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Shoulder Rebalancing Surgery for Brachial Plexus Birth Palsy (BPBP)

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Abstract

Shoulder dysplasia is common constituting 80% of sequelae secondary to BPBP. Muscle imbalance due to uninhibited co-contractions and eccentric forces across the shoulder joint are the main causes of shoulder sequelae. This leads to limitation of shoulder movements and gleno-humeral deformity. This article discusses the presentations, investigations and treatment approach to various severities of deformities resulting from the muscular imbalance about the shoulder in BPBP.

Key words: Shoulder sequelae; Rebalancing conjoint transfer; Salvage surgery.

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Introduction

Shoulder dysplasia secondary to BPBP and its management is a vast subject. 80 percent of BPBP sequelae occur at the shoulder. Muscle imbalance due to uninhibited co-contractions and eccentric forces across the shoulder joint are the main causes of shoulder sequelae. This leads to limitation of shoulder movements and gleno-humeral deformity.

Traditionally, the focus was on improving function of the upper limb, but progress in imaging modalities and reconstructive techniques has changed the emphasis to restoration of normal shoulder anatomy. Early and proactive management right from infancy to pre-school age group can eventually result in a near complete restoration of morphology and shoulder movements. This article is an overview touching upon important clinical signs, imaging and surgical procedures. However, there is a substantial learning curve in both decision making and operative techniques. We urge the reader to go through the recommended reading list for a detailed understanding.

Primary Pathology

The powerful shoulder abductors i.e. deltoid, supraspinatus, and infra-spinatus, which are innervated principally by C5-6 are rendered weak in BPBP. Shoulder abduction and external rotation are diminished. Internal rotation is carried out primarily by the subscapularis muscle. Latissimus dorsi, Pectoralis major, and Teres major muscles are secondary internal rotators. The internal rotators are spared in C5-6 palsy and hence

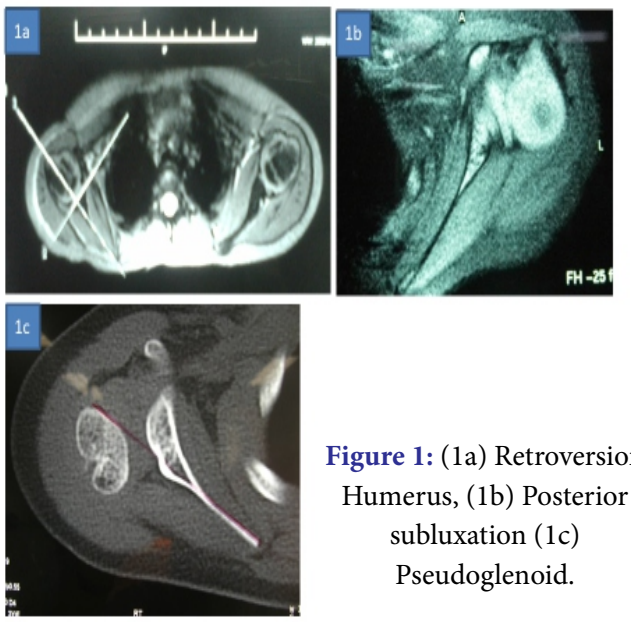


Figure 1: (1a) Retroversion Humerus, (1b) Posterior subluxation (1c) Pseudoglenoid.

Figure 2: The angle made between arm and trunk on attempted hand-to-mouth movement is termed as "Trumpeting"

Figure 3: Prominence of supero-medial angle of the scapula on attempted passive shoulder adduction and external rotation is termed as "Putti's sign".

unopposed. Normal internal rotators and weak external rotators leads to asymmetrical force transmission across the gleno-humeral joint (GHJ). An internal rotation contracture results from such persistent unequal forces.

These unchecked deforming forces and subsequent growth result in progressive glenohumeral joint dysplasia (GHJD) [1, 2]. GHJD progresses gradually from excessive retroversion of glenoid deformity to humeral head subluxation to the development of a false glenoid (Fig. 1).

During neuronal recovery, cross-innervation takes place due to neuroma formation leading to a mix-up of regenerating axonal sprouts. This often causes co-contraction of muscles. i.e. simultaneous firing of agonist and antagonist across a particular joint. Co-contraction between biceps and deltoid is very common resulting in the 'Trumpet Sign', but can exist across many subgroups of upper limb muscles. This gives rise to compensatory composite movements rather than original single movements of a muscle for a particular action. Asymmetric abnormal movement of the scapula called Scapular Dyskinesia, usually accompanies restricted motion of glenohumeral joint. 3-D kinematic analysis demonstrated increased scapula-thoracic joint motion in BPBP compared to normal children [3].

Clinical Presentation

Infants with residual BPBP present with internal rotation posture of arm and pronated forearm. Gradual reduction in passive external rotation (ER) at the shoulder is the hallmark sign of an evolving internal rotation contracture. With further progression, the infant demonstrates a deep anterior arm crease and an apparently shortened arm segment [4]. Children with an established posterior subluxation show hemi-neglect of the extremity [4, 5].

When babies lack active ER and muscular co-contractions predominate, any attempt to bring the hand to mouth causes trumpeting. The severity of trumpeting increases with the magnitude of the contracture (Fig. 2) [6].

Some patients demonstrate prominence of supero-medial angle of scapula with passive shoulder adduction & external rotation. This is termed as "Putti's sign" (Fig. 3) [7]. It is indicative of an abduction contracture with global capsular tightness. Patients with severe global capsular contracture compensate glenohumeral movement at thoraco-scapular level by winging the scapula and tilting of the torso to elevate the arm. Continuous imbalance between IR and ER leads to inability to perform activities of daily leaving like reaching behind the head, reaching out to hold objects and carry out overhead functions.

Classification and Scoring

Passive external rotation (ER) at the shoulder with arm in adduction is considered the most reliable measure correlating with gleno-humeral dysplasia. Kozin reported that children with a concentric glenoid averaged 15° of

Figure 4: Passive External Rotation at shoulder is measured with arm adducted and flush to the trunk. Examiner gently moves the limb in possible external rotation without any compensatory scapular movement.

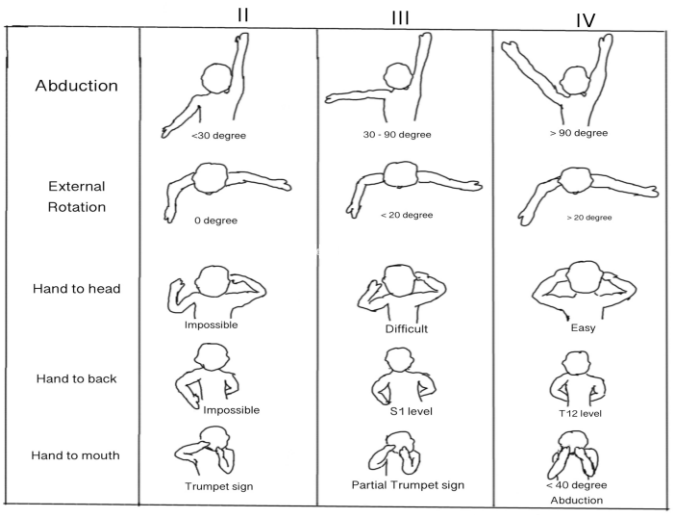


Figure 5: Mallet Score

external rotation, those with a flat glenoid averaged 8°, and children with a pseudo-glenoid averaged minus 21 degrees [7]. In younger children who cannot follow commands, passive ER should be used (Fig. 4). Modified Mallet Score is a reliable and reproducible five-point score denoting five active movements at the shoulder joint [8] (Fig. 5, article 1). It checks global external rotation, global abduction, hand-to-mouth, hand-to-spine and hand-to-neck movements. Russo et. al [9] described that hand-to-spine motion is a combination of shoulder internal rotation and extension. Thus, they added hand-to-midline as the sixth movement denoting isolated Subscapularis strength. Nath & colleagues used arm position at rest as seventh component of the score [10].

Gilbert (1993) described a alternate method of assessing shoulder function in BPBP patients based on degrees of

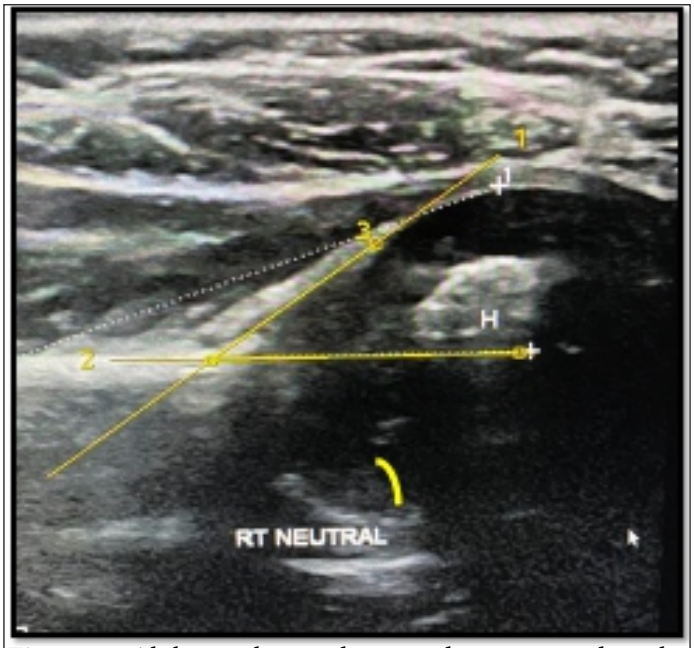


Figure 6: Alpha angle on ultrasound is measured as the angle between the mid-scapular line and the line intersecting it from the most prominent part of posterior cartilaginous humeral head at the spino-labral junction. Alpha angle of more than 30° along with ossified humeral head behind mid-scapular line is considered as posterior subluxation.

shoulder abduction and lateral rotation, ranging from Stage 0 denoting a flail shoulder to stage VI suggestive of a normal functioning shoulder [11].

Imaging

1. Ultrasonography

Hunter and colleagues (1998) described ultrasound to detect posterior dislocation of shoulder in brachial plexus palsy [12] with high intra-observer reproducibility and inter-observer reliability [13]. The position of the humeral head is measured by the intersection of a line through the axis of the scapula and a line tangential to the most prominent posterior part of the cartilaginous humeral head. The resultant angle is termed Alpha angle and denotes posterior shoulder dislocation if it exceeds 30° (Fig. 6). Sanchez et. al. reported the use of dynamic and real time ultrasound to detect subluxation of humeral head [14]. Passive shoulder external rotation with the arm adduction is the most correlated clinical finding with an abnormal ultrasound examination. Shoulder ultrasound is advised when clinical passive external rotation decreases below 60 degrees [15]. Ultrasound is preferred over CT or MRI in children below 1 year of age [16].

3. Computed Tomography (CT) and Magnetic

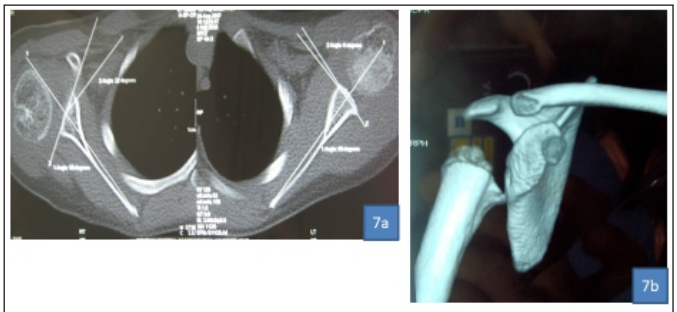


Figure 7: (7a) showing retroversion Ct scan which increases with severe posterior subluxation. (7b) showing 3 D reconstruction of the same.

Resonance Imaging (MRI)

Axial MRI images are preferred over CT scan in children less than five years as MRI detects cartilaginous outlines more reliably. Waters et al. reported an MRI axial image-based classification of glenohumeral deformity. It reliably measured the amount of glenoid retroversion and the percentage of the humeral head anterior to the mid-scapular line. Correlation was found between clinical parameters and MRI findings [18, 19]. A congruent gleno-humeral joint (glenoid retroversion with subluxation) is amenable to soft tissue rebalancing, whilst bony procedures are usually recommended for non-congruent (pseudo-glenoid formation and dislocations) joints. Van der Sluijis et al. found humeral head retroversion in children with BPBP on simultaneous axial imaging of the shoulder and the distal humerus [17, 18]. However, Pearl et al. recently reported that retroversion of humeral head on the affected side is usually less compared to the normal side and discussed its merits in surgical planning [18] (Fig. 7).

Early Interventions

Initial non-operative management includes regular passive stretching, splinting, muscle relaxation by botulinum toxin, percutaneous subscapularis release and closed reduction of the humeral head under anaesthesia (Hoffer et al). Verchere et. al. advocated use of a splint to maintain the extremity of infants in forearm supination and shoulder external rotation, starting as early as 6 weeks of life [19].

Botulinum toxin has been used to relax the tight internal rotators, thereby aiding in stretching of the contracture and allowing shoulder external rotators to recover strength. It also helps to relieve co-contraction between antagonist muscles. Botulinum toxin is injected in the subscapularis, pectoralis major, teres major and triceps in the dose of 2-3 units/Kg per muscle (maximum 10 units/Kg body weight) through anatomic localization or with the help of nerve stimulator. Ezaki et. al. found botulinum toxin to be an important adjunct in preventing and treating early posterior subluxation of shoulder [5].

Various methods are described to lengthen subscapularis. Minimally invasive subscapularis release (MISR) involves subscapularis slide from the medial border of the scapula through a 1-centimetre incision [20]. Arthroscopic anterior release of subscapularis combined with closed reduction and spica is an alternative method. Closed reduction is initiated by slow and steady manipulation under anaesthesia. By stabilizing the scapula and maximally adducting the arm, the humeral head is reduced into the glenoid. Achieving passive ER of ≥60° and palpable forward translation of the humeral head indicates good reduction [4].

Surgical Management

Children older than 18 months of age presenting with an internal rotation contracture and gleno-humeral joint incongruity require surgical intervention. Soft tissue lengthening of the pectoralis and sub scapularis tendon with closed reduction of the joint have been shown to arrest progressive dysplasia if done between the ages of 9 months to 18 months [21, 22].

In the absence of gleno-humeral deformity, soft-tissue surgery is preferred and involves lengthening of the tendon of subscapularis or sliding the muscle from the scapula. Sever originally described release of the subscapularis and the pectoralis major. L'Episcopo described a conjoint transfer of the latissimus dorsi and teres major to the rotator cuff. This procedure significantly improves active external rotation and abduction strength [25, 26]. The tendinous origin of the teres major and latissimus dorsi were divided proximally in the arm and repositioned under the lateral head of the triceps along the posterior lip of the humerus. Various modifications of this technique have been described over the years. Since the patho-anatomy is both anterior and posterior, it is logical that both anterior and posterior aspects of the shoulder joint will require correction.

Chuang [27] from Taiwan described transfer of the clavicular portion of pectoralis major to the anterior deltoid resulting in an augmentation of abduction. This was combined with a conjoint latissimus and teres major transfer to the rotator cuff posteriorly. Abduction improves as a result of removal of the chief adductor i.e. teres major.

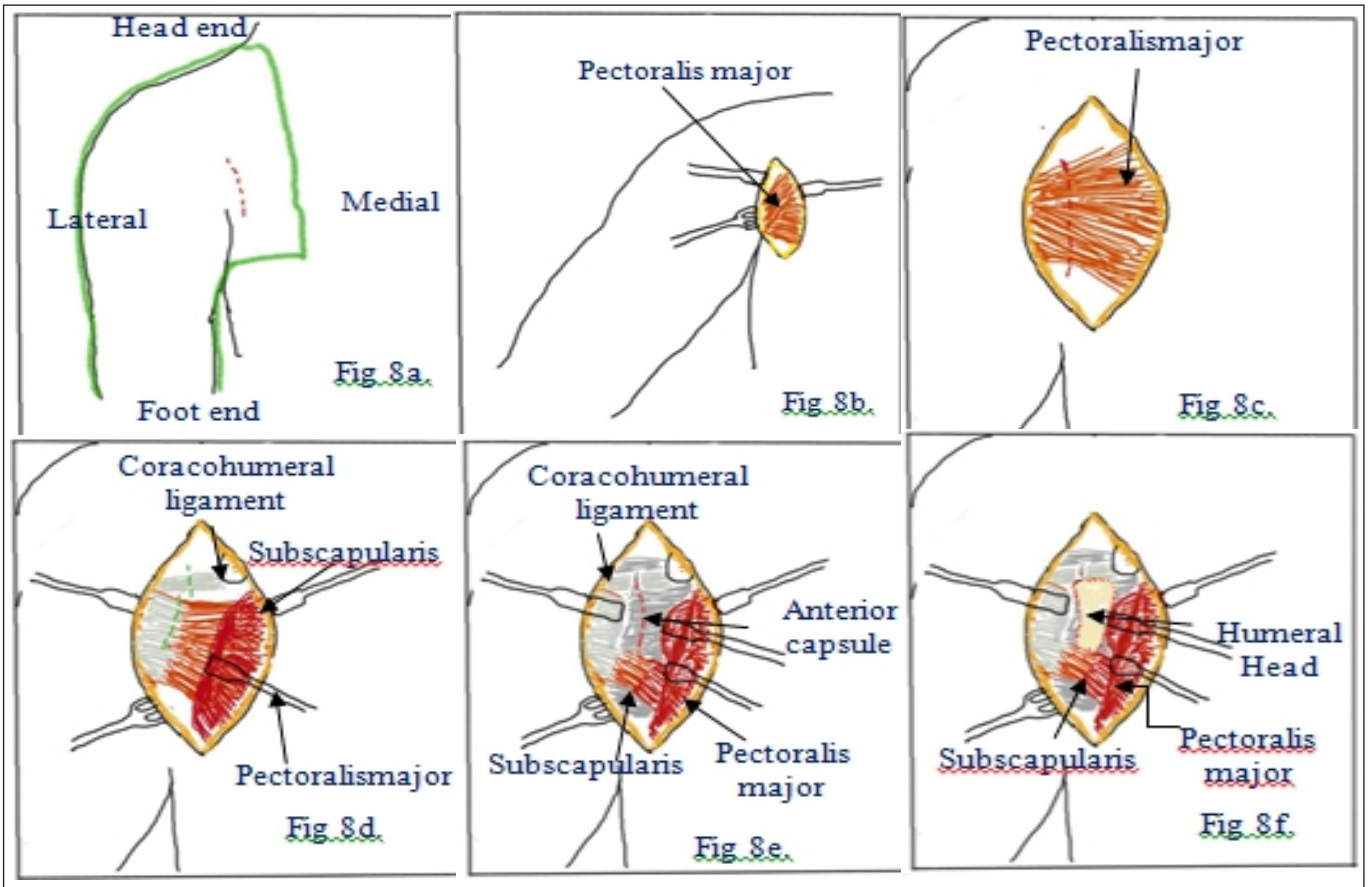


Figure 8: Anterior release of shoulder joint. (8a) Incision along the anterior axillary fold; (8b) Exposure of pectoralis major; (8c) Pectoralis major tenotomy; (8d) Subscapularis and coraco humeral ligaments were exposed and fractional lengthening of subscapularis and coracohumeral ligament release were done; (8e) Exposure of anterior capsule of the shoulder joint and anterior capsulotomy of the shoulder joint; (8f) Humeral head exposed and reduction achieved through anterior capsulotomy. (Acknowledgment: Digital artist D. Vigneshwaram Pragadesan for figure 8)

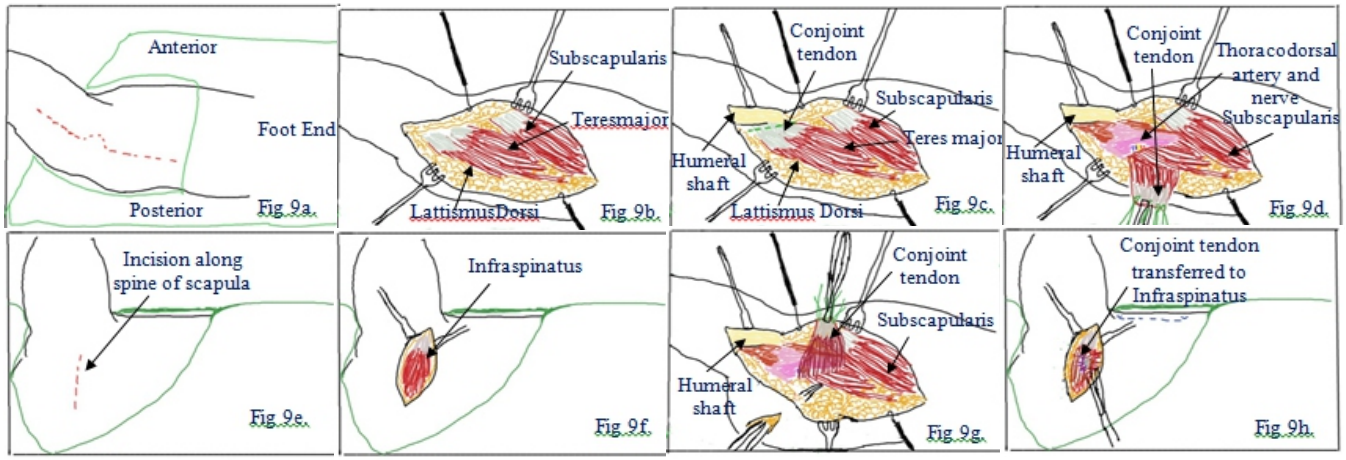


Figure 9: Shoulder Muscle Transfer. (9a) Patient in lateral position and incision along the posterior axillary fold; (9b) After subcutaneous dissection, Latissimus dorsi and Teres major are exposed; (9c) Conjoint tendon released from the humeral shaft protecting the radial nerve and axillary nerve; (9d) Conjoint tendon tagged and mobilized from the underlying soft tissue taking care not to injure the nutrient vessels; (9e) Second incision along the spine of the scapula; (9f) Infraspinatus muscle was exposed; (9g) Subcutaneous tunnel was created from the posterior to anterior incision; (9h) Conjoint tendon mobilized through the subcutaneous tunnel and sutured to the infraspinatus. (Acknowledgment: Digital artist D. Vigneshwaram Pragadesan for figure 9)

However, winging of the scapula is exaggerated, leading to an unsightly protrusion, since scapula-thoracic movement now compensates for over-head abduction.

Combined open reduction of the shoulder, capsulorrhaphy and tendon transfers attempts to restore normal gleno-humeral anatomy and rebalance the muscle forces [28].

Open Surgical technique highlights

Excessive release of the subscapularis causes an external rotation contracture of the shoulder with reduced hand-to-back movement and loss of midline function. Therefore, subscapularis muscle slide or a partial release (25% of the total muscle bulk) is advocated. Anatomically, the subscapularis is continuous with the anterior capsule and careful dissection should be undertaken to distinguish between the two (Fig. 8). The coraco-humeral ligament is released, pectoralis major fractional lengthening should be performed and not sectioned completely to prevent external rotation contracture and facilitate independent undergarment use especially in grown-up girls. The anterior capsule should be opened if the percentage of humeral head subluxation exceeds 60%. Humerus is to be reduced under vision. Coracoid nibbling is done if there is impingement in long standing cases especially if there is an overhang and abutment during external rotation. Teres major tendon transfer to restore external rotation as the chief motor always is to be done for external rotation deficit; latissimus may be spared or used as a conjoint transfer if both the deficits are severe. i.e. abduction and external rotation. In that case one muscle is used at each insertion, latissimus dorsi to deltoid tuberosity and teres major to rotator cuff (Fig. 9).

Open surgeries have stood the test of time [29] and yielded good results (Fig. 10 a-d) even for Waters grade 5 and from 1 to 14 years of age [30].

Other Surgeries

In the rare case of a pure deltoid paralysis with isolated abduction deficit, few authors have tried Saha's transfer [31, 32]. Anterior transfer of pectoralis as described by Chuang may be added [27]. The prerequisite for this is absence of a Putti's sign or internal rotation contracture. In flail shoulder, there are isolated reports of combining a levator scapulae transfer in conjunction with the trapezius but the outcomes are guarded and it is worthwhile considering an arthrodesis of the shoulder joint at skeletal maturity.

Figure 10a: Pre-operative 3-year-old mallet 2, restricted abduction and external rotation.

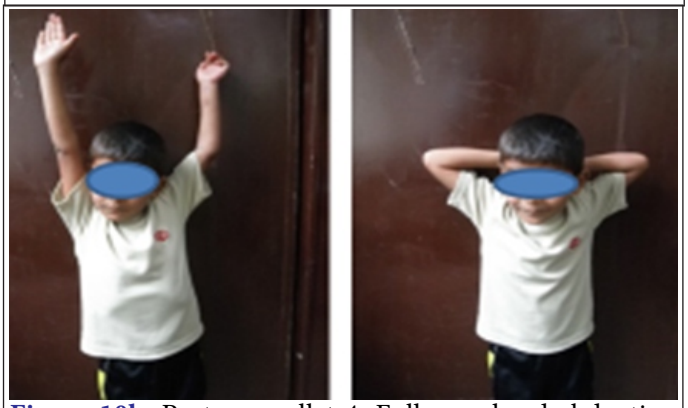


Figure 10b: Post op mallet 4. Full over head abduction with good external rotation.

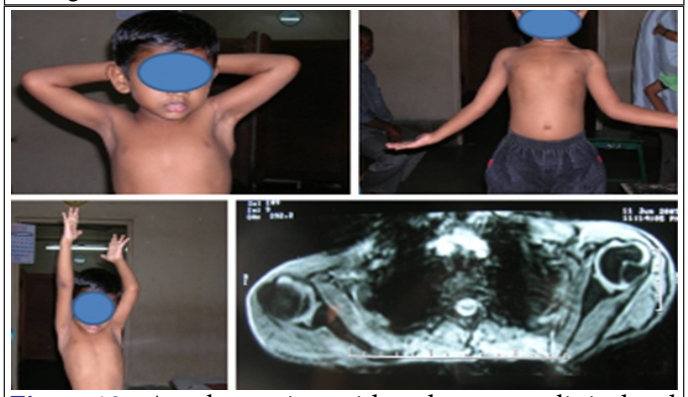


Figure 10c: Another patient with early surgery clinical and MRI result.

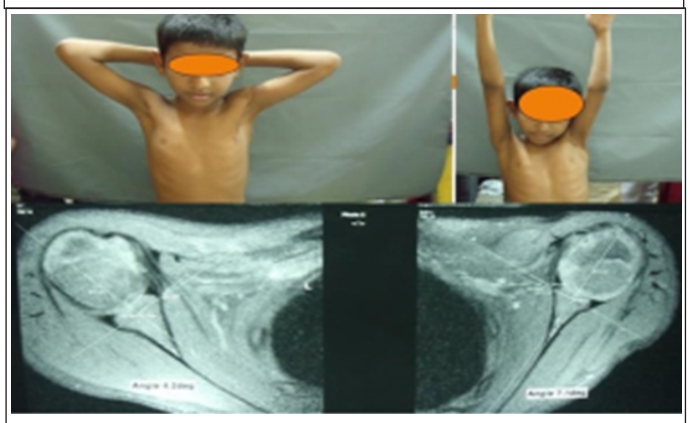


Figure 10d: Result maintained 8 years later.

Patients with advanced GHJD may not be candidates for tendon transfers alone and require hybrid procedures or palliative procedures [33].

Role of Humerus Osteotomy in children with BBPP

Indications for derotation osteotomy of the humerus include: older age at presentation, limited passive external rotation, and significant glenohumeral joint deformation i.e. Waters grade 4 onwards. It is also indicated in weak but useful contracted donors in flail limbs [34, 35]. The osteotomy can be performed at the proximal humerus (above the deltoid insertion), or at the lower humerus level; the latter being aesthetically better as the scar is on the medial aspect of the arm [34, 36, 37]. Besides, the biomechanical advantage of an osteotomy done proximal to the deltoid insertion by lateralizing the deltoid contributes to improved shoulder abduction after this procedure [38-39] (Fig. 11 a, b).

Calculation of the amount of external rotation required depends on the external rotation deficit and available internal rotation. It is imperative to understand that internal rotation is essential for midline activities like buttoning and dressing and its loss could be disabling [38, 39]. The objective is to achieve external rotation of at least 15-20 degrees without losing midline reach. This is confirmed intra-operatively after preliminary fixation of the osteotomy. If limited, the rotation angle is reduced. In general, a child who is able to reach the back preoperatively is suitable for 60 degrees of derotation without losing midline reach. The osteotomy is performed through a standard delto-pectoral approach and stabilised with a 6-holed plate (Fig. 11 c). We have occasionally used K-wires or external fixation, but plating is better tolerated and allows early use of the limb.

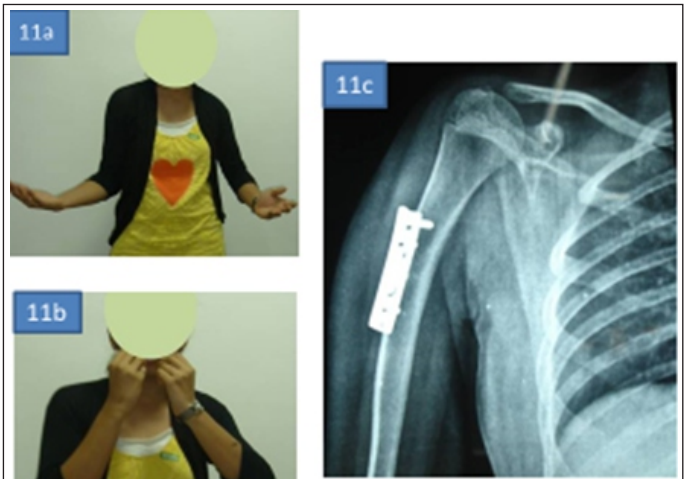


Figure 11 a-c: Clinical and radiographic outcome of an adolescent who underwent derotation humeral osteotomy.

Scapular Neck Osteotomy

Hopyan and colleagues combined glenoid neck osteotomy with soft-tissue rebalancing surgery [40]. They found improved Mallet scores for global external rotation and hand-to-neck movements. Waters grade improved from average of 4.3 preoperatively to 1.6 postoperatively. This novel technique was proposed as an alternative to humeral derotation osteotomy. Other authors have observed that this procedure should be considered for older children with severe gleno-humeral dysplasia (Waters-IV and beyond), where soft tissue rebalancing might not be effective due to non-congruent underlying joint surface and limited remodelling potential (Fig. 12 a-h).

Outcomes and complications

In spite of many variables, most papers about open surgical intervention showed improvement in shoulder abduction and external rotation with improvement in the MMS. Abduction, external rotation, hand to neck, and hand to mouth subsets of MMS improved by 1 to 2 grades after

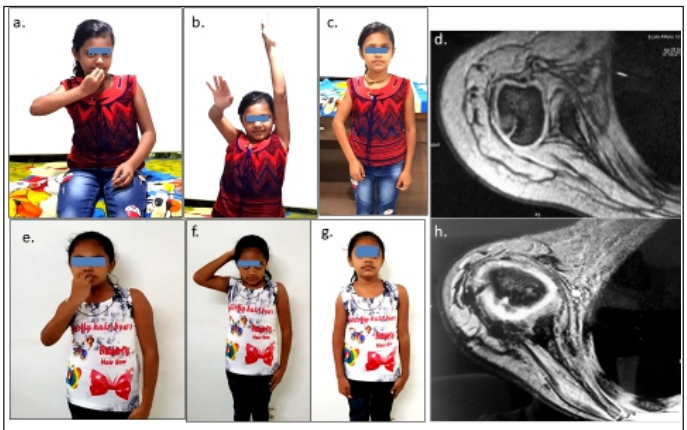


Figure 12(a-h): (12 a-c) 10-year-old girl with residual BPBP and limited shoulder abduction, external rotation, persistent internal rotation posture of arm and marked trumpeting. (12 d) Axial MRI revealed pseudo-glenoid formation with posterior subluxation correlating with Waters IV gleno-humeral deformity. Combined Scapular neck anteversion osteotomy and conjoined tendon transfer was performed. (12 e-g) Patient had improved trumpeting, resting posture of limb and active external rotation range. (12 h) Anteverted glenoid with graft at the neck and well-reduced humeral head is visible on axial imaging at one-year follow-up.

surgery [28]. However, diminished internal rotation has been observed postoperatively. To avoid this, transfer of only teres major or tendon transfer without subscapularis lengthening can be considered. Where the functional impairment is significant, an internal rotation osteotomy may become necesssary. There is a growing concern that the early improvements with tendon transfers can fade over time, with one study showing progressive loss of ROM at 30-year follow-up. Gradual degeneration of the transferred muscles, contracture of the surrounding soft tissues, and degenerative changes in the glenohumeral joint may account for the observed decline [24].

Effect of tendon transfer on GHD

There have been mixed results with regard to improvement in GHD following tendon transfer surgery [28, 41].

Outcomes of humeral derotation osteotomy

Waters et al noted improvement in global shoulder function as indicated by an increase in the Mallet score from 13 to 18 after external rotation osteotomy [36]. Kirkos et al also reported satisfactory outcome and increase in shoulder abduction by 27 degrees [33]. Most patients report high satisfaction in terms of improved posture of the limb and function (Fig. 12).

Outcomes of glenoid anteversion osteotomy:

Short term results showed improvement in active and passive ER. Both active and passive IR reduced postoperatively. The aggregate Mallet score improved by a mean of 4.0 points. Glenoid retroversion improved by a mean of 26 degrees. The percentage of the humeral head anterior to the mid-scapular line improved by a mean of 35%.

Complications

Complications associated with tendon transfers are - injury to any of the neurological structures including the radial nerve, axillary nerve, subscapular nerve, and thoracodorsal nerve. Thoracodorsal vessels, subscapular vessels, and the posterior humeral circumflex vessels are vulnerable. In addition, when donor tendons are not adequately mobilised, adequate fixation of the tendon is not achieved. This may cause insertional failure and suboptimal improvement.

Abduction contracture can occur if the tendons are inserted under too much tension. Intraoperative examination of passive motion before closure lessens the risk of this complication [28].

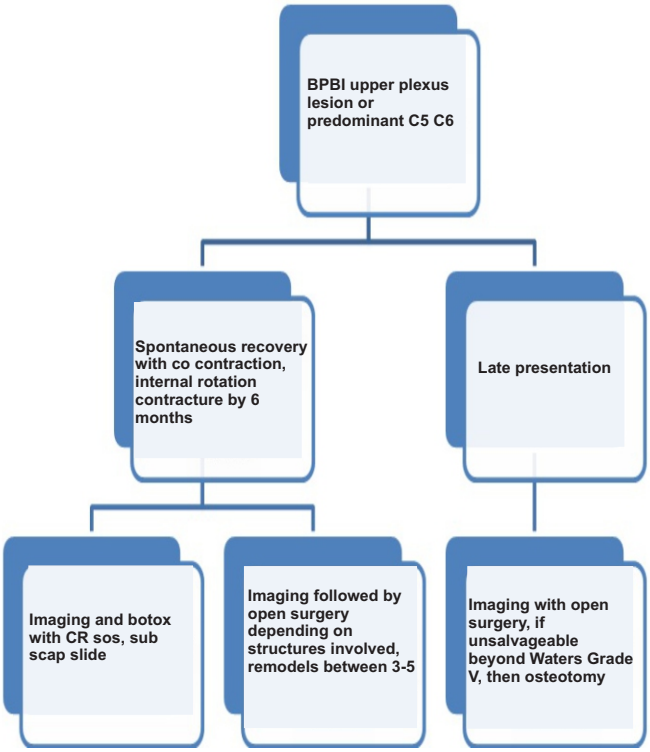
During humeral osteotomy radial and ulnar nerves are susceptible [39]. Most of the iatrogenic palsies recover without further interventions. Other reported complications are non-union, malunion, periprosthetic fracture, hypertrophic scar, and implant failure [37]. Under-correction may necessitate revision surgery.

Glenoid osteotomy: An initial report mentioned inadvertent completion of the osteotomy in few patients. The suprascapular bundle exiting the spinoglenoid notch is more vulnerable in the setting of severe posterior glenoid deficiency [40].

Optimum Timing of Surgical treatment.

Muscle rebalancing procedures around the shoulder are best undertaken before structural bony changes set in. Advancing bony changes call for restoration of shoulder anatomy through bony surgery in addition to tendon transfers.

In children older than 11 years of age; muscle transfers are recommended for improvement in abduction and external rotation, in addition to derotation osteotomy for waters Grade VI and VII dysplasia. Gleno-humeral dysplasia can remodel if rebalancing is performed early in life. The



Recommended algorithm of management

optimum time for re-establishing congruency of the gleno-humeral joint is in the early years of life from 2-5 years of age.

Conclusion

Recent MRI based studies have proven that gleno-humeral deformity is best treated early with muscle rebalancing procedures before irreversible changes develop. Extra-articular muscle transfers and release of antagonistic muscles improve range of motion and rebalance forces around the shoulder. Injection of Botulinum toxin type A has proved to be an effective adjunct to bracing and physical therapy. Humeral derotation osteotomy is an effective salvage procedure if the gleno-humeral joint is non-congruent.

Summary

- Early recognition of co-contractions and gleno-humeral dysplasia.
- Clinical scoring and age-appropriate imaging.
- Closed procedures in early infancy to prevent malformation of the shoulder joint.
- Early open surgery converting adductors and internal rotators i.e. deforming forces into corrective forces while maintaining midline function.
- Bony procedures for late presenting cases with advanced shoulder dysplasia or poor motors.

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