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## Imaging of osteonecrosis of the femoral head in sickle cell patients: About 36 cases

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### Abstract

**Purpose:** To study the epidemiological, clinical and radiological aspects of osteonecrosis of the head of the femur in sickle cell patients.

**Methods:** This was a retrospective study carried out at the Amirou Boubacar Diallo National Hospital in Niamey from January 2016 to December 2021. It involved 36 sickle cell patients aged 4 to 42 years (mean age = 16 years 7 months). All patients had a frontal x-ray of the pelvis. Scintigraphy was performed in 5 symptomatic patients due to persistence of pain while the pelvis x-ray was normal. The lesions were grouped according to their evolution according to the Arlet and FICAT classification.

**Results:** In 36 patients, radiography revealed 26 cases of unilateral osteonecrosis of the head of the femur and 5 cases of bilateral involvement. In 5 patients who had a normal x-ray, the scintigraphy showed 3 cases of unilateral osteonecrosis of the femoral head at stage I. In total, 34 patients had osteoarticular lesions. These lesions were unilateral in 29 patients (85.29%) and bilateral in 5 patients (14.71%). In the majority of cases (79.41%) the lesions were diagnosed at late stages (III and IV of ARLET and FICAT classification).

**Conclusion:** Osteonecrosis of the femoral head in sickle cell patients is a serious complication because of the often-significant after-effects. In our context, radiography remains the basic examination for diagnosis. In children, MRI, which is a non-irradiating modality, remains the examination of choice.

**Keywords:** Sickle cell disease, osteonecrosis, femoral head, imaging, Niger

### Introduction

Sickle cell disease is a hereditary structural haemoglobin disease with autosomal recessive transmission <sup>[1]</sup>. According to the WHO in 2006, 5% of the world population had hemoglobinopathy <sup>[2]</sup>. Black Africa, India and the Arabian Peninsula constitute the main centers. In Niger, the prevalence of sickle cell disease in the population was estimated at between 18 and 22% in 2009 <sup>[3]</sup>. The progression of this disease is characterized by the occurrence of complications, particularly osteoarticular complications <sup>[4, 5]</sup>. Osteonecrosis of the femoral head most often occurs insidiously, resulting in the development of advanced osteoarticular lesions.

### Aim

The aim of this work was to study the epidemiological, clinical and radiological aspects of osteonecrosis of the head of the femur in sickle cell patients.

### Materials and Methods

This was a retrospective study carried out at the Lamordé National Hospital in Niamey from January 2016 to December 2021 (72 months). It involved 36 sickle cell patients aged 4 to 42 years (Mean age = 16 years 7 months). All patients had an X-ray of the pelvis. Scintigraphy was performed in 5 symptomatic patients whose radiographs were normal. The lesions were grouped according to their evolution according to the ARLET and FICAT classification.

### Results

#### Epidemiologically

During this period 86 sickle cell patients were consulted. 36 patients presented clinical symptoms (pain and/or lameness) for an incidence of 41.86%.

Among these 36 symptomatic patients, 34 presented necrosis of the head of the femur, i.e. 94.44% of cases. 20 patients were male (58.82%), a ratio of 1.42. The age of the patients ranged from 4 to 42 years, an average of 16 years 7 months. The most affected age groups were 5-14 years (32.35%) and 15-24 years (38.23%). The SS genotype was predominant with 26 cases (76.47%) and 8 cases (23.53%) for SC. Hip pain was the main reason for consultation in 31 patients (91.17%). Lameness was found in only 3 cases. The other locations of the pain were the shoulder (3 cases) and the spine (1 case).

**Radiologically**

Among 31 patients, radiography revealed 26 cases of unilateral osteonecrosis of the head of the femur and 5 cases of bilateral involvement.

Among 5 patients with a normal x-ray, the scintigraphy (figure No. 1) showed 3 cases of unilateral osteonecrosis of the femoral head at stage I.

The cases diagnosed at stage II were 4.

In the majority of cases (27 out of 34 cases) the lesions were diagnosed at late stages (III and IV of ARLET and FICAT classification) (figure No. 3; figure No. 4).

In total, the osteoarticular lesions were unilateral in 29 patients (85.29%) and bilateral in 5 patients (14.71%).

The radiological aspects are summarized in the following tables:

**Table 1:** Distribution of cases according to radiographic stage (ARLET and FICAT classification)

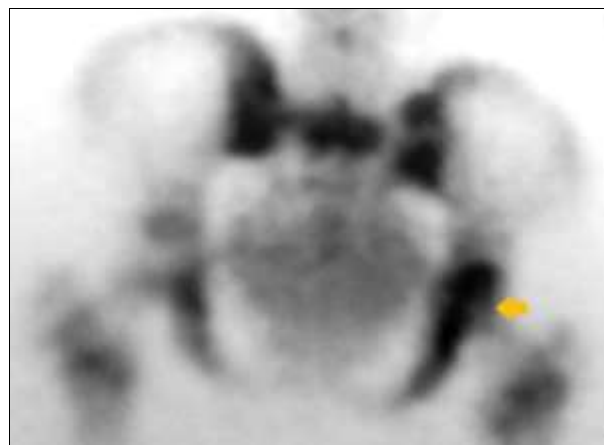
Staging	Number of affected hips	Percentage
Stade I	03	8,8
Stade II	04	11,8
Stade III	07	20,6
Stade IV	20	58,8
Total	34	100

**Table 2:** Distribution of cases according to age group and radiographic stage.

Age (year)	Stade I	Stade II	Stade III	Stade IV	Total
<5	0	0	1	0	1
[5-14]	2	2	2	5	11
[15-24]	1	2	4	6	13
[25-34]	0	0	0	6	6
[35-44]	0	0	0	3	3
Total	3	4	7	20	34

**Table 3:** Distribution of bone lesions according to stage and genotype.

Type	Stade I	Stade II	Stade III	Stade IV	Total
SS	3	4	4	12	23
SC	0	0	3	8	11
SβThal	0	0	0	0	0
Total	3	4	7	20	34



**Fig 1:** Bone scan showing left coxofemoral hyperuptake in a sickle cell patient during an episode of pain while the pelvis x-ray was normal (stage 1 of ARLET and FICAT classification).



**Fig 2:** Frontal x-ray of the pelvis showing a subchondral border (“eggshell” appearance) without loss of sphericity of the head of the left femur (stage 2 of ARLET and FICAT classification).



**Fig 3:** Frontal x-ray of the pelvis showing significant compression with loss of sphericity of the head of the right femur (stage 3 of ARLET and FICAT classification).



**Fig 4:** Frontal x-ray of the pelvis showing major collapse of the head of the right femur with coxarthrosis (stage 4 of ARLET and FICAT classification).

### Discussions

The incidence of osteonecrosis of the femoral head was around 41.86%. This result is close to that of Mukisi *et al.* who found 36.50% [6]. Elsewhere, Hernigou *et al.* estimated the prevalence of necrosis of the femoral head at 46% [7, 8]. The average age of our patients was 16 years 7 months while in RCI and Senegal it was 22.7 years and 22 years respectively. We did not find the cause of this difference but in all cases, it concerned young populations. The sex ratio was 1.42, comparable to that found in RCI and Senegal (1.43 and 1.7) [9, 10]. Regarding the genotype, 76.47% of our patients were SS type and 23.53% SC type. Coulibaly *et al.* found 37.50% and 28.10% respectively [11]. Hernigou *et al.* found 42% and 20% [7] unlike Homawoo *et al.* who found twice as many SC heterozygous subjects [12]. Hip pain was the most frequent reason for consultation (91.17%) followed by lameness (7.14%), close to that found by Rouvillain *et al.* with 92.59% lameness [13]. In Mali their frequency was 65.6% and 15.6% according to Coulibaly [11]. The evolution of pain was greater than 6 months in 92.86% of our patients, comparable to that observed in Mali [11].

Standard radiography was the most used diagnostic method during our study. However, it had shown its limits at the beginning stage. Our study showed that osteonecrosis of the femoral head was unilateral in 83.38% of cases seen on standard radiography, a rate relatively higher than that observed by Coulibaly in Mali with 65.6% of cases [11]. Some authors have highlighted the interest of magnetic resonance imaging (MRI) [9] whose use remains very limited in our context in Niger. These same authors, using CT and MRI, found 90.2% bilaterality of osteoarticular lesions [9]. This shows that MRI remains the most sensitive examination and is of major interest in children because it is non-irradiating. The high cost of scintigraphy had limited its performance and the use of this technique was only 3 cases in our series. At the late stage (III and IV), 27 out of 34 cases of osteonecrosis of the head of the femur (79.41%) were observed, which implies an evolution of the disease over more than 12 months on average. In our study population, 3 out of 34 patients (8.82%) presented with associated humeral head necrosis. The correlations between the age of the patients, the clinical course and the radiological signs show that 42.42% of the hips were affected before adolescence. Between 15 and 24 years of

age, the majority of lesions were diagnosed at late stages (III and VI). Beyond the age of 24, coxarthrosis was evident. In our series, stage I (3 cases) was encountered in patients with painful symptoms lasting less than 6 months. Stages II and III (11 cases) appeared over a course of 6 to 12 months and stage IV over a course of 92 months. Hernigou *et al.* found an average duration of 42 months between the transition from stage I to stage IV [7-8]. Confined to a series of 58 hips, 40% of secondary coxarthrosis was noted after 13 years of evolution [14]. In our context where medical follow-up is irregular, the natural evolution of osteonecrosis of the femoral head would begin at 11 years and 2 months to reach stage IV at 20 years 4 months on average without orthopedic and surgical treatment. In SC sickle cell patients, osteoarticular lesions were diagnosed at the late stage, including 72.73% at stage IV; this may probably be related to less noticeable clinical symptoms than in SS forms.

### Conclusion

Osteonecrosis of the femoral head in sickle cell patients is a serious complication because of the often-significant sequelae. A radiological assessment is necessary in order to classify the lesions for better therapeutic management. In our context, radiography remains the basic examination for diagnosis. Scintigraphy is necessary for early diagnosis of lesions. In children, non-radiating MRI remains the examination of choice.

**Conflict of interest:** The authors declare no conflict of interest.

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