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Case Series

Shoulder Arthroplasty in Patients with Sickle Cell Disease: A French Specialized Center Case Series and Literature Review

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Abstract

Background : Humeral head osteonecrosis is a frequent complication of Sickle Cell Disease (SCD). Little is known about shoulder arthroplasty indications and results in these patients. The aim of this retrospective review is to compare the actual indications and results for SCD to a literature review.

Methods : All shoulder arthroplasties performed on SCD patients between 2016 and 2022 have been evaluated. 17 shoulders in 15 patients were followed up for 2.5 years. Indications, functional and radiological results, and complications were compared to a literature review.

Results : The Constant score significantly improved by 35 points (p =0.016). Patients were finally satisfied with a mean SSV evaluation at 72%. Post-operative complications due to SCD were frequent. There was a bilateral dramatic reverse prosthesis loosening in a patient with severe bone sclerosis due to necrosis. The use of pyrocarbon hemi-arthroplasty is promising and was never described in the literature for SCD shoulder osteonecrosis.

Discussion : Shoulder arthroplasty for collapse humeral head in SCD patients should be considered because of the significant functional improvement that can be obtained. Patients with SCD should be operated in a center with specialized surgeons surrounded by heamatologists to prevent specific complications.

Keywords: Sickle cell disease; Osteonecrosis; Shoulder arthroplasty.

INTRODUCTION

Sickle cell disease is a frequent genetic disorder, affecting around 300,000 births a year worldwide, particularly in sub-Saharan Africa. The latest available data indicate that 466 children with sickle cell disease were born in France in 2015, corresponding to a prevalence of 1/1736 births. However, this prevalence is much higher in overseas departments (1/499) and in the Paris region (1/765), where the at-risk population is concentrated [1].

This autosomal recessive disease causes hemolytic anemia characterized by abnormally shaped red blood cells. Sickle red cells cause vascular occlusion, leading to coagulation in the small vessels, with tissue ischemia complicated by tissue infarction (vaso-occlusive crisis, stroke, vascular nephropathy, etc.) in the form of necrosis in the bone, preferentially affecting the hips and shoulders [2].

Several conservative surgical procedures exist to treat osteonecrosis. Core decompression, with or without bone marrow grafting, is the most common. As in the case of the hip, it is proposed for early stages, but does not cure necrosis [3-6].

Shoulder arthroplasties are being performed with increasing frequency in the general population, and the frequency of their indication is likely to continue to increase over the coming years [7]. Shoulder hemiarthroplasties have shown good long-term results, particularly in cases of avascular osteonecrosis [8,9]. Pyrocarbon hemiarthroplasties have also shown good results at a minimum two-year follow-up in young subjects with humeral head osteonecrosis requiring arthroplasty [10].

Few studies have evaluated the results of shoulder arthroplasty in humeral head osteonecrosis secondary to sickle cell disease. They also report inconsistent results [11,12].

The aim of this study is to report the current indications of a reference center, as well as the functional and radiological results obtained, and to compare them with a review of the literature.

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MATERIALS AND METHODS

Population

This is a case series of sickle cell patients operated on for shoulder arthroplasty in a specialized center in collaboration with the Genetic Red Blood Cell Disease Unit, between 2016 and 2022. All sickle cell patients who had shoulder arthroplasty over this period and with a follow-up of at least 12 months were included in this study, i.e. 17 patients. Two patients died of complications due to sickle cell disease (unrelated to the shoulder surgery) without being seen again. The first, aged 51, of a mesenteric infarction after total hip arthroplasty 9 months after anatomic total shoulder arthroplasty. The second, aged 54, of an acute chest syndrome 2 years after pyrocarbon hemiarthroplasty of the shoulder without having been seen again due to confinement during the Covid pandemic.

Fifteen patients were monitored, including two who had undergone surgery on both shoulders (2 total anatomical prostheses and 2 reversed prostheses), i.e. 17 operated shoulders monitored for an average of 28.5 months (range, 12-62 months).

The sample included 10 women and 5 men [Table 1]. All patients had major sickle cell disease, homozygous SS (12 patients) or heterozygous SC (3 patients). The mean age of patients was 38.6 years at the date of data collection (min 26, max 63).

Because of their sickle cell disease, these patients had numerous comorbidities, such as chronic anemia with a history of alloimmunization,

Table 1: Patient's characteristics

Patient	Age(y)	Sexe	SCD type	AVN stage	Comorbidities	Previous core decompression(s) and delay before arthroplasty (months)
1	35	M	SS	4	Pulmonary infarct, SCCs, 1 ACS	0
2	40	M	SS	4	right THR D, SCCs	1/17
3	28	F	SS	4	SCCs, ACS, 2 THR	1/10
4	48	F	SS	3	ACS, SCCs	0
5 right side	38	M	SS	6	SCCs, 2 THR, cholecystectomy, pneumonia	0
5 left side	38	M	SS	6		0
6	56	F	SS	5	right THR, SCCs	0
7 right side	56	F	SC	5	left THR, colic surgery	1/31
7 left side	56	F	SC	5		1/15
8	63	F	SS	5	Chondrocalcinosis, obesity	1/9
9	40	F	SS	5	SCCs, ACS, HBP, cholecystectomy	1/58
10	60	F	SC	5	SCCs	0
11	29	F	SS	4	SCCs	0
12	31	M	SS	5	SCCs, ACS, liver and kidneys transplantations	0
13	37	F	SC	4	SCCs	0
14	31	M	SS	4	SCCs, ACS, THR, cholecystectomy	0
15	26	F	SS	4	SCCs, cholécystectomy	0

SCD : sickle cell disease, AVN : avascular necrosis, SCC : sickle cell crisis, ACS : acute chest syndrom, THR : total hip replacement

sometimes precluding any possibility of blood transfusion, Sickle Cell Crisis (SCC), Acute Chest Syndromes (ACS), pulmonary infarction, High Blood Pressure (HBP), cholecystitis treated by cholecystectomy, and osteonecrosis of the femoral head. The comorbidities presented by the patients are listed in Table 1.

Six patients had undergone humeral head core decompression with reinjection of hematopoietic stem cells (bone marrow) an average of 23 months prior to arthroplasty [4]. Two patients received exchange transfusions in the days prior to surgery.

A Constant score, which is a functional shoulder score (up to 15 points for the pain, 20 points for daily activities, 40 points for range of motion and 25 points for the strength; a healthy young male scores 100 points) was calculated preoperatively for each patient [13].

Standard radiographs were taken preoperatively to classify the stage of osteonecrosis of the humeral head according to Steinberg's 7-stages classification for the hip [Table 2] [14]. A CT arthrogram was also performed to search for possible cuff lesions and to analyze the state of the glenoid cartilage in order to optimize the surgical indication.

Choice of Prosthesis Type

The indication for surgery was based on the stage of the osteonecrosis and the patient's age. All osteonecroses were at least stage 3 (loss of sphericity of the humeral head). Stage 3 and 4 osteonecroses were treated with a Pyrocarbon Hemiarthroplasty (PYC). Stage 5 osteonecroses were treated with a Total Anatomical Prosthesis (TAP). Stage 6 osteonecrosis and revision prostheses were treated with Reversed Prosthesis (RP).

Among the 17 shoulders studied, a hemiarthroplasty with pyrocarbon head was performed on 8 shoulders [Figure 1], an anatomic total arthroplasty on 5 shoulders [Figure 2], a reversed arthroplasty on 3 shoulders [Figure 3] and on a failed metal hemiarthroplasty implanted 5 years previously [Figure 4].

A short humeral stem, cemented or uncemented depending on bone quality (Ascend stem), was always used with pyrocarbon head for hemiarthroplasty, metal head opposite an all-polyethylene glenoid with cemented keel for TAP, cementless metaglenoid with central stud and centered glenosphere diameter 36 for RP.

Table 2: Surgical choice according to the necrosis stages (Steinberg [14])

Necrosis stage	Imaging findings	Surgical choice
0	none	None
1	Normal X-ray, necrosis on MRI or CT-scan	Core decompression
2	Sclerosis on standard X-ray	
3	Crescent sign, Subchondral collapse less than 1 cm	Pyrocarbon hemi-arthroplasty
4	Head flattering or fragmentation	
5	Articular space narrowing	Total anatomic arthroplasty
6	Advanced articular destruction	Reversed prothesis

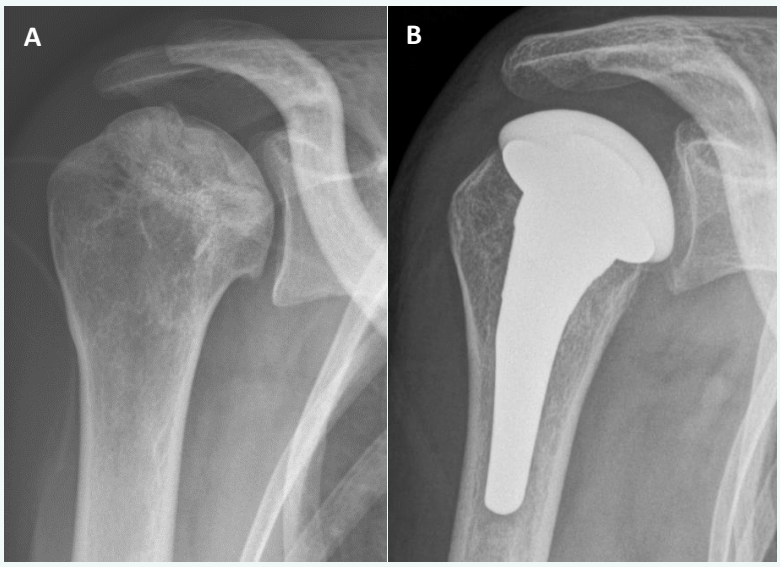

Figure 1: humeral head stage 3 osteonecrosis (A), hemi-arthroplasty with pyrocarbon head and uncemented stem (B).

Figure 2: humeral head stage 5 osteonecrosis (A), anatomic total shoulder arthroplasty with a cemented polyethylene glenoid component and a metallic head on an uncemented stem (B).

Surgical Technique

The patients' surgery was supervised by hematologists specializing in sickle cell disease.

Surgeries were performed under general anesthesia in the half-seated position. A deltopectoral approach with tenotomy of the subscapularis was used, with transosseous reinsertion at the end of the procedure for hemiarthroplasties and anatomic total arthroplasties. A superior-lateral approach was used for reversed prostheses. Patients were immobilized for 4 weeks postoperatively. Rehabilitation management consisted of pure passive mobilization without external rotation beyond 0° for the first 4 weeks, to protect subscapularis muscle repair on the anterior approaches. Immobilization was removed at 1 month postoperatively, and active rehabilitation without limitation was then initiated.

Postoperative Follow-Up and Statistical Analysis

Constant score [13] and subjective shoulder value (SSV: patients have answered the question "if a normal shoulder is 100%, how much do you scale yours ?") [15] were assessed after a minimum of 12 months postoperatively. Preoperative Constant scores and those at latest follow-up were compared using the Wilcoxon statistical test (non-parametric rank test), with significance set at $p \leq 0.05$. Any radiological lucent lines or implant loosening was recorded on the radiographic images at latest follow up.

RESULTS

Six patients required reaming of the humeral shaft during the procedure. Intraoperative fracture of the humeral metaphysis occurred in two patients, necessitating additional humeral wiring. The humeral stem was cemented in 6 cases and cementless in the others. All polyethylene

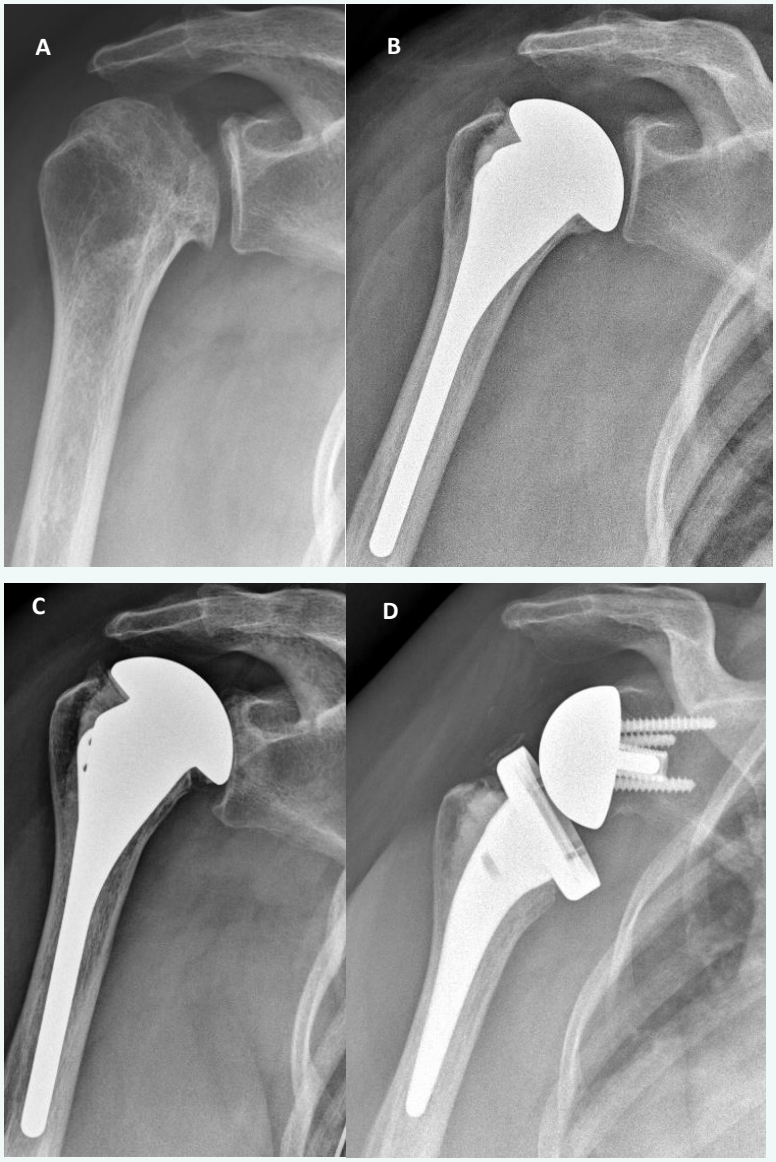

Figure 3: stage 6 bilateral gleno-humeral necrosis with extend humeral shaft and scapular body necrosis (A,B), bilateral reversed arthroplasty with cemented stems (B) with peroperative fracture on the left side requiring synthesis with a metallic wire (C), dramatic right glenoid loosening (D) and left humeral loosening (E) at 5 years follow-up.

glenoid components were cemented and the reversed baseplate components were uncemented. The long biceps was preserved in 8 patients, always during hemiarthroplasty. Tenotomy was performed in all other patients.

Of the 15 patients studied, two required postoperative blood transfusion. One patient developed an acute chest syndrome 2 days after surgery, with no signs of severity, and did not require intensive care. Another developed a SCC 9 days later, and the last one a SCC at 10 days post-op, complicated by a segmental pulmonary embolism. The average hospital stay was 8.6 days. Among patients who did not develop complications, the average hospital stay was 4.8 days, whereas it was 19 days for the three patients who did develop complications [Table 3].

The mean Constant score was 28.6 preoperatively (SD±12.2, min 6, max 57) and 64.1 at maximum follow up (SD±14.2, min 27, max 80), with a significant statistical difference (p=0.016, [Tables 3]. The mean (SSV) was 71.8% (min 50, max 100) at maximum follow up.

The patient's reversed prostheses operated on both sides came loose 5 years after surgery [Figure 3]. This patient had major necrotic bone involvement, with cortico-medullary dedifferentiation of his scapula and the entire humerus. His main complaints at last follow-up were right shoulder pain, with a 43-point reduction in the right Constant score (from 70/100 at 24 months to 27/100 at 57 months). On the left side, the stem mobilized with the tip of the stem migrating away from the humerus. His Constant score on the left side decreased by 21 points (from 72/100 to 51/100 between months 24 and 57). However, Constant scores remained higher than preoperative scores of 6/100 on the right and 10/100 on the left. A prosthesis replacement is scheduled, starting with the right side.

No other implant did not demonstrated signs of gross radiological loosening at latest follow up. Two TAP showed usual lucent lines around the glenoid implant. No lucent lines was observed on the humeral stems.

Table 3: Clinical results and peri-operative events

Patients	Pre-op Constant score	Prosthesis type	Post-opérative complications	Pre-op blood exchange	Post-op blood transfusion	Time of stay (days)	Follow-up (months)	Final Constant score	Final SSV (%)
1	33	PYC	none	yes	no	6	54	80	70
2	25	PYC	SCC at day 9	no	no	10	42	78	100
3	39	PYC	ACS at day 2	no	no	7	30	73	70
4	17	PYC	none	no	no	4	12	62	60
5 right side	6	RP	none	no	no	3	62	27	50
5 left side	10	RP	none	no	no	4	57	51	75
6	26	TAP	none	no	yes	7	36	68	80
7 right side	32	TAP	none	no	no	4	46	67	75
7 left side	30	TAP	none	no	no	5	30	69	80
8	28	TAP	none	no	no	12	31	59	70
9	29	TAP	none	no	no	4	12	37	50
10	37	RP	none	no	yes	4	13	63	90
11	30	PYC	none	no	no	5	13	69	50
12	30	RP	none	yes	no	7	12	68	80
13	15	PYC	none	no	no	5	12	69	60
14	41	PYC	none	no	no	2	12	80	90
15	57	PYC	SCC + PE	no	yes	40	12	70	70
mean	**28.6**						**28.5**	**64.1**	**71.8**

PYC : pyrocarbon head replacement, RP : reversed prosthesis, TAP : total anatomic prosthesis, SCC : sickle cell crisis, ACS : acute chest syndrom, PE : pulmonary embolism; SSV : subjective shoulder value

LITERATURE REVIEW AND DISCUSSION

Literature Review

Sickle cell disease is often absent from the etiologies of shoulder osteonecrosis treated by arthroplasty in the literature [16-18]. Hattrup and Cofield [19], report the results of 127 shoulders with osteonecrosis, of which only 3 were of sickle-cell origin, although it is not known whether these were among the 88 shoulders followed up at the minimum 2-year follow-up.

A Pubmed search for "shoulder arthroplasty" or "shoulder replacement" and "sickle cell disease" yields 3 articles describing the results of prosthetic shoulder replacement in sickle cell patients.

The first article dates back to 2007 [11], and reports on the clinical and radiological results of 8 sickle cell patients (7 hemiarthroplasties with metal heads and one TAP) at a mean follow-up of 4 years. Blood transfusion was required in 2 patients preoperatively, one intraoperatively and one intra- and postoperatively. The average length of stay was 5 days, with 2 patients experiencing a SCC immediately post-operatively. Clinical results were mixed, and the authors identified 3 groups of patients;

-A first group of 2 patients (2 hemiarthroplasties) with excellent results in every respect,

-A second group of 4 patients (4 hemiarthroplasties) with good functional results but persistent pain,

-A final group of 2 patients (a hemiarthroplasty and a PTEA) with a poor outcome in every respect.

There was no radiological loosening of the humeral stems, 4 of which were cemented and 3 cementless. One patient had a progressive deterioration of the clinical result due to glenoiditis at 5 years: he was resumed for totalization.

The second article dates from 2021, and is a review of the literature [3], reporting the results of 6 articles, including Lau's paper described just above and that of Harreld et al. [20], which reports only the results of conservative treatment.

Feeley at al. [21], compared the results of hemiarthroplasty with PTEA in 64 patients, of whom only 4 were sickle-cell patients, allowing no particular conclusions to be drawn with regard to this etiology.

Kennon et al. [5], report the results of 7 resurfacings and 1 TAP at 1 year minimum follow-up in sickle-cell patients among 19 arthroplasties for atraumatic osteonecrosis of the humeral head with good results without distinction between etiologies (sickle-cell or cortico-induced).

Colegate-Stone et al. [22], report the results of only 2 hemiarthroplasties and one PTEI on the visual analogue scale of pre- and postoperative pain, without specifying the postoperative delay, among 45 sickle cell osteonecroses treated mainly conservatively.

Ristow et al. [23], report the good clinical results of shoulder

arthroplasty on 29 osteonecroses at 1 year minimal follow-up, of which only 8 were of sickle-cell origin, without specifying the type of prosthesis implanted specifically in these patients (hemiarthroplasty or TAP).

The third article dates from 2023 [12], and compares the clinical results of 17 shoulder arthroplasties (9 hemiarthroplasties, 7 TAP and 1 RP) for sickle-cell osteonecrosis at 5 years mean follow-up with 34 arthroplasties for shoulder osteoarthritis in a matched group (age, sex, BMI, type of prosthesis and postoperative time). The authors found more blood transfusions and a longer mean length of stay in sickle cell patients. Functional results were good in both groups, with a significant difference only in pre- and postoperative pain, which was greater in the sickle-cell patients, although the improvement was also very significant in this group. Two sickle cell patients had a SCC immediately post-operatively. There were 3 times more complications in the sickle-cell group, although this was not statistically significant probably due to the small number of patients.

DISCUSSION

Arthroplasty of sickle cell osteonecrosis of the humeral head has long been considered disappointing [11,24], however the literature is very weak. Reserved for advanced stages where the humeral head is no longer round, treatment mainly involved implantation of a hemiarthroplasty with a metal head [5,11,13]. This metal head in contact with the glenoid cartilage in these young patients could be the cause of the pain described in relation to other etiologies. Like Lau et al. [7], our study includes one patient who required revision of her metal hemiarthroplasty because of symptomatic articular space narrowing at 5 years' follow-up [Figure 4]. We believe that shoulder prosthesis revision should preferentially be carried out as an RP, given the osteoarticular and soft-tissue changes resulting from the first operation.

The use of a pyrocarbon head for hemiarthroplasty, which was never published in this etiology, probably limits the risk of articular

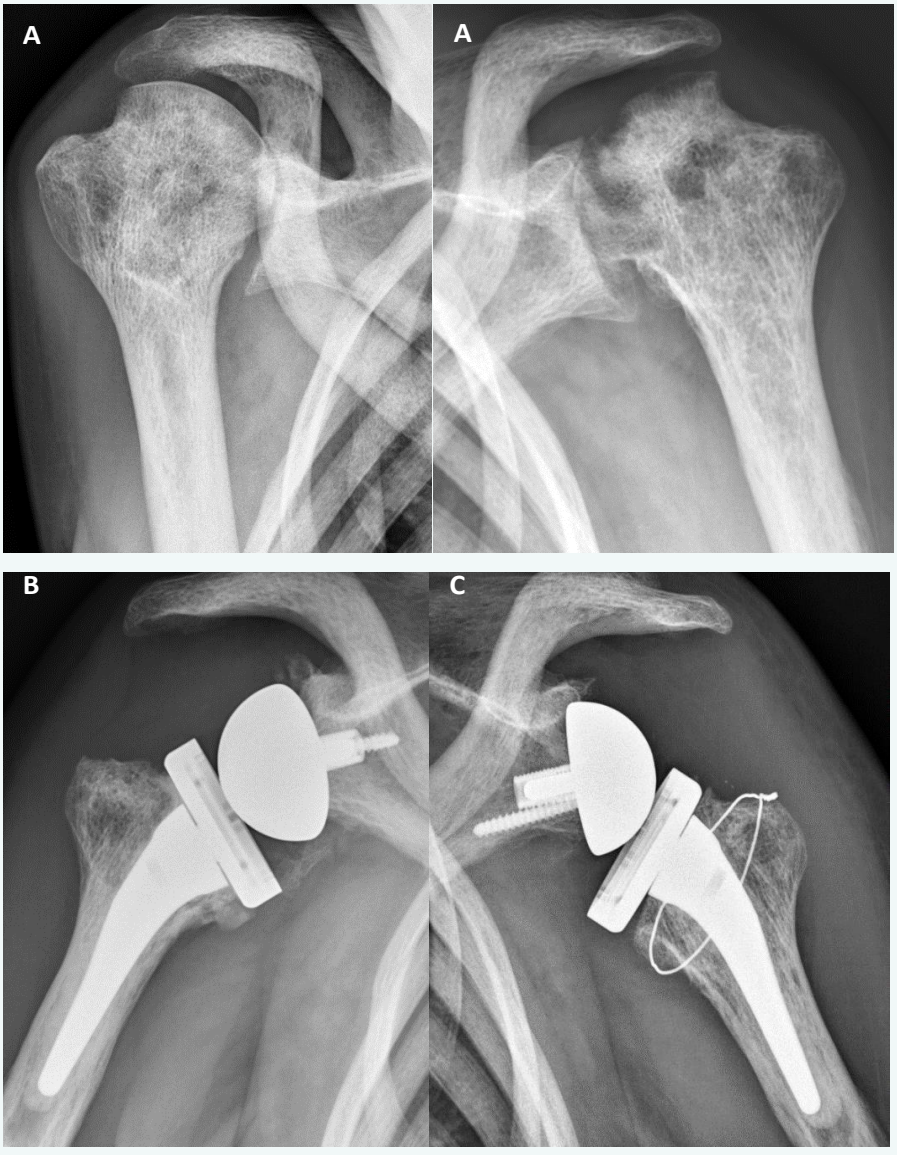

Figure 4: humeral head stage 3 osteonecrosis (A), hemi-arthroplasty with metallic head and a cemented stem (B), painful articular space narrowing at 5 years follow-up (C), prosthesis replacement for a reversed arthroplasty (D).

space narrowing in the medium term and reduces residual pain due to its biomechanical characteristics [10]. Contrary to Lau et al. [11], and Marigi et al. [12], we found a clear improvement in pain comparable to the results described in the literature for other etiologies [10].

Our indications, in the event of failure of medical treatment, are similar to those in the literature [3,4], and are currently as follows [Table 2]

-Symptomatic stage 1 and 2 osteonecrosis: conservative treatment with core decompression +/- bone marrow autograft,

-Stage 3 and 4 osteonecrosis: pyrocarbon hemiarthroplasty,

-Stage 5 osteonecrosis: hemiarthroplasty in PYC or TAP depending on patient's age and remaining glenoid cartilage,

-Stage 6 osteonecrosis: RP or isolated reverse glenoid resurfacing (hemi-reversed) [14].

The characteristics of necrotic bone, which is hard and brittle, make surgery difficult. In advanced osteonecrosis with epiphyseal-metaphyseal-diaphyseal necrosis, the humeral shaft cannot be catheterized due to cortico-medullary dedifferentiation. In such cases, false routes and intra-operative fractures are to be feared, and humeral preparation must be carried out with a motorized burr from proximal to distal. A patient with bilateral stage 6 necrosis initially benefited greatly from RP implantation on both sides, with constant increasing from 8/100 to 70/100 at 2 years' follow-up. Painful functional deterioration then progressively appeared, with glenoid loosening on one side (-40 points on Constant score) and humeral loosening on the other side (-20 points on Constant score) at 5 years' follow-up [Figure 3]. The humeral loosening occurred on the shoulder that had undergone additional wiring during arthroplasty, due to an intraoperative epiphyso-metaphyseal fracture.

This study had several limitations. First, there is no control group nor randomization. Indeed, it was not adapted to this kind of study as shoulder arthroplasty is already well known for its efficacy in gleno-humeral degenerative disease [25]. Second, the retrospective design makes the study prone to type I error. Finally, the follow-up is short. Mid and long-term studies should be considered particularly for pyrocarbon hemi-arthroplasties which survival needs to be confirmed.

CONCLUSION

Shoulder arthroplasty of sickle cell osteonecrosis leads to a marked improvement in shoulder function in the majority of cases. The indications for surgery depend on the stage of necrosis, with pyrocarbon hemiarthroplasty, anatomic total prosthesis or reversed prosthesis progressively proposed. Very advanced lesions greatly complicate the surgical procedure and compromise mid-term results. These patients need to be managed in a specialized center in collaboration with hematologists to prevent peri-operative complications such as sickle cell crisis [26].

INFORMED CONSENT

Written informed consent was obtained from the patients for their anonymised information to be published in this article.

ETHICAL APPROVAL

Henri Mondor teaching hospital does not require ethical approval for reporting case series.

CONTRIBUTORSHIP

SZ conceived the study, SZ and VH researched the literature, VB was involved in protocol development, gaining informed consent, patient recruitment and data analysis. VB wrote the first draft of the manuscript. SZ and VH reviewed and edited the manuscript.

AVAILABILITY OF DATA AND MATERIAL

All data and material are available on demand.

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