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Cerebral Aneurysm Rupture during Pregnancy Resulting in Subdural Hematoma without Subarachnoid Hemorrhage

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Subarachnoid hemorrhage is typically present in cerebral aneurysm rupture, whereas acute subdural hematoma without subarachnoid hemorrhage is rare. We herein report a case of cerebral aneurysm rupture during pregnancy resulting in acute subdural hematoma without subarachnoid hemorrhage. A 37-year-old gravida 4 para 3 pregnant woman was admitted for threatened preterm labor at $29^{4/7}$ weeks of gestation. At $29^{6/7}$ weeks of gestation (day −14), she developed mild left eye pain, which disappeared within one day. At $31^{6/7}$ weeks of gestation (day 0), she developed the sudden onset of severe headache and nausea. A neurological examination revealed no abnormal findings, and analgesics ameliorated her headache. At $32^{1/7}$ weeks of gestation (day 2), after consultations with neurosurgeons, magnetic resonance imaging showed acute subdural hematoma without subarachnoid hemorrhage. Further examinations revealed a cerebral aneurysm. Emergent clipping surgery was performed with the fetus *in utero* in consideration of the immaturity of the fetus and stable maternal/fetal general conditions. At $35^{6/7}$ weeks of gestation (day 28), her headache of unknown cause recurred. Considering the maturity of the fetus, the patient underwent cesarean section with good maternal and neonatal outcomes. The absence of subarachnoid hemorrhage does not eliminate cerebral aneurysm rupture.

Keywords: cerebral aneurysm rupture; clipping surgery; pregnancy; solitary acute subdural hematoma; subarachnoid hemorrhage
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Introduction

Pregnancy-associated hemorrhagic stroke often causes maternal/fetal/neonatal morbidities and/or mortalities. Cerebral aneurysm rupture is a common cause of this type of stroke (Takahashi et al. 2014, Yoshida et al. 2017). Cerebral aneurysm rupture generally accompanies subarachnoid hemorrhage (SAH) and sometimes acute subdural hematoma (aSDH) (Inamasu et al. 2002, Gelabert-Gonzalez et al. 2004, Marebacher et al. 2010, Mrfka et al. 2013); however, aSDH without SAH (solitary aSDH) has rarely been reported. We herein present a case of cerebral aneurysm rupture during pregnancy resulting in solitary aSDH. Emergent clipping surgery was performed with the fetus *in utero* and cesarean section later yielded good maternal and neonatal outcomes.

Case Presentation

A 37-year-old gravida 4 para 3 pregnant woman, with an unremarkable previous medical history, was admitted for threatened preterm labor at $29^{4/7}$ weeks of gestation (Fig. 1). At $29^{6/7}$ weeks of gestation (day −14), she developed mild left eye pain, which disappeared within one day. At $31^{6/7}$ weeks of gestation (day 0), she had the sudden onset of severe headache and nausea. She was alert with a blood pressure of 151/93 mmHg and pulse of 93 bpm. She felt transient weakness of the right leg that lasted for several hours. The Glasgow Coma Scale was measured as 15 points (with a full mark of 15 indicating the least abnormalities), and a neurological examination revealed no abnormal findings. Laboratory tests showed anemia (hemoglobin 7.5 g/dL), with an otherwise normal complete blood count.

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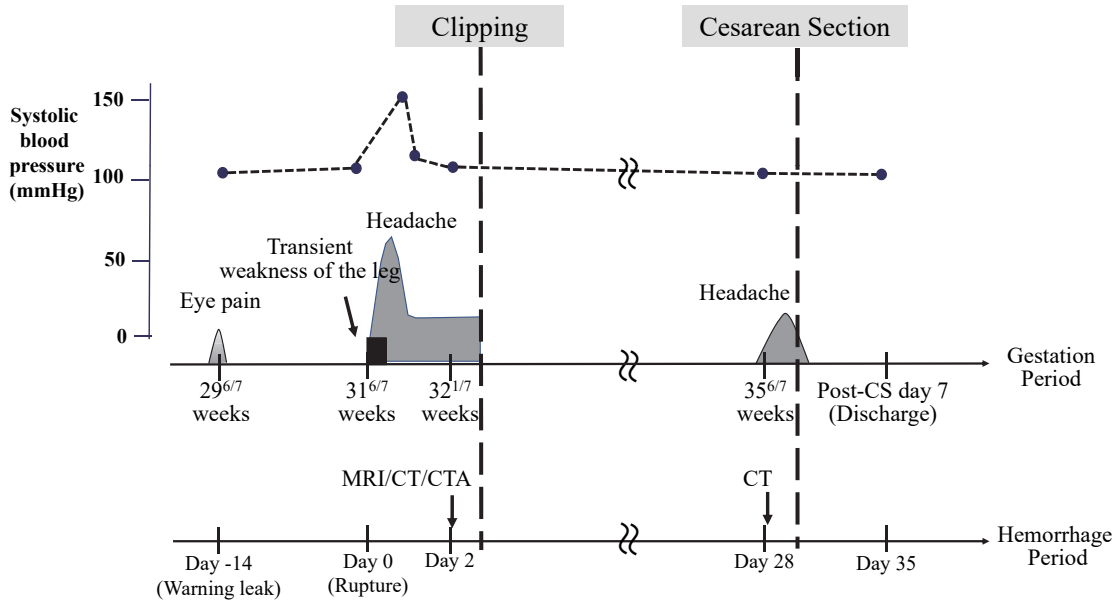

Fig. 1. Timelines of the present case.
The upper and lower lines indicate the gestation period and hemorrhage period, respectively. In the uppermost panel, systolic blood pressure together with neurological symptoms with the superimposition of major events (emergent clipping and cesarean section) are described. Day 0 indicates the major rupture day of the aneurysm.
MRI, magnetic resonance imaging; CT, computed tomography; CTA, computed tomography angiography; CS, cesarean section.

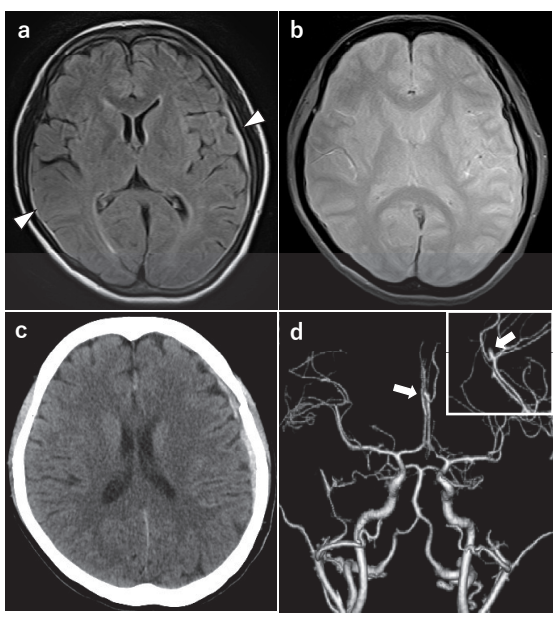

Fig. 2. Magnetic resonance imaging (a, b) and computed tomography (c, d) of the brain. (a) Magnetic resonance imaging of fluid-attenuated inversion recovery (FLAIR) showed subdural hematoma (SDH) without subarachnoid hemorrhage (SAH) on the bilateral convexities (white arrowheads). (b) T2*-weighted imaging showed the same findings as those observed in (a). Note that there was no intraventricular hemorrhage. (c) Computed tomography showed acute SDH without SAH. (d) Computed tomography angiography showed a saccular cerebral aneurysm at the left distal anterior cerebral artery (white arrow). The inset figure shows a lateral view of the aneurysm.

Kidney, liver, and coagulation functions were all within normal levels. Cardiotocography showed a reassuring fetal status. Analgesics ameliorated her headache.

At $32^{1/7}$ weeks of gestation (day 2), after consultations with neurosurgeons, magnetic resonance imaging (MRI) (Fig. 2a, b) and computed tomography (CT) (Fig. 2c) were performed, which revealed slight aSDH bilaterally without SAH. Although SDH generally occurs after head trauma, she had no previous history. CT with contrast enhancement (CT angiography (CTA)) was performed to detect any cerebrovascular lesions, and revealed a saccular cerebral aneurysm (left distal anterior cerebral artery (ACA) aneurysm, diameter of 5 mm) (Fig. 2d). Emergent clipping surgery was immediately performed with the fetus *in utero* in consideration of the immaturity of the fetus and stable maternal/fetal general conditions. Intraoperative findings revealed aSDH; the aneurysm had strongly adhered to the adjacent arachnoid membrane and distal artery (Fig. 3), indicating repeat ruptures. SAH was absent.

There were no complications after surgery. Her headache improved, but recurred at $35^{6/7}$ weeks of gestation (day 28). A neurological examination and CT revealed no abnormal findings: no vasospasms or hydrocephalus was observed. We consulted with multidisciplinary team members and also with the patient and her family members regarding whether the patient should wait until full term (37 weeks of gestation) or undergo an immediate preterm delivery. The latter was selected because i) it was not possible to completely eliminate the presence of other cerebrovascular lesions, ii) at that time, vasospasms and re-rupture were

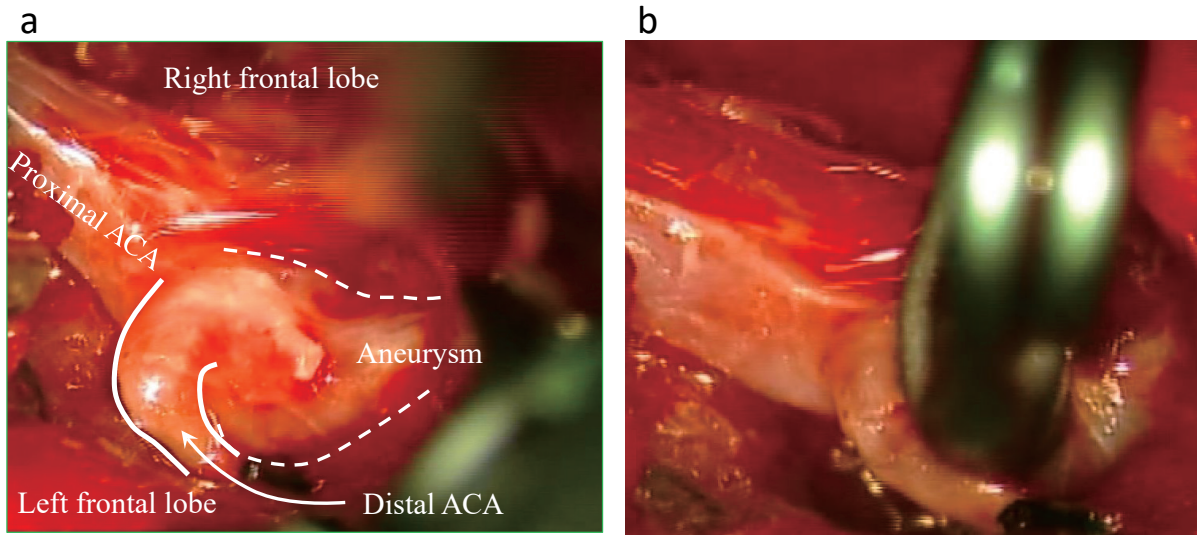

Fig. 3. Intraoperative findings during dissection of the arachnoid membrane at emergent clipping surgery. (a) Before clipping of the aneurysm. The aneurysm (dotted lines) adhered to the arachnoid membrane. Solid lines indicate an artery distal to the aneurysm to which the aneurysm also adhered. (b) The aneurysm was pinched with a clip. ACA, anterior cerebral artery.

denied, and iii) fetal intact survival was highly expected. Cesarean section was performed due to an immature cervix, yielding a 2,025 g neonate with an Apgar score 8/9 (1/5 minutes). Her headache subsequently disappeared and its cause was not identified. She was discharged post-Cesarean section day 7 without any neurological sequelae. The baby was intact without sequelae at 1 year of age.

Discussion

The patient developed the acute onset of severe headache and transient weakness of the right leg. Brain MRI and CT revealed solitary aSDH. CTA showed a ruptured aneurysm in the left distal ACA. Emergent clipping surgery led to good maternal/neonatal outcomes. This case suggests that solitary aSDH is a manifestation of cerebral aneurysm rupture in pregnancy. Solitary aSDH in cerebral aneurysm rupture is rare; to the best of our knowledge, 52 cases have been reported to date (Katati et al. 2018), among which only one was pregnant (Hubert 1994). Therefore, this patient is the second pregnant case of solitary aSDH in cerebral aneurysm rupture to be reported.

In a review of 41 non-pregnant patients with solitary aSDH, ruptured aneurysms were located, in descending order, in the internal carotid artery-posterior communicating artery (22 cases, 43%), middle cerebral artery (16 cases, 31%), anterior communicating artery (Acom, 6 cases, 12%), and distal ACA (4 cases, 7%) (Gong et al. 2014). This aneurysmal topography is similar to that of ruptured aneurysms reported in a pregnant population with SAH (Dias and Sekhar 1990, Kataoka et al. 2013), with the internal carotid artery being the most common site (49%). However, it differs from that of the general population, in which the most common site of rupture is Acom (39%) (Kassell et al. 1990). Aneurysmal rupture in the distal ACA is also rare in the general population (6%) (Lehecka et al. 2008). The present pregnant case had a ruptured aneurysm in the distal ACA, while that in the first pregnant case was in the Acom (Hubert 1994). These Acom/distal-ACA sites were rare in the pregnant population. Therefore, Acom/distal-ACA sites may be related to the underlying mechanism of solitary aSDH during pregnancy; however, since data are only currently available on two cases, concrete conclusions cannot yet be reached.

In a review of non-pregnant cases (Gong et al. 2014), the characteristics of solitary aSDH in distal ACA (4 cases) were as follows: (1) SDH was located in the convexity and/or interhemispheric fissure, (2) outcomes were poor when symptoms at onset were severe (coma), and (3) outcomes were good when symptoms at onset were mild (headache or nausea). The present pregnant case had (1) and (3). Although the site of aSDH revealed by CT was not adjacent to the ruptured aneurysm (distal ACA), but in the bilateral convexities, the initial manifestation of transient weakness in the right leg strongly suggested damage to the medial surface of the left hemisphere due to minor ruptures of the aneurysm (left distal ACA). Her neurological symptoms included headache and nausea, and she had a favorable outcome after emergent clipping surgery and Cesarean section. Early detection and interventions may be important for a good outcome. Therefore, this disease entity needs to be considered by the primary physician as well as obstetricians.

One possible explanation for solitary aSDH is as follows (Gong et al. 2014). The brain is covered by three structures: the skull, dura, and arachnoid membrane. The cerebral artery, and thus, the aneurysm in the present case, exist under (the inner side of) the arachnoid membrane. The rupture of an aneurysm generally causes hemorrhage

under the arachnoid membrane, namely, SAH. Repeated hemorrhage due to minor aneurysmal rupture results in adhesion between the aneurysmal wall and arachnoid membrane, and thus, the aneurysmal walls make direct contact with the subdural space. A large rupture then occurs, with blood directly entering the subdural space (not the subarachnoid space), causing aSDH without SAH. In this case, surgery revealed that the aneurysm strongly adhered to the adjacent arachnoid membrane. Furthermore, mild eye pain on day −14 indicated minor recurrent ruptures of the aneurysm. Some pathophysiological similarities exist in hemorrhagic disorders other than cerebral aneurysms. In aortobronchopulmonary and aorto-enteric fistulas, the aortic aneurysm adheres to the bronchus or intestinal wall, respectively, and the fistula that forms is in direct contact with them. Bleeding sometimes occurs in the bronchus lumen and intestinal lumen (not in the mediastinum or abdomen), respectively. Therefore, ruptured vessels may not always hemorrhage to the site at which vessels are actually located.

In the present case, emergent clipping was performed at $32^{1/7}$ weeks of gestation (day 2) with the fetus *in utero*. Headache of unknown cause recurred at $35^{6/7}$ weeks of gestation (day 28), which prompted us to perform preterm Cesarean section. Due to the gestational weeks of the events and maternal/fetal conditions, clipping surgery with the fetus *in utero* and later preterm Cesarean section were natural choices. The first pregnant case reported by Hubert (1994) had solitary aSDH at 39 weeks of gestation, by which stage the fetus had matured, and thus, clipping surgery was performed after Cesarean section. The strategy selected, namely, clipping surgery with the fetus *in utero* versus after delivery, depends on various factors, i.e., infant maturity, neonatal treatment level, and the neurological/systemic status/condition of the mother. The treatment strategy for ruptured aneurysms and the timing of delivery are still controversial. Therefore, treatment scenarios need to be individualized.

The following result from the present case is important: aSDH without SAH does not preclude cerebral aneurysm rupture. A survey of cerebral vascular disorders needs to be considered in order to give warranty to the primary physician and obstetricians to consider this disease entity.

Conflict of Interest

The authors declare no conflict of interest.

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