Scientific Research Publishing

Open Journal of Medical Imaging, 2024, 14, 72-78
https://www.scirp.org/journal/ojmi
ISSN Online: 2164-2796
ISSN Print: 2164-2788

Bithalamic Ischemic Stroke in Sickle Cell Patient at the Kamenge University Hospital in Burundi: Case Report

Sébastien Manirakiza[1*], Daniel Nduwayo[2], Patrice Barasukana[2], Chantal Murekatete[1], Jean Bosco Bizimana[3], Alice Ndayishimiye[4], Léopold Nzisabira[2], Hélène Bukuru[4]

[1]Department of Radiology, Kamenge University Hospital Center, University of Burundi, Bujumbura, Burundi
[2]Department of Internal Medicine, Neurology, Kamenge University Hospital Center, University of Burundi, Bujumbura, Burundi
[3]Department of Surgery, Neurosurgery, Kamenge University Hospital Center, University of Burundi, Bujumbura, Burundi
[4]Department of Pediatrics, Kamenge University Hospital Center, University of Burundi, Bujumbura, Burundi
Email: *sebastien.manirakiza@ub.edu.bi, danielnduwayo@yahoo.fr, pbarasukana@gmail.com, muhire70@yahoo.fr, jeanbizim@gmail.com, shimyali@yahoo.fr, nzisabiraleo@yahoo.fr, bukhelene2004@gmail.com

How to cite this paper: Manirakiza, S., Nduwayo, D., Barasukana, P., Murekatete, C., Bizimana, J.B., Ndayishimiye, A., Nzisabira, L. and Bukuru, H. (2024) Bithalamic Ischemic Stroke in Sickle Cell Patient at the Kamenge University Hospital in Burundi: Case Report. *Open Journal of Medical Imaging*, **14**, 72-78.
https://doi.org/10.4236/ojmi.2024.142007

Received: January 31, 2024
Accepted: May 25, 2024
Published: May 28, 2024

Abstract

Aim: To describe the clinical and radiological diagnostic for a sickle cell patient who got a bithalamic vasculature accident. **Observation:** This was a 4-year-old female child admitted to the pediatric ward for convulsive seizures in a feverish context. The physical examination did not find signs of meningeal irritation. The effective workup including thick gout, lumbar puncture, complete blood count, and C reactive protein was normal. An electrocencephalogram was not performed due to lack of availability of the device. Based on clinical data and the fact that Burundi is an area of high malaria prevalence, antimalarial and anticonvulsant treatment has been started. An alteration of consciousness (the Glasgow score of 7) had motivated a CT scan. Bithalamic hypodensities in favor of a vascular accident have been demonstrated. Etiological investigation revealed homozygous sickle cell disease ss during hemoglobin electrophoresis. The final evolution has been characterized by a return to normal consciousness. **Conclusion:** The complexity of the clinical picture following occlusion of Percheron's artery makes it difficult to diagnose. Sickle cell disease may favour the occurrence of ischaemic lesions. Cerebral imaging enables the diagnosis to be made with certainty and avoids delays in treatment.

Keywords

Stroke, Bithalamic Infarction, Sickel Cell, Percheron Artery

1. Introduction

Bithalamic infarctions are relatively rare types of brain injuries, which are often characterized by diagnostic delay. Clinical evaluation, imaging, and correlation with other findings are essential for accurate diagnosis and management. Bithalamic infarctions represent around 0.6% of cerebral infarctions [1]. For this type of brain injury, both thalami are affected by an infarct. They are consecutive to an occlusion of the thalamic paramedian arteries united by a common trunk. This anatomical variant of the thalamo-mesencephalic vascular network is called the Percheron artery because of the anatomical description he made of it in 1977 [2]. This type of anatomical variant is present in a range of 4% to 12% of the general population [3].

For both adults and children's populations, bithalamic infarctions are rare, but significant type of brain injury. Early detection and appropriate management are crucial for better outcomes in children. However, accurate diagnosis can be challenging [4].

The clinical presentation is polymorphic, due to the anatomical variability of the vascular territories of the thalamus and the extension of the territories infarcted to the midbrain. This clinical polymorphism suggests an etiology other than stroke. Brain imaging, in particular MRI, and a well-conducted etiologic workup allow diagnosis and treatment to be adapted [5]. We report the case of a child who had had a bithalamic infarction in the pediatric department of the Kamenge University Hospital center in Burundi.

2. Observation

A 4-year-old female child was hospitalized in the pediatric ward for convulsive seizures in a feverish context.

The physical examination did not reveal any signs of meningeal irritation and no focal neurological signs. The blood count revealed anemia with a hemoglobin level of 8 grs/dl. Despite the infectious assessment including thick gout, lumbar puncture, and C-reactive protein which was found to be normal, the child had been treated with an anticonvulsant (phenobarbital) and antimalarial (quinine) based only on epidemiological criteria. Under this treatment, apyrexia was observed but with an alteration of consciousness (the Glasgow score of 7). The deterioration of the state of consciousness motivated the performance of a cranioencephalic scan carried out nine (09) days after admission. Bilateral thalamic hypodensities suggestive of ischemic vascular injury were identified. as we can see below at **Figure 1**. Unfortunately, we could not perform Angiography and/or MRI, since we do not have technical capacity at our hospital or at any nearby hospital.

During the etiological investigation, hemoglobin electrophoresis revealed homozygous SS sickle cell disease, previously unrecognized. Anticoagulant therapy was started immediately and the disturbance in consciousness resolved after 6 days.

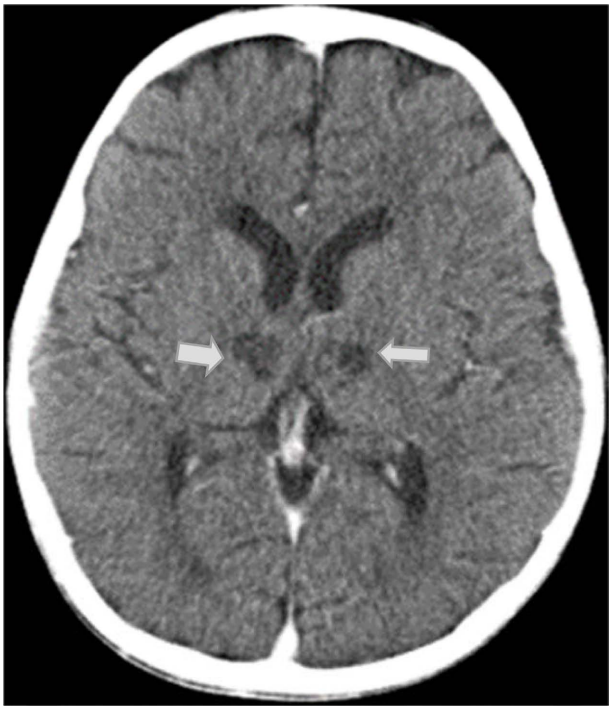

Figure 1. Cranioencephalic computed tomography: Axial sections showing bithalamic hypodensity (white arrow) compatible with a stroke in the subacute phase. Bifrontal cerebral atrophy is noted.

3. Discussion

The thalamic arterial blood supply is provided by a network of perforating arteries, originating from the posterior communicating arteries. There are four major vascular territories in the thalamus: tuberothalamic, inferolateral, paramedian, and posterior choroid [2].

The anatomical variants of thalamic perfusion had been studied by Percheron in 1977; the latter had described three types of vascularization (see **Figure 2**), starting from the P1 segment of the posterior cerebral artery. Arteries can arise on either side of P1 (type I); they can also come from the same artery (type IIa) or come from a common trunk (type IIb) which will supply the middle part of the two thalami. This common core thus bears the eponym of the Percheron artery [4].

Percheron artery occlusion (type IIb) always results in bilateral and medial infarction. In addition, in a third of cases, the artery irrigating the tuberothalamic territory may be absent and replaced by the paramedian artery, thus extending the infarcted territory in the event of occlusion of the Percheron artery [6]. In addition, the superior mesencephalic artery, which supplies the midbrain, and the paramedian thalamo-subthalamic artery may arise from a common trunk. Thus, in addition to this paramedian bithalamic infarction, the occlusion of the Percheron artery may in some cases lead to thalamopeduncular ischemia [4].

Thalamic infarctions represent 11% of ischemic vascular accidents in the vertebrobasilar territory, of which 22% - 35% are localized in the paramedian territory of the thalamus. They are distributed as follows: bilateral paramedian

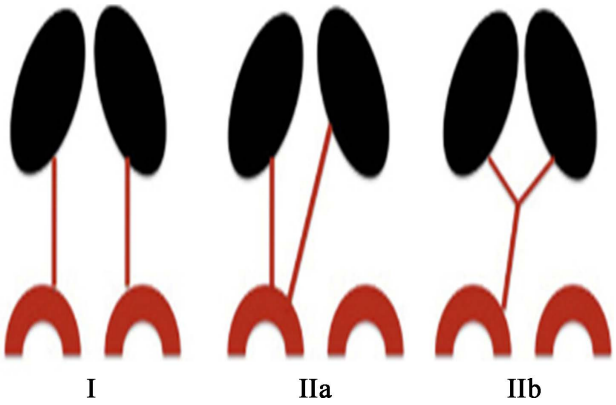

Figure 2. Variants of the paramedian thalamic arterial vascularization according to the classification established by Percheron [7]. Type I: the paramedian arteries originate each of the P1 segments of the posterior cerebral arteries; Type IIa: the two paramedi. Type IIb: single main trunk stemming from the posterior cerebral artery to supply the paramedian nuclear groups of each side of the thalamus.

infarction with mesencephalic stroke (43%), isolated bilateral paramedian infarction (38%), and bilateral paramedian infarction with involvement of the anterior thalamus and midbrain (14%) [8].

The clinical picture of paramedian bithalamic infarction is extremely variable and complex. The most frequently observed clinical signs combine vigilance disorders (decreased alertness, coma, hypersomnia), cognitive disorders: memory (anterograde and retrograde) or behavioral (confusion, behavioral and mood disorders) and paralysis of verticality [3].

Regarding the disturbances of consciousness which were the reason for carrying out the brain imaging of our patient, coma was found in 42%, hypersomnia in 35%, and reduced vigilance in 12% of cases. For cognitive and behavioral impairments, memory disorders (anterograde and retrograde) are reported at 63%, confusion in 53%, and 28% in behavior and mood disorders. Oculomotor disorders concern the verticality of the gaze with Parinaud's syndrome in 65% of cases, 32% of convergence paralysis, and 35% of deficit of the common occulomotor nerve. Other signs that can be found are motor disturbances (30%), ataxia (19%), and 12% dysarthria [2].

The variability of this clinical picture is explained by the anotomo-functional correlation of the areas affected by the infarction in the event of Percheron artery occlusion. For alertness disorders, hypersomnia is commonly explained by involvement of the intralaminar nucleus of the thalamus, which is part of the rostral extension of the ascending reticular activation system of the subthalamic and internal thalamic region. Coma also reflects mesencephalic extension.

The memory disorders are explained by involvement of the mamillothalamic tract leading to disconnection of the Papez circuit when there is a replacement of the tuberothalamic artery by the paramedian artery. Occulomotor disorders reflect an impairment of the supranuclear pathways by involvement of the interstitial rostral nucleus of the median longitudinal strip in the tectal region. Vertical

paralysis, loss of convergence, III impairment, internuclear ophthalmoplegia, and miosis are due to an extension of the infarction to the midbrain [6] [9] [10].

Behavioral and mood disorders mimic a frontal syndrome and are most frequently explained by an interruption of the thalamo-frontolimbic loop by involvement of the tuberothalamic territory [11]. The evolution of bilateral thalamic infarctions is characterized by a total regression of signs to 14%, which was observed in our patient, 30% of residual memory and/or psychiatric disorders, 19% of residual visual disorders, 12% of disorders with residual sleep and finally 7% of deaths [2].

This clinical and evolutionary complexity of the Percheron artery occlusion explains the difficulty in making the diagnosis based on physical examination data alone. Complementary radiological examinations, in particular brain MRI, make it possible to correct the diagnosis, in particular thanks to diffusion imaging, which is a technique with better sensitivity allowing an early diagnosis and early diagnosis of ischemic stroke [9] [12]. Computed tomography may show bithalamic hypodensity, but this exam is most often taken failing in the acute phase [13]. This makes it often show the ischemic lesion at the late phase as in our patient where it was done on the ninth day of hospitalization.

The most common etiologies of infarction in patients with bilateral thalamic involvement are small vessel disease and embolic origin. Arauz A *et al.* had identified embologenic heart disease as the main cause of bithalamic ischemic stroke (34%) in a series of 64 patients [3].

The etiology that was favored in our patient is sickle cell disease that was discovered during hospitalization. The cerebrovascular risk is a real threat that weighs on children with sickle cell disease at an early stage. Indeed, cerebral infarctions are frequent in this population. Studies put the spontaneous risk of symptomatic stroke at 11% before the age of 20, with these acute events mostly occurring before the age of 10, peaking between 2 and 5 years [14] [15], the age group to which our 4-year-old patient belonged. The risk is therefore major, approximately 300 times greater than that of children of the same age without sickle cell disease, in whom the incidence of ischemic stroke is estimated at 2-3/100,000 children/year [16].

This child's sickle cell disease background should be known from the time of admission once the anemia of 8 g/dl has been noted. Unfortunately, in our context, hemoglobin electrophoresis can only be carried out in a private laboratory, where prices are prohibitive for our low-income population, which often has no health insurance. In the case of our patient, it remains difficult to know whether the ischemic vascular accident occurred at the very beginning of the disease or during hospitalization. Faced with the epileptic seizure that our patient presented, a CT-scan or MRI exploration was necessary to highlight the type of lesion that could be at the origin of the symptoms [9]. This was not possible due to the lack of financial resources as well as the deficit of the technical platform (three CT-scan machines and no MRI machine for the whole country). All of

this demonstrates how difficult it is to diagnose complications related to sickle cell disease in low-income countries like Burundi.

4. Conclusion

Bithalamic infarcts are often secondary to occlusion of a common trunk of the paramedian artery of the thalamus called the Percheron artery. The topographic and lesional diagnosis is difficult to evoke according to the arguments of the clinic, often polymorphic in addition. MRI, an imaging method not yet available in our country, remains the reference examination, especially in the acute phase. Computed tomography allows suggestive images to be obtained late. Brain imaging is recommended for any suggestive sign that is not justified by other pathological situations, especially in subjects with cardiovascular risk factors such as sickle cell anemia.

Data Availability

The data for this case are available in the patient file of the pediatrics department. They can be obtained on request from the corresponding author.

Ethical Approval

This study was approved by the ethics committee of the Faculty of Medicine at the University of Burundi.

Consent

Written consent was obtained from the child's parents. The parents were informed of the purpose of the article.

Authors' Contributions

All authors have read and agreed to the final manuscript.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

References

[1] Decourcelle, A., Kalsoum, E., Dequatre-Ponchelle, N., Aguettaz, P. and Cordonnier, C. (2013) Fistule durale révélée par un infarctus bithalamique. *Pratique Neurologique-FMC*, **4**, 178-183. https://doi.org/10.1016/j.praneu.2013.07.005

[2] Monet, P., Garcia, P., Saliou, G., Spagnolo, S., Desblache, J., Franc, J., *et al.* (2008) Bithalamic Infarct: Is There An Evocative Aspect? Radioclinical Study. *Revue neuRologique*, **165**, 178-184. https://doi.org/10.1016/j.neurol.2008.08.012

[3] Arauz, A., Patiño-Rodríguez, H.M., Vargas-González, J.C., Arguelles-Morales, N., Silos, H., Ruiz-Franco, A., *et al.* (2014) Clinical Spectrum of Artery of Percheron Infarct: Clinical-Radiological Correlations. *Journal of Stroke and Cerebrovascular Diseases*, **23**, 1083-1088. https://doi.org/10.1016/j.jstrokecerebrovasdis.2013.09.011

[4] Lamboley, J., Le Moigne, F., Have, L., Tsouka, G., Drouet, A., Salamand, P., *et al.* (2011) Occlusion de l'artère de Percheron: Difficultés du diagnostic clinique et place de l'IRM. À propos de six cas. *Journal de Radiologie*, **92**, 1113-1121. https://doi.org/10.1016/j.jradio.2011.08.007

[5] Mahfoudhi, M. and Turki, S. (2015) Occlusion of Percheron Artery Revealing Systemic Lupus Erythematosus. *The Pan African Medical Journal*, **22**, Article No. 30. https://doi.org/10.11604/pamj.2015.22.30.7760

[6] Schmahmann, J.D. (2003) Vascular Syndromes of the Thalamus. *Stroke*, **34**, 2264-2278. https://doi.org/10.1161/01.STR.0000087786.38997.9E

[7] Percheron, G. (1976) Arteries of the Human Thalamus. II. Arteries and Paramedian Thalamic Territory of the Communicating Basilar Artery. *Revue Neurologique*, **132**, 309-324.

[8] Payenok, A.V., Shevaha, V.M., Kulyk, A.R., Netliukh, A.M. and Kulmatytskyi, A.V, (2019) Stroke in the Artery of Percheron Territory: The Two Edges of One Diagnosis. *Wiadomosci Lekarskie* (*Warsaw*, *Poland*: 1960), **72**, 1851-1853. https://doi.org/10.36740/WLek201909226

[9] Kumral, E., Evyapan, D., Balkır, K. and Kutluhan, S. (2001) Bilateral Thalamic Infarction: Clinical, Etiological and MRI Correlates. *Acta Neurologica Scandinavica*, **103**, 35-42. https://doi.org/10.1034/j.1600-0404.2001.00141.x

[10] Josseaume, T., Calvier, E.A., Duport, B.D., Lebouvier, T., Mathieu, G.P., de Kersaint Gilly, A., *et al.* (2007) Syndrome amnésique aigu par infarctus des faisceaux mamillothalamiques de Vicq d'Azir. *Journal of Neuroradiology*, **34**, 59-62. https://doi.org/10.1016/j.neurad.2007.01.003

[11] Carrera, E. and Bogousslavsky, J. (2006) The Thalamus and Behavior: Effects of Anatomically Distinct Strokes. *Neurology*, **66**, 1817-1823. https://doi.org/10.1212/01.wnl.0000219679.95223.4c

[12] Matheus, M.G. and Castillo, M. (2003) Imaging of Acute Bilateral Paramedian Thalamic and Mesencephalic Infarcts. *American Journal of Neuroradiology*, **24**, 2005-2008.

[13] Caballero, P.E.J. (2010) Bilateral Paramedian Thalamic Artery Infarcts: Report of 10 Cases. *Journal of Stroke and Cerebrovascular Diseases*, **19**, 283-289. https://doi.org/10.1016/j.jstrokecerebrovasdis.2009.07.003

[14] Verlhac, S. (2008) Doppler transcrânien et protocole de prévention des accidents vasculaires cérébraux de l'enfant drépanocytaire. *Archives de Pédiatrie*, **15**, 636-638. https://doi.org/10.1016/S0929-693X(08)71858-X

[15] Kossorotoff, M., Grevent, D. and de Montalembert, M. (2014) Drépanocytose et atteinte vasculaire cérébrale chez l'enfant. *Archives de Pédiatrie*, **21**, 404-414. https://doi.org/10.1016/j.arcped.2014.01.005

[16] Roach, E.S., Golomb, M.R., Adams, R., Biller, J., Daniels, S., Deveber, G., *et al.* (2008) Management of Stroke in Infants and Children: A Scientific Statement from a Special Writing Group of the American Heart Association Stroke Council and the Council on Cardiovascular Disease in the Young. *Stroke*, **39**, 2644-2691. https://doi.org/10.1161/STROKEAHA.108.189696