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# *Evaluation of unipolar transfer of the latissimus dorsi to flexor antebrachii in patients with arthrogryposis*

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**Objective** To evaluate active elbow flexion restored with latissimus dorsi (LD) transfer in patients with arthrogryposis and determine the correlation with the level of segmental injury to the spinal cord. **Material and methods** Active elbow flexion was restored in 30 patients with arthrogryposis (44 upper limbs) using unipolar LD transfer performed between 2011 and 2018 at the Turner Scientific and Research Institute for Children's Orthopaedics. The patients' age at the time of surgery ranged from 1 year to 10 years with the mean age of 3.98 ± 2.35 years. Clinical and neurological assessment was performed for the patients. Statistical data analysis was produced. **Results** The patients were subdivided into three groups with regard to the level of segmental injury to the spinal cord including C6–C7 (n = 8, 29.6 %, 13 limbs), C5–C7 (n = 17, 54.5 %, 24 limbs) and C6 (n = 5, 15.9 %, 7 limbs) levels. The patients were followed from 1 year to 7 years (3.2 ± 1.9). Postoperative passive and active elbow flexion was 100° ± 7.0° (min 80°, max 110°) and 90.5° ± 14.7° (min 40°, max 110°), respectively. Extension deficit of the elbow increased by 12.8° ± 4.8° (min 10°, max 20°) in 18 (51%) cases but made no impact on activities of daily living. The results of 20 patients (55.6 %) were rated good, 12 (33.3 %) were satisfied and 4 (11.1 %) had poor results. No correlation could be found between postoperative active flexion, extension deficit of the elbow and the level of segmental injury to the spinal cord in patients with involved levels of C6–C7 and C5–C7. **Conclusions** The LD can be regarded as the choice flap for restoration of active elbow flexion in patients with arthrogryposis and segmental involvement at C6, C6–C7, C5–C7 levels with baseline donor muscle strength grading 4 and over and passive elbow flexion of at least 90°.
**Keywords**: arthrogryposis, elbow joint, latissimus dorsi, graft

## INTRODUCTION

The latissimus dorsi (LD) muscle is a unique flap for reconstruction of shoulder and elbow soft tissue defects and, unlike most other autologous grafts, allows a single-stage soft-tissue coverage. The world literature search performed by A. Sood et al. (2017) yielded 13 relevant studies with a total of 52 patients who received functional LD flaps for upper-extremity reconstruction produced for injuries of various etiology [1].

The LD muscle was initially transferred to achieve elbow extension in 1949. Transposition of the whole LD to restore active elbow flexion was described by E.R. Schottstaedt et al. in 1955 [2, 3]. The technique was further developed by Zancolli and Mitre who performed a bipolar LD transfer in 8 patients with poliomyelitis and brachial plexus injury to restore elbow flexion [4]. Since then, the LD flap has been a workhorse for reconstruction surgery of the upper extremity to restore motion to the elbow and shoulder [1].

The LD flap is widely used for reconstruction of active elbow flexion in patients with posttraumatic conditions of brachial plexus or poliomyelitis. The literature review yielded only 3 publications describing the LD to biceps transfer in patients with arthrogryposis with 3 to 11 cases reported [5–7]. There are controversies in the literature discussing the quality of LD in patients with arthrogryposis that can pose limitations for the use as an autologous graft in the cohort of cases. No reports can be found on correlation between results of LD transfer improving elbow flexion and the level of segmental injury to the spinal cord. So, the topic appears to be relevant and requires further investigation.

**The purpose** of the study is to evaluate active elbow flexion restored with unipolar transposition of the LD flap in patients with arthrogryposis and determine the correlation with the level of segmental injury to the spinal cord.

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## MATERIAL AND METHODS

Restoration of active elbow flexion was performed for 30 patients with arthrogryposis (44 upper limbs) using unipolar LD transfer between 2011 and 2018 at the Turner Scientific and Research Institute for Children's Orthopaedics. All patients and/or their statutory representatives voluntarily provided an informed consent to participate in the study, receive surgical treatment and publication of the findings.

Patients were examined preoperatively, at one-year-and-over follow-up. Clinical parameters evaluated in the patients included active and passive range of motion in the elbow and shoulder joints, muscle strength of elbow flexors and the donor site, ability to practice self-care skills and use of compensatory adaptive mechanisms.

Muscle strength was measured using the 6-point scale (from 0 to 5) moving on a flat plane, against gravity and manual resistance. The LD muscle was tested by palpation at shoulder adduction, extension and internal rotation. Classification offered by Agranovich, Lahina (2013) was used to identify a level of segmental injury to the spinal cord based on clinical and neurological examination. The findings were not included in the series due to complexity of neurological assessment in younger children [8].

The patients' age at the time of surgery ranged from 1 year to 10 years with the mean age of 3.98 ± 2.35 years.

The Wilcoxon signed rank test was used to assess changes in pre- and postoperative elbow flexion. The Spearman Rank correlation test was used to analyze the correlation between changes in ranges of motion and levels of injury. The Wilcoxon signed rank test was applied for correlations between pre- and postoperative parameters for groups with different levels of injury to the spinal cord. The Bonferroni correction was used as an adjustment for multiple comparisons. The hypotheses were tested with a significance level of $p < 0.05$ (95 %).

## RESULTS

The patients were subdivided into three groups with regard to the level of segmental injury to the spinal cord including C6–C7 (n = 8; 29.6 %; 13 limbs), C5–C7 (n = 17; 54.5 %; 24 limbs) and C6 (n = 5; 15.9 %; 7 limbs) levels. No case of segmental injury to C5–Th1 was presented. Each of the groups showed different muscle strength of the shoulder, shoulder girdle, LD, and range of motion in the shoulder and elbow joints that correlated with a level of segmental injury to the spinal cord. Compensatory adaptive movements the patients used for passive elbow flexion were assessed in all the groups.

Group I exhibited injury primarily to C6–C7 segments of the spinal cord. The glenohumeral joint maintained its full range of active motion that was moderately limited in abduction measuring ≥ 70º with the strength of shoulder girdle preserved or the muscles being moderately hypoplastic. Passive movements persisted in the majority of elbow joints, active flexion was either absent or limited with biceps brachii strength scored 2 and active supination of the forearm limited. The LD strength scored 4 to 5.

Group II exhibited injury primarily to C5–C7 segments of the spinal cord. The glenohumeral joint demonstrated limitation in active motion with abduction measuring 30º to 45º, maintained or moderately limited passive motion. The upper limb was positioned in internal rotation with muscles of shoulder girdle being evidently hypoplastic. Passive movements persisted in the majority of elbow joints, active flexion was either absent or limited with biceps brachii strength scored up to 2 and no active supination of the forearm recorded. The LD strength scored 3 to 4.

Group III exhibited injury primarily to C6 segments of the spinal cord. Elbow joints were mostly involved in the patients who developed either extension or flexion-extension contractures. No limitations were observed in the glenohumeral and radiocarpal joints with grip hand function maintained. Active elbow flexion was either severely limited or absent with passive elbow flexion being either normal or limited and active supination of the forearm being absent. Range of motion in the glenohumeral joint was physiologically normal with biceps brachii strength scored up to 2. The LD strength scored 4 to 5.

Two types of compensatory adaptive movements were noted in patients with injuries to C6–C7 and C6 segments and maintained active motion in the glenohumeral joint to produce active elbow flexion.

The first type involved active flexion and abduction in the glenohumeral joint, passive elbow flexion against gravity of the limb, relaxation of triceps brachii and radiocarpal flexion in a child's attempt to move from hand to mouth. Hand-to-mouth movement of the second type was produced by a forearm pressed against another forearm with flexed and abducted shoulder and the head tilted forward towards the hand.

Patients with injuries to C5–C7 segments and evident hypoplasia of shoulder girdle muscles had different compensatory adaptive movements for passive elbow flexion of the arm as compared to the above groups that were dependent on patient's posture. A child would tend to tilt his/her torso forward, flex the leg in the knee and hip joints and press the femur against the forearm, thus flexing the arm in the elbow and bringing the hand to the face while sitting. Passive elbow flexion was produced by pressing the forearm against an object (a table) and tilting the torso and the head towards the hand while standing or sitting

Absent or limited active elbow flexion of less than 90°, passive elbow flexion of 90° and over, LD score of 3 and over were an indication to restoration of elbow flexion using monopolar LD transfer.

Twenty six patients (36 upper extremities) were followed from 1 year to 7 years (3.2 ± 1.9 months) postoperatively. Preoperative passive and active elbow flexion was 99.7° ± 7.7° (min 80°, max 110°) and 17.5° ± 11.9° (min 0°, max 40°), respectively, with elbow extension deficiency of 7.7 ± 10° (min 0°, max 30°), elbow flexor and LD strength scored 0-2 and 3-5, respectively. At long-term follow-up, passive and active elbow flexion was 100° ± 7.0° (min 80°, max 110°) and 90.5° ± 14.7° (min 40°, max 110°), respectively, with elbow extension deficiency of 14.0 ± 12.9° (min 0°, max 45°) and elbow flexor strength scored 2 to 5. Elbow extension deficiency increased by 12.8° ± 4.8° (min 10°, max 20°) in 18 (51 %) cases but made no impact on activities of daily living. Active range of motion of the elbow joint measured 75.4 ± 18.0° (min 40°, max 110°).

Modified A. Van Heest Scale was used to evaluate postoperative active elbow flexion, deficiency of elbow extension, power of elbow flexors and a need in compensatory adaptive mechanisms performing self-care activities [6]. Normal flexion of the elbow joint as reported by B.F. Morrey et al (1981) was defined as zero to 145° although an arc of elbow flexion from 60° to 120° (so called 'practical arc') was adequate to performed most of activities of daily living. Deficiency of elbow extension of 60° is within the limits of the positions assumed during personal care and hygiene, using crutches and wheel-chair [9]. A. Van Heest et al (1998) suggested that passive elbow flexion of more than 90° was sufficient to allow most common daily functions in arthrogryposis patients with an elbow flexion restorative procedure [6].

Results of treatment were subdivided into three groups.

Good outcomes included grade 4/5 muscle strength, a 'practical arc' of active motion, an active elbow flexion of more than 90°, elbow extension deficiency of less than 60°, none or rare use of compensatory adaptive mechanisms performing most of self-care activities.

Fair outcomes included muscle 3 strength, a 'practical arc' of active motion, an active elbow flexion of less than 90°, elbow extension deficiency of less than 60°, frequent use of compensatory adaptive mechanisms performing most of self-care activities.

Poor outcomes included muscle 0-2 strength, an amplitude of active motion being less that 'practical arc', an active elbow flexion of less than 90° and/or elbow extension deficiency of more than 60° and frequent use of compensatory adaptive mechanisms.

The results of 20 patients (55.6 %) were rated good, 12 (33.3 %) were satisfied and 4 (11.1 %) had poor results (**Fig. 1, 2**).

Comparative measurements of elbow arc of motion, elbow flexors' power before and after LD transposition to improve elbow flexion in patients with arthrogryposis are presented in Table 1.

The Wilcoxon signed rank test was applied for correlations between pre- and postoperative parameters in all the patients and in each group. Statistically significant results in postoperative active elbow flexion and postoperative deficiency of elbow extension were obtained in all the patients (Table 2).

The Table indicates to significant changes seen with the Bonferroni correction applied for multiple comparisons in:

1) postoperative active elbow flexion;

2) postoperative deficiency of elbow extension (**Fig. 3, 4**).

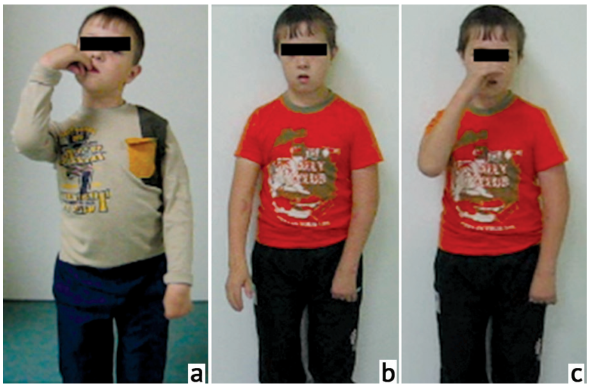


**Fig. 1** A good result of active elbow flexion (right side) restored in an 8-year-old patient with arthrogryposis and segmental involvement of C6-C7 at one year postsurgery showing (a) preoperative elbow flexion using compensatory adaptive mechanisms; (b, c) active elbow flexion and extension following LD transfer to restore elbow


**Fig. 2** A good result of active elbow flexion (right side) restored in a 3-year-old patient M. with arthrogryposis and segmental involvement of C6-C7 at three years postsurgery showing (a) preoperative elbow flexion using compensatory adaptive mechanisms; (b, c) active elbow flexion and extension following LD transfer to restore elbow flexion

Table 1

Results of LD transfer to improve elbow flexion in patients with arthrogryposis

| Comparison criterion | Preoperative | Postoperative |
|---|---|---|
| passive elbow flexion | 99.7 ± 7.7° (min 80°, max 110°) | 100 ± 7.0° from 80° (min 10°, max 20°) |
| active elbow flexion | 17.5 ± 11.9° (min 0°, max 40°) | 90.5 ± 14.7° (min 40°, max 110°) |
| deficiency of elbow extension | 7.7 ± 10 ° (min 0°, max 30°) | 14.0 ± 12.9° (min 0°, max 45°) |
| elbow flexors power | 0–2 | 2–5 |

Table 2

The Wilcoxon signed rank test applied for comparison of pre- and postoperative parameters

| | Postoperative active elbow flexion | Postoperative deficiency of elbow extension |
|---|---|---|
| level | C5–C7 | |
| h | 1 | 1 |
| p | 0.000083* | 0.0166** |
| level | C6–C7 | |
| h | 1 | 1 |
| p | 0.000244* | 0.007800* |
| level | all levels | |
| h | 1 | 1 |
| p | 0.0000001* | 0.000124085* |

Value of accepted null hypothesis with the Wilcoxon signed rank test (for "pre/post" conditions) (h) and the significance level (p). * – all significant values with the Bonferroni correction for multiple comparisons; ** – values being less than the significance level with no Bonferroni correction withstood, i.e. being at a level of 'tendency'

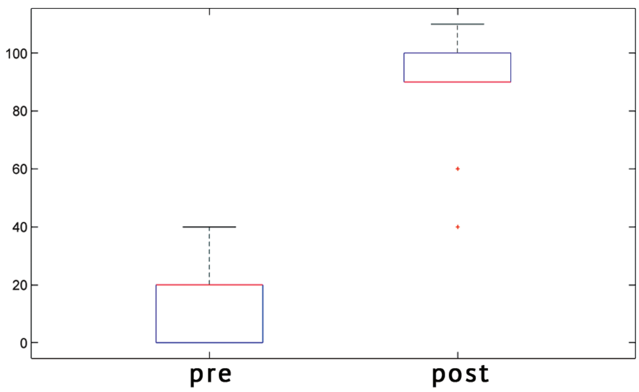


**Fig. 3** Diagram showing a range of active elbow flexion in patients with arthrogryposis before and after LD transposition

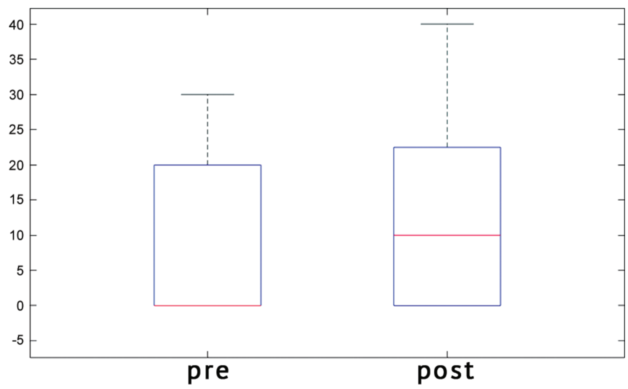


**Fig. 4** Diagram showing a range of deficiency of elbow extension in patients with arthrogryposis after LD transposition

There is a clear tendency for involved C5–C7 level taking into consideration the Bonferroni correction whereas other measurements show statistically significant differences. Due to a low incidence of the spinal cord involvement at C6 level and a small sample size the amount of information was insufficient for proper statistical analysis with the group of patients, however, elbow flexion and ability to practice self-care skills were restored in all the cases. We aimed at testing a hypothesis about relationship between outcomes of active elbow flexion restored with LD to biceps unipolar transposition in patients with arthrogryposis the level of segmental injury to the spinal cord. The Wilcoxon signed rank test was used to evaluate results of treatment with regard to the level of segmental injury to the spinal cord including C6–C7 and C5–C7 levels (Table 3). No correlation could be found between postoperative active elbow flexion, deficiency of elbow extension and the level of segmental injury to the spinal cord in patients with involved levels of C6–C7 and C5–C7.

Table 3

The Wilcoxon signed rank test applied for comparison of pre- and postoperative measurements at different levels of segmental injury to spinal cord

| Postoperative active elbow flexion | |
|---|---|
| level | C5–C7 vs C6–C7 |
| h | 0 |
| p | 0.94 |
| Postoperative deficiency of elbow extension | |
| level | C5–C7 vs C6–C7 |
| h | 0 |
| p | 0.36 |

## DISCUSSION

Controversies can be found in the reported literature regarding the quality of the LD muscle in patients with arthrogryposis: E. Mercuri et al (2009) indicates to trunk muscles being intact in the pathology whereas J. Philpot et al (2001), A. Van Heest et al (1998) reported a possibility of absent LD in patients with arthrogryposis. Palpating the muscle manually to assess the muscle mass is not a reliable technique since the mass can be added by fat infiltration or thick subcutaneous tissue. It is suggested that ultrasound or MRI might be useful for preoperative planning [6, 10, 11].

Due to complexity of MRI assessment in younger children O. Agranovich (2017) recommends producing CT scans of the shoulder and the chest using a variety of regimens to explore the quality of the donor muscle, bone and joint deformities. Intraoperative diagnostic test employing a 2-cm skin incision on the posterior axillary aspect in the LD projection can be rather practical. The subcutaneous fat tissue is dissected using an electrical knife and muscle contraction and the underlying structures can be evaluated. Minimal or absent contractions, fat or fibrous infiltrations are suggestive of the LD muscle being not suitable for transfer to restore an active elbow flexion [12].

E. Boven (2017) analyzed volumes of the LD measured on MR images in patients with arthrogryposis and found that until the age of 10, all LD volumes were similar, and after the age of 10 there was more variation in LD volume, and after the age of 14 this variation increased. The patients that had a normal volume of the LD had the biggest decrease in passive range of motion after transfer caused by an extension deficit. No extension deficiency was reported in the cases when the LD was of less volume [5].

Functional transfer of the LD flap may be unipolar or bipolar. The LD muscle can be transferred by detaching a single end (unipolar) or as a bipolar transfer. The insertion of the LD muscle is proximally attached to the coracoid or acromion/clavicle with the bipolar transfer. The origin of the muscle is attached distally to the radial tuberosity or biceps tendon, or to the ulna in case of dislocated radial head [1, 4, 6, 13–15]. In the small case series described by K. Kawamura and colleagues (2007), there was no difference seen in outcomes of the two types of LD transfer [16]. According to A. Sood (2017) the choice between unipolar and bipolar transfer of the LD flap is mainly determined by surgeon preference [1].

K. Kawamura et al. (2007) reported sufficient restoration of elbow flexion with LD transfer avoiding severe elbow flexion contractures [16]. Patients are normally presented with postoperative deficiency of elbow extension of not more than 10-15° that is in line with our findings [3, 17–20]. S. Chaudhry et al. (2013) reported transfer being most likely to be successful if there was full passive range of motion of the elbow and the shoulder was stable. Poor results could be attributed to atrophic LD [3]. The literature search yielded 3 relevant studies of foreign authors including A. Van Heest et al. (1998), E. Boven (2017), R. Zargarbashi et al. (2017) who analyzed outcomes of LD to biceps transfer in a small series of patients with arthrogryposis [5–7]. A. Van Heest et al (1998) performed bipolar LD transfer for 3 patients (4 flaps) with an average age 11 years (range, 6–14 years) and an average follow-up period of 1.5 years (range, 1–3 years). Postoperative muscle strength scored 4 (n = 2) and 3 (n = 2). Postoperative loss of LD strength was observed in two cases with at least grade 4/5 strength of the muscle to be transferred in all the patients. Postoperative active range of motion of the elbow measured 84° on average (range, 70°-100°). No postoperative deficiency of the elbow extension was noted in the cases and passive range of motion of the elbow maintained. The results of two patients were rated good, and two had fair outcomes [6].

In the retrospective case series produced by E. Boven, 6 patients (8 flaps) received a unipolar and bipolar LD transfers to restore active elbow flexion. The patients' age ranged from 7.8 to 23 years with an average follow-up period of 4.5 years (range, 1.6–8.3 years). Postoperative active range of motion of the elbow improved from 4° to 80° (by 43° on average) and measured an average of 46.8°. Postoperative deficiency of the elbow extension ranged from 4° to 46° (22.3°, on average). With active elbow flexion restored a passive range of motion decreased in 50% of the cases without loss of function. The results of 4 patients were rated good, two were satisfied and two had poor results [5].

R. Zargarbashi et al (2017) produced a retrospective analysis of bipolar LD to biceps transfer to restore active elbow flexion in 11 patients (13 limbs) with arthrogryposis. The patients' age was 5.69 ± 2.49 years and follow-up period was 27.3 ± 17.8 months. Postoperative active elbow flexion measured 97.7° ± 34.5°. Of 13 patients, 10 showed improvements in activities of daily living but 11 still used compensatory adaptive mechanisms. Overall patient satisfaction with surgical intervention was 92.3 % [7].

Retrospective analysis of unipolar LD transfer to restore active elbow flexion in 26 patients (34 upper limbs) followed from 1 to 7 years (3.15 ± 1.9) showed complete restoration of ability to practice self-care skills in the cases with at least grade 4 strength of the muscle to be transferred, passive elbow flexion of more than 90° and well maintained active shoulder range of motion. Patients who received LD to biceps transfers presented with segmental injury to the spinal cord at C6, C6–C7 and C5–C7 levels. No procedure was performed for segmental injury to C5–Th1 in the series due to fibrous and fibrous-fat infiltrations in the LD because the LD muscle is innervated by the thoracodorsal nerve including C6, C7 and C8. No correlation could be found between the level of segmental injury to the spinal cord and outcomes of LD to biceps transposition to restore elbow flexion in patients with involved levels of C6–C7 and C5–C7.

## CONCLUSION

The LD is a reliable option for restoration of active elbow flexion in patients with arthrogryposis and segmental involvement at C6, C6–C7, C5–C7 levels with baseline donor muscle strength grading 4 and over and passive elbow flexion of at least 90°.

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