal intracranial hematoma. MRI with angiographic sequences show a left parietal hematoma without visible arteriovenous malformation. The patient underwent emergency surgery to evacuate her intracranial hematoma, then the patient was transferred to a gynecological unit for emergency fetal extraction. Histo-pathological analysis of the removed fragments showed an intracranial cavernoma appearance. Urgent surgical evacuation is only necessary if the prognosis is vital.

Keywords: Intracranial cavernoma, hemorrhagic stroke, pregnancy.

I. INTRODUCTION

Cavernomas of the central nervous system are defined by the presence of capillary-type vascular malformations without interposition of nervous tissue. Very few cases of cavernoma in pregnancy have been reported in the literature and management is poorly codified. We report a case of cavernoma in a pregnant woman and review the literature.

II. CASE REPORT

A 35-year-old female patient, right-handed, 30 weeks pregnant, was admitted to the emergency room with consciousness problems. Clinical examination found a somnolent patient with 14/15 GSC and right hemiplegia. The obstetrical examination was normal. Obstetrical ultrasound noted an evolving monofetal pregnancy with cardiac activity present. The cerebral CT scan (Fig. 1A) showed spontaneous hyperdensity of the left temporoparietooccipital junction with perilesional hypodensity. Brain MRI (Fig. 1) showed a hematoma of the left temporoparietooccipital junction with no clear image of arterio venous malformation. We operated on the patient in emergency to evacuate her intracranial hematoma, then the patient was transferred to a gynecological unit for emergency fetal extraction. Histopathological exam confirmed the diagnosis of cavernoma.
III. DISCUSSION

This is a relatively rare condition but is nevertheless a common etiology of intracranial hemorrhage in pregnant women [1]. Intracranial cavernomas in pregnant women is associated with increased morbidity and mortality for both the woman and the fetus [2]. The incidence of intracranial cavernoma tends to increase during the puerperal period, as shown by many studies [3]-[6]. The incidence of cerebral cavernoma in pregnant women is 1.15%. The increase in this incidence can be explained theoretically by hormonal stimulation. Numerous studies have shown an increase in the size of cavernomas during gestation [7]. However, recent studies have demonstrated the absence of estrogen and progesterone receptors in cavernous malformations and therefore the risk of bleeding is not increased during pregnancy as originally thought [8].

Risk factors can be divided into two groups: pregnancy-related and non-pregnancy-related [9]. The risk factors for intracranial hemorrhage in pregnancy are mainly represented by advanced maternal age, African-American race, pre-existing or incipient hypertension, pre-eclampsia/eclampsia, and smoking [9]. Risk factors unrelated to the puerperal state are like those of the normal population: hypertension, coagulopathies, smoking, vasculitis [2].

The risk of bleeding was somewhat higher in patients with multiple familial lesions than in those with single lesions [8]. In our case, the lesion was unique without a family history of cavernoma.

The mode of onset can be either a haemorrhage or an epilepsy. Table I summarizes the different manifestations found in the literature. The most common clinical manifestation in patients with sustentorial cerebral cavernomas is epilepsy (40 to 80%). In our case, the mode of onset was hemorrhage.

MRI is currently the most performing imaging technique for both diagnosis and follow-up. As well as for monitoring the evolution of the disease. The typical appearance is that of a lesion in heterogeneous hypersignal on T1 and T2 sequences, surrounded by a peripheral hypointense peripheral zone in T1 and T2, without mass effect or perilesional oedema. However, this characteristic appearance is not pathognomonic. MRI also establishes the indication for surgery, guides to the best surgical approach and can provide non-invasive follow-up.

The management strategy for intracranial cavernomas is as follows: asymptomatic cavernomas should be respected, cavernomas that have bled, or rebled or with refractory epilepsy should be evaluated for possible surgical management. Termination of pregnancy has no added value in reducing the risk of bleeding [8].

Delivery needs to be expedited in case of severe preeclampsia and eclampsia, if the source of the bleeding has been controlled, the gynecologist can induce labor as well as delivery. In addition, there is no advantage of a caesarean section over a vaginal delivery, either for the mother or for the child if the lesion has already ruptured [14].

Management depends on gestational age, symptomatology and fetal and maternal risk benefit. Thus, symptomatic cavernomas require management during pregnancy or deferred [15]-[18]. For cavernomas in pregnancy, there is no established and codified strategy that can be used to treat them. Burkhardt et al. propose in their study a therapeutic strategy in the form of an algorithm as shown in Fig. 3. Some authors have suggested that an accessible cavernoma in women planning a future pregnancy is an indication for surgical intervention [20]. In our case we decided to save the mother first and then do a cesarean section to extract the fetus.

TABLE I: LITERATURE REVIEW OF CASES OF CEREBRAL CAVERNOUS IN PARTURIENTS

Fig. 1. Axial CT sections without PDC injection (A) showing a spontaneously hyperdense lesion with perilesional hypodensity and mass effect on the left lateral ventricle with subfalcral involvement. MRI (B, C, D, E) showing a left parietal hematoma without visible arteriovenous malformation.

Fig. 2. Postoperative brain CT scan showing some haemorrhagic remodelling with periwound oedema.
IV. CONCLUSION

Cerebral cavernomas are vascular malformations with a hemorrhagic potential and a prevalence of 0.5% in the general population. The advent of MRI has considerably changed our knowledge of this pathology. Management depends on gestational age, symptomatology, and fetal and maternal risk-benefit.

REFERENCES

[1] S. Yamada, H. Nakase, I. Nakagawa, F. Nishimura, Y. Motoyama, and Y. S. Park, “Cavernous malformations in pregnancy,” Neurol Med Chir 53(8):555-60, 2013.
[2] M. Khan and M. Wasay, “Haemorrhagic strokes in Pregnancy and Puerperium,” Int J Stroke 8(4):265-272, 2013.
[3] A. H. James, C. D. Bushnell, M. G. Jamison, and E. R. Myers, “Incidence and riskfactors for stroke in pregnancy and the puerperium,” Obstet Gynecol 106:509-16, 2005.
[4] C. A. Davie and P. O’Brien, “Stroke and pregnancy,” J Neurol Neurosurg Psychiatry 79:240-5, 2008.
[5] H. Deutsch, G. I. Jallo, A. Faktorovich and F. Epstein, “Spinal intramedullary cavernoma: clinical presentation and surgical outcome,” J Neurosurg 93(Suppl.):65–70, 2000.
[6] Y. Katayama, T. Tsukobawa, T. Maeda and T. Yamamoto, “Surgical management of cavernous malformations of the third ventricle,” J Neurosurg 80(1):64-72, 1994.
[7] H. Zauherman and M. Feinsod, “Orbital hemangioma growth during pregnancy,” Acta Ophthalmol 48:929–33, 1970.
[8] M. Y. Kalani and J. M. Zabramski, “Risk for symptomatic hemorrhage of cerebral cavernous malformations during pregnancy,” J Neurosurg 118:50–5, 2013.
[9] B. T. Bateman, H. C. Schumacher, C. D. Bushnell, J. Pile-Spellman, L. L. Simpson, R. L. Sacco and M. F. Berman, “Intracerebral hemorrhage in pregnancy: frequency, risk factors, and outcome,” Neurology 67(3), 424-429, 2006.
[10] J. E. Warner, J. F. Rizzo 3rd, E. W. Brown and C. S. Ogilvy, “Recurrent chiasmal apoplexy due to cavernous malformation,” J Neuroophthalmol 16:99–106, 1996.
[11] A. Awaada, T. Watson and T. Obeid, “Cavernous angioma presenting as pregnancy-related seizures,” Epilepsia 38:844–6, 1997.
[12] E. Pozzati, N. Acciarri, F. Tognetti, F. Mariani and F. Giangaspero, “Growth, subsequent bleeding, and de novo appearance of cerebral cavernous angiomas,” Neurosurgery 38(4), 662-670, 1996.
[13] E. Nossek, R. E. EksteinM and M. J. Kupferminc, “RamZ,” Neurosurgery and Pregnancy 153(09), 1727-1735, 2011.