Should shoulder instability be operated in children?

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Introduction

The glenohumeral joint is an enarthrosis with 3 degrees of freedom and is especially unstable due to the shallow nature of the glenoid. The ossification centers of the proximal humerus (i.e., humeral head, greater trochanter, and lesser trochanter) fuse around the ages of 4-6 years to form the epiphyses of the proximal humerus. They fuse with the metaphysis of the humerus around age 18 years in girls and 19 years in boys. The physis at the level of the glenoid fuses around the age of 19 years. This literature review will be limited to patients in whom the glenohumeral growth plates are still open.

Pediatric shoulder instability is a rare pathology [1,2]. Between 2010 and 2014, an Italian national study reported an average incidence of glenohumeral dislocation in children younger than 14 years old of 0.3/100,000 individuals [1]. The main issue is etiological. Contrary to adults, instability is rarely due to trauma and is generally non-traumatic.

The multiple studies published on pediatric shoulder instability are difficult to generalize for clinical practice due to their lack of distinction between children (i.e., <10 years old), adolescents with and without an open physis, and the young adult.

However, functional conservative treatment is always indicated as a first line treatment, regardless of the etiology. There is a lack of consensus regarding indications for stabilizing surgery in chronic glenohumeral instability in children and adolescents. After having detailed the anatomopathological specificities of pediatric glenohumeral instability and exposed the diagnostic elements of the etiological assessment, we will conduct a literature review of the different arguments in favor or against stabilizating surgery.

Anatomopathological notions specific to the pediatric shoulder

Static elements of glenohumeral stability

The static elements are the glenoid, labrum, capsule, coracohumeral ligament, and superior, middle, and inferior glenohumeral ligaments. Fetal cadaveric studies [3] have shown that the morphology of the shoulder is similar to the postnatal period. The glenohumeral and coracohumeral ligaments are present at a very early stage of fetal life.

The analysis of second-trimester specimens shows a possible immature capsule at the level of the rotator interval. Biomechanically, a notable capsular defect in this area seems to be related to excessive inferior glenohumeral laxity while in abduction [4]. Furthermore, in utero capsulo-ligamentous collagen fibers are irregularly organized [3]. In the postnatal period, constraints from constant shoulder use induces organization the collagen fibers. During growth, the collagen content of the capsulo-ligamentous structures varies. Elastic type III collagen is predominant at birth before being replaced during development by stronger, type I collage [5]. In fact, patients conserving a higher proportion of type III collagen are at increased risk of recurrent instability without major trauma [6].

Anthropometric data showed an evolution of glenoid version. In its early form, the glenoid is comprised of two ossification centers. The superior center appears around age 10 and fuses around age 16. The inferior center appears around puberty and constitutes 70% of the inferior portion of the glenoid and forms its concavity. A defect in this ossification center may lead to posteroinferior hypoplasia, commonly referred to as glenoid dysplasia. Excessive glenoid retroversion is a risk factor for recurrent posterior instability. A low retroversion of the proximal humerus could itself be a potential cause of anterior instability.

The presence of a fine inferior capsular recess that is attached to a hypotrophic labrum in patients with excessive joint laxity [7] may increase the risk of uni-, bi- or multidirectional instability. The hammocking effect of the inferior glenohumeral ligament can also be seen during shoulder arthroscopy. The anterior portion extends in a fanlike pattern and the posterior bands are shaped of a cord in external rotation, or inversely into internal rotation. In young patients with uni- or multidirectional instability [7] there is often a defect in this ligament.

Dynamic elements of glenohumeral stability

Dynamic elements include the rotator cuff, stabilizing muscles of the scapula, and the deltoid. Alterations in muscle balance by hypo- or hyperactivity could lead to glenoid and proximal humeral dysplasia by mechanisms similar to the hip. These muscular and bony disorders lead to shoulder instability, ranging from subluxation to inveterate dislocations.

Clinical definition of glenohumeral instability

An episode of instability is defined as excessive translation of two joint surfaces leading to a loss of contact between these surfaces.

Caracteristics of instability

- **The direction:** anterior, posterior, or inferior. It is unidirectional in most cases, but certain instabilities are multidirectional.
- **The frequency:** unique, recurrent, or inveterate (diagnosed after 3 weeks).
- **The type**
    - Subluxation: A partial and/or transitory loss of contact between articular surfaces that spontaneously resolves without the help of a third party.
    - Dislocation: A complete loss of contact between articular surfaces requiring a maneuver for reduction by a third party.
- **The unilateral or bilateral**
- **Voluntary or involuntary:** Voluntary instability is defined as controlled and painless recurrent subluxations, without apprehension, and without radiographic anomalies, whereas involuntary instability is characterized by episodes of uncontrolled recurrent subluxations [8].
- **Etiology**

Traumatic
Neuromuscular: - **Peripheral involvement:** Sequelae of neonatal or traumatic brachial plexus palsy, transverse myelitis, Parsonage-Turner syndrome. - **Central involvement:** traumatic or vascular sequalae, fetal distress; dystonia.
Congenital bony malformations - **Glenoid dysplasia:** idiopathic, constitutional bone disease, mucopolysaccharidosis - **Congenital anomalies of the shoulder girdle**
Connective tissue anomalies - **Syndromic:** joint hypermobility syndromes, Ehlers-Danlos syndrome - **Congenital**
Psychological disturbances

The search for an etiology

Children with shoulder instability generally undergo multiple examinations and/or reduction maneuvers. As a result, they have severe apprehension and wariness from the medical community. Patients should be examined in a calm and familial environment.

- Clinical exam:

Central or peripheral neurological involvement must be ruled out. A sensory and motor exam of the upper limb should rule out possible injury of the axillary nerve.
Cutaneous fragility or hyper elasticity, a deformity of the trunk or the extremities, an instability or excessive laxity of another joint should also be assessed. A Beighton score ≥ 6-7 points is in favor of connective tissue disease. Physical exam should assess anterior and posterior constitutional laxity (in the seated position with elbow to the trunk according to Rockwood, bending forward according to Rodineau, or supine with the arm in abduction according to

Gerber), inferior constitutional laxity (Neer's sulcus test) and especially pathological laxity of the inferior glenohumeral ligament (Gagey's or Boileau's tests). Multidirectional constitutional hyperlaxity is a risk factor for episodic instability and is assessed by >85° of passive shoulder external rotation with the upper arm in a neutral position at the patient's side and the elbow in 90 degrees of flexion [8].

Assessment for an underlying psychological derangement is primordial in patients with voluntary instability. Intervention by a psychologist or a social worker could prove useful in such cases.

- Imaging

Required imaging studies include three coronal (neutral, internal rotation, and external rotation views), a Lamy sagittal (45° false profile view), and an axillary view. The axillary view visualizes a well-centered humeral head over the glenoid.

Instability can be accompanied by a labral tear (i.e., Bankart lesion), periosteal-labral sleeve, or, rarely, a superior or inferior glenohumeral ligamentous injury. Bone loss or a glenoid or humeral deformity (e.g., bony Bankart lesions, Hill-Sachs fracture in anterior dislocations, or a McLaughlin fracture in posterior dislocations) can often be identified on radiographs or on an MRI, but sometimes require a CT-scan [7]. Recent studies have shown that magnetic resonance imaging can evaluate glenoid bone loss with a precision similar to CT-scans [9]. An Arthro-MRI is the preferred imaging modality in children outside of the acute phase, evaluating cartilaginous structures, labral tears, and the capsule.

A medullary MRI, especially of the cervical spine, aids in the assessment of a plexus or medullary injury (e.g., myelitis).

- Electromyogram

An electromyogram may be used to rule out a Parsonage-Turner syndrome and to determine the location of a peripheral neurological injury.

What are the treatment options in pediatric shoulder instability?

post-traumatic recurrent shoulder instability

The primary risk, after a first episode of traumatic instability, is recurrence. The recurrence rate in children varies from 21 to 100% based on the child's age, with a higher risk in adolescents [10-12].

Different studies use different definitions, and some have even included subluxations and dislocations within the same group, thus rendering the interpretation of the literature difficult. As a result, indications for surgical treatment remain controversial. Analyses of the effectiveness of conservative treatment and MRI screening of a structural injury are an essential part of preoperative planning.

Postacchini et al. published a study where an MRI was obtained 7 years after a first episode of shoulder dislocation in 28 adolescents aged 12 to 17 years old. In adolescents who were aged 14 to 17 years old at the time of dislocation, the rate of recurrence was of 92%. In adolescents with recurrence, the authors observed a Bankart lesion on imaging, which required surgical intervention. However, in patients younger than 13 years old, the recurrence rate was 33% and no Bankart lesion was found on MRI [13]. The hypothesis upheld by some authors is that prepubertal patients have higher capsuloligamentous elasticity and are thus at a lower risk of developing capsulo-labral injuries [14]. The ratio of type III to type I collagen is higher in this group compared to post-pubertal patients [6].

Some authors have suggested surgical treatment after a first episode of traumatic instability in adolescents. This approach may be justified if patients and their families are unwilling to modify the child's sporting activities [7]. Joes et al. [15] reported the results of 32 anterior arthroscopic repairs based on the Bankart technique in 30 adolescents with an average age of 15 years. Half of the patients presented recurrent instability despite rehabilitation, while the other half underwent primary surgical stabilization. At 2 years follow-up, in the rehabilitation group, the recurrence rate was of 19% compared to 12.5% in the primary surgery group. Kraus et al. [16] reported on a series of 6 patients with an average age of 12 years, of which 5 patients had been treated by arthroscopic capsuloligamentous stabilization. No recurrence was noted after an average follow-up of 26 months. Risk factors for treatment failure by soft tissue stabilization procedures included Hill-Sachs lesions and a preexisting glenoid bone loss [16]. In such cases, a Latarjet procedure and/or a Hill-Sachs "remplissage" could be considered (Figure 1). Khan et al. published a case series in skeletally immature adolescents who had had at least one episode of anterior dislocation. Twenty-three patients had been treated by conservative treatment and 26 patients had been operated with a Latarjet procedure. The rate of recurrence was of 56% in the conservative group compared to 7% after surgical stabilization [17]. No differences were found in terms of functional scores or pain. Post-traumatic posterior instability in skeletally immature patients is rare and its treatment is controversial, similar to adults.

Non-traumatic recurrent shoulder instability

This type of instability is most often bilateral. These cases are usually unidirectional and are associated with a constitutional hyperlaxity of the shoulder [18]. In rare cases, instability may be multidirectional and associated with hyperlaxity. Voluntary and involuntary forms should be distinguished since outcomes differ. Contrary to traumatic instability in children, which is usually anterior, non-traumatic instability could be either anterior or posterior. In posterior forms, pain is the principal symptom [18], while anterior forms are dominated by episodes of subluxation.

These types of instability are generally due to connective tissue diseases, either congenital or syndromic. In the voluntary form, a psychological component is often found. However, biological and psychological components are almost always intertwined. In fact, multiple episodes of instability, both voluntary or not, facilitated by connective tissue disease may lead to psychological disturbances. The voluntary component usually starts before the age of 10 years while in school where the instability develops into a game or a tic [18]. Voluntary instability is

achieved by a mechanism of muscular co-contraction. Regardless of the etiology or the type of instability, conservative treatment should be attempted for at least 6 months, including modifying sporting activities, relearning daily “non-dislocating” movements, rehabilitation, and painkillers in patients with painful subtypes. This is especially true since some types of hyperlaxity are transitory and regress after the puberty. Conservative treatment must be maintained as long as possible until reaching capsulo-ligamentous maturation.

In voluntary forms, a multidisciplinary approach (psychologist, physiotherapist, pain specialist) is advised for both children and their families (Figure 2). Furthermore, children should be made responsible for their own therapy, and the risks involved with voluntarily subluxation of the shoulder made clear. Surgical treatment was, historically, contraindicated in voluntary forms. Recent studies have shown that such approaches be discussed in patients with uncontrollable and painful shoulder instability after failure of a proper conservative treatment program, and with objective anatomical lesions [18,19]. In most cases, the uncontrollable nature coincides with the start of manual professional activity [18]. Surgical treatment is thus discussed after skeletal maturity has been achieved. In non-traumatic, uni- or multidirectional types, surgical treatment is based on the principle of reducing capsular volume along with a reduction of the dislocation pouch, which is most often at the inferior axillary region. If there is glenoid bone loss, a Latarjet procedure may be considered. Lefort et al. [20] analyzed a series of children aged 5 to 15 years old complaining of voluntary instability, which was posterior for 15 children and anterior for 4. Eleven patients had undergone posterior and/or anterior capsulorrhaphy based on the direction of the instability. No episodes of recurrence were noted after an average follow up of 8 years. Vavken et al. reported the outcomes after a follow up of 7.5 years in a series of 15 adolescents with an average age of 17 years who had undergone inferior capsular shift [21]. All patients had multidirectional instability with a Beighton score > 6. Five patients underwent genetic testing confirming Ehlers-Danlos syndrome. Thirteen patients did not present any episode of recurrence. 7 other patients occasionally presented with further subluxation. One patient reported a higher number of instability episodes than pre-operatively. Furthermore, the authors did not find any influence of the number of preoperative episodes of dislocation and a diagnosed Ehlers-Danlos syndrome on the subjective and objective clinical results. The median number of dislocations reported was 30. However, Ehlers-Danlos syndrome had a significant negative effect on functional scores (ASES, Quick-DASH), but not on satisfaction.

The role of Botulinic toxin A has been studied in the treatment of injuries of the brachial plexus, of spasticity/dystonia, and voluntary recurrent instability. The expected effect is that of reducing muscle tone and muscle hyperactivity, thus restoring axial muscle balance. This effect is only temporary and may be considered a therapeutic test. Alternative treatments described in young adults, such as “biofeedback” by electromyography or a pacemaker, have not been evaluated in children [22].

Recurrent instability with psychological impact

Management by a psychologist in an adapted structure is fundamental in case of voluntary instability and/or significant psychological impact. It allows the management of the somatic

effect by a pain specialists and mental suffering by a psychologist. This multidisciplinary approach also includes familial support with possible intervention by a social worker.

What about episodes of shoulder instability starting in childhood?

In some cases, episodes of instability become less frequent. As these children become adults, non-manual workers participate in less risky sporting activities leads to an improvement of symptoms. The maturation of the capsulo-ligamentous system also plays a role.

Episodes of recurrent gleno-humeral instability may lead to arthritic degeneration. Ogawa et al. [23] published a radio/tomographic study of 282 subjects with unilateral instability without previous surgery, whose age was younger than 40 years and who had no possibility of nontraumatic secondary osteoarthritis.
Radiographs and CT-scans showed osteoarthritis in 11.3 and 31.2% of cases. The total number of episodes of instability was significantly higher in patients with arthritis.
Boileau et al. [18] reported a series of children suffering from posterior voluntary instability starting around the age of 10 years that had become involuntary, uncontrollable, and painful by 16 years old. The authors argued that the anatomical gleno-humeral lesions shared similar characteristics to a cohort of adolescents suffering from pure involuntary posterior instability. Although gleno-humeral stabilization may be more difficult to achieve, the authors concluded that stabilization surgery was still indicated in patients with voluntary posterior instability that had become involuntary and painful.

Conclusion

Shoulder instability in children and skeletally immature adolescents is rare, the etiologic diagnosis of which remains problematic. Contrary to adults, non-traumatic and recurrent forms are more frequently encountered. The maturation of the capsulo-ligamentous system, anomalies of connective tissues or muscles, and psychological disturbances are elements that should be considered in the multidisciplinary management of these patients. Treatment is most often functional and may be accompanied by a psychological follow-up; nevertheless, during follow-up, an MRI should be obtained in order to assess for structural lesions and adapt the therapeutic strategy. The etiology, the direction of instability, and associated lesions should guide treatment. As a primary management, treatment should always be functional and, in case of voluntary instability with psychological impact, could be supported by a multidisciplinary team. Surgical stabilization strategies should be reserved for symptomatic forms with an impact of daily life.

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Figure legends

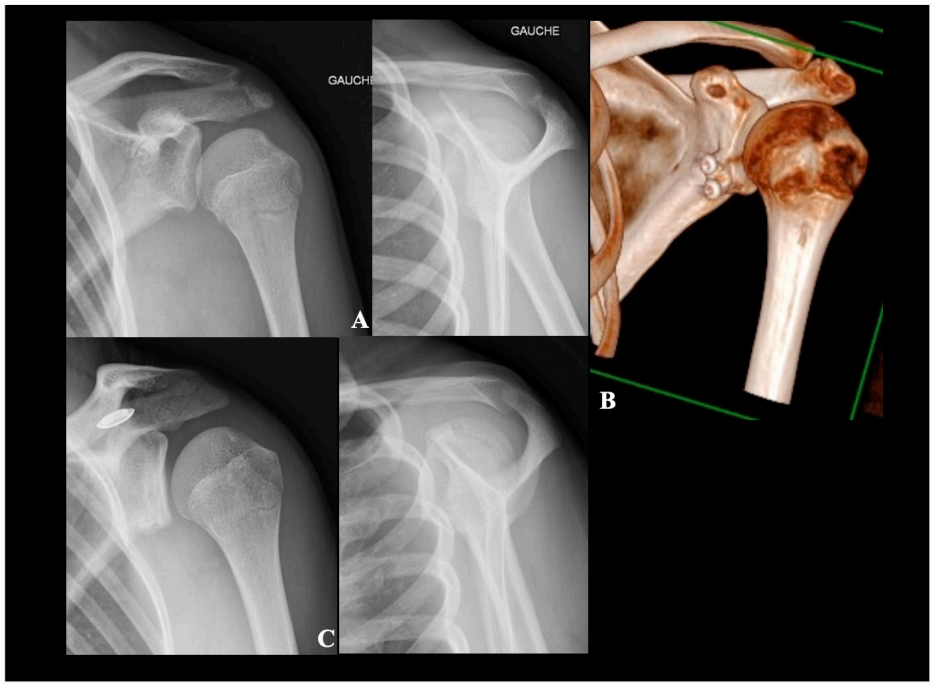

Figure 1: A case of post-traumatic unilateral involuntary and recurrent instability in an adolescent patient. Coronal and Lamy sagittal radiographs (A) as well as an MRI showed a Hill-Sachs lesion. Stabilization with an open Latarjet procedure was undergone. 3D reconstruction of a CT-scan showing proper healing of the Latarjet procedure 5 months post-operatively. No episodes of recurrence were recorded at 18 months follow-up and the hardware was removed (C).

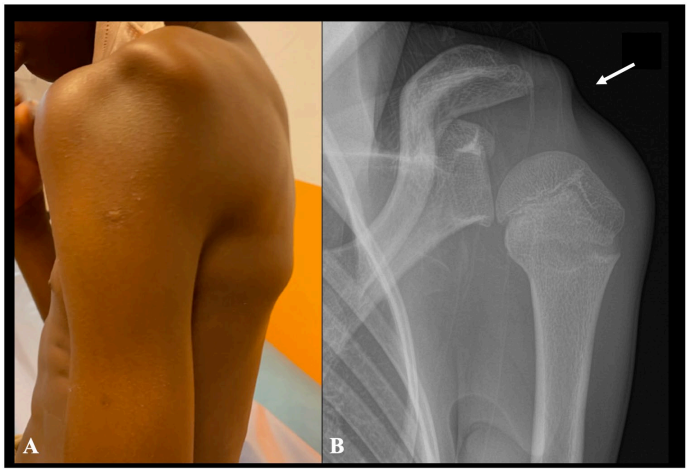

Figure 2: 9 year-old boy presenting with non-traumatic anterior unilateral voluntary and recurrent instability of the shoulder. Neuro-muscular and genetic testing were normal. Clinical evaluation showed numerous anterior voluntary and reproducible subluxations in the clinic (A). Excessive anterior and inferior laxity were found. Neer's sulcus test is illustrated on the coronal radiograph (B) by sub-acromial depression of soft tissues, identified by the arrow. Symptoms completely regressed after multidisciplinary functional management.