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# Osteitis and cerebral empyema in sickle cell disease: About two cases

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**Abstract**
Subdural empyema or brain abscess are infectious lesions which can complicated meningitis or frontal sinusitis infection. Spontaneous cranial vault osteomyelitis or osteitis followed by cerebral empyema are rare intracranial complications which can occured in sickle cell disease and immune compromise due to intravenous drug abuse. Sickle cell disease is a hemoglobinopathy responsible for many complications that are sometimes severe. Importance or early diagnosis and proper treatment is the key for the managment for these complications. The authors reported two cases of subdural empyema which were surgically evacuated and treated with adapted antibiotics with good outcomes.

**Keywords:** subdural empyema, sickle cell disease, surgery, antibiotic, osteitis

## Introduction

Subdural empyema or brain abscess are infectious lesions which can complicated meningitis or frontal sinusitis infection. Spontaneous cranial vault osteomyelitis or osteitis followed by cerebral empyema are rare intracranial complications which can occured in sickle cell disease and immune compromise [1, 2]. Sickle cell disease is an autosomal recessive genetic hemoglobin disorder characterized by the presence of an abnormal hemoglobin (hemoglobin S) [3, 4]. African countries are the most affected countries. The major complications after vaso occlusives crises are neurological and represented by ischemic stroke, spontaneous hemorrhagic. Intracranial infections are rare and engage the vital and functional prognosis [2, 3, 11, 14].

## Atients and Methods

**Case 1:** A 14-year-old-patient, known homozygous sickle cell disease SS, was admitted to our neurosurgical department with 2 weeks history of headache with fever. On admission, he present loss consciousness with a Glasgow Coma Scale 11/15 and left hemiparesis. The diagnosis of meningoencephalitis was retain and he underwent medical treatment after brain CT scan and lumbar punction. Worsening of the symptomatology and progressive onset of right frontal swelling (a), a week later, with Glasgow coma scale about 8/15 allowed another brain CT scan with contrast which showed an image of right frontal epidural empyema (b) associated with interhemispheric empyema (c) with mass effect and frontal bone osteitis (Figure 1). Biological examination showed anemia at 4.4g/dL, hematocrit at 15.8%, white blood cells at 10 Giga/L, platelets at 470 Giga/L, and Prothrombin at 84%. Before the intervention, the patient received a partial transfusion protocol with the aim of stabilizing the hemoglobin value over 10 g/l and reducing the percentage of pathologic HbSC below 30%. No transfusion complications such as hyperhemolysis or alloimmunization were observe. He underwent surgery with an evacuation of pus and bacteriological examination identified pseudomonas aeruginosa infection. An double adapted antibiotic therapy was started with clinical improvement after 4 weeks intravenous treatment.

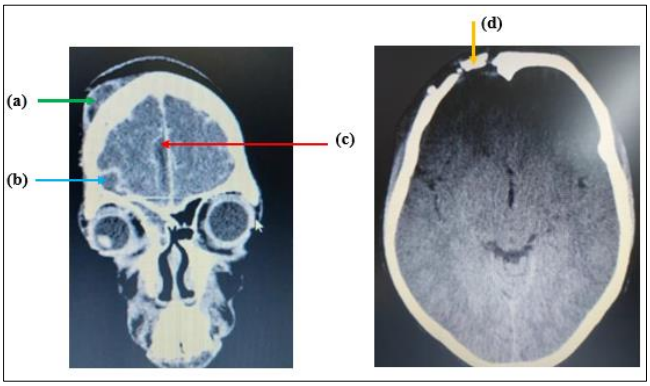


**Fig 1:** Brain CT with contrast showing frontal swelling, (a:green), right epidural empyema (B: Blue) and interhemispheric empyema (c:red) and osteitis on axial view (d:yellow)

**Case 2:** A patient aged of 8 years, homozygous sickle cell disease known since the age of 6 months, with many recent blood transfusion who presented a sudden headache and progressive

disorder of consciouness. On physical examination the patient had fever at 39°2C, with a Glasgow score of 13/15 and there was left side deficit, with a painful frontal swelling. A brain CT scan with contrast was performed and showed an image in favor of a right frontal brain absces and interhemispheric subdural empyema associated with frontal osteitis that had intracranial communication (Figure 2) and fluid content of frontal swelling and axial view of CT scan showing parietal empyema (j), per operating view of empyema (k) and post-operative control CT scan (l) (figure 3).

The patient had undergone neurosurgical intervention to evacuate the empyema by craniotomy and ponction of brain abcess. The outcome was favourable under antibiotic adapted for staphylococcus.

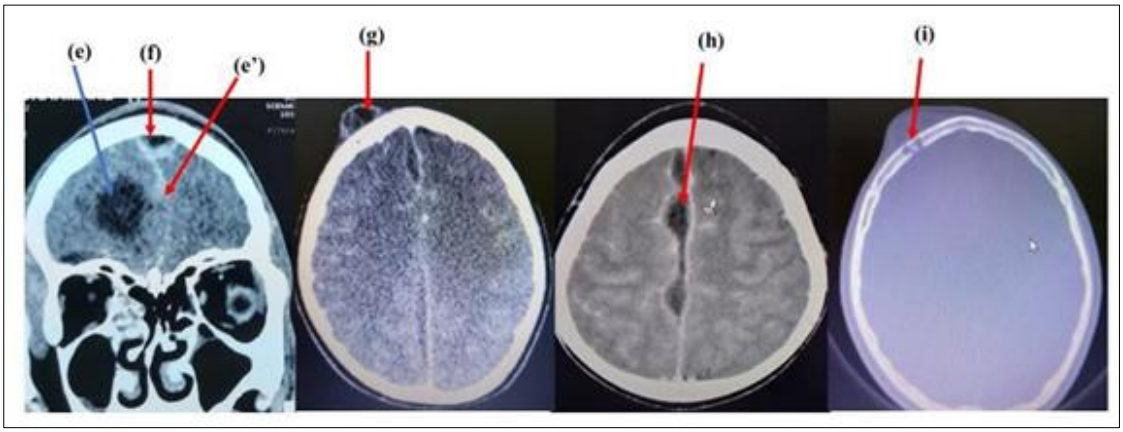


**Fig 2:** A brain CT scan with contrast showing a right frontal brain abcess (e) with mass effect (e'); with longitudinal superior sinus thrombosis (f), frontal swelling (g), interhemspheric subdural empyema (h) associated with frontal osteitis (i) that had intracranial communication.

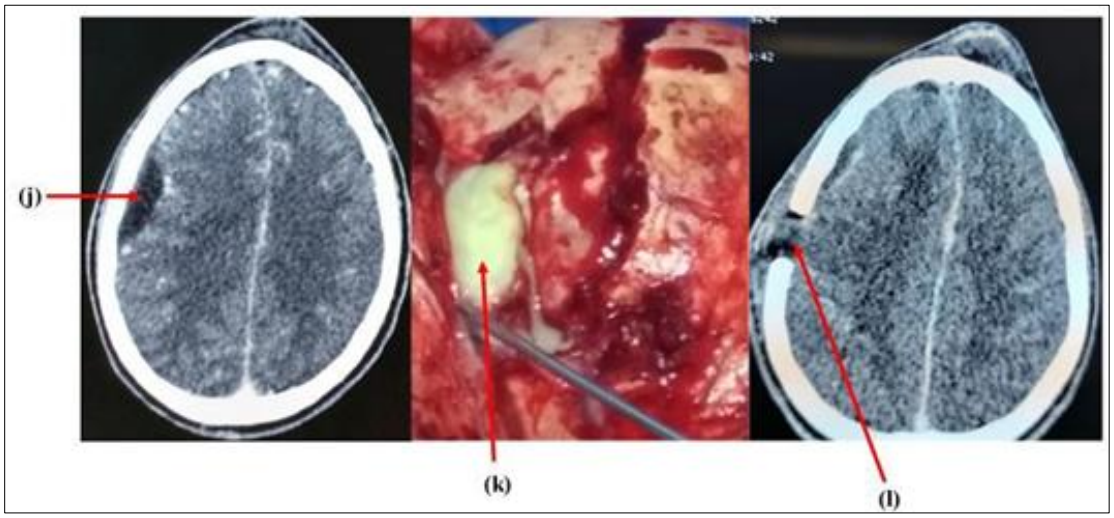


**Fig 3:** axial view of CT scan showing parietal empyema (j). Per operating view of empyema (k) and post-operative control CT scan (1)

## Discussion

Neurological complications are one of the major causes of morbidity and mortality in sickle cell disease, represented by ischemic stroke, intracranial hematomas and infections [4]. Entrance of pathogens into cerebral parenchyma can occur through multiple pathways. The first is haematogenic from emboli via intracranial vessels such as diploe veins. The second is from the suppurative process within sinus and ear. The third is direct innoculation after trauma or surgical procedures. As explained above, due to anatomical differences, for our first case, it was an extradural empyema type infection and a frontal subcutaneous collection due to the pseudomonas germ. According to the literature, pneumococcus is the first suspected germ before Haemophilus influenzae B in cases of intracranial infection [5]. Infections are common in sickle cell disease and can be severe. This susceptibility to infections can be promoted by functional asplenism, alteration of the complement pathway, which are proteins that participate in the phagocytosis of intruding germs, repeated vasoocclusive crises that lead to venous stasis and promote infections, especially of the skin [3, 5, 7]. In the second case, it was a rare intracranial complication of a type of associated brain abcess and empyema. The mechanism of this infection combined haematogenic pathway and endolymphatic channels and infection caused osteomyelitis and direct invasion into the intracranial compartiment with identifiable areas of bone and dural defects adjacents (figure 2).

According to the literature, risk factors for intracranial infection in children with sickle cell disease are favoured by recent blood transfusion, high blood pressure and corticosteroid or non-steroidal anti-inflammatory drug use [8] and subcutaneous

infection. In the reported case, this patient has a history of recent repeated transfusion [2, 4, 9]. The inflammatory phenomenon secondary to the adhesion of sickle cell adhesion to the vascular endothelium, which will be responsible for vascular fragility, is also responsible for the occurrence of this infectious complication.

The gold standard of the treatment like many infections of central nervous system, antibiotics should not be delayed. Follow by surgical procedures of drainage which permitted to release intracranial pressure, evacuate pus and allow for antibiotic spectrum. Strecococcus species and anaerobic gram positive. Use of combined intraveinous third generation cephalosporin with metronidazole and gentamicin contributed to resolve the infection [13, 19]. Intra operatively, decision to remove the bone flap is done in the cases becauses of osteomyelitis [5, 6, 7, 11, 12, 14].

In the littérature, the preoperative use of exchange transfusions for sickle cell patients is still discussed controversely and standard recommendations are not available [16, 19]. The benefit of an improved oxygene transport capacity has to be weighed against the risk of transfusion complications [15, 16, 18, 20]. In our case the patient received partial exchange tranfusion with a good outcome.

## Conclusion

Sickle cell disease is a challenge in terms of public health, because of its complications such as anemia and infection. Neurological complications are rare and often serious but should be evoked in front of neurological signs and should benefit from an emergency brain CT scan. Early detection of complications and multidisciplanry approach and rapid treatment can allows for good outcome for the young patients.

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