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CASE REPORT Nigerian Journal of Paediatrics 2024; Volume 51(3):361-368. <u>https://dx.doi.org/10.4314/njp.v51i4.06</u> Chronic Encapsulated Intracerebral Haematoma in a Two-Month-Old Infant Following Forceps-Assisted

**Delivery: A Case Report** 

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#### Abstract

Chronic encapsulated intracerebral haematoma is a rare form of intracerebral haematoma that grows progressively. It is usually laden with diagnostic challenges, and only a few cases have been diagnosed preoperatively. This report is about an eight-week-old male infant who presented with a history of fever, excessive crying, multiple seizure episodes and loss of consciousness. He was delivered with forceps assistance. Transfontanelle ultrasonography showed an oval encapsulated hyperechoic mass in the left frontal lobe, associated compression of the adjacent limb of the anterior ventricle, and mild intraventricular hyperechoic collection. Magnetic resonance imaging of the brain revealed a well-defined encapsulated mass in the left frontal lobe extending and compressing on the adjacent lateral ventricles with a significant midline shift to the contralateral side. Thus, a diagnosis of encapsulated intracerebral haematoma was made. The infant had a craniotomy, and the lesion was excised *en bloc*. The histological analysis was consistent with blood clots. This case report highlights one of the possible complications of forceps-assisted delivery. A comprehensive history and thorough clinical examination with a multidisciplinary approach should be constituted for prompt and appropriate treatment.

Keywords: Chronic Encapsulated Intracerebral Haematoma, Craniotomy, Forceps-assisted Delivery, Magnetic Resonance Imaging.

#### Introduction

Chronic encapsulated intracerebral haematoma is a rare form that progresses slowly, mimics intracranial tumours or other space-occupying lesions and presents diagnostic challenges.<sup>1</sup> It was first reported in 1981 by Hirsh, and it was characterized by the presence of a fibrotic (fibrin) capsule that resembles the outer capsule of chronic subdural haematoma.<sup>2</sup> Chronic encapsulated intracerebral haematoma increases in size slowly while forming the capsule. It may present with increased intracranial pressure, neurologic deficit, and obstructive hydrocephalus.<sup>3</sup> The aetiology of chronic encapsulated intracerebral haematoma is unknown. However, it is thought to be associated with trauma, vascular malformation, bleeding disorder, and neoplasm.<sup>4,5</sup> The mechanism of slow and progressive growth of the lesion is believed to be due to repeated bleeding from the new blood vessels in the capsule. It can occur at any age and has been described in patients aged two months to 80 years and above.<sup>1,3-5</sup>

The index patient was evaluated using trans fontanelle ultrasonography and magnetic resonance imaging. This report is important because of the rarity of this disease entity and the paucity of literature reviews in Nigeria and some African countries. Notably, it is necessary to consider it in the differential diagnoses of intracerebral space-occupying lesions and ensure early diagnoses.

Case Presentation: Baby UP, an 8-week-old male infant, presented to the Paediatric Emergency Unit of the National Hospital, Abujawith a history of sudden onset of fever of four days duration, excessive crying, and multiple seizure episodes of three days duration and loss of consciousness of 15 hours duration, all prior to presentation to the hospital. He was well until four days before the presentation when his mother noticed he was febrile to touch. A day later, he became irritable and restless and cried excessively. There was associated feed refusal, evidenced by the baby's inability to suckle. A few hours following this restlessness, the baby had upward rolling of eyeballs, with contraction of his limb's muscles. This occurred multiple times (mother estimated more than 20 times), with each episode lasting between 5-10 seconds. The baby subsequently lost consciousness as he was being rushed to a peripheral hospital. He briefly regained consciousness after 30 minutes and relapsed into unconsciousness. The baby was given some medications (intramuscular and intravenous injections) to abort the seizure before referral to the National Hospital.

The baby's conception was desired and achieved naturally. His mother was 33 years, and his father was 45 years. The mother booked for antenatal care at a gestational age of 18 weeks in a private hospital. There were no significant ante-natal events. The patient is the second child of both parents and was delivered vaginally at term. There was a history of prolonged labour and forceps-assisted delivery. The baby cried immediately after birth. The immediate neonatal period was uneventful. The patient was on exclusive breastfeeding.

Physical examination revealed an acutely ill baby, unconscious, febrile, pale, anicteric, and acyanosed, with reduced muscle tone. The occipitofrontal circumference was 37.5cm (mildly increased), the anterior fontanelle was not bulging and there were no lateralizing signs.

The patient was anaemic with a Packed Cell Volume of 21%. The remaining laboratory investigation findings were unremarkable. Transfontannelle ultrasonography showed a fairly oval encapsulated hyperechoic mass in the left frontal lobe with associated compression of the adjacent limb of the anterior ventricle, mild intraventricular hyperechoic collection, but no obvious dilatation of the other parts of the ventricles as shown in Figure 1. Magnetic resonance imaging of the brain revealed a well-defined encapsulated mass in the left frontal lobe extending and compressing on the adjacent lateral ventricles with a significant midline shift to the contralateral side, as illustrated in Figures 2 and 3 in different planes. The mass showed heterogenous intensity with predominantly hyperintensity on T1-weighted, heterogenous intensity with hypointense capsule on T2-weighted, and marked blooming in gradient recall echo image. There was no significant enhancement in the post-gadolinium image. Magnetic resonance angiography showed hyperintense vascular flush with a deviation of surrounding vessels but no obvious evidence of vascular rupture or malformation, as shown in Figures 4 and 5. Based on the imaging findings, a diagnosis of complex cystic mass with features suggestive of encapsulated intracerebral haematoma was made. Haemorrhagic/calcific tumours like teratoma and vascular malformations were also possible differential diagnoses that were entertained.

The baby was selected for surgical treatment but was not booked for surgery immediately due to financial incapacitation; he was therefore admitted and placed on 0.9 mg dexamethasone q8-hours, phenytoin 44 mg *stat* and 7.5mg q12-hours, and ceftriaxone 148 mg q12-hours, all for five days. This conservative management protocol was meant to control seizures, reduce the raised intracranial pressure, and treat infections.

One month later, while waiting for surgery, the patient's condition deteriorated; his head progressively increased in circumference with associated seizures and loss of consciousness. This necessitated emergency attention at the intensive care unit, where he was stabilized and prepared for surgery. A craniotomy was done, and the lesion was excised *en bloc*, as illustrated

in Figure 6. The surgical findings included 20-30 mls of altered blood within the encapsulated brownish mass at the frontal lobe with extension into the ipsilateral lateral ventricles, as shown in Figure 7. The specimen was sent for histopathological analysis with 10% neutral buffered formalin. The histology report revealed fibrin clots with red blood cells only. There was no histopathologic evidence of vascular malformation. The final diagnosis of encapsulated haematoma was established. The patient made dramatic improvement postsurgery and was discharged two weeks later. He is currently on monthly follow-up and has no new complaints.



Figure 1: A fairly oval heterogenous but predominantly hyperechoic mass with a hyperechoic capsule was noted within the frontal lobe (long white arrows) with associated compression of the adjacent lateral ventricle and mild intraventricular haemorrhage (short white arrow).

#### Ethical considerations

The case report was conducted in compliance with the guidelines of the Helsinki Declaration on biomedical research in human subjects. The parents also gave informed consent for using the child's data for research.

#### Discussion

Chronic encapsulated intracerebral haematoma (CEIH) is a rare type of haematoma first

described by Hirsh in 1981.<sup>2</sup> It progressively enlarges over time and encapsulates and mimics intracerebral space-occupying lesions. It is associated with diagnostic difficulty such that only about 20% of this lesion is diagnosed preoperatively, as documented by Nishiyama et al.<sup>1</sup>. The clinical diagnosis of the disease is even harder because of non-specific history, symptoms and signs in the setting of a wide age range that can be affected.<sup>1,3,4,6</sup>

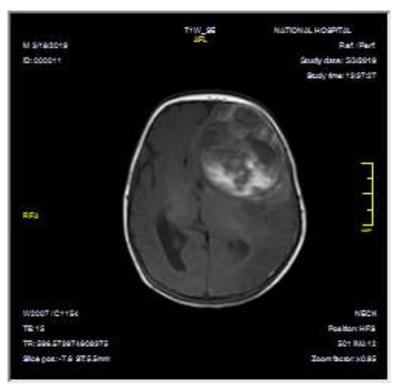


Figure 2: An axial T1 weighted magnetic resonance image of the brain at the level of the Sylvian fissures showing heterogenous intensity mass at the left frontal lobe with effacement of the ipsilateral Sylvian fissure and frontal horn of the lateral ventricle and associated severe subfalcine herniation.



Figure 3: An axial T2-weighted magnetic resonance image of the same patient showing the lesion as a heterogeneously hyperintense left frontal lobe mass with hypointense capsular rim.

Many theories have been proposed to explain the aetiology and mechanism of this disease. The exact aetiology, however, is still unclear. The histological findings reported by Hirsh *et al.* indicated that vascular malformation due to fibroblasts may be responsible for the development of the capsule in this lesion. It has been widely accepted that angiographically-occult intracranial vascular malformations (AOIVMs) may cause headache, intracranial haemorrhage, and refractory epilepsy; and has been classified into arteriovenous malformations, cavernous angiomas, capillary telangiectasias, and venous angiomas.<sup>6,7</sup>

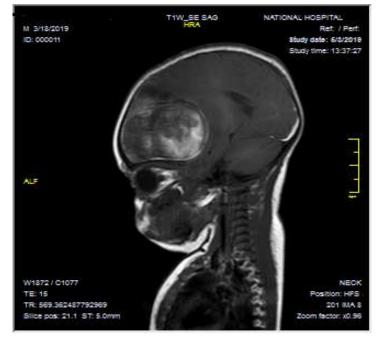


Figure 4: A sagittal T1-weighted magnetic resonance image of the index patient showing heterogenous intensity mass with hyperintense rim.



Figure 5: Magnetic resonance cerebral angiography revealed vascular flush in the left frontal region with displacement of surrounding vessels.

Takeuchi *et al.* reported that vascular endothelial growth factor may also play a role

in encapsulated intracerebral haematoma's evolution since new blood vessel and

lymphocyte infiltrations are observed.<sup>8</sup> There was no demonstrable evidence of vascular malformation on imaging in the index case.

Histological studies also reported the presence of fibrin dots with red blood cells, consistent with the reports of other reviewers.<sup>1-8</sup>



Figure 6: Gross intraoperative image taken during craniotomy.



Figure 7: Gross specimen of the mass excised *en-block*.

Clinically, encapsulated intracerebral haematomas are usually encountered in adults with a history of hypertension.<sup>9</sup> It is rare among infants and is usually due to head trauma, bleeding diasthesis, arteriovenous malformation, and neoplasm.<sup>4,5,9</sup> Unlike other types of intracerebral haematoma, which are

usually sudden in onset and may resolve spontaneously within 6-8 months, chronic encapsulated intracerebral haematoma is characterized by gradual clinical onset, and spontaneous resolution does not usually occur.<sup>1,3-5,7,9</sup> Furthermore, Cakir *et al.* <sup>9</sup> reported a case of ruptured encapsulated intracerebral haematoma in a 2-month-old female with a history of perinatal head injury.9 Cakir's patient shared clinical and radiological features with the index patient. Furthermore, Cakir et al. documented a history of perinatal head injury that was asymptomatic in the first month and presented with a seizure later. The present case also occurred in a two-month-old male with a history of birth trauma who was asymptomatic until a few days prior to presentation and also had a seizure. Cakir and colleagues reported magnetic resonance imaging features of the lesion as a well-circumscribed heterogeneous intensity mass with the hyperintense rim on the T1-weighted image, the heterogeneous intensity with a hypointense rim on the T2weighted image, and faint enhancement with gadolinium.9 These features were similar to those seen in the index case. However, unlike the case documented by Cakir and colleagues, the capsule of the index case was intact at the time of surgery.

In addition, Aoki et al.<sup>10</sup> also reported a case of encapsulated intracerebral haematoma in a sixmonth-old infant who presented with an inability to achieve head control. They documented a rounded hyperdense intracerebral mass using computed tomography. Subsequently, the mass was surgically excised en bloc with a good prognosis. Finally, Nishiyama et al.<sup>1</sup> reported a series of three cases of encapsulated intracerebral haematoma in patients aged 58, 59, and 62 years. Although these cases were seen in adults, the imaging findings in magnetic resonance imaging were similar to those of the index patient. In addition, Nishiyama et al.<sup>1</sup> documented а consistent computed tomographic finding of hyperdense mass with perilesional oedema in all their cases. The index patient was not evaluated with computed tomography due to concerns about ionizing radiation and because it was not necessary. The treatment of encapsulated haematoma entails surgical excision of mass en bloc, as was done in the cases reviewed 1,3-9 The treatment

outcome in reviewed works shows a good prognosis, which is even better in infants.<sup>1,3-7,9</sup>

#### Conclusion

Chronic encapsulated intracerebral haematoma is rare in infants, but it is associated with trauma, vascular malformations, bleeding diathesis, or neoplasm. It mimics other intracranial space-occupying lesions, making diagnosing the condition preoperatively difficult. A detailed clinical history and optimal imaging evaluation are key to making a preoperative diagnosis of CEIH. Chronic encapsulated intracerebral haematoma can be treated by surgical excision of the mass *en bloc* and with a good prognosis. Clinicians and radiologists should develop a high index of suspicion in patients with risk factors for this disease entity to ensure early diagnosis and prompt treatment.

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