Open inferior capsular shift for multidirectional shoulder instability in adolescents with generalized ligamentous hyperlaxity or Ehlers-Danlos syndrome

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\textbf{Background:} The objective of this study was to assess the outcome of open inferior capsular shift for multidirectional shoulder instability in patients with generalized ligamentous hyperlaxity or Ehlers-Danlos syndrome.

\textbf{Methods:} Data were obtained for 18 open inferior capsular shift surgeries in 15 adolescent patients with generalized ligamentous hyperlaxity or Ehlers-Danlos syndrome with a mean follow-up of 7.5 years. End points were subjective clinical outcome (pain, stability, satisfaction, return to sport), objective clinical outcome (recurrence, complications), and functional outcome scores (American Shoulder and Elbow Surgeons, 11-item version of the Disabilities of Arm, Shoulder and Hand).

\textbf{Results:} Thirteen patients (87\%) reported improved pain and stability and were satisfied with the procedure. Nine patients (64\%) were able to return to sports. One patient (7\%) was dissatisfied with continuous pain and recurrent instability and considered a surgical failure. Seven patients (47\%) reported no further episodes of instability. The mean American Shoulder and Elbow Surgeons score at a mean of 7.5 years of follow-up was 88 ± 10 points, and the mean score for the 11-item version of the Disabilities of Arm, Shoulder and Hand was 14 ± 14 points.

\textbf{Discussion:} The management of multidirectional shoulder instability in adolescent patients with generalized ligamentous hyperlaxity or Ehlers-Danlos syndrome is challenging. Open inferior capsular shift results in improvement in subjective and objective shoulder function and stability in adolescent patients with ligamentous hyperlaxity or Ehlers-Danlos who have failed nonoperative treatment. We found no effect of the recalled number of prior dislocations, laterality, and type of hyperlaxity on subjective and objective clinical outcomes.

\textbf{Level of evidence:} Level IV; Case Series; Treatment Study

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\textbf{Keywords:} MDI; shoulder instability; pediatric; hyperlaxity; Ehlers-Danlos syndrome; open shift
Hyperlaxity and resultant joint hypermobility is a fairly frequent finding in adolescent patients. Current estimates are that between 4% and 13% of the general pediatric population exhibit some degree of joint hypermobility, albeit most not in association with a defined soft-tissue disease. The prevalence of this condition is somewhat higher in adolescent athletes because individuals with hyperlaxity succeed in gymnastics, throwing, racquet sports, and swimming as a result of increased flexibility and range of motion.

Ligamentous hyperlaxity and shoulder instability are distinct entities and can occur independently from each other. Most affected athletes embrace their joint hypermobility and are able to maintain shoulder stability through dynamic compensation. Nevertheless, these patients can suffer traumatic shoulder dislocations, with typical structural damage resulting in unidirectional instability. More often, however, these patients experience recurrent subluxations and subsequent gradual lengthening of the static restraints of the shoulder that will lead to symptomatic multidirectional instability (MDI), that is, atraumatic shoulder instability in multiple directions.

The mainstay of treatment for MDI in the hyperlax adolescent patient is nonoperative treatment with physical therapy and activity modification. In addition, laxity typically diminishes with age, and instability symptoms may improve as patients reach skeletal maturity. Therefore, conservative treatment aimed at muscular control and a temporizing effect can be successful in these patients. When conscientious nonoperative treatment fails, surgical stabilization may be required. Arthroscopic capsular plication and open capsulorrhaphy have both been described for management of MDI.

The literature regarding surgical treatment for MDI in adolescent patients with hyperlaxity or Ehlers-Danlos syndrome (EDS) is limited and mostly extrapolated from data on MDI without hyperlaxity. The objective of this study was to assess the outcome of open inferior capsular shifts for multidirectional shoulder instability in adolescent patients with generalized ligamentous hyperlaxity or EDS.

### Methods

Patients surgically treated for multidirectional shoulder instability at a tertiary care pediatric hospital were identified from a computerized database. Patients were included if they had undergone open inferior capsular shift for atraumatic shoulder instability with a positive diagnosis of generalized ligamentous hyperlaxity and had at least 24 months of follow-up. All patients had failed nonoperative treatment consisting of physical therapy and activity modification for a minimum of 6 months for AMBRI-type instability (atraumatic, multidirectional, frequently bilateral, initially treated with rehabilitation protocol, inferior capsular shift recommended if surgery is required). All patients exhibited instability in at least anterior and inferior directions with positive load-and-shift as well as sulcus sign testing. For the purpose of this study we defined hyperlaxity as clinical or genetic diagnosis of a connective tissue disease, such as EDS or Marfan syndrome, or a Beighton Score >6 points. We excluded patients who had language, psychiatric, or cognitive difficulties that prevented reliable completion of the questionnaire and patients with multiple medical comorbidities or neurologic conditions that prevented them from participating in sports or other physical activities.

Open inferior capsular shift was performed as described by Neer and Foster. All surgeries were performed by the senior author (M.S.K.) with the patient in the beach chair position under general anesthesia with an interscalene block. An anterior, deltopectoral approach was performed through a limited vertical incision in the anterior axillary crease. The subscapularis was dissected from the capsule and split vertically. A laterally based T-capsulotomy was performed. A humeral head retractor was used, and the humeral head, glenoid, and labrum were inspected. No labral tears were noted in this group, and therefore, no labral repair was performed.

The inferior capsular flap was mobilized inferiorly to approximately the 6 o’clock position. In 30° of abduction and external rotation, the inferior flap was mobilized superiorly and sutured to the superior part of the lateral capsular remnant with #1 Ethibond (Ethicon, Somerville, NJ, USA) sutures. This obliterated the redundant axillary pouch. The superior flap was mobilized distally to the inferior leaf of the lateral capsular remnant and sutured with #1 Ethibond suture as well. The subscapularis was repaired end to end. The wound was irrigated and closed in layers.

All patients followed a standardized postoperative rehabilitation regimen consisting of sling use and pendulums for 4 weeks, followed by discontinuation of the sling and active range of motion exercise at 4 weeks, strengthening at 6 weeks, and return to sports at 4 months.

Data collected included demographic information, such as gender, age, and laterality, and disease-specific information. Patients were contacted directly to obtain further data. Outcome data were collected for the following end points: subjective clinical outcome, objective clinical outcome, and clinical scores.

The subjective outcomes were perceived improvement of pain and stability, subjective satisfaction with shoulder performance postoperatively as ordinal variables (much better, better, same, worse, or much worse), and as return to sport (same level or higher level, lower level, not able to return).

For objective clinical outcomes, we collected data on recurrence of instability (dislocations) and subluxations as well as perioperative complications. We also assessed subscapularis function and degenerative changes at the last follow-up. Subscapularis function was tested by belly press and lift-off testing. Degenerative changes were assessed on anteroposterior radiographs using the Samilson-Prieto classification, which has been shown to be the most reliable classification for postoperative arthritis in shoulder instability.

Clinical outcome scores used for this study were the American Shoulder and Elbow Surgeons (ASES) score, and the 11-item version of the Disabilities of Arm, Shoulder and Hand (QuickDASH). The ASES is a validated functional
shoulder outcome score with a defined minimal clinically important difference of 6.4 points. The QuickDASH is a validated self-administered outcome measure for upper limb function.

We were interested in 3 covariables that are proven risk factors for surgical failure and poor long-term outcome in shoulder stabilization:

First, we wanted to assess the effect of the recalled number of prior dislocations on surgical outcome. Because many hyperlaxity and EDS patients will stop counting their dislocations once they go into the higher double-digit numbers, this should be considered a trend rather than a very precise assessment numerically.

Second, we tested for differences in outcome between those patients with a genetic diagnosis of EDS vs those diagnosed as hyperlax without a known genetic disorder.

Last, we analyzed whether surgical outcome was affected by laterality (ie, surgery on the dominant, nondominant, or both shoulders) or rotator interval closure.

Statistical analysis included descriptive statistics for patient characteristics and surgical outcomes. Multivariate regression analysis was used to assess the effect of the 4 covariates. “Prior dislocations” was used as a continuous variable, “EDS” and “rotator interval closure” as binary variables, and “laterality” as a categoric variable. The ordinal outcomes of pain, stability, satisfaction, and return to sport were assessed in ordinal logistic regression, and the continuous outcomes recurrences, ASES, and QuickDASH in linear regression analysis. All calculations were done using Stata 10 software (StataCorp LP, College Station, TX, USA). A P value of .05 was considered significant. Results for continuous variables are given as means ± standard deviation.

Results

We treated 18 shoulders in 15 patients (4 males and 11 females). Treatment was unilateral in 12 patients (1 left, 11 right) and bilateral in 3. The operation in 13 patients (87%) was on the dominant side. All patients exhibited clinical criteria of hyperlaxity (Beighton Score >6 points) and multidirectional shoulder instability. Genetic testing for EDS was positive in 5 patients (36%).

Nine patients (60%) also had surgery on other joints based on an EDS-related diagnoses, including 7 patella stabilizations, 4 hip arthroscopies, and 1 peroneal tendon stabilization. Three patients (20%) underwent interventions by general surgery, including 1 gastrointestinal/vascular procedure and 2 gastrointestinal/gynecologic procedures. The average age at the time of the capsular shift was 17 ± 2.9 years. The mean follow-up after shoulder stabilization was 7.5 ± 3.5 years, with a minimum follow-up of 32 months. Patient characteristics are reported in Table I.

In terms of subjective outcome, 6 patients (40%) graded their postoperative pain as much better and 7 (47%) as better. One patient (6%) did not observe a change in pain level, and 1 patient (6%) reported a deterioration of pain. Eleven patients (73%) felt that their shoulders were much more stable after the surgery, 2 patients (13%) reported improved stability, and 2 (13%) reported no change in shoulder stability. Nine patients (60%) were very satisfied, 4 (27%) were satisfied with their shoulder performance, and 1 patient (6%) was dissatisfied with the unilateral stabilization of his dominant right shoulder. All patients undergoing bilateral procedures reported improved pain and shoulder stability and were satisfied with the outcome.

Nine patients (60%) returned to sports: 5 (36%) at the same level and 4 (28%) returned to sports at a lower level than before the procedure. Four patients (28%) were not able to return to sports, and 2 patients (14%) did not respond. The mean time until return to sports was 7.0 ± 3.3 months. Of the 3 patients treated bilaterally, 2 returned to sports at the same level as before the procedure; however, 1 patient was not able to do so.

As for the objective clinical outcomes, 7 patients (47%) had no recurrence of instability or dislocation after the procedure, and 7 (47%) experienced recurrent subluxation. Only 1 patient (7%) reported an increase in instability after the procedure. One perioperative complication occurred in the form of reflex sympathetic dystrophy, which resolved with physical therapy and pain clinic management. No patient was found to have subscapularis dysfunction at final follow-up. Five patients underwent postoperative magnetic resonance imaging (MRI) after a new injury, and all 5 showed intact subscapularis tendon on their report by a board-certified, musculoskeletal radiologist. Postoperative imaging for arthritis was available for 6 patients (40%), and none showed signs of arthropathy as defined by the Samilson classification.

In terms of functional outcome scores, the mean postoperative ASES score for the treated shoulders was 87.7 ± 9.4 points. The mean QuickDASH score was 13.9 ± 13.7 points. There was no statistical difference in ASES scores (P = .731) or QuickDASH (P = .707) between unilateral and bilateral stabilization.

Table I Patient characteristics

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>Result</th>
</tr>
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<tbody>
<tr>
<td>Patients, No.</td>
<td>15</td>
</tr>
<tr>
<td>Shoulders, No.</td>
<td>18</td>
</tr>
<tr>
<td>Age, mean (SD) y</td>
<td>17 (3)</td>
</tr>
<tr>
<td>Females, No. (%)</td>
<td>11 (73)</td>
</tr>
<tr>
<td>Follow-up, mean (SD) y</td>
<td>7.5 (3.5)</td>
</tr>
<tr>
<td>Shoulder treated</td>
<td></td>
</tr>
<tr>
<td>Dominant, No. (%)</td>
<td>10 (67)</td>
</tr>
<tr>
<td>Nondominant, No. (%)</td>
<td>2 (13)</td>
</tr>
<tr>
<td>Bilateral, No. (%)</td>
<td>3 (20)</td>
</tr>
<tr>
<td>Recalled prior dislocations, median (range) No.</td>
<td>30 (1-180)</td>
</tr>
<tr>
<td>Genetic diagnosis of Ehlers-Danlos syndrome, No. (%)</td>
<td>5 (33)</td>
</tr>
</tbody>
</table>

SD, standard deviation.
Regression analyses showed that the recalled number of dislocations before surgery, the genetic diagnosis of EDS, and laterality had no influence on subjective and objective clinical outcomes (Table II). The median recalled number of prior dislocations was 30 (range, 1-180). For clinical scores, however, we did see a significant negative effect of a genetic diagnosis of EDS on ASES and QuickDASH scoring compared with those patients deemed hyperlax without a diagnosis of EDS. There was no association between recurrence and a diagnosis of EDS ($P = .178$) or the recalled number of prior dislocations ($P = .341$). Interval closure did not affect postoperative pain ($P = .621$), stability ($P = .202$), subjective satisfaction ($P = .417$), return to sports ($P = .753$), or recurrence ($P = .237$). Rotator interval closure did not show an association with ASES ($P = .706$) or QuickDASH ($P = .605$) scores.

**Discussion**

The management of MDI in adolescent patients with generalized ligamentous hyperlaxity or EDS is challenging. Nonoperative treatment is emphasized; however, surgical treatment may be necessary in those patients who fail nonoperative treatment with symptomatic instability and functional limitations. Surgical treatment has traditionally been thought to have poor outcomes in adolescents with EDS or generalized ligamentous hyperlaxity. Our data show that open inferior capsular shift for MDI in adolescent patients with generalized ligamentous hyperlaxity or EDS results in a clinically relevant reduction of symptoms of pain and instability, reduction in instability episodes, good patient satisfaction, and good functional outcome scores at a mean follow-up of 7.5 years.

Although the data for surgical treatment of MDI in adolescent patients, be it with or without hyperlaxity, is sparse, there is good evidence in support of open and arthroscopic treatment of MDI in adults. The open inferior capsular shift, as reported by Neer and Foster in 1980, has been used, with consistent success rates exceeding 85% in a number of studies since. More recently, arthroscopic capsular plication has gained popularity and has demonstrated good results if capsular laxity and redundancy are adequately addressed. Open inferior capsular shift was used in the patients in this study because of the senior author’s preference for open capsulorrhaphy in patients with generalized ligamentous laxity or EDS. In these patients, arthroscopic capsular plication may not adequately control instability given the extensive capsular laxity. Cohen et al were able to show that open capsular shifts will reduce joint volume by approximately 50%, compared with only 23% in arthroscopic plication consisting of three 1-cm anteroinferior tucks. Karas et al found a 19% reduction of joint volume with four 1-cm tucks.

For an athletic population, return to sports in a timely manner and at an adequate level can be a more meaningful outcome than subjective shoulder stability and reduced pain postoperatively. Hyperlax athletes may succeed in their chosen sport because of an advantageously increased range of motion or level of flexibility. In this study, roughly one-third of all patients returned to sports at a level similar than before surgery, one-third returned to a level lower than before, and one-third did not return to sports. The mean duration until return to sports was 6 months. Return to sports after surgical treatment of patients with MDI is challenging. In a recent study, Ma et al reported the surgical stabilization of 23 patients with MDI, 18 of whom were not able to return to sports.

Patients in this study reported good functional outcome after surgery. At a mean follow-up of 7.5 years after surgery, our patients reported mean ASES scoring of 88.4 points. Ozturk et al in 2013, reported their results for a group of patients younger than 25 years (mean, 19.5 years) undergoing anterior stabilization and reported mean ASES scores of 83.2 points at 2.25 years of follow-up.

We consider postoperative rehabilitation a crucial factor in functional outcome. Hence, we focus our rehabilitation protocol, after a period of immobilization, on proprioceptive exercises and strengthening of the kinetic chain.

**Table II** $P$ values from the regression analysis of covariates

<table>
<thead>
<tr>
<th>Covariates</th>
<th>Recalled number of dislocations before surgery</th>
<th>Genetic diagnosis of Ehlers-Danlos syndrome</th>
<th>Laterality of surgery (dominant, nondominant, bilateral)</th>
<th>Rotator interval closure performed</th>
</tr>
</thead>
<tbody>
<tr>
<td>$P$ value</td>
<td>$P$ value</td>
<td>$P$ value</td>
<td>$P$ value</td>
<td>$P$ value</td>
</tr>
<tr>
<td>Pain after surgery</td>
<td>.076</td>
<td>.278</td>
<td>.833</td>
<td>.278</td>
</tr>
<tr>
<td>Stability after surgery</td>
<td>.149</td>
<td>.205</td>
<td>.586</td>
<td>.205</td>
</tr>
<tr>
<td>Satisfaction with surgery</td>
<td>.200</td>
<td>.635</td>
<td>.698</td>
<td>.635</td>
</tr>
<tr>
<td>Return to sports</td>
<td>.483</td>
<td>.154</td>
<td>.092</td>
<td>.154</td>
</tr>
<tr>
<td>Recurrent dislocations</td>
<td>.682</td>
<td>.344</td>
<td>.902</td>
<td>.344</td>
</tr>
<tr>
<td>ASES score</td>
<td>.606</td>
<td>.027*</td>
<td>.174</td>
<td>.027*</td>
</tr>
<tr>
<td>QuickDASH score</td>
<td>.875</td>
<td>.044*</td>
<td>.133</td>
<td>.044*</td>
</tr>
</tbody>
</table>

ASES, American Shoulder and Elbow Surgeons; QuickDASH, 11-item version of the Disabilities of Arm, Shoulder and Hand.

* Statistically significant.
core-scapulothoracic joint-glenohumeral joint. We also prefer active and active-resisted range of motion over passive range of motion or stretching exercises to avoid decoupling of static and dynamic stabilizers.

Half of our patients reported some recurrence of instability while at the same time reporting satisfaction with the procedure. Early studies of shoulder stabilizations reported but did not specifically analyze subgroups of patients with hyperlaxity. What is known from this prior research is that hyperlax patients have a higher risk of recurrent instability; however, this did not seem to influence clinical scoring and subjective satisfaction. In fact, we consider one of the most important findings of this study is that in this patient population, subjective satisfaction is high despite low objective stability. This point proves that classic definition of stability is unreliable in hyperlax patients because these patients function at a level of laxity that would be pathologic in a soft tissue–competent person.

Blackman et al reviewed 90 adolescent patients requiring revision anterior stabilization and found a 17% failure rate for primary stabilization in this group (18% for arthroscopic and 11% for open stabilization). The mean interval between the primary procedure and revision surgery was 17.1 months. At approximately 5-times that follow-up, we did see recurrent instability, but no revision surgery was required.

The only perioperative complication in this study was 1 patient with reflex sympathetic dystrophy. In a study of 64 patients undergoing an open Bankart procedure, Boileau et al found reflex sympathetic dystrophy in 8 patients (13%), necessitating extended physical therapy beyond 6 months.

Subscapularis recovery after shoulder stabilization is a pivotal factor in surgical outcome. Slabaugh et al studied return of subscapularis function after open shifts in 16 patients and found full return of function by 8.8 weeks. In our group, we found good subscapularis function in clinical assessment. Five of our patients (33%) underwent MRI after a new trauma in the postoperative period, a time where we use MRI with a very low threshold. For all 5, a musculo-skeletal radiologist read the subscapularis tendon intact. Interestingly, 3 of these patients showed posterior labral fraying on MRI. The question from these findings that stands out is whether this is a result of their years of instability or a consequence of anterior overtightening.

We found no evidence for postoperative arthritis in our population by assessment of radiographs. A prior study found 17% of patients had early arthritic changes (Samilson stages I-III) at a median follow-up of 40 months, but in an older population with a mean age of 27 years of age. Plath et al showed some arthritic changes had developed in 69% of their patients 13 years after arthroscopic Bankart repair. Our data are limited by the fact that the return rate for radiographs was only 40%. However, given the young age of our patients, mostly young women in reproductive age, radiation is often an issue. Our population might progress down a similar path with longer follow-up. However, arthritic changes might manifest at a lower rate in the hyperlax patient because dislocations and subluxations occur less forcefully.

As for confounders and covariates for surgical outcome, we were surprised to see that the number of prior dislocations did not affect any of the outcome parameters, even if a meaningful recall bias is considered. However, an argument could be made that whatever “critical defect” it is that causes instability in this patient population, its nature and extent will not change considerably after the first few dislocations. Our population recalled a median of 30 prior dislocations. Interestingly, the genetic diagnosis of EDS was a negative factor on clinical scoring, albeit not for subjective satisfactions. This fact may speak to both the enduring mindset of these patients as well as a true, biologic difference between EDS and hyperlaxity.

Our study has some limitations. First, it is retrospective and cross-sectional in nature; thus, we cannot make recommendations for choosing treatment. Second, our study lacks a formal control group; however, our intention was not to compare treatments but to longitudinally describe the midterm outcome of inferior capsular shifts in patients with MDI and hyperlaxity. Last, our sample size is small, but the highly specific nature of our population put constraints on sample size.

### Conclusion

Our data show that inferior capsular shifts will produce predictable improvement in subjective and objective shoulder function and stability in adolescent patients with ligamentous hyperlaxity or EDS during a 7.5-year follow-up. These outcomes and recurrence rates are in line with the literature for patients with normal soft-tissue competence. We found no effect of the recalled number of prior dislocations, laterality, or type of hyperlaxity on subjective and objective clinical outcomes.

### Disclaimer

The authors, their immediate families, and any research foundation with which they are affiliated did not receive any financial payments or other benefits from any commercial entity related to the subject of this article.

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