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

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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.
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).
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.
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.
Level of evidence: Level IV; Case Series; Treatment Study

Keywords: MDI; shoulder instability; pediatric; hyperlaxity; Ehlers-Danlos syndrome; open shift

The Institutional Review Board at Boston Children's Hospital approved this study (IRB Protocol Number P00008337).

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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.[2] 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.[11,21]

Ligamentous hyperlaxity and shoulder instability are distinct entities and can occur independently from each other.[12] Most affected athletes embrace their joint hypermobility and are able to maintain shoulder stability through dynamic compensation.[29] 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.[12,30]

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.[5,12] Therefore, conservative treatment aimed at muscular control and a temporizing effect can be successful in these patients.[7,12] When conscientious nonoperative treatment fails, surgical stabilization may be required. Arthroscopic capsular plication and open capsulorrhaphy have both been described for management of MDI.[9]

The literature regarding surgical treatment for MDI in adolescent patients with hyperlaxity or Ehlers-Danlos syndrome (EDS) is limited and is mostly extrapolated from data on MDI without hyperlaxity.[12] 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 (*a*traumatic, *m*ultidirectional, frequently *b*ilateral, initially treated with *r*ehabilitation protocol, *i*nferior capsular shift recommended if surgery is required).[15] 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.[1] 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.[18] 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.[28] 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.[6]

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