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Case Report

Neonatal subpial hemorrhage along the medial side of the temporal lobe: Two case reports ☆,☆☆

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ABSTRACT

Neonatal subpial hemorrhage has been underrecognized until recently and its pathophysiology remains unclear. Advances in magnetic resonance imaging have facilitated the identification of hemorrhage within the subpial space and cohort studies recently reported its imaging and clinical features. We encountered two cases of neonatal subpial hemorrhage along the medial side of the temporal lobe. Case 1: A 1-day-old boy had repeated apneic attacks with cyanosis from 2 hours after birth at 39 weeks of gestation by vacuum extraction delivery. Computed tomography and magnetic resonance imaging showed subpial hemorrhage from the medial to caudal side of the right temporal lobe with T2 prolongation in the underlying cerebral parenchyma. Case 2: A 0-day-old boy had repeated apneic attacks with cyanosis from 3 hours after birth at 39 weeks of gestation by vaginal delivery. Subpial hemorrhage was observed from the anterior to medial side of the left temporal lobe on computed tomography and magnetic resonance imaging. On magnetic resonance imaging, the adjacent brain parenchyma showed a hyperintense signal on T2-weighted imaging. No abnormalities or signs of fetal distress were noted in the course of delivery. A mildly prolonged activated partial thromboplastin clotting time, an elevated D-dimer level, and low fibrinogen level were detected in a blood examination after birth in both cases. Both cases had subpial hemorrhage along the medial side of the temporal lobe, which suggested that an external mechanical force with fetal head molding during delivery caused subpial hemorrhage; however, other factors, including coagulopathy, may be involved in its pathophysiology.

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Introduction

Neonatal subpial hemorrhage is a subtype of intracranial hemorrhage that was recently reported in a series of cohort studies; however, it has been underrecognized [1–4]. The clinical findings and long-term neurological prognosis of subpial hemorrhage have been shown to vary [1–4]. Since difficulties are associated with accurately diagnosing subpial hemorrhage using imaging modalities other than magnetic resonance imaging (MRI), most cases have historically been grouped with subarachnoid hemorrhage under the broader term of leptomeningeal hemorrhage [1,2,5]; therefore, its frequency may be underestimated. Although the exact pathophysiology of subpial hemorrhage has not yet been clarified, several theories have been proposed. In a study of seven newborns, Huang et al. confirmed leptomeningeal hemorrhage with MRI and argued that the underlying cause was related to a brain contusion or venous compression due to birth related-injury or fetal skull molding [5]. Cain et al. suggested neonatal subpial hemorrhage as a pre-delivery rather than peri- or post-delivery occurrence based on findings showing fetal distress and acute coagulation abnormalities in many newborns with subpial hemorrhage [3].

We encountered two cases of neonatal subpial hemorrhage along the medial side of the temporal lobe on MRI for apneic attacks. Previous studies indicated that subpial hemorrhage is more common in the temporal lobe, and all reported cases of subpial hemorrhage have been detected along the lateral side of the temporal lobe [1–5]. We herein describe these cases in consideration of the underlying pathophysiology.

Case report

Case 1: A full-term male newborn was born at 39 w 4 d by vacuum extraction delivery. His birth weight was 3478 g, and Apgar scores were 9 and 10 at 1 and 5 minutes, respectively. There were no abnormalities or sign of fetal distress during delivery. He developed repeated apneic attacks with cyanosis from 2 hours after birth and was transported to our hospital. Abnormal waves were confirmed on electroencephalography (EEG) and treatment, such as anticonvulsants, was initiated. A hematoma from the medial to caudal side of the right temporal lobe was detected on head computed tomography (CT) on day 1. MRI on day 12 showed a hematoma at the same site with a high signal intensity on T1-weighted images (Fig. 1A, B) and high and partially low signal intensi-

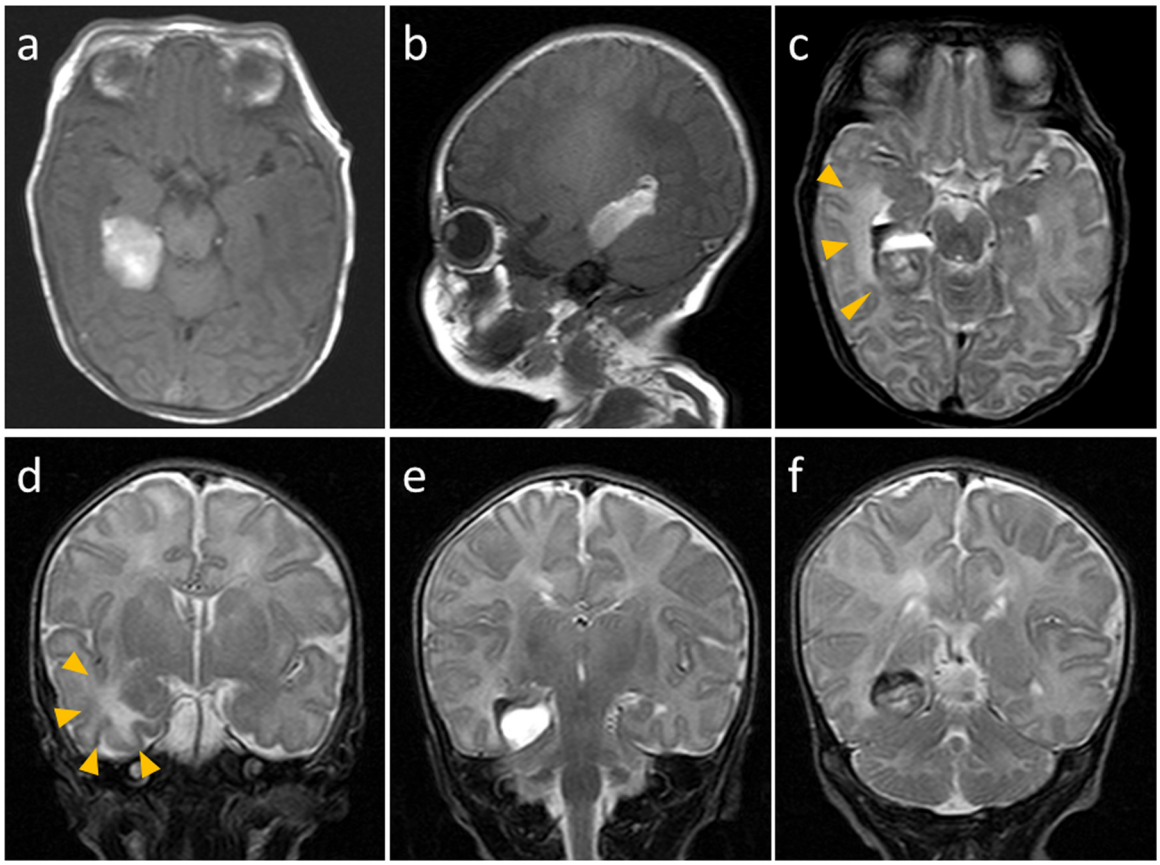

Fig. 1 – Case 1: Brain MRI on day 12. Axial T1-weighted image (A) and sagittal T1-weighted image (B) showing a hematoma with a high signal intensity along the medial side of the temporal lobe. A mixed signal intensity on T2-weighted images was observed along the medial side of right temporal lobe overlying the cortex (C-F). T2 prolongation was noted in the adjacent brain parenchyma (C, D: arrowhead).

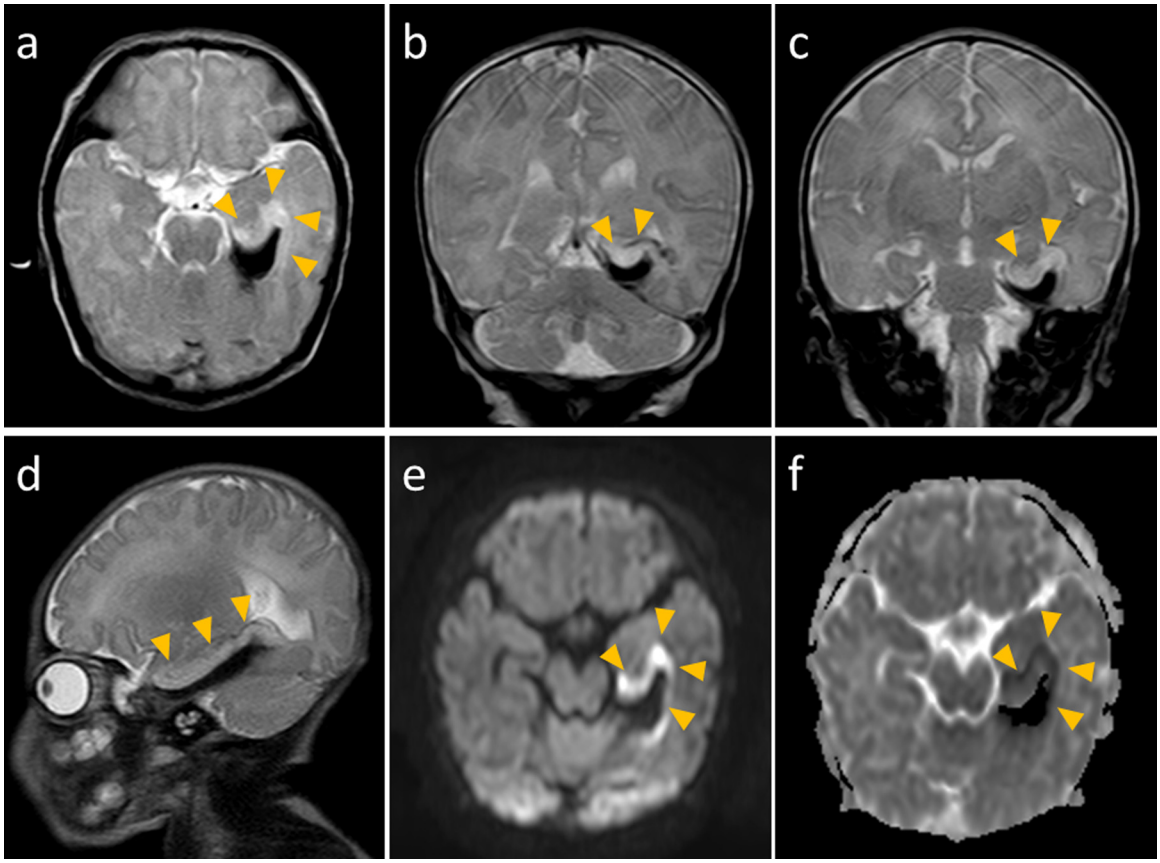

Fig. 2 – Case 2: Brain MRI on day 4. Axial (A), coronal (B, C), and sagittal (D) T2-weighted images showing a hematoma with a low signal intensity along the medial side of the left temporal lobe overlying the cortex with a high signal intensity at the underlying brain parenchyma (arrowheads). A diffusion-weighed image (E) and ADC map (F) indicated diffusion restriction, suggesting venous infarction (arrowheads).

ties on T2-weighed images overlying the cortex to deep into the sulcus (Fig. 1 C-F). Furthermore, T2 prolongation was observed in the adjacent brain parenchyma (Fig. 1C, D). Based on the localization of the hematoma and parenchymal signal changes, the patient was diagnosed with subpial hemorrhage and parenchymal edema. Blood examinations showed a prolonged activated partial thromboplastin clotting time (APTT), low fibrinogen level, and elevated D-dimer level. His platelet count and the prothrombin time and international normalized ratio were normal. These abnormal values had returned to within the normal ranges in the follow-up. Seizures subsequently disappeared and the patient was discharged on day 14. He has not had any seizures by the age of 6 years and is in normal development. Head CT for repeated vomiting at the age of 4 years showed no abnormal findings, such as parenchymal atrophy at the site of postnatal hematoma.

Case 2: A 0-day-old boy had repeated apneic attacks with cyanosis from 3 hours after birth at 39 w 2 d by vaginal delivery. His birth weight was 2718 g, and Apgar scores were 9 and 10 at 1 and 5 minutes, respectively. No abnormalities, such as fetal distress, were observed during delivery. EEG showed abnormal waves. Head MRI on day 4 showed a hematoma with a high signal intensity on T1-weighted images and low signal intensity on T2-weighted images from the anterior to medial side of the left temporal lobe, overlying the cortex, but separated from the adjacent CSF by a smooth border (Fig. 2A-D). Diffusion restriction was detected in the underlying cortex (Fig. 2E, F). The patient was diagnosed with subpial hemorrhage based on the localization of the hematoma and the presence of diffusion restriction in the underlying cortex. Transient APTT prolongation, a low fibrinogen level, and elevated D-dimer level were observed after birth. The patient was also treated with medication and seizures disappeared. He was discharged on day 28. He has had no seizures by the age of 3 years and is in normal development.

In both cases, hemorrhagic diathesis and postnatal trauma were ruled out and considered to be birth-related trauma.

Discussion

Neonatal subpial hemorrhage has been underrecognized until recently and pooly understood. Friede et al. initially discussed subpial hemorrhage in the autopsy findings of nine infants [6]. Advances in MRI have resulted in subpial hemorrhage becoming more recognizable, with blood collection along the margin of the cerebral parenchyma extending into the cerebral sulci often deeply displacing the underlying cortical ribbon, which is accompanied by venous congestion or infarction in the un-

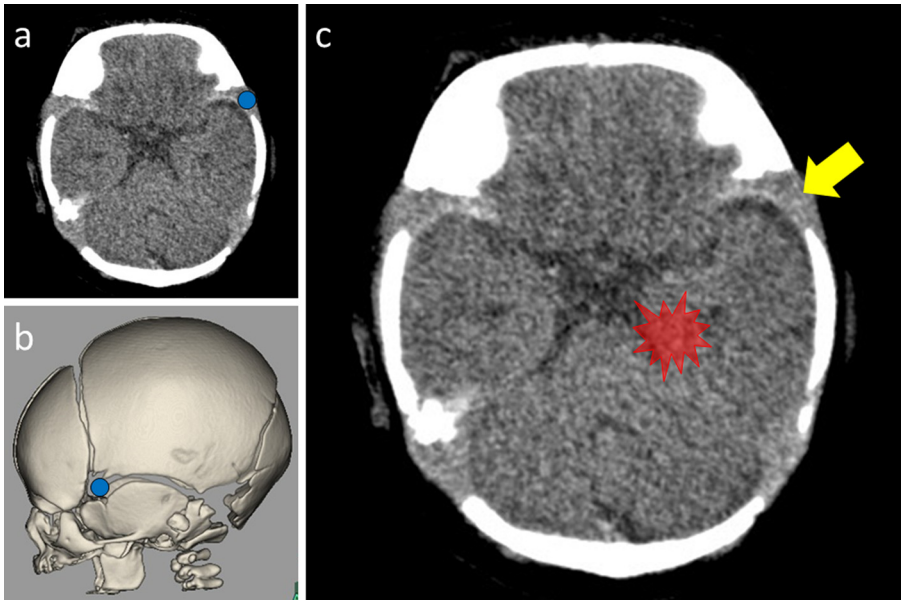

Fig. 3 – The pterion, or sphenoidal fontanelle, is marked with • on the axial image (A) and 3-D reconstruction image of newborn head CT (B). If a force is applied at the pterion and sphenoidal fontanelle (C, yellow arrow), the opposite side of the temporal lobe collides with the tent (C, red), and damage to the basilar venous plexus may result in subpial hemorrhage and underlying brain edema due to venous stasis.

derlying parenchyma [1–5]. The appearance of hypointense subpial hemorrhage and the underlying hyperintense cerebral cortex on T2-weighed images resemble the Yin-yang symbol in Chinese philosophy, which is a characteristic finding [4]. Huang et al. described the MRI findings and clinical course of seven neonates confirmed to have spontaneous superficial parenchymal and leptomeningeal hemorrhage. They found that 4 out of the 7 cases had hematomas near the pterion, which is a large, relatively unprotected sutural confluence in the neonate, while another two hematomas were close to the suture, and the cause of spontaneous superficial parenchymal and leptomeningeal hemorrhage was considered to be local trauma with contusions or venous compression/occlusion [5]. Other risk factors have been proposed for neonatal leptomeningeal hemorrhage, including neonatal asphyxia, clotting disorders, venous sinus compression, intracranial pressure abnormality, and incomplete regression of the primary vascular network [1]. Cain et al. recently summarized 17 cases of neonatal subpial hemorrhage, which included 2 cases (12%) of assisted delivery, 8 (47%) of fetal distress, and 13 (77%) of acute coagulation abnormalities. Based on clinical findings, they suggested that neonatal subpial hemorrhage occurred predelivery rather than peri- or post-delivery [3].

Evidence for a birth-related injury or fetal head molding as the cause of subpial hemorrhage is that most cases of subpial hemorrhage occurred near the pterion or suture, as reported by Huang et al [5].. On the other hand, Cain et al. reported that it is unlikely that mechanical external forces during delivery is the main cause of subpial hemorrhage, because the frequency of assisted delivery was low (12%) in their series [3]. However, subdural hematoma, a common type of intracranial hemorrhage in neonates, is generally attributed to a tear in the falx, tentorium, or bridging vein due to cranial extension and distortion during delivery and, thus, is regarded as a mechanical birth-related injury. Subdural hematoma is considered to be caused by physical head molding based on previous findings showing no relationship between the occurrence of subdural hematoma and the mode of delivery, whether spontaneous or assisted, or the time of delivery, and vaginal delivery was the only risk factor [7–9]. Therefore, trauma due to mechanical external force cannot be denied in vaginal delivery.

Coagulopathy has also been proposed as one of the causes of subpial hemorrhage. Cain et al. found that transient coagulation abnormalities were common in their cohort study [3]. Fetal distress and coagulation abnormalities were also commonly associated with neonatal hemorrhagic stroke in other cohort study [10]. On the other hand, Dabrowski et al. showed that only 7 out of 31 cases of subpial hemorrhage (23%) had an abnormal coagulation status [2], while Assis et al. reported that none of their 16 cases of subpial hemorrhage had coagulopathy [4]. Therefore, coagulopathy does not appear to be the main factor causing neonatal subpial hemorrhage and, thus, its etiology may be multifactorial.

Subpial hemorrhage was previously reported to be located just underneath the skull; however, hemorrhage in the present cases was located along the medial side of the temporal lobe. Although the presence of coagulopathy or intracranial pressure fluctuations may have contributed to some extent, we suspect that local trauma with a contusion or venous compression/occlusion was the main causative factor for subpial hemorrhage. We propose the following pathophysiology for subpial hemorrhage on the medial side of the temporal lobe. When a strong external force is applied to the pterion and squamosal suture, the brain parenchyma just under the pterion is compressed and the temporal lobe is medially deformed inwardly with compression or obstruction of the basal

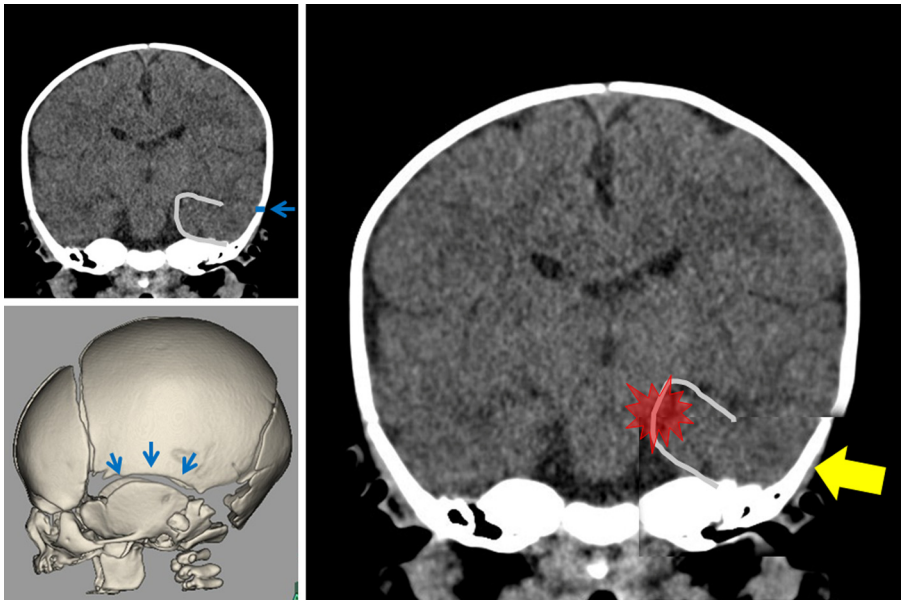

Fig. 4 – The squamosal suture is indicated by the arrow on the coronal CT image (A) and 3-D reconstruction image of newborn head CT (B). When the squamosal suture is compressed (C, yellow arrow), the temporal bone is deformed and displaced inwardly, and the pushed temporal lobe may cause compression of the basal vein (C, red), resulting in subpial hemorrhage and venous infarction.

vein, which causes subpial hemorrhage and venous infarction of the medial temporal lobe (Figs. 3C and 4C).

Previous studies demonstrated that the clinical features and prognosis of subpial hemorrhage varied among cases, particularly those born term or preterm. The present cases had very similar clinical courses. No abnormalities, such as fetal distress, were observed during delivery in either case. Both cases were born by vaginal delivery, one with suction and the other by spontaneous delivery. Apneic attacks occurred several hours after birth and soon disappeared. These patients have been followed for at least 3 years without further seizures and their development has been normal. In both cases, CT or MRI was performed during the clinical course, and no abnormalities, such as atrophy, were observed in the brain parenchyma around the site at which subpial hemorrhage occurred.

Conclusion

We encountered two cases of neonatal subpial hemorrhage along the medial side of the temporal lobe. Although the influence of coagulopathy cannot be ruled out, we suspected that mechanical force with fetal head molding during delivery caused subpial hemorrhage. However, multiple factors may contribute to the occurrence of subpial hemorrhage and, thus, the accumulation of more cases is needed to clarify its exact pathophysiology.

Availability of data and material

Not applicable

Code availability

Not applicable

Patient consent

Written informed consent was obtained from both patients' parents in this study.

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