Management of bipolar shoulder injuries with humeral head allograft in patients with active, uncontrolled seizure disorder: case series and review of literature

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Background: The purpose of this study is to present surgical outcomes after humeral head allograft augmentation and glenoid-based procedures in patients with active, uncontrolled seizure activity and anterior shoulder instability.

Methods: A retrospective review of a surgical database for patients with active seizure disorder and with recurrent shoulder instability managed with humeral head augmentation was performed. All patients underwent surgical intervention. Postoperative outcomes including Shoulder Pain and Disability Index, Simple Shoulder Test, American Shoulder and Elbow Surgeons questionnaire, and the Short Form Health Survey (SF-12) were recorded at a minimum of 2 years. We hypothesized that appropriate management of the bony defects in these bipolar injuries would result in low recurrence and satisfactory outcomes.

Results: Ten patients including 8 males and 2 females (15 shoulders) with active seizure-related shoulder instability underwent surgical intervention including allograft bone grafting of the Hill-Sachs lesion for anterior shoulder instability. The average age was 27 years. All patients reported recurrent seizures postoperatively, but only one sustained a shoulder dislocation after surgery that was unrelated to seizure activity.

Self-reported satisfaction was "much better" or "better" in 92% of shoulders. Average outcome scores were as follows: American Shoulder and Elbow Surgeons score = 67 (33-100), Shoulder Pain and Disability Index = 32.5 (0-83), Simple Shoulder Test = 9.4 (5-12), SF-12 PCS = 44.1 (21-65), and SF-12 MCS = 50.6 (21-61). The average follow-up was 4.8 years.

Conclusion: Management of bipolar shoulder injuries with humeral head allograft augmentation and glenoid based surgery leads to low recurrence rates and good clinical outcomes in patients with uncontrolled, seizure-related shoulder instability.

Epilepsy is a complex seizure disorder with many debilitating sequelae, including recurrent shoulder instability.[1,2,6,8,10,13,14] Shoulder dislocation after a seizure event occurs at a rate of 0.6%,[4] although the true incidence is unknown as many dislocations go undiagnosed.[12] Direction of shoulder instability associated with major motor seizures can be anterior or posterior. Posterior instability is traditionally ascribed to seizure activity; however, anterior instability is also common.[2,5,6,11,14] These patients present several unique challenges that must be considered for successful treatment. Management of seizure activity is critical to reducing shoulder instability events and requires a multidisciplinary approach including medical and sometimes neurosurgical intervention. Despite appropriate antiepileptic treatment, seizure recurrence is possible. In addition, these patients are often young and have large bony defects. Size and location of bone defects define the track and engagement of these lesions which correlate with risk of recurrent instability.[7] Together, these factors create a high risk for recurrent shoulder instability after surgical intervention.

There are no clear guidelines on the appropriate orthopedic surgical management of patients with seizure-related shoulder instability. In the general shoulder instability literature,
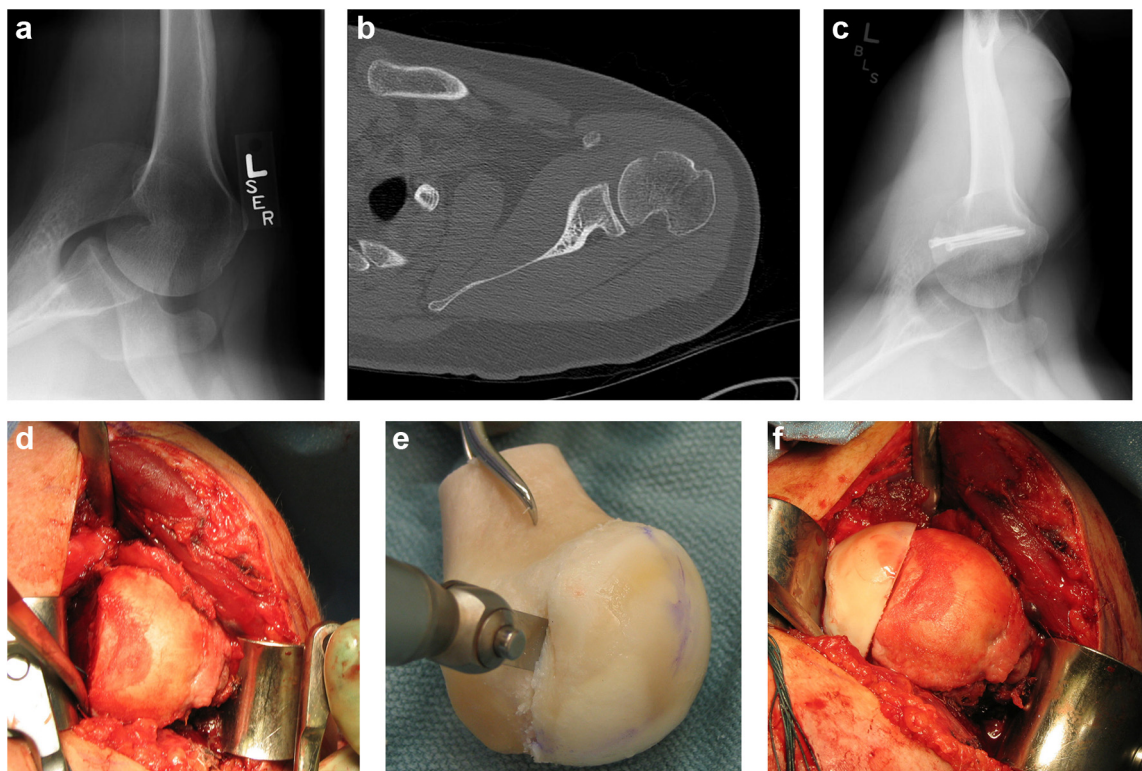
Figure 1 (**a**) Preoperative axillary radiograph of the left shoulder with a large humeral head defect. (**b**) Preoperative axial CT of the left shoulder with a large humeral head defect. (**c**) Postoperative axillary of the left shoulder. (**d**) The intraoperative photograph of large Hill-Sachs deformity. (**e**) The intraoperative photograph of humeral head allograft preparation. (**f**) The intraoperative photograph after fixation of humeral head allograft. *CT*, computed tomography.

management of associated bone loss leads to lower instability rates than soft tissue repair.[3,7] This would intuitively apply to seizure-related instability; however, few studies have reported surgical outcomes specific to seizure-induced instability, and even fewer studies have reported on bony augmentation procedures. Available data focus on isolated glenoid-based augmentation procedures and conflict with one study reporting no residual instability[8] and others with high rates of recurrent instability.[2,5,11,14] To our knowledge, no studies have reported on humeral head augmentation procedures in this unique patient population. The purpose of this study is to present surgical outcomes after humeral head allograft augmentation and glenoid-based procedures in patients with active, uncontrolled seizure activity and anterior shoulder instability. Our hypothesis is that these patients will have large humeral head defects and surgical management will lead to low recurrence rate despite recurrent seizures.

Methods

This study was approved by our institutional review board. All patients who underwent humeral head augmentation for seizure-related shoulder instability by the two senior authors (TW and MM) were identified from each surgeon's surgical database. The inclusion criteria were bipolar shoulder injuries managed with surgical intervention including the bone-graft procedure for a humeral-head defect in patients with seizure-related shoulder instability with a minimal follow-up of 2 years.

Exclusion criteria included nonseizure-related shoulder instability and patients who were managed nonoperatively. Medical records were then retrospectively reviewed for demographic and surgical data. Patient-reported outcomes were collected at the time of the present study in person or via telephone.

Surgical procedure

Surgical technique involved addressing the pathology of both the humerus and the glenoid (Fig. 1). The resulting large humeral-head defects were addressed with an allograft bone graft fashioned to fill the entire defect. Fresh, frozen humeral head osteochondral allograft was used to treat the humerus defect in all patients. A deltopectoral approach was used in all cases, and all patients were positioned in the beach chair position. The humeral head defect was exposed with extension, traction, and external rotation of the arm. Retractors are placed deep to the rotator cuff. The humeral head defect is cleared of nonviable tissue and shaped into a chevron. Bone wax is then placed in the defect to create a three-dimensional mold. Humeral head allograft is then shaped to match the mold. Allograft is then fixed with headless compression screws. Fluoroscopy is utilized to confirm the position of graft and orthopedic hardware. It is critical to avoid excess tension on the arm during exposure of the humeral head defect to avoid neurological traction injury. The glenoid was then surveyed for the extent of bone and soft-tissue damage. Surgeon discretion was used to determine glenoid-based procedure and included no intervention, capsulolabral repair, or bone augmentation procedure with either coracoid transfer or allograft augmentation.

Subjects were followed postoperatively and asked to complete a questionnaire either in person or via telephone that allows physicians to derive several functional outcome scores. These outcome scores include the Shoulder Pain and Disability Index, Simple Shoulder Test, American Shoulder and Elbow Surgeons questionnaire, and the Short Form Health Survey. In addition, we inquired

about epilepsy-control methods used, subsequent dislocation history, and additional surgeries performed elsewhere using a separate questionnaire.

A statistical analysis was not performed because of the small numbers and lack of a valid comparison group.

Results

Eleven patients (16 shoulders) met the inclusion criteria. One patient (one shoulder) declined to participate. This left 10 patients (15 shoulders), including 8 males and 2 females. One patient (2 shoulders) expired 4 years after his left and 5 years after his right shoulder surgery. This patient's data were included in demographic and complication sections but were excluded from the outcome data, as scores were not available for this patient. Demographic information is included in Table I. The average age at surgery was 27 years (16-50 years) with follow-up averaging 4.8 years (2-9 years). Five patients who were included in the study had bilateral shoulder involvement. Seven patients had the dominant shoulder involved.

Of the included shoulders, 1 sustained both anterior and posterior dislocations, whereas the remaining 14 sustained only an anterior dislocation. Ten glenoids were addressed with capsulolabral repair with suture anchors only. An additional number of 4 shoulders underwent concomitant bone grafting with 3 undergoing allograft bone grafting and 1 undergoing open Latarjet procedures. One patient underwent an isolated humeral head procedure. Glenoid-based procedures were not performed because of significant glenohumeral arthritis. Five patients had undergone previous surgery. Four underwent failed capsulolabral repair, and

1 underwent fixation of a tuberosity fracture. All reported recurrent shoulder instability due to recurrent seizure.

Bone loss was common. All patients had large humeral head defects; the average humeral head defect was $20 \times 15 \times 24$ mm. The percent glenoid bone loss ranged from 0% to 40% (mean 20.8%). Seven patients had less than 10% glenoid bone loss (mean 3.7%). All patients demonstrated some degree of glenohumeral arthritis mostly noted on the glenoid (Table I).

Although all patients reported being treated with medication for seizures after surgery, all continued to have seizures. Despite this, only 1 shoulder sustained an additional dislocation after surgery (redislocation rate of 7.7%), which occurred 3 months postoperatively and which the patient stated was not seizure related. One shoulder required two reoperations at our institution for hardware removal within 1 year and a second surgery 4 years later for a posterior labral tear obtained while playing hockey (reoperation rate of 8%). No other complications were encountered.

Ten shoulders were reported as "much better" and 2 as "better". One was reported as "unchanged". The Simple Shoulder Test, American Shoulder and Elbow Surgeons score, and Shoulder Pain and Disability Index scores indicate that in terms of disability, pain, function, strength, and range of motion, the patients had reasonably good functional results (Table II). The results of the Short Form Health Survey indicate that the patient population in this study exhibited relatively poor mental and physical health given their young age. Shoulder outcome scores are summarized in Table II.

Discussion

This study sought to add to the current, limited body of literature specific to management of seizure-related shoulder instability.

Table I
Patient demographics and surgical data.

Subject	Sex	Age at surgery	Involved side	Direction of dislocation	Corocoid fracture	Previous surgery	Humeral head defect size (mm)	Glenoid defect size (%)	Operative treatment humerus	Operative treatment glenoid
1L	Male	16	Left	Anterior	N	No	$15 \times 10 \times 24$	8	Humeral head allograft with AccuTrak screws[*]	Suture anchor
1R	Male	19	Right	Anterior	N	No	$19 \times 9 \times 21$	40	Humeral head allograft with AccuTrak screws[*]	Allograft bone grafting
2R	Female	29	Right	Anterior	N	3 Bankarts	$16 \times 25 \times 20$	38	Humeral head allograft with AccuTrak screws[*]	Allograft bone grafting
2L	Female	30	Left	Anterior	N	Bankart failed	$18 \times 11 \times 19$	30	Humeral head allograft with Synthes screws[†]	Latarjet with suture anchors
3R	Male	28	Right	Anterior	N	Bankart failed	$25 \times 23 \times 15$	33	Humeral head allograft with AccuTrak screws[*]	Suture anchor
3L	Male	28	Left	Anterior	N	No	$14 \times 28 \times 19$	35	Humeral head allograft with AccuTrak screws[*]	Suture anchor
4	Male	26	Left	Anterior	Y	No	$19 \times 15 \times 22$	0	Humeral head allograft with TriMed headless screws[‡]	Suture anchor
5	Male	41	Left	Anterior	Y	No	$29 \times 14 \times 37$	0	Humeral head allograft with AccuTrak screws[*]	Suture anchor
6R	Male	19	Right	Anterior	N	No	$19 \times 13 \times 32$	0	Humeral head allograft with TriMed headless screws[‡]	Suture anchor
6L	Male	20	Left	Anterior and posterior	N	No	$18 \times 15 \times 32$	5	Humeral head allograft with TriMed headless screws[‡]	Suture anchor
7	Male	50	Right	Anterior	N	Tuberosity fracture	$17 \times 15 \times 27$	40	Humeral head allograft with AccuTrak screws[*]	Suture anchor
8	Male	23	Right	Anterior	N	Bankart	$20 \times 16 \times 20$	31	Humeral head allograft with AccuTrak screws[*]	Allograft bone grafting
9R	Male	23	Right	Anterior	N	No	$20 \times 8 \times 24$	5	Humeral head allograft with AccuTrak screws[*]	Suture anchors
9L	Male	24	Left	Anterior	N	No	$28 \times 10 \times 26$	8	Humeral head allograft with TriMed headless screws[‡]	Suture anchors
10	Female	31	Right	Anterior	N	No	$19 \times 15 \times 25$	39	Humeral head allograft with TriMed headless screws[‡]	None (severe OA)

[*] Acumed, Hillsboro, Oregon.
[†] DePuy Sythnes, Warsaw, Indiana.
[‡] Trimed, Santa Clarita, California.

Table II
Patient outcomes.

Subject	Follow-up time (yrs)	Seizure after surgery	Redislocation	Reoperation	Self-reported overall satisfaction	SPADI 130	SST-12	ASES	SF-12 (PCS)	SF-12 (MCS)
1L	5	Yes	No	No	Much better	14	12	93	40	53
1R	2	Yes	Yes	No	Unchanged	55	8	58	40	53
2R	8	Yes	No	No	Much better	82	9	38	21	62
2L	7	Yes	No	No	Much better	82	9	38	21	62
3R	-	-	-	-	-	-	-	-	-	-
3L	-	-	-	-	-	-	-	-	-	-
4	2	Yes	No	No	Better	12	9	72	38	48
5	7	Yes	No	No	Much better	83	5	52	40	55
6R	3	Yes	No	No	Much better	3	12	97	52	53
6L	2	Yes	No	No	Better	51	6	33	30	61
7	9	Yes	No	No	Much better	0	12	100	57	54
8	8	Yes	No	Yes	Much better	34	8	62	51	58
9R	3	Yes	No	No	Much better	14	12	87	65	21
9L	4	Yes	No	No	Much better	28	11	75	65	21
10	2	Yes	No	No	Much better	47	9	73	53	57

SPADI, Shoulder Pain and Disability Index; *SST-12*, Simple Shoulder Test; *ASES*, American Shoulder and Elbow Surgeons; *SF-12*, Short Form Health Survey.

We report on a unique and challenging cohort of seizure-related shoulder instability cases managed with surgical intervention who continued to have active seizure activity postoperatively. Several important findings were encountered. Large humeral head bone defects were common, whereas glenoid bone defects were encountered but less frequently. Bone augmentation of the humeral head lesion with an appropriate glenoid-based procedure led to low recurrence rates despite recurrent, active seizures. The overall recurrence rate was 1 of 13 with this approach. This rate is lower than much of the available data.

To our knowledge, one study has reported on humeral head augmentation procedures in seizure-related shoulder instability. Thangarajah et al[14] reported outcomes on 33 patients with 49 unstable shoulders related to seizure activity. Surgical intervention was performed in 31 patients and included isolated soft tissue repair, glenoid bone augmentation, and isolated humeral head allograft augmentation. Twenty-seven patients experienced recurrent seizures after surgical intervention. Within their cohort, 24 patients and 36 shoulders experienced isolated anterior instability. Bone loss was noted in 86% of anterior instability cases, with 28 shoulders having a Hill-Sachs lesion, of which 21 were described as large and 7 as small. Hill-Sachs lesions were described as small if less than 20% of the humeral head and large if greater than 20%. Recurrent instability was noted in 61% of patients who underwent procedures for anterior instability. The authors found that isolated soft tissue repair was associated with higher rates of failure than bone augmentation procedures. Two patients underwent isolated allograft augmentation of the humeral head defect, and recurrent instability occurred in one of these patients.

In a review of glenoid-based procedures in seizure-related shoulder instability, Hutchinson et al[9] reported outcomes of 13 patients and 15 shoulders with recurrent anterior shoulder instability related to grand mal epilepsy. All patients underwent glenoid bone buttress augmentation with either iliac crest or femoral head allograft or autograft. At a mean follow-up of 2.7 years, no recurrent instability events were reported despite eight patients experiencing recurrent seizure activity postoperatively. Comparable studies have not been able to reproduce these results.

Buhler et al[2] reported on 34 shoulders, 17 with anterior and 17 with posterior instability related to seizure. Of the patients with anterior shoulder instability, 2 were treated nonoperatively, 6 with isolated soft tissue repair, 7 with humeral head derotational osteotomy, and 7 with glenoid bone block augmentation. No humeral head augmentation procedures were performed despite humeral bone defects being common. They noted 12 large and 4 small Hill-Sachs lesions and 5 large and 10 small glenoid lesions. Recurrent shoulder instability occurred in 47% of patients after primary surgery with 5 due to recurrent seizures. Reoperation was required in 6 shoulders.

Raiss et al[11] reported outcomes of the Laterjet procedure in 12 patients with 14 anterior dislocations due to seizure with 1-year minimum follow-up and a mean of 8.3 years. All patients had glenoid rim and Hill-Sachs defects; however, the Hill-Sachs lesion was not addressed in this group. Eight patients continued to have seizures, and 6 experienced recurrent dislocations. The authors stated "an associated allograft reconstruction of the humeral head might decrease the rate of redislocation".

Most recently, Ersen et al[5] reported outcomes of the Laterjet procedure in patients with epilepsy. Their cohort consisted of 9 patients and 11 shoulders with recurrent anterior instability due to seizure with a minimum of 2-year follow-up. All patients underwent Laterjet procedures with no humeral head augmentation procedures. Three patients sustained recurrent seizures, and one shoulder experienced a recurrent shoulder dislocation.

To date, the few studies specific to surgical outcomes after seizure-induced anterior instability have focused on glenoid-based procedures despite the presence of humeral head bone loss. With this approach, recurrent instability rates range from 0% to 61%. Hutchinson et al initially reported on his glenoid bone augmentation and reported no recurrent instability. These results have not been duplicated. Other authors report much higher rates of recurrent instability after glenoid-based procedures.[2,5,11,14] The authors stress the importance of medical co-management to reduce frequency of postoperative seizures. Ongoing seizure activity presents a unique challenge that will continue to stress surgical repair. Within the available literature, recurrent seizure activity occurred at rates of 33% to 82%.[2,5,10,11,14] In addition, not all postoperative instability events were related to seizure activity. In our patient population, all subjects experienced postoperative seizures, creating a uniquely challenging cohort. In addition to medical co-management, one author suggested the importance of adding a humeral head augmentation procedure to lower instability recurrence. In the present study, large Hill-Sachs lesions were found in all patients, whereas large glenoid-based lesions were less common. This variance is consistent with the abovementioned studies that reported on glenoid defect size. All patients underwent surgical intervention with humeral head allograft augmentation in addition to a glenoid procedure. Glenoid procedures varied from capsulolabral repair to a Laterjet. One patient underwent isolated humeral head augmentation without associated glenoid surgery. This was secondary to advanced arthritic changes of the

glenohumeral joint. With this approach, one patient experienced postoperative instability. The one reported redislocation in our cohort did not have a previous shoulder surgery and underwent three-anchor capsulolabral repair with humeral head augmentation. Although the patient did experience recurrent seizure activity postoperatively, the mechanism of recurrent instability was not related to seizure activity. The patient sustained a traumatic injury after a fall while fishing.

Although definitive conclusions cannot be drawn based on the present study, our results hint at the importance of appropriate management of bony defects, including humeral head allograft augmentation. Available literature of glenoid-based procedures reports failure rates as high as 61% percent. Patients with active seizure disorder present a truly challenging cohort as recurrent seizures will continuously stress any surgical repair, especially isolated soft-tissue repairs. In addition, the unique pathophysiology of seizure-induced shoulder instability creates other challenges that limit surgical options. Coracoid fractures can occur after seizure-induced instability events, which may limit availability for transfer. In addition, powerful muscle contracture during seizure events will continuously stress a transferred coracoid process if the muscular attachments are still preserved. Given this, we suggest consideration of humeral head allograft augmentation in patients with recurrent, seizure-induced shoulder instability with large Hill-Sachs lesions.

Several limitations should be noted. First, this is a relatively small cohort. The study is retrospective in nature, which imparts variables and bias that are difficult to control. Although all patients underwent humeral head allograft procedures, glenoid procedures varied which prevents standardization. As a case series, we lack comparison against a control group. Variation in preoperative imaging prevents defect track calculations. In addition, preoperative baseline scores were not collected, and clinical assessment data are not available as the final follow-up was conducted by telephone for some patients. These limitations prevent definitive conclusions about the optimum treatment for these challenging patients. However, our results add to the overall low number of outcome studies specific to this cohort and suggest the importance of addressing bipolar lesions.

Conclusion

Shoulder dislocations associated with recurrent motor seizures are particularly challenging to manage. Successful management is often achieved via multispecialty co-management, including medical and surgical intervention. In this retrospective study, humeral head allograft bone grafting with an appropriate glenoid-based procedure was able to minimize seizure-related recurrent dislocations in a patient population with poor seizure control. Patients also exhibited favorable outcomes with regard to disability, pain, function, strength, and range of motion, while preserving their native anatomy.

Disclaimers:

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