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RESTORATION OF ELBOW ACTIVE FLEXION VIA LATISSIMUS DORSII TRANSFER IN PATIENTS WITH ARTHROGRYPOSIS

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Background. Severe hypoplasia (or aplasia) of the biceps brachii is a primary cause of restriction in activities of daily living in patients with arthrogryposis.
Aim. To estimate the possibility of restoring elbow active flexion via a latissimus dorsii transfer in patients with arthrogryposis.
Materials and methods. From 2011 to 2018, we restored active flexion of the elbow via a latissimus dorsi transfer to the biceps brachii in 30 patients with arthrogryposis (44 upper limbs). We used different regimes including clinical examinations, EMG donor and recipient sites, and CT of the chest wall and shoulder.
Results. The mean age of the patients was 4.0 ± 2.4 years, and the follow-up period was 3.2 ± 1.9 months. Follow-up results were available for 26 patients (30 upper limbs). The active postoperative elbow motion was 90.5 ± 14.9°. Elbow extension limitation occurred in 51% of cases (12.8 ± 4.3°) without any problems in activities of daily living. In total, 55.6% of patients had good results, 33.3% had satisfactory results, and 11.1% had poor results.
Discussion. Our latissimus dorsi transfer results were comparable with those of other authors. Transposition of the latissimus dorsi to the biceps brachii restores sufficient flexion of the elbow without severe elbow flexion contractures.
Conclusions. We suggest pedicle monopolar latissimus dorsi transfer as a reliable therapeutic option to restore active elbow flexion in patients with arthrogryposis having passive elbow flexion of 90° or higher before operation and donor muscle strain grade 4 or higher.

Keywords: arthrogryposis; elbow; flexion; muscle transfers.

ИСПОЛЬЗОВАНИЕ ШИРОЧАЙШЕЙ МЫШЦЫ СПИНЫ ДЛЯ ВОССТАНОВЛЕНИЯ АКТИВНОГО СГИБАНИЯ В ЛОКТЕВОМ СУСТАВЕ У БОЛЬНЫХ С АРТРОГРИПОЗОМ

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Введение. Одной из основных проблем, обусловливающих ограничение или невозможность самообслуживания пациентов с артрогрипозом, является отсутствие активного сгибания в локтевом суставе в связи с тяжелой гипоплазией (аплазией) сгибателей предплечья, и прежде всего двуглавой мышцы плеча.
Цель — оценить возможность восстановления активного сгибания предплечья у больных с артрогрипозом путем монополярной пересадки широчайшей мышцы спины (ШМС).
Материалы и методы. С 2011 по 2018 г. в ФБГУ «НИДОИ им. Г.И. Турнера» было выполнено восстановление активного сгибания в локтевом суставе у 30 больных с артрогрипозом (44 верхние конечности) путем монополярной пересадки ШМС. Проводилось клиническое, нейрофизиологическое исследование (ЭМГ) донорской и реципиентной областей, КТ-исследование грудной клетки, плеча в разных режимах.
Результаты. Возраст пациентов на момент операции составил от 1 года до 10 лет (4,0 ± 2,4). Результаты лечения были изучены у 26 человек (36 верхних конечностей) в сроки от 1 до 7 лет после операции (3,2 ± 1,9). При оценке в отдаленные сроки после операции активное сгибание в локтевом суставе составило 90,5 ± 14,9°. Дефицит разгибания в локтевом суставе после хирургического вмешательства увеличился в 51 % случаев и со-

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ставил 12,8 ± 4,3, что не привело к ограничению возможности пациента осуществлять основные бытовые действия. Хорошие результаты были отмечены в 55,6 % случаях, удовлетворительные — в 33,3 %, неудовлетворительные — в 11,1 %.

Обсуждение. Данное исследование показало, что транспозиция ШМС в позицию сгибателей предплечья позволяет восстановить достаточное сгибание предплечья без формирования тяжелых сгибательных контрактур в локтевом суставе, что соответствует данным других авторов.

Заключение. Монополярная пересадка ШМС дает возможность восстановить активное сгибание в локтевом суставе у больных артрогрипозом в тех случаях, когда сила донорской мышцы составляет 4 балла и более, а пассивное сгибание в локтевом суставе — не менее 90°.

Ключевые слова: артрогрипоз; локтевой сустав; сгибание; пересадки мышц.

Introduction

Arthrogryposis is one of the most severe congenital malformations of the musculoskeletal system, characterized by congenital contractures of two or more joints, muscle hypotrophy or atrophy, and damage to the motor neurons of the spinal cord.

One of the primary problems that determine the limitation or inability to perform activities of daily living independently in these patients is the lack of active flexion in the elbow joint due to severe hypoplasia (aplasia) of the forearm flexors, especially that of the shoulder biceps. The absence of only active flexion in the elbow joint compromises the function of the upper limb by 30%, which, in combination with impairment to adjacent segments, results in severe disability [1].

Restoration of the lost function of forearm flexion is possible via autografting of the muscles of various (the most intact) donor areas; the broadest muscle of the back (SMC) is commonly used [2–6].

Most studies describe the restoration of active flexion in the elbow joint via grafting of BMB, primarily in patients with brachial plexus injuries or poliomyelitis consequences. Few studies have examined patients with arthrogryposis, warranting further research in this field [4–6].

The present study aimed to evaluate the possibility of restoring active forearm flexion in patients with arthrogryposis via monopolar BMB grafting.

Material and methods

Restoration of active flexion in the elbow joint was performed for 30 patients with arthrogryposis (44 upper limbs) using monopolar grafting of BMB between 2011 and 2018 in the Turner Scientific and Research Institute for Children's Orthopedics.

The patients were examined preoperatively and ≥1 year after the intervention. The following indicators were examined in a clinical examination: the amplitude of movements in the elbow joint (active and passive), the strength of the flexor muscles of the forearm and the donor area, and the ability to perform daily activities independently. The amplitude of elbow joint movements was determined using a goniometer. Muscle strength was evaluated on a six-point scale (0–5 points) when moving on a plane, while overcoming gravity, and with manual resistance. BMB was examined via palpation in the position of adduction, extension, and internal rotation of the shoulder. When surgery was planned for children >4 years of age, electromyography (EMG) of the muscles of the flexors and extensors of the forearm and the broadest muscle of the back was performed. Due to the complexity of the neurophysiological study and magnetic resonance imaging in young children, a computed tomography examination of the chest and shoulder in different modes was performed to assess the condition of the muscles of potential donor areas; this enabled a simultaneous understanding regarding the state of soft tissues, bones, and joints of the segment under study (Fig. 1).

Statistical analyses were performed using parametric and non-parametric tests with the software package Statistica 8.0. The normality of the data was assessed using the Kolmogorov-Smirnov test and the Shapiro–Wilk test. Average indicators were determined in the groups. When comparing the dependent pairs of groups, the nonparametric Wilcoxon criteria were used. Correlation analysis was applied with the calculation of paired Pearson correlation coefficients. When testing statistical hypotheses, the difference in the indicators at the significance level of the criterion $p < 0.05$ (95%) was considered statistically significant. The interval and point estimates of the parameters were used.

The indication for the restoration of flexors of the forearm via monopolar grafting of SMS was the

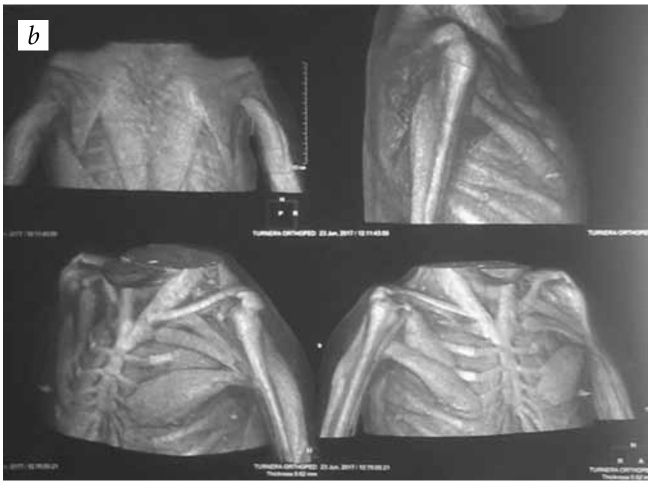

Fig. 1. CT scan of the chest and shoulder in different modes for the simultaneous visualization of the soft tissues, bones, and joints of the segment under study: *a* — visualization of the bones and the joints; *b* — visualization of the muscles of the shoulder and the chest

absence or limitation of active flexion in the elbow joint (< 90°), passive flexion of ≥90°, and strength of BMB of ≥3 points.

After the BMB was isolated on the thoracodorsal neurovascular bundle, the proximal point of the flap attachment to the upper arm bone was preserved, while the distal one was dissected. The lateral and medial margins of the muscle were sutured together, allowing more effective muscle contracture. Thereafter, the autograft was transferred through the subcutaneous canal to the front surface of the forearm and was fixed with transosseous sutures to the diaphysis of the radial bone with light tension in the supine position of the forearm and flexion at the elbow joint at 150°–160°. The limb was immobilized using a plaster cast from the fingertips of the operated hand to the upper third of the contralateral shoulder for 4 weeks. Then, the patients were prescribed rehabilitation treatment that included exercise therapy, massage, muscle electrical stimulation, and robotic mechanotherapy (ARMEO).

Results

A monopolar BMB grafting to the shoulder biceps position was performed in 30 patients with arthrogryposis (44 upper limbs). The age of the patients at the time of surgery ranged from 1–10 years (average age 4.0 ± 2.4 years).

Before the surgery, the passive flexion in the elbow joint ranged from 80°–110° (99.7° ± 7.7°), active flexion ranged from 0°–40° (17.5° ± 11.9°), extension deficit in the elbow joint ranged from 0°–30° (7.7° ± 10°), strength of the forearm flexors was 0–2 points, and the strength of BMB was 3–5 points in the patients.

Long-term postoperative assessment revealed that passive flexion in the elbow joint ranged from 80°–110° (100° ± 7.0°), active flexion ranged from 40°–110° (90.5° ± 14.7°), deficit of extension in the elbow joint ranged from 0°–45° (14.0° ± 12.9°), and the strength of the forearm flexors was 2–5 points. The deficit of extension in the elbow joint increased in 18 patients (51%) by 10°–20° (12.8° ± 4.8°) and did not limit the ability of the patient to perform basic household activities. The amplitude of the active movements in the elbow joint ranged from 40°–110° (75.4° ± 18.0°).

The results of surgical treatment of patients with arthrogryposis revealed the efficiency of restoration of active flexion in the elbow joint via monopolar grafting of the BMB in the position of the forearm flexors (Fig. 2). The Wilcoxon *t*-test for the evaluation of pre- and postoperative active flexion in the elbow joint showed a p value of 0.0072 ($p < 0.05$).

The treatment results were assessed using a modified A. Van Heest scale that included the definition of active flexion in the elbow joint,

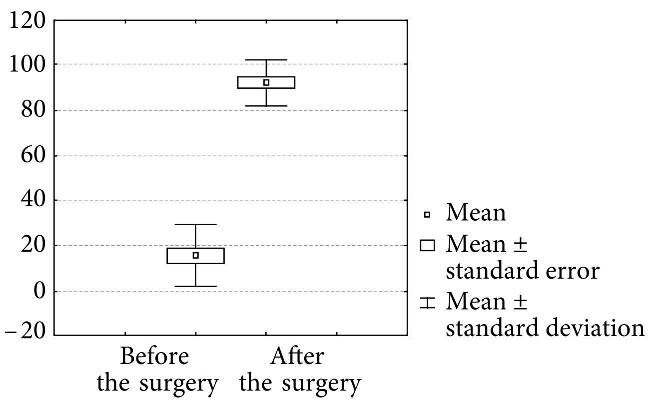

Рис. 2. Diagram of the amplitude of active flexion in the elbow joint in patients with arthrogryposis before and after the surgery

deficit of extension in the elbow joint, muscle strength of the forearm flexor, and the need to use compensatory and adaptive mechanisms during the performance of basic self-care activities [4]. According to B.F. Morrey et al. (1981), the range of movements in the elbow joint is normally 0°–145°; however, most daily activities are performed in the range of 60°–120° (the so-called "useful range"). The deficit of extension of the forearm of 60° allows the patient to use crutches, a wheelchair, and to perform activities necessary for maintaining personal hygiene [7]. According to J. Chomiak et al. (2008), in patients with arthrogryposis, active flexion in the elbow joint of 120° is usually impossible to achieve via surgery. Restoration of active flexion to 90° allows patients to eat independently. Moreover, the authors agree with A. Van Heest et al. (1998) in that passive flexion in the elbow joint of >90° is sufficient for performing surgery to restore the forearm flexors and enables arthrogryposis patients to perform self-care activities [8].

We divided the treatment results into the following three groups:

Good results: muscle strength is 4–5 points, amplitude of active movements is within the "useful" range, active flexion in the elbow joint is >90°, deficit of extension is <60°, the patient does not use or rarely uses compensatory and adaptive mechanisms for performing basic self-care activities (Fig. 3).

Satisfactory results: muscle strength is 3 points, amplitude of active movements is within the "useful" range, active flexion in the elbow joint is <90°, deficit of extension is <60°, and there is frequent use of compensatory and adaptive mechanisms when performing basic self-service activities.

Unsatisfactory results: muscle strength is 0–2 points, the amplitude of active movements is less than "useful", active flexion in the elbow joint is <90° and/or deficit of extension is >60°, and there is frequent use of compensatory and adaptive mechanisms.

Good results were observed in 20 cases (55.6%), satisfactory results were noted in 12 (33.3%) cases, and unsatisfactory results were registered in 4 (11.1%) cases.

Immediate and long-term postoperative complications were observed in 2 patients; in one case, it was neuropathy of the radial nerve that was arrested after 2 weeks with conservative treatment; the other case involved hypertrophic scars in the recipient area that required subsequent excision. Repeated intervention was performed in 1 child and consisted in the tenolysis and reinsertion of the BMB due to improper muscle strain.

Discussion

According to L. Chang et al. (1993), the indication for BMB transposition is the paralysis of the forearm flexors with intact shoulder adduction [9]. E. Zancolli and E. Miter (1973), M. Vekris (2008) recommend performing BMB transposition for the biceps of the shoulder in cases where its strength is ≥4 points [3, 10]. BMB transposition enables the restoration of sufficient flexion of the forearm without the formation of severe flexion contractures in the elbow joint [11]. Typically, patients have a postoperative deficit of extension in the elbow joint of ≤10°–15° [12–16]. According to S. Chaundry et al. (2013), best outcomes are observed in cases where before the surgery, the patient has overall amplitude of passive movements in the elbow joint and a stable shoulder joint. Poor results are associated with BMB atrophy [16].

a *b* *c* *d* *e*

Fig. 3. Good result of monopolar grafting of the broadest muscle of the back in the position of the forearm flexors bilaterally in patient C., 3 years: *a* — passive flexion in the right elbow joint using compensatory and adaptive movements; *b*, *c* — stages of the surgery; *d*, *e* — active flexion in the elbow joints 4 years after the surgery

Monopolar and bipolar BMB grafting are distinguished. With monopolar grafting, one attachment points is retained, while with bipolar, both are changed. In bipolar grafting, the proximal fixation point of the BMB is the coracoid process of the scapula (less commonly, the acromion or the collarbone), the distal one is the radial tuberosity or the biceps brachii tendon, and in the case of dislocation of the head of the radius, it is the ulnar bone [3, 4, 9, 10, 17, 18]. Recent reports mention bipolar grafting of BMB most frequently because this surgery contributes to shoulder joint stabilization, provides more physiological traction of the muscles, and enables the restoration of the normal anatomy of the reconstructed shoulder biceps [6]. Moreover, as per S. Chaundry and S. Hapyan (2013), this surgery enables improvement in the flexion in the shoulder joint. With monopolar grafting, it is impossible to improve the supination of the forearm; however, after a bipolar transplant, the postoperative muscle strength does not reach the preoperative value [16]. These methods have certain differences; however, based on the results of their comparative analysis of the treatment results in patients who have undergone bipolar and monopolar grafting of BMB, K. Kawamura et al. (2007) revealed no significant differences between them [11]. According to Sood (2017), the choice of the surgical method depends only on the surgeon's preference [17].

It is important to determine the graft length during BMB transposition. The optimal length of the graft can be determined after its mobilization with the preservation of the attachment points. In case of bipolar grafting, most authors calculate the distance from the coracoid process to the radial tuberosity in the position of flexion of the elbow joint at an angle of 90° and supination of the forearm to resolve the issue of graft length [6, 17, 18]. A. Van Heest et al. (1998) prefer to perform fixation of the translocated muscle with flexion in the elbow joint by 70°–90°; E. Zancolli, E. Miter (1973), and K. Kawamura et al. (2007) perform it a 100° flexion; M. Vekris et al. (2008) perform it at 120° flexion; and T.D. Pierce (2000) perform it with full extension [3, 4, 10, 11, 17, 19]. In cases where the BMB size is larger than necessary to restore the shoulder biceps, it is possible to take only its lateral portion on the neurovascular bundle [20].

We were able to identify only three studies performed by foreign authors that analyzed the results of BMB transposition to the position of the shoulder biceps in arthrogryposis patients [4, 6, 21].

The functional results of the surgery were evaluated by all authors on the ADL scale that considers the performance in basic household self-care activities (the ability to eat independently, drink out of a cup, comb hair, and write). Moreover, they also took into account the use of compensatory and adaptive mechanisms during the performance of these actions described by A. Van Heest et al. [4].

A. Van Heest et al. (1998) performed the BMS transposition in a bipolar version in 3 patients (4 grafts). Patient age ranged from 6–14 years (average: 11 years), and the follow-up period ranged from 1–3 years (average 1.5 years). Postoperatively, the strength of the muscles in 2 children was 4 points and that in 2 patients was 3 points. The loss of strength of the BMB after this intervention was registered in 2 patients (4–5 points before the surgery in all patients). The postoperative amplitude of active movements in the elbow joint ranged from 70°–100° (average 84°). After the intervention, the deficit of extension in the elbow joint was not observed in any case, and the amplitude of passive movements remained unchanged. In 2 cases, the treatment results were judged to be good and in 2 cases, they were satisfactory [4].

In the group of patients examined by E. Boven (2017), active flexion of the forearm was restored via monopolar and bipolar grafting of BMB in 6 patients (8 grafts). Patient age ranged from 7.8–23 years, and the follow-up period was 1.6– 8.3 years (average 4.5 years). The postoperative amplitude of active movements in the elbow joint increased from 4°–80° (average increase 43°) and averaged 46.8°. After the intervention, a deficit of extension in the elbow joint was observed from 4°–46° (average 22.3°). The amplitude of passive movements in the elbow joint after the restoration of active flexion was reduced in 50% of the patients without loss of function. Postoperatively, 2 patients had complications; in one case, it was venostasis in the "buoy" graft that required the surgical revision; in the other case, it was muscle separation. After the intervention, 2 patients showed impairment of sensitivity in 1 interdigital space. Good results were noted in 4 cases, satisfactory results were observed in 2 cases, and unsatisfactory results were noted in 2 cases [21].

R. Zargarbashi et al. (2017) retrospectively analyzed the results of restoration of active flexion in the elbow joint in 11 patients with arthrogryposis (13 limbs) via bipolar grafting of the BMB in the position of the shoulder biceps muscle. The average patient age was 5.69 ± 2.49 years, and the follow-up period was 27.3 ± 17.8 months. The postoperative amplitude of active movements was 97.7° ± 34.5°. Ten out of the 13 patients showed improved ability to perform self-care; however, compensatory and adaptive mechanisms were preserved in 11 patients. Overall satisfaction with treatment results after surgery was reported by 92.3% of the subjects. In 2 cases, muscle re-insertion was required (in one case, it was attributable to muscle separation, while in the other, it occurred because of improper direction of muscle traction) [6].

Retrospective analyses of the results for the restoration of the forearm active flexion via monopolar BMB grafting in 26 patients (34 upper limbs) for 1–7 years postoperatively (average 3.15 ± 1.9 years) enabled us to evaluate the possibility of using this autograft in arthrogryposis patients and compare our date with previous reports. Full recovery of the ability to perform self-care was observed in patients where the strength of the BMB was ≥4 points, passive flexion in the elbow was >90°, and active movements in the shoulder joint approached the physiological norm. Unsatisfactory treatment results were observed in cases where the strength of the BMB was <4 points, and active flexion and abduction in the shoulder joint was <45°. Moreover, this study revealed that the transposition of the BMB into the position of the flexors of the forearm enables the restoration of sufficient flexion of the forearm without the formation of severe flexion contractures in the elbow joint; this finding is consistent with previous reports [11–16].

Conclusion

Monopolar grafting of BMB enables the restoration of active flexion in the elbow joint in arthrogryposis patients where the strength of the donor muscle is ≥4 points and passive flexion in the elbow joint is ≥90°.

Additional information

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Conflict of interest. The authors declare no obvious and potential conflicts of interest related to the publication of this article.

Ethical review. All patients and/or their legal representatives voluntarily signed an informed consent for participation in the study, the surgical intervention, and the publication of personal data.

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