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Surgical treatment of humeral head avascular necrosis in patients with sickle cell disease: a systematic review

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Background: Sickle cell disease is the leading etiology for atraumatic humeral head avascular necrosis worldwide. Treatment of this condition is not standardized, with only few studies evaluating clinical outcomes after surgical interventions. The aim of this study was to review the available evidence on the results of surgical intervention for humeral head avascular necrosis in the sickle cell disease population.
Methods: A systematic electronic search was conducted using PubMed (MEDLINE), EMBASE, and Cochrane Library databases. Relevant studies that reported the outcomes of surgical intervention for humeral head avascular necrosis for patients with sickle cell disease were reviewed. Outcome parameters were pain, range of motion, specific shoulder outcome scores, and complications.
Results: Six studies, three retrospective cohorts (2 level III and 1 level IV) and three case series (level IV), were included in this review. A total of forty-three patients with sickle cell disease, comprising forty-nine shoulders, underwent different surgical procedures. Surgical procedures were core decompression, arthroscopic intervention, humeral head resurfacing, shoulder hemiarthroplasty, and total shoulder arthroplasty.
Conclusion: Surgical intervention for humeral head avascular necrosis in patients with sickle cell disease is selected based on the osteonecrosis stage. In the precollapse stage, core decompression is regarded as the first surgical option. However, in the light of current evidence, it has not been confirmed to prevent or delay natural progression of the disease. Shoulder arthroplasty is reserved for late stages, which despite the fairly good outcomes, data for long-term implant survival and complications are not well documented.

Sickle cell disease (SCD) is a common inherited hematological disorder, caused by single amino acid substitution in the hemoglobin beta chain (HbS).[21,27] In addition to the homozygotic HbSS disease (sickle cell anemia), it includes all heterozygous genotypes containing at least 1 sickle gene in which HbS makes up at least half the present hemoglobin.[27] In the United States, SCD affects more than 90,000 individuals.[2] Worldwide, it is predominantly prevalent in the Arab-India region and distinct areas in Africa.[27] The overall spectrum of disease expression varies greatly among patients with different genetic variants.[1,3,27] Geographical and cultural specificities have also been recognized to be of an important influence on disease morbidity.[23]

Skeletal manifestations of SCD have been well documented with avascular necrosis (AVN) reported as the most frequent complication.[1,3,20] Worldwide, SCD is considered the most common etiology of AVN.[1,12] In terms of large joint involvement, humeral head AVN (HHAVN) comes second to the femoral head,[12,22] with an estimated prevalence rate of 5.6%.[18] The primary risk factors for the development of HHAVN include the presence of femoral head AVN and genotype HbSC or HbS thalassemia, whereas the risk for lesion size, rate of progression, and joint collapse are attributed to the presence of the HbSS homozygous genotype.[3,13,22]

Clinically, patients present with shoulder pain and decreased range of motion (ROM).[6] Initially, symptoms are mild and transient; therefore, it can go unrecognized for a long time.[6,22] The glenohumeral joint is less conforming and bears less significant weight than the hip; therefore, deformity can be tolerated as long as ROM is maintained.[3] Shoulder disability occurs slightly before structural failure develops.[6] Most of the affected patients will progress into

Institutional review board approval was not required for the systematic review.

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head collapse within an average of 6 years from the onset of pain, if left untreated.[22]

Diagnosis of HHAVN is confirmed by radiographic images. The most commonly used classification to evaluate extent of joint involvement is Cruess modification[5] of the Ficat and Alert classification,[11] which is a radiological classification staged (1-5) based on the shoulder's progression to collapse (Table I). Several surgical interventions have been reported[27]; they include joint-preserving procedures (arthroscopic débridement, core decompression, vascularized bone grafting, humeral head resurfacing) or shoulder arthroplasty. Considering treatment options is based on the stage of HHAVN, extent of joint involvement, and patient overall clinical condition.[8] In precollapse stages, core decompression is regarded as the first surgical option, whereas total shoulder arthroplasty is to be reserved for end-stage HHAVN.[8,11]

Given special considerations for SCD as a leading etiology to HHAVN worldwide,[1,12] literature is scarce with studies evaluating effectiveness of surgical treatment for HHAVN in patients with SCD. David et al[6] was the first to report complications after shoulder arthroplasty in two patients with SCD. Since then, several studies have described the outcomes of surgical treatment in patients with HHAVN of different etiologies.[25] However, these studies are few and have limited patients. In addition, they fail to compare surgical outcomes of different etiologies, with the patients with SCD being notably underrepresented.[25]

The purpose of this systematic review is to present the available evidence of clinical outcomes after surgical intervention in the SCD population suffering from HHAVN. The primary objective is to investigate functional outcome scores and complication rates for each surgical procedure. We hypothesized that surgical intervention would yield significant improvement in shoulder functional scores and overall patient satisfaction.

Materials and methods

Search strategy

We used the Preferred Reporting Items for Systematic review and Meta-Analysis (PRISMA) protocol in the reporting of this review.[19] A search strategy was designed to retrieve all articles related to any surgical interventions for the shoulder in patients with SCD. The search strategy was conducted by a medical librarian and peer-reviewed by a second qualified medical librarian using the PRESS (Peer Review of Electronic Search Strategies) checklist.[24] To maximize sensitivity, no study design filters were applied. The search was not limited by language or publication year. The search strategy was devised on OVID MEDLINE and was adapted for the other databases. In all cases, the databases were searched from database inception to March 8, 2020. Duplicates were removed manually. Complete search strategy from all databases is available (Supplementary Appendix S1).

Eligibility criteria

The research question and eligibility criteria were determined a priori. A pilot test was performed before screening on a random sample of twenty studies to ensure applicability of those criteria. Studies were included if they (1) involved adult patients with SCD diagnosed with HHAVN, (2) reported surgical intervention, and (3) reported clinical outcomes. In regard to the study type, randomized controlled trials, prospective and retrospective comparative studies, and case series of three or more patients were included. There were minimal exclusion criteria to ensure comprehensive results. Studies were excluded if they (1) did not involve any surgical intervention, (2) did not specify clinical outcomes for patients with SCD as a separate entity, (3) were case reports, and (4) were review articles.

Table I
Cruess HHAVN classification—modified Ficat and Arlet classification.

Stage	Description
Stage I	• Diffuse clinical signs and symptoms • Normal X-rays • MRI may identify and quantify precollapse disease especially in symptomatic shoulders
Stage II	• Sclerosis (wedged, mottled) • Osteopenia • Humeral head sphericity is maintained
Stage III	• Crescent sign indicating a subchondral fracture • Minimal depression of articular surface
Stage IV	• Flattening and collapse of joint surface and subchondral bone • Fragmentation • Loose bodies • Secondary arthritis
Stage V	• Degenerative disease involving the glenoid

HHAVN, humeral head avascular necrosis; *MRI*, magnetic resonance imaging.

Data collection and analysis

Selection of studies

Two independent reviewers (J.A. and M.A.) conducted all levels of screening. For any disagreement, it was resolved via discussion between the two reviewers, with any potential unresolved conflicts mediated by a third senior reviewer (S.Q.). The reviewers were not blinded to the year, authors, and journal of publication.

Quality assessment of the included studies

We used the methodological index for nonrandomized studies (MINORS) criteria to evaluate the quality of included studies.[17,26] This was carried out and discussed in duplicate (J.K. and M.A.). MINORS is a validated scoring tool for nonrandomized studies including 12-item assessment. Each item is given a score from 0 to 2, with an ideal score of 16 for noncomparative studies and a score of 24 for comparative studies. This tool was compatible with the adopted inclusion and exclusion criteria.

Data abstraction

Data were abstracted in duplicate by the two reviewers and recorded into a Microsoft Excel spreadsheet. The abstracted data included author, year of publication, study design, patient demographics (sample size, number of patients/shoulders with SCD, age, and sex), type of surgical intervention, length of follow-up, loss of follow-up, and revision surgery. The outcome data were preoperative and postoperative measurements of pain, ROM, upper extremity functional outcome scores, and complications. Any discrepancies were to be resolved by discussion with a senior reviewer to minimize selection bias and errors.

Statistical analysis

Assessment of inter-rater agreement was carried by calculating a weighted κ (kappa) for each stage of title, abstract, and full-text screening.[17] The intraclass correlation coefficient was used for evaluating the quality assessment score agreement.[17] Reviewers leaned toward including studies (ie, if 1 reviewer thought a study should be included at the title screening stage, it was included). A kappa value of $\kappa > 0.61$ indicates substantial agreement, $0.21 < \kappa < 0.60$ indicates moderate agreement, and $\kappa < 0.20$ indicates slight agreement.[17]

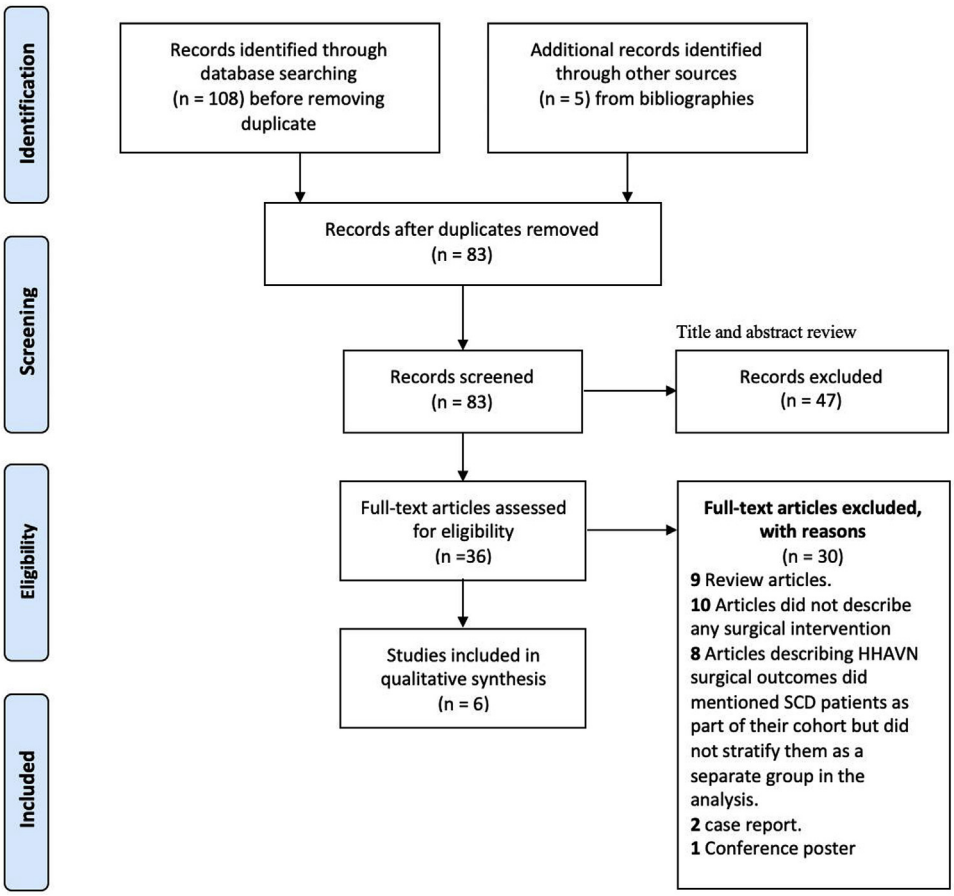

Fig. 1 The flow diagram of included studies. *HHAVN*, humeral head avascular necrosis; *SCD*, sickle cell disease.

Results

A total of 108 studies were found across the following databases: MEDLINE, EMBASE via Ovid, Cochrane Library via Wiley, and CINAHL via Ebsco. After duplicates were removed, seventy-nine records were retrieved. Five additional studies were identified through screening bibliographies, resulting in a total of eighty-three studies. Of the initial eighty-three studies found, thirty-six proceeded to full-text screening after title and abstract screening. Thirty studies were excluded: nine review articles and ten articles did not involve surgical intervention, eight articles did not describe outcomes of patients with SCD in their results as a separate entity, two were case reports of a single patient, and 1 was a conference poster. Ultimately, six studies were included in this review (Fig. 1): three retrospective cohort studies (2 level III and 1 level IV evidence)[4,7,25] and three case series (level IV evidence).[10,13,14] In those six studies, forty-three (SCD) patients, comprising forty-nine shoulders, underwent different surgical interventions for HHAVN (Table II).

Description of agreements between reviewers

In this review, the reviewers had a substantial agreement for selecting articles for inclusion at the title and abstract stage with an inter-rater agreement value of 0.92 and full-text screening agreement of 1.0. A third reviewer for resolving discrepancies was not required. The agreement among quality assessment scores of included studies was 0.783 (95% confidence interval, 0.661-0.864). Three comparative studies were scored out of 24 points, and 3 noncomparative studies were scored out of 16 points. The average MINORS score was 21 of 26 and 13 of 16 for comparative and noncomparative studies, respectively (Table III)

Type of surgical intervention

All included studies in this review reported the outcomes of surgical intervention for treating HHAVN in patients with SCD. All studies except 1 described the results of more than 1 intervention for their cohort. Two studies investigated the role of percutaneous decompression; one study investigated the role of arthroscopic intervention. The role of shoulder arthroplasty was reported in all except 1 article; hemiarthroplasty (HA) was mentioned in five articles, total shoulder arthroplasty (TSA) was mentioned in four articles, reverse TSA (RTSA) was mentioned in two articles, and humeral head resurfacing was mentioned in 1 article. In regard to comparative studies, two studies compared results of HA vs. TSA, and 1 study compared results of core decompression with nonoperative intervention using a historical control group (Table II).

Classification and outcome measures

All studies except 1 used the Cruess modification of the Ficat and Arlet classification.[5] A single study used the Association Research Circulation Osseous international classification of HHAVN, which uses multiple radiographic modalities and histologic findings to describe five stages of osteonecrosis (0-4) based on the progression to collapse.[9] However, only 4 studies reported the disease stages of their cohort.

Table II
Characteristics of included studies.

Author	Yr of publication	Level of evidence	Type of study	Procedures	All patients		Patients with SCD		Disease stage	MINORS score
					No. of shoulders (patients)	Mean age (range)	No. of shoulders (patients)	% of SCD shoulders		
Ristow et al[25]	2019	III	Retrospective cohort	TSA vs. HA	29 (25)	49 (16-77)	8 (6)	27.6		22
Colegate-Stone et al[4]	2018	IV	Retrospective cohort	Arthroscopic intervention*, HA, and RTSA	45 (45)	40 (21-62)	11 (11)	24.4	II, III, IV	11
Kennon et al[13]	2016	IV	Case series	Decompression, resurfacing, HA, TSA, and RTSA	25 (20)	37 ± 15	13 (11)	52	I, II, III, IV	14
Harreld et al[10]	2009	IV	Case series	Decompression	26 (15)	37 (15-50)	5 (3)	19.2	I, II	20
Feeley et al[7]	2008	III	Retrospective cohort	TSA vs. HA	64 (64)	57 (46-77)	4 (4)	6.2		22
Lau et al.[14]	2007	IV	Case series	TSA, HA	8 (8)	37 (25-47)	8 (8)	100	II, III, IV, V	14
					N = 49					

HA, hemiarthroplasty; *MINORS*, methodological index for the nonrandomized studies; *RTSA*, reverse total shoulder arthroplasty; *SCD*, sickle cell disease; *TSA*, total shoulder arthroplasty.
Decompression: Small-diameter percutaneous decompression.
Arthroscopic intervention: Arthroscopic debridement, capsular release, and bursectomy ± subacromial decompression.
N: total number of SCD shoulders in all included studies.

The main clinical scores were the American Shoulder and Elbow Surgeons (ASES) score, which evaluates pain and function of the shoulder,[28] University of California Los Angeles (UCLA) score, and Constant score, which each combines findings of physical examinations with patient-reported measures including pain, function, and patient satisfaction, visual analog scale (VAS) for subjective evaluation of pain, L'Insalata score, which evaluates pain, daily activities, recreational and athletic activities, work, and overall satisfaction.[15] Although ROM was mentioned in two studies, preoperative and postoperative degrees were incomplete and therefore were not reported in our tables. Main outcome scores are summarized in Table IV.

Decompression outcomes

Two studies discussed the effect of percutaneous decompression of the humeral head in patients with SCD with HHAVN. Both studies were similar in the following: multiple cohorts with different etiologies were evaluated, included patients were at stage I or II and similar technique described by Harreld et al was used. Harreld et al studied twenty-six shoulders; five shoulders (three patients with SCD) were retrospectively reviewed with a mean follow-up of 2.7 years. The UCLA score showed significant improvement in all cohorts (from 14 to 27, $P < .0001$), with significant reduction in pain (from 2.9 to 8.8, $P < .0001$). Kennon et al evaluated six shoulders (six patients with SCD); all patients progressed to humeral head collapse and required either resurfacing or TSA at a mean follow-up of 17 months.

Arthroscopic intervention outcomes

The role of arthroscopic intervention was described in a single study. Colegate-Stone et al retrospectively looked at improvement in the pain score and subjective patient satisfaction after a combination of arthroscopic procedures for eight shoulders with stages II, III, and IV on the Association Research Circulation Osseous staging system. Significant improvement in the VAS pain score was noticed in patients with stage II who underwent arthroscopic core decompression and subacromial decompression (from 8.8 to 3) However, patients with stage III who underwent arthroscopic débridement, capsular release bursectomy, and subacromial decompression showed no significant improvement in pain and were generally unsatisfied.

Arthroplasty outcomes

Five studies analyzed the role of shoulder arthroplasty in patients with SCD suffering from HHAVN.

A total of thirty-one shoulders included in this review underwent different arthroplasty procedures: humeral head resurfacing, HA, TSA, and RTSA. Alongside discussing the role of percutaneous decompression, Kennon et al discussed the role of humeral head resurfacing as an alternative option in early stages and arthroplasty in advanced stages. Seven patients with SCD (stages II, III) underwent resurfacing as a primary procedure, or as revision to decompression, and 1 patient (stage IV) underwent TSA. The outcome measures of both resurfacing and arthroplasty were combined for different etiologies: SCD and steroid-induced osteonecrosis at the 2-year follow-up. All outcome scores improved: UCLA (from 9.6 to 29), ASES (from 19.7 to 81.4), and Constant scores (from 28 to 87).

Colegate-Stone et al also described the role of arthroplasty in advanced stages of osteonecrosis. Three patients with SCD (stage IV) underwent arthroplasty (2 HA and 1 RTSA). Results were assessed using the VAS for the pain score, which showed an average

Table III
The methodological index for the nonrandomized studies (MINORS) score.

MINORS score	Author's name																	
Items	Ristow et al[25]			Colegate-Stone et al[4]			Kennon et al[13]			Harreld et al[10]			Feeley et al[7]			Lau et al[14]		
	R1	R2	C	R1	R2	C	R1	R2	C	R1	R2	C	R1	R2	C	R1	R2	C
Clear aim	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2
Inclusion of consecutive patients	2	2	2	2	2	2	2	0	2	2	2	2	2	2	2	2	2	2
Prospective collection of data	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2
Endpoints appropriate to the aim	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2	2
Unbiased opinion of endpoints	2	2	2	1	2	1	2	2	2	2	2	2	2	2	2	2	2	2
Appropriate F/U period	2	2	2	1	0	0	2	2	2	2	2	2	2	2	2	2	2	2
Loss to F/U <5%	2	2	2	2	2	2	0	0	0	2	2	2	2	2	2	2	2	2
Prospective calculation of the study size	2	2	2	0	0	0	2	2	2	2	2	2	2	2	2	1	0	0
Adequate control group	1	0	0							0	1	0	0	0	0			
Contemporary groups	2	2	2							0	0	0	2	2	2			
Baseline equivelance of groups	1	2	2							1	2	2	2	2	2			
Adequate statistical analysis	2	2	2							2	2	2	2	2	2			
Total score	22	22	22	12	12	11	14	12	14	19	21	20	22	22	22	15	14	14
Ideal score	24			16			16			24			24			16		

Scoring: 0, not reported; 1, reported but inadequate; 2, reported adequate.
C, consensus; R1, reviewer 1 (J.K.); R2, reviewer 2 (M.A.).

pain reduction from 9.5 to 4.1 and patient satisfaction score of 8.5 of 10.

Ristow et al retrospectively studied the role of shoulder arthroplasty in different patients' etiologies including trauma, chronic corticosteroid, and SCD. Outcomes of HA vs. TSA were then compared regardless of the etiology. Eight shoulders (six patients with SCD) underwent either HA or TSA. Outcomes were evaluated using the UCLA, ASES, and the Constant scores, which all showed improvement in the median scores from 11.5, 27.3, and 42.6 to 25, 84.2, and 96.6, respectively. Feeley et al also retrospectively analyzed results of shoulder arthroplasty for four patients with SCD (2 HA, 2 TSA) as part of a larger cohort (sixty-four patients) with various etiologies of HHAVN. Outcome measures (ASES and L'Insalata scores) were stratified as per etiology. Postoperative ASES and L'Insalata scores for patients with SCD were 77 and 75, respectively (±8 standard deviation). Postoperative degrees of ROM were 53.3 (±3.4) external rotation, 143 (±7.9) flexion, and an internal rotation level up to T12 vertebrae.

Finally, Lau et al solely reviewed surgical outcomes of HHAVN in patients with SCD independently. The study followed up eight patients (7 HA, 1 TSA) for an average of 4.25 years. Outcome measures were assessed using ASES and VAS for pain. All patients showed improvement on both scores, from 15 and 9.25 to 46.9 and 6.38, respectively. In addition, the degree of ROM improved as follows: 20 degrees external rotation, 3 degrees glenohumoral abduction, and 35 degrees forward flexion. This improvement was also reflected on activity of daily living.

Comparative studies

Harreld et al compared the outcomes of patients suffering from atraumatic HHAVN (stages I, II), who underwent percutaneous decompression, with a historical control group established from 4 articles that described the natural progression of the disease. Compared with the control group where most patients progressed and required arthroplasty, all patients in the cohort reported in the study by Harreld et al had improvement in pain and outcome measures, requiring no arthroplasty after 3 years of follow-up. It should be noted that in the historical control group, some patients were at more advanced stages at presentation. Another two articles[6,18] focused solely on patients suffering from SCD and were discussed separately as they were not representative of the population reported in the study by Harreld et al. In both studies, more benign disease progression was observed than that of osteonecrosis of other etiologies. Only three of 428 patients with SCD with HHAVN included in both articles required arthroplasty.

Two studies compared the outcomes of HA vs. TSA. Feeley et al compared thirty-seven patients with HHAVN who underwent HA with twenty-seven patients treated with TSA, regardless of their etiology. There was no significant difference in ASES or L'Insalata

Table IV
Main outcome scores.

Author	Mean follow-up (range in yr)	Lost F/U	UCLA		Constant score		VAS pain		ASES		L'Insalata	
			Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Ristow et al[25]	3.9 (1-8.5)	0	11.5	25	42.6	96.6	–	–	27.3	84.2	–	–
Colegate-Stone et al[4]	N/A	0	–	–	–	–	Arth CD: 8.8 Arth+: 9.5 Arthroplasty: 9.5	3.5 8.5 4.1	–	–	–	–
Kennon et al[13]	at 1 and 2 yrs	3	9.56 (2.58)	29 (9.4)	28.06	87 (18.52)	–	–	19.69 (19.2)	81.43 (27.49)	–	–
Harreld et al[10]	2.7 (2-3.4)	0	14 (10-22)	27 (14-30)	–	–	–	–	–	–	–	–
Feeley et al[7]	4.8 (2-7)	0	–	–	–	–	–	–	–	77 (9.2)	–	75 (±8)
Lau et al[14]	4.25 (2-10)	0	–	–	–	–	9.25	6.38	15	46.9	–	–

ASES, American Shoulder and Elbow Surgeons; *UCLA*, University of California Los Angeles; *VAS*, visual analog scale.
Arth CD: arthroscopic core decompression + SAD subacromial decompression; stage II.
Arth+: arthroscopic debridement, capsular release, bursectomy + SAD; stage III.

Table V
Complications.

Author	Procedure	Complications (no. of patients)	Revision/Other intervention
Ristow et al[25]	TSA and HA	–	–
Colegate-Stone et al[4]	Arthroscopic interventions, HA, and RTSA	–	–
Kennon et al[13]	Decompression	Progression (6)*	Resurfacing (5) TSA (1)
	Resurfacing	Stiffness (1)	HA
		Glenoid wear (1)	TSA
		Scapular insufficiency (1)	RTSA
	TSA	–	–
Harreld et al[10]	Decompression	–	–
Feeley et al[7]	TSA vs. HA	–	–
Lau et al[14]	TSA	–	–
	HA	Required blood transfusion (4)	Medical management
		Sickle cell crises (2)	Medical management
		Intra-op rotator cuff tear (1)	Repaired during the same procedure
		Stiffness (1)	Arthroscopic capsular release
		Septic loosening (1)	Antibiotics suppression

HA, hemiarthroplasty; *RTSA*, reverse total shoulder arthroplasty; *TSA*, total shoulder arthroplasty.
* All the patients who underwent small-diameter percutaneous decompression in the study by Kennon et al progressed and required revision surgeries.

scores between the two groups. The only significant difference was better flexion ROM in the HA group ($P < .007$). When stratified based on etiology, steroids vs. traumatic versus sickle cell, patients with SCD had higher outcome scores. However, sample size was too small (four shoulders) to show significance. Ristow et al had nineteen patients treated with HA and ten patients treated with TSA. Both groups showed significant overall improvement in functional outcome scores regardless of implant selection, disease stage, or etiology. The TSA group had higher outcome scores than the HA group. However, these differences were statistically insignificant ($P > .05$).

Complications

Three studies reported complications after surgical intervention for HHAVN in patients with SCD. Described complications include progression of osteonecrosis, septic loosening, glenoid wear, scapular insufficiency, and joint stiffness (Table V). Colegate-Stone et al reported progression of 1 patient with SCD (stage III) after arthroscopic débridement and capsular release. This patient was subsequently considered for arthroplasty. Kennon et al reported a 100% progression rate in six patients with SCD after percutaneous decompression at a mean follow-up of 17 months. All patients were revised by humeral head resurfacing except 1 patient who required TSA owing to advanced degenerative changes (stage IV). Three of those patients who underwent humeral head resurfacing had different complications requiring further revision surgeries; one patient had osteonecrosis changes at the resurfacing stem at the 2-year follow-up, at which HA was planned for. Another patient who had stiffness and glenoid wear at the 1-year follow-up was revised to TSA. The third patient presented with scapular insufficiency and was revised to RTSA.

Finally, Lau et al particularly described perioperative complications that are unique to the SCD population. Four patients required blood transfusion during their hospital stay which ranged between three and 8 days. Two patients had sickle cell crises immediately after operation. In regard to complications after HA, three patients had complications that required intervention. An intraoperative rotator cuff avulsion was encountered in 1 patient. Another patient had persistent pain and stiffness that necessitated arthroscopic capsular release that was performed at 22 months from primary surgery. However, the patient reported no significant improvement. The last patient had a more complicated course; she initially presented with an infected total knee arthroplasty and was also found to have shoulder HA septic loosening, with significant bone loss along the medial calcar, and glenoid erosion 7 years after the operation. However, because of her very poor medical condition related to her SCD, she was not fit for further shoulder revision and was treated with antibiotic suppression. Four years later, her pain score was similar to her initial post-HA procedure. She later passed away from SCD complications.

Discussion

Treatment for osteonecrosis in patients with SCD is not standardized, with less experience in treating the humeral head than the femoral head.[16] This review aimed to evaluate the effectiveness of surgical intervention in treating humeral head osteonecrosis in patients with SCD. There is no randomized controlled trial available for this review. Despite the absence of high-level articles, our work provides a comprehensive review of available literature.

It is important to emphasize that all included studies but 1 had the SCD group as part of a larger cohort. During data extraction, we experienced difficulties in separating outcome results for those with SCD from other etiologies of osteonecrosis. In 1 study, the outcome measures were calculated for the overall cohort which could be a confounding factor.

In addition, included studies had very small number of patients with SCD, making it difficult to compare results. Initially, we thought there could be room for quantitative statistical analysis given that data were reasonably homogenous in which more than 1 article used a similar outcome measure for a similar cohort undergoing similar intervention. However, important data, normal distribution measures, and some preprocedure outcome scores were incomplete. We attempted to reach to authors to provide us with the missing data; unfortunately, we had no response. As a result, it was not possible to provide an official meta-analysis.

All studies but 1 had a decent follow-up period of no less than 2 years after the primary procedure. There is consensus regarding indications for procedure selection. All studies agreed that selecting a procedure is based on disease progression; in the precollapse stage, core decompression was regarded as a first option, whereas arthroplasty was reserved for late stages. All studies but 1 used a similar radiographic staging system.

Arthroscopic intervention was mentioned in 1 study[4]; the study lacks objective assessment. A single subjective outcome measure was used: VAS for pain with no proper statistical significance parameters. A procedure was called successful depending solely on the patient satisfaction score at 1 follow-up. This makes it impossible to draw respectful conclusion.

The role of core decompression as an early intervention is still uncertain. Although it has been argued to have beneficial results in 1 cohort,[10] it showed no effect on disease progression in another cohort.[13] Humeral head resurfacing was selected as either an alternative or revision procedure for those who continued to progress after decompression. Although resurfacing initially yielded significant improvement in functional scores, revision was required for the following complications: the presence of osteonecrosis at the resurfacing stem (revised with HA), glenoid wear (revised with HA), and scapular insufficiency (revised with RTSA).

It is agreed that arthroplasty is valuable in advanced stages of the disease. Majority of patients included in this review were treated with HA. All authors agreed that both procedures (HA and TSA) yielded significant benefits in terms of pain, ROM, function, and patient satisfaction. When outcome measures were compared between the two procedures, no significant difference was shown. This is most likely owing to the small number of patients in each group. Nevertheless, 1 author[17] noted better forward flexion in the HA group ($P < .007$). Reoperation rates were higher in the TSA group regardless of etiology. However, when patients with SCD were stratified, none had reoperations or complications on a short term. After HA, three complications that required intervention were reported: rotator cuff avulsion, stiffness, and deep infection. The use of RTSA was indicated in managing scapular insufficiency complication after the head resurfacing procedure.

Finally, specific concerns for patients with SCD undergoing shoulder arthroplasty should be anticipated. Lau et al[14] pointed out the increased demand for narcotics and blood transfusion in the perioperative period. Pain control after shoulder arthroplasty is less predictive than those with other etiologies of HHAVN. It has been suggested that individual response to surgery maybe genetically determined. Of course, further research is needed to look if a potential relationship between SCD genetic variance and shoulder arthroplasty outcomes exists.

Conclusion

Considering the low level of evidence of included articles in this review, there is a clear necessity for larger high-quality prospective and comparative trials to further evaluate the effectiveness of surgery in treating humeral head osteonecrosis in the SCD population. For patients with SCD suffering from early stages of HHAVN, core decompression has not yet been confirmed to prevent or delay natural progression of the disease. The role of arthroplasty on the other hand is promising for advanced stages. Superiority of one arthroplasty procedure over another cannot be concluded in the light of the evidence in the current review. In addition, its effectiveness compared with other shoulder etiologies and implant long-term survival data are not available. Future studies should aim to standardize data, provide longer follow-ups, and pay attention to SCD-related perioperative complications and implant survival.

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Supplementary data

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