European Journal of Physical and Rehabilitation Medicine
EDIZIONI MINERVA MEDICA

EVALUATION OF HAND FUNCTION IN PATIENTS WITH UNILATERAL CEREBRAL PALSY WHO UNDERWENT MULTILEVEL FUNCTIONAL SURGERY: A RETROSPECTIVE OBSERVATIONAL STUDY

Giovanna CRISTELLA, Mariacristina FILIPPI, Maurizio MORI, Silvia ALBORESI, Adriano FERRARI

European Journal of Physical and Rehabilitation Medicine 2018 Aug 27
DOI: 10.23736/S1973-9087.18.04904-3

Article type: Original Article

Article first published online: August 27, 2018
Manuscript accepted: July 27, 2018
Manuscript revised: July 11, 2018
Manuscript received: July 9, 2017

EVALUATION OF HAND FUNCTION IN PATIENTS WITH UNILATERAL CEREBRAL PALSY WHO UNDERWENT MULTILEVEL FUNCTIONAL SURGERY: A RETROSPECTIVE OBSERVATIONAL STUDY

Giovanna Cristella*[1], Maria C. Filippi[2], Maurizio Mori[2], Silvia Alboresi[2], Adriano Ferrari[2]

[1] Children Rehabilitation Unit, IRCCS Fondazione Don Gnocchi, Firenze, Italy; [2] Children Rehabilitation Unit, Arcispedale Santa Maria Nuova, Reggio Emilia, Italy

*Corresponding author: Giovanna Cristella, Children Rehabilitation Unit, IRCCS Fondazione Don Gnocchi, via di Scandicci 267, 50143 Firenze, Italy. Email: gcristella@dongnocchi.it

1. **ABSTRACT**

Background. Hemiplegia is the most common form of Cerebral Palsy. Upper Limb is generally more affected than lower one. In fact, hemiplegic children can spontaneously acquire standing and walking ability, while manipulation remains uncertain, with severe limitations in activity and participation, which define child's functional status (International Classification of Functioning - ICF).

Several non-surgical tools are currently available to approach upper limb impairments. Studies regarding upper limb multilevel surgery in Hemiplegic Cerebral Palsy are relatively few and inhomogeneous.

Aim. The aim of this study is to propose a surgical approach based on upper limb functional level and manipulation strategy and establish whether multilevel surgery can improve segmental alignment, performance and capacity, that ICF defines as activities and participation qualifiers.

Design. This study is an observational retrospective study.

Setting. This study involves patients who referred to Children Rehabilitation Unit of IRCCS S. Maria Nuova in Reggio Emilia (Italy), along a period of four years.

Population. It involves children affected by hemiplegic cerebral palsy who underwent upper limb multilevel surgery.

Method. For each patient, we previously defined functional use of affected upper limb applying the House classification and the Ferrari one of manipulation pattern. Patients are divided into three groups: synergic hand (House 4, 5), imprisoned hand (House 3), excluded hand (House 0). We recorded goals achievement through Goal Attainment Scale and unimanual and bimanual abilities through Melbourne Assessment of Unilateral Upper Limb Function and through Assisting Hand Assessment respectively.

Results. We record 16 upper limb multilevel surgical interventions in 13 children and report their results.

Conclusion. This study suggests that surgery can induce a segmental and/or aesthetic and/or a functional change depending on manipulation pattern. It also underlines the importance to analyse results in term of spontaneous manipulation abilities and daily use.

Clinical rehabilitation impact. This study provides a preliminary guide to plan surgery in relation to segmental deformities and overall manipulation pattern and describes their feasible improvement measures. It also suggests the most useful tools to record goal achievements in modifying manipulation function.

Further controlled, randomized and prospective studies are required to support this idea.

Key words: upper limb, hand deformity, hemiplegia.

2. INTRODUCTION

Cerebral Palsy (CP) is the principle cause of childhood physical disability in industrialized societies (1/500 live births) (1). Recently, Himmelman recorded a turnaround in term of prevalence of CP forms: hemiplegic cerebral palsy is the most frequent (38%), followed by diplegia (32%) (2). Hemiplegia is the most common form of CP in term infants (more than 50%) and the second one in preterm ones (20%) (3).

The Surveillance of Cerebral Palsy in Europe terms Hemiplegic Cerebral Palsy as Unilateral Cerebral Palsy (UCP) and suggests the following diagnostic criteria: involvement of limbs on one side of the body and at least two of these: i) abnormal pattern of posture and movement, ii) increased tone (not necessarily constant) and iii) pathological reflexes (4).

The main clinical characteristic of hemiplegia is the reduction of motor repertoire on the affected side in terms of modules (meant as the elementary components of movement the child is provided with), combinations (possibility to organize the individual modules into different patterns according to space relations), and sequences (ability to assemble the individual modules according to different time relations). These early clinical signs allow a prompt diagnosis of UCP (5). Other clinical signs often reported in UCP are: the presence of associated movements, sensory and perceptive defects, attention disorders, alteration of muscle tone, muscle retractions and (early or late) bone growth alterations, elements more or less frequent, according to the different clinical forms.

In UCP, the Upper Limb (UL) is generally more affected than the lower one. Unimanual abilities of the affected UL and manipulation strategies are heterogeneous; consequently, activity and participation could be severely impaired (6). For this reason, rehabilitation should focus on improving UL competence in executing a task or an action (capacity) and in daily activity (performance). UL spasticity and/or weakness, muscle contracture and/or retraction,

limitation in joint range of motion, forearm, wrist and finger deformation, poor dexterity and motor control, lack of sensation and perception in affected limb, cause functional impairment that can be amplified by learned non-use, and auxiliary grips (7).

Several non-surgical tools are currently available; a recent systematic review describes their efficacy (8).

Studies about functional outcome after surgery in UCP are relatively few and inhomogeneous. In a recent review, Van Muster indicates that many of them consist of case series providing a low level of evidence. These studies suggest a positive effect on hand position, manipulation strategy, grip repertoire and spontaneous use, but it remains unclear whether surgery influences daily activity (9).

Van Heest explores change in functional use by House classification in a heterogeneous group (spastic/athetoid CP, quadriplegia/hemiplegia/triplegia) over a 25-year period; this author reports a functional improvement for all patients and identifies good voluntary control as a positive prediction factor (10). In a recent study, the same author compares efficacy of tendon transfer surgery versus botulinum toxin injection and ongoing therapy and records an improvement at twelve months of follow-up for surgical group in Shriners Hospital Upper Extremity Evaluation, dynamic positional analysis, Paediatric Quality of Life Inventory - module domain of movement- and Canadian Occupational Performance Measure score for satisfaction (11). This research shows surgery benefit in UL function, but mixes three different kinds of rehabilitation instruments that, according to us, have different indications: physiotherapy to modify function, botulinum toxin injection to reduce spasticity and surgery to reduce/modified muscle retraction, bone and joint deformities. Smitherman et al. compare functional outcomes following UL surgery in a retrospective case control series in hemiplegic CP, showing a significant improvement in dynamic segmental alignment and in spontaneous use (12).

The aim of this study is to establish whether multilevel UL surgery in children with UCP improves segmental alignment, UL function (capacity) and UL daily use (performance) when indication for surgical treatment is performed in relation to the assessed functional level and not only to segmental deformity. Specific and individual goals were recorded through Goal Atteinment Scale (GAS). When possible, we record unimanual and bimanual abilities through

Melbourne Assessment of Unilateral Upper Limb (MUUL) and Assisting Hand Assessment (AHA) respectively.

3. METHODS

3.1 Study design

This is a retrospective observational study conducted in the Children Rehabilitation Unit of IRCSS S. Maria Nuova Hospital in Reggio Emilia, Italy, along a four years period. Approval was obtained by ethics Committee on research.

a. Participants

The inclusion criteria were: Clinical (UCP as defined by Surveillance of Cerebral Palsy in Europe, previously described), and instrumental (brain MRI) diagnosis of UCP; history of multilevel upper limb surgery performed at IRCSS S. Maria Nuova Hospital in Reggio Emilia, Italy; video-recording about spontaneous manipulation activity before and after multilevel surgery, to establish whether surgical goals were achieved. Patients with previous diagnosis of mental retardation and/or behavioural disorder were excluded.

While assessment and surgical interventions took place in Children Rehabilitation Unit, IRCCS S. Maria Nuova Hospital in Reggio Emilia, the following rehabilitation program was delivered at the rehabilitation centres, where each child came from. In fact, UL multilevel surgery was followed by an individualized physiotherapy treatment and/or personalised orthosis.

For each patient, before surgery (T0), we defined manipulation ability referring to House classification, reported in table 3.1.1 (House Functional Classification System; House et al, 1981) (13).

Tab. 3.1. 1 House Functional Classification

In recent years, we attempted to validate a classification of manipulation in UCP. It describes five patterns of manipulation by analysing hand kinematic profile and functional use (Ferrari et al. 2016) (14): Integrated, Semi-functional, Synergic, Imprisoned, Excluded.

Main characteristics of each class and corresponding clinical approach are summarized in the table 3.1.2

Tab. 3.1. 2 Ferrari manipulation pattern

As shown in the table above, Synergic, Imprisoned and Excluded hands have surgical indications.

It is possible to establish a correspondence between Ferrari manipulation pattern classification and House classification: Integrated hand corresponds to House level 8, Semifunctional hand to House level 7, Synergic hand to House level 4- 5- 6, Imprisoned hand to House level 1- 2- 3, Excluded hand to House level 0.

b. Outcome

Outcomes were recorded using GAS. GAS is a method of measuring individual progress towards specific, measurable, acceptable, relevant and time-related goals. It is a 5-point scale: “0” represents the expected level of success, “+1” and “+2” the achievement of more than the expected, “-1” and “-2” a worse result than expected. Each goal has a weight defined as Importance x Difficulty (15, 16).

By assigning “0”, GAS assumes the value 50. By assigning “+1” and “+2” it assumes values progressively greater than 50, while assigning “-1” and “-2” it assumes values progressively less than 50 (17).

For each patient, we defined three goals: GASP (goal concerning postural and segmental alignment), GASF (goal concerning spontaneous functional use of affected UL) and GASA (goal describing patient satisfaction in term of autonomy in daily activities). They are summarized in GASTOT, which expresses a weighted average of all three (in other words, an average that considers the weight of each goal).

GAS was defined considering not only limitation in ROM and deformation, but also House level and Ferrari manipulation pattern, and specific patient demand.

We estimated goal achievement by calculating GAS after three (T1) and twelve (T2) months. For some patients we have intermediate data. We compare mean values of GASP, GASF, GAS A and GASTOT.

As a secondary outcome, when possible, some patients were assessed through MUUL and AHA. MUUL (18) and AHA (19) are scales frequently used in rehabilitation: the first measures unilateral upper limb function while the second one measures bimanual activities.

4. RESULTS

We recorded 16 UL multilevel surgical interventions in 13 children with UCP. One patient underwent 3 surgical interventions, but we only have video-recording of two of them; another patient underwent 2 surgical interventions. Two patients were excluded due to mental retardation, which required a different rehabilitation approach. Children came from different Italian Rehabilitation centres.

Tab. 4. 1 Sample features

Table 4.1 summarizes the sample characteristics.

House level distribution (level: percentage): 8:0%; 7:0%, 6:0%, 5:15%, 4:31%, 3:39%, 2:0%, 1:0%, 0:15%.

Ferrari manipulation pattern (pattern: percentage): integrated hand: 0%, semifunctional hand: 0%, synergic hand: 46%, imprisoned hand: 36%, excluded hand: 18%.

Following, multilevel surgical interventions are briefly described group by group.

Synergic hand group. Patient 4 was subjected to pronator teres release. The other patients with synergic hand (6, 7, 9.1, 9.2, 11) received a more substantial approach to their wrist deformities: patient 6 was subjected to epitrochlear muscle tenotomy and ulnar flexor carpi release, patient 7 and 11 to transfer of ulnar flexor carpi to extensor carpi radial brevis, patient

9 to carpus arthrodesis and ulnar flexor carpi tenotomy first and to plate removal and flexor pollicis brevis myotomy in a second time.

Imprisoned hand group. Patient 1 underwent surgical intervention in two time: first, flexor pollicis brevis, ulnar flexor carpi, radial flexor carpi, brachialis biceps release together with extensor brevis carpi radialis retention and in a second time, because of relapse in term of segmental alignment, adhesion release, adductor pollicis tenotomy and transfer of ulnar flexor carpi and radial flexor carpi to extensor brevis carpi radialis. Patient 8 required surgical approach to wrist flexion deformity through ulnar flexor carpi and radial flexor carpi release, together with pronator teres and finger flexor digitorum profundus release and palmar gracilis tenotomy. The other patients with imprisoned hand were subjected to palmar gracilis tenotomy (patient 5) and to brachialis biceps fasciotomy (patient 10).

Excluded hand group. Patient 2 and 3 were respectively subjected to brachialis biceps fasciotomy, pronator teres and superficial finger flexor release, transfer of ulnar flexor carpi to extensor carpi radial brevis, extensor ulnaris carpi brevis tenotomy the first and metacarpophalangeal joint arthrodesis, brachialis biceps release, ulnar flexor carpi, flexor pollicis brevis, pronator teres release, palmar gracilis release the second.

We can notice that patient with synergic hand were subjected to wrist and forearm surgery, but no surgery was performed for fingers, except for thumb. Imprisoned hand patients required surgical approach to wrist or thumb or finger deformities with a case of relapse when surgery is more conservative overall for thumb and wrist. Both patients with excluded hand required intervention to obtain wrist alignment and to reduce forearm pronation.

After surgery, physiotherapy and splinting were defined considering House level, Ferrari manipulation pattern, spasticity, weakness and surgical techniques. This constituted a first phase, based on daily specific stretching manoeuvres and a second phase based on therapeutic activities consisting of a series of goal-directed actions, spontaneously and voluntarily executed by patients, under therapist guide, in order to achieve functional solution in manipulation activity. Exercises take into account each child coping solutions. When suitable, such as in case of tendon transfer, arthrodesis and muscle weakness, in early stages, continuous positional splint use was recommended.

Physiotherapeutic programme was planned by an experienced paediatric research physiotherapist and delivered to case physiotherapist.

Table 4.2. Results.

Table 4.2 reports results for each group. All patient with synergic hand achieve all goals (GASP, GASF, GASA) in T1 (except for patient 4) and maintained the results at follow up (T2) in fact GAS mean values are greater than 50 for all GAS at T1 and T2, except for GASA at T1, influenced by negative result of patient 7, not confirmed at follow up. Patient 4 negative result is non-acceptance of hand appearance, resulting in a very poor spontaneous hand use and compliance to physiotherapeutic programme, instead of a good motor repertoire. In other words, patients' expectations were satisfied, together with both better segmental alignment and improvement of manipulation abilities. GASTOT mean values confirm that preoperative goals were achieved.

We have data about MUUL at T0 and T1 for patients 6 and 7 but not about MUUL at T2. MUUL variation score is considered significant when equal to 12% or more (20). An intermediate MUUL was recorded for patient 7 after six months from surgery, its score is 92.44%. We record a positive but not significant variation for patient 7 after three months and a significant variation after six months. A significant modification was recorded in MUUL for patient 6 at T1. Patients 4 and 11 were evaluated with AHA. AHA variation score can be considered significant when equal or greater than 4 in raw score (21). At T1, both patients recorded a significant variation. Patient 11 repeated evaluation in T2, recording a non-significant improvement.

Into imprisoned hand group, postural goals were achieved for all patient in T1 and maintained at follow up, with a GASP mean value greater than 50 at T1 and T2. Patient 1 needed a second-time surgery (1.2) because of loss of segmental alignment after first surgical time (1.1), while maintaining good functional results. In other words, Patient 1 required two surgical intervention to achieve segmental and functional goals. GASF mean value is very positive in T1 (overall because of the great results of patient 1) but this result is not confirmed at T2, with a negative GASF mean value. All patients' expectation was achieved at T2, except for patient 10 (this can be explained by relapse of segmental deformities), with GASA mean values greater than 50 at T1 and T2. GASTOT mean values confirm that preoperative goals were achieved.

Patient 1 was also evaluated by MUUL. After the first surgical session, the patient showed a positive but insignificant difference in T1 and T2. After second surgical session (1.2), patient recorded a positive but not significant difference after three months; comparing this to the MUUL score in T1, the difference was 11.48%, very close to 12% (limit of statistical significance). Patient 5 recorded a positive score in MUUL in an intermediate time -six months after surgery- (not reported in the table 4.2) confirmed at T2, but with a not significant score.

About excluded hand group**,** both patients satisfied segmental alignment goals in T1, but both recorded a relapse in T2. Positive GASP and GASF mean value at T1 was not confirmed at follow up. GASA follows an opposite trend, with a very positive mean value at T2. Overall results (GASTOT) were satisfactory and were influenced overall by GASA positive values. Patient 2 was evaluated by MUUL, recording a positive but not significant improvement in T2.

5. DISCUSSION

By results, we can describe some differences between three groups.

As shown, into synergic hand group, multilevel surgery gives a gain in term of segmental alignment and manipulation ability. Fulcrum joint in synergic strategy manipulation seems to be wrist, so it's important to obtain an optimal wrist alignment. In these patients, pattern manipulation contains a substantial modifiability and surgery allows to express it. According to us, the better surgical approach to obtain a good wrist alignment and to express functional modifiability is transferring ulnar flexor carpi to extensor carpi radial brevis (arthrodesis hinders wrist motion and can make synergic strategy difficult). In fact, the results obtained in two patients with synergic hand (patient 7 and 11) are remarkable. Probably this transfer gives a better wrist alignment so that fingers flexor muscles are in a favourable position to take advantage in synergic strategy; furthermore, it breaks pathological synergies by putting a muscle to function with its original antagonist and maybe induces cortical reorganization by accessing to motor modules previously unused and reorganizing them in more complex combinations and sequences. In other words, if there are grounds (modifiability, learning ability and motivation), it allows the maximum expression of manipulation function for each patient. This happens regardless of surgical technique, in fact in these two patients we used

two different techniques: Carlson and Green transfer. Realistically, the grater is patient age, the less is function modifiability. It could be useful to carry out transfer in earlier age (our patients are 11 and 13) but there are two main risks: manipulation mistakes are not yet permanent, so results cannot be predictable, technique difficulties related to small tendons and co-contraction in case of high level of spasticity.

Transfer is useful to modify function in patients with synergic hand but not in patient with imprisoned and excluded hand because of their lower modifiability.

GASA generally follows the positive trend of GASP and GASF (except for patient 4, who always refers trouble in accepting his disability).

Into imprisoned hand group we describe high risk or recurrence and we obtain the best results in patient who underwent surgery in two times. For this reason, it seems reasonable to propose surgery as later as possible in order to plan a one-time surgery by a "more aggressive" technique.

Looking at Patient 10, he records negative results at T2 in all three GAS and so in GASTOT. These negative results could be explained by considering the "minimal" surgical approach that allowed a precocious recurrence. In other words, to prevent recurrence, tenotomy seems to be better than fasciotomy, wrist transfer (for example transferring ulnar flexor carpi to extensor carpi radial brevis) seems to be better than flexor lengthening to obtain wrist tendon-suspension. For example, patient 1.2 records a small but gradual gain suggesting efficacy of transferring ulnar flexor carpi and radial flexor carpi to extensor carpi radial brevis in getting wrist tendon- suspension. Looking at surgery plan, we can suppose that thumb and wrist are the fulcrum of imprisoned hand manipulation ability. In fact, when thumb is imprisoned into palm, it induces grasp reflex and limits the possibility of passive hand grip. Good wrist alignment puts fingers flexor in a more advantageous position to grasp. Both Patient 8 and 10 recorded a negative trend in GASF; this suggest that, even if it is possible to obtain a functional change in imprisoned hand, it is hard to stabilize the results, suggesting changeability in segmental alignment but not in functional spontaneous use. In particular, the outcome of patient 8, whose negative trend in GASF is opposed to positive trend in GASP confirms this: even if multilevel surgical approach can modify local alignment, the lack of motor repertoire, typical of imprisoned hand, severely conditions functional prognosis. In other words, obtaining a good segmental alignment does not ensure obtaining a manipulation ability gain

because of the narrow space of modifiability. In Patient 1 functional gain was probably due to adductor pollicis tenotomy, which reduces self-stimulation of grasping, and to ulnar flexor carpi and radial flexor carpi transfer onto extensor brevis carpi radialis because it puts finger flexor muscles in a more advantageous position to execute grasp. We have also to underline that this patient showed a very high level of compliance and motivation in rehabilitation programme; those give a great guarantee of functional gain.

About GASA, its trend generally follows GASP: imprisoned hand has a strong visual impact and patients seem to be satisfied of getting it better. This remind us firstly the strong imprisoned hand aesthetic impact on patients, probably more than functional ones (because of hyper-specialization of unaffected hand), and secondly the great awareness of its poor modifiability. So, changing hand appearance could be a good reason to surgery.

About excluded hand group, differently from the others, we record no possibility to induce a functional long-term gain. GASF results confirm that is not possible to modify functional use because of poor motor and sensitive repertoire. The recurrence of segmental deformities suggests a more extended use of positional splint since early age and a more aggressive surgical approach (i.e. wrist arthrodesis). Looking at GASA, we can say that patient expectations are satisfied probably because of lower functional expectations and a greater awareness of narrow modifiability, compared to patients belonging to the other two groups. These patients indeed early develop very effective strategies of manipulation by unaffected UL hyper- specialization.

Comparing these groups, we can underline the positive results of GASP mean values after three months and at follow up (T2), except for excluded hand patient who recorded relapse and we can observe the grater results in synergic hand. Comparing GASF mean values between group, we can conclude that significant change at follow up is possible only in synergic hand group. GASA mean values record negative results at T1 for synergic and excluded hand, maybe due to difficulties related to post-operative time; at T2, we record GASA mean values greater than 50 in all groups, with the best result in the excluded hand group, probably because of the great awareness of low modifiability, as previously described. GASTOT mean records positive values at T1 and T2 for all groups, confirming goals achievement when they are established according to House level and Ferrari manipulation pattern.

In conclusion, we can suppose that surgery can positively modify segmental alignment and spontaneous hand function in synergic hand group, sometimes leading to unexpected results; surgery can also favourably influence imprisoned hand segmental alignment, carrying sometimes a narrow functional gain. Excluded hand group has no space of functional modifiability and has a high risk of segmental deformities recurrence. Generally patient satisfaction is related to function in synergic hand and to segmental alignment and aesthetic appearance in the other two groups; the grater is poorness in motor repertoire, the greater is awareness of little modifiability.

Compliance to rehabilitation programme and its appropriateness affect final result; for this reason, it requires a specialized team and patient motivation should be always considered before surgery.

Following, some observations about evaluation instrument. We can suppose some differences between GAS, MUUL and AHA in term of recording changes following treatment. Six patients were evaluated by MUUL. One of them records a significant change in MUUL score, three of them a better but no significant change, one of them a worse score (one patient did not perfume MUUL at follow up) whereas GAS values show the achievement of surgical goals (GASTOT) for all of them (except for patient 5 who came back to pre-surgical situation but did not have a worsening; patient 1.1 achieves goals by two-time surgery). In other words, this study suggests that MUUL has low specificity in recording manipulation abilities change probably because it is made up of mono-manual tasks in which unaffected UL is never involved. This manipulation strategy is not congruent with hemiplegic child strategy: UCP child employs hemiplegic hand only in supporting other hand's activity or when dominant hand is not available for manipulation. Only two patients performed AHA; both recorded significant results in T1, and positive but insignificant result in T2. AHA seems to be a more specific scale because it measures how effectively these children use the affected hand in bimanual performance; indeed, it provides bimanual tasks and probes also affected UL initiative of use and coordination. However, while the MUUL scale evaluates the capacity (the person's ability to execute a task or an action on the highest probable level of functioning that a person may reach in a standardized environment), AHA measures performance (the person's ability to execute a task or an action in a real-life environment, e.g. semi-structured play

session). AHA tasks are always performed after examiner request so do not describe spontaneous UL use. Hence, new scales, based on spontaneous play observation, is needed. Video-recording spontaneous manipulation activities is a good instrument to evaluate impact on daily living.

GAS is a good evaluation instrument because it allows rehabilitation team to establish for each patient tailored goals, but it requires a proper training to avoid over-estimation and under-estimation mistakes. Training on House and Ferrari classification is important to establish manipulation ability modifiability and the better instrument (physiotherapy, botulinum toxin and multilevel surgery) to obtain it.

5.1 Study limit

Sample size is little because of the recent experience of our group in UL multilevel surgery. We lost some patient at follow up because patients referring to Children Rehabilitation Unit IRCSS S. Maria Nuova Hospital, Reggio Emilia come from all part of Italy, so some of them has organizational difficulties and were followed by local hospital. Some patient did not have MUUL or AHA evaluation because they consider them repetitive and boring and were less engage in them, with risk of a worse score.

Instrument used for evaluation are sometimes little sensitive to record changes; this leads to the need of a more appropriate scale and to a more extensive use of narrative medicine.

6 CONCLUSION

This study provides a preliminary guide to plan surgery as a function of manipulation pattern and not only of segmental deformities.

It suggests that surgery induces: a) a segmental and functional gain in synergic hand, b) a segmental, aesthetic and sometimes minimal functional gain in imprisoned hand and c) a segmental, aesthetic gain in excluded hand. Surgery induce a positive change in capacity and

performance in synergic hand UCP and sometimes in imprisoned hand group. However hand appearance should be consider as indication for surgery.

This study also underlines the importance to analyse results in term of spontaneous manipulation abilities and daily use; for this reason, according to us, video-recording spontaneous hand use is the gold standard.

GAS is a good instrument to identify tailored goals for each patient and to establish if they are achieved or not. Other scales do not consider CP form and their modifiability space and sometimes they do not follow CP children strategy.

Further controlled prospective studies are required to justify this kind of surgical approach.

REFERENCES

1. Surman G, Hemming K, Platt MJ, Parkes J, Green A, Hutton J, Kurinczuk JJ. Children with cerebral palsy: severity and trends over time. Paediatr Perinat Epidemiol. 2009; 23(6):513-21.

2. Himmelmann K, Hagberg G, Uvebrant P. The changing panorama of cerebral palsy in Sweden. X. Prevalence and origin in the birth-year period 1999–2002. ActaPædiatrica 2010 Sep;99(9):1337-43

3. Hagberg B, Hagberg G, Olow I. The changing panorama in cerebral palsy in Sweden. VII Prevalence and origin during the birth period 1987- 1990. ActaPaediatrica 1996 Aug;85(8):954-60

4. Surveillance of Cerebral Palsy in Europe (SCPE) Surveillance of cerebral Palsy in Europe: a collaboration of cerebral palsy surveys and register. DevMed Child Neurol 2000 Dec;42(12):816-24.

5. Ferrari A., Cioni G Le forme spastiche della PCI. (2005) Milano: Springer,2005

6. Sgandurra G, Ferrari A, Cossu G, Guzzetta A, Fogassi L, CioniG. Randomized trial of observation and execution of upper extremity actions versus action alone in children with unilateral cerebral palsy. NeurorehabilNeuralRepair. 2013 Nov-Dec;27(9):808-15.

7. Ferrari A, Maoret A.R, Muzzini S, Alboresi S, Lombardi F, Sgandurra G, Paolicelli PB, Sicola E, Cioni G. A randomized trial of upper limb botulinum toxin versus placebo injection combined with physiotherapy in children with hemiplegia. Research in developmental disabilities 2014; (35):2505 - 2513

8. Sakzewski L, Ziviani J, Boyd RN. Efficacy of upper limb therapies for unilateral cerebral palsy: a meta-analysis. Pediatrics. 2014 Jan;133(1):e175-204. doi: 10.1542/peds.2013-0675. Epub 2013 Dec 23.

9. van Munster JC, Maathuis KG, Haga N, Verheij NP, Nicolai JP, Hadders-Algra M. Does surgical management of the hand in children with spastic unilateral cerebral palsy affect functional outcome? Dev Med Child Neurol. 2007 May;49(5):385-9

10. Van Heest AE1, House JH, Cariello C. Upper extremity surgical treatment of cerebral palsy.J Hand Surg Am. 1999 Mar;24(2):323-30

11. Van Heest AE, Bagley A, Molitor F, James MA. Tendon Transfer Surgery in Upper-Extremity Cerebral Palsy Is More Effective Than Botulinum Toxin Injections or Regular, Ongoing Therapy. J Bone Joint Surg Am 2015 Apr 1;97(7):529-36

12. Smitherman JA, Davids JR, Tanner S, Hardin JW, Wagner LV, Peace LC, Gidewall MA. Functional Outcomes Following single-event Multilevel Surgery of the Upper Extremity for Children with Hemiplegic Cerebral Palsy. J Bone Joint Surg Am. 2011 Apr 6;93(7):655-61

13. House JH, Gwathmey FW, Fidler MO. A dynamic approach to thumb-in-palm deformity in cerebral palsy. J Bone Joint Surg (Am) 1981 Feb;63(2):216-25

14. Ferrari A., Benedetti M.G., Mori M., Alboresi S. L'arto superiore nella paralisi cerebrale infantile. (2016) Padova: Piccin Nuova Libraria, 2016

15. Kiresuk T, Sherman R. Goal attainment scaling: a general method of evaluating comprehensive community mental health programs. Community Ment Health J 1968 Dec;4(6):443-53

16. Steenbeek D, Ketelaar M, Lindeman E, Galama K, Gorter JW. Interrater reliability of goal attainment scaling in rehabilitation of children with cerebral palsy. Arch Phys Med Rehabil. 2010 Mar;91(3):429-35.

17. Forbes DA. Goal Attainment Scaling. A responsive measure of clinical outcomes. J Gerontol Nurs 1998; 24 (12): 34-40.

18. Johnson LM, Randall MJ, Reddihough DS, Oke LE, Byrt TA, Bach TM. Development of a clinical assessment of quality of movement for unilateral upper-limb function. Dev Med Child Neurol. 1994 Nov;36(11):965-73.

19. Holmefur M, Aarts P, Hoare B, Krumlinde-Sundholm L. Test-retest and alternate forms reliability of the assisting hand assessment. J RehabilMed 2009; 41: 886–891

20. Johnson LM, Randall MJ, Reddihough DS, Oke LE, Byrt TA, Bach TM. Development of a clinical assessment of quality of movement for unilateral upper-limb function. Dev Med Child Neurol. 1994 Nov 36(11):965-73

21. Holmefur M, Aarts P, Hoare B, Krumlinde-Sundholm L. Test-retest and alternate forms reliability of the assisting hand assessment. J RehabilMed 2009; 41: 886–891

TITLES OF TABLES

Tab. 3.1. 3 House Functional Classification

Tab. 3.1. 4 Ferrari manipulation pattern

Tab. 4. 2 Sample features

Tab. 4. 3 Data “group by group”

TABLES

Tab. 3.1. 5 ***House Functional Classification***

House Functional Classification		
Grade	Designation	Activity Level
0	Does not use	Does not use
1	Poor passive assist	Uses as stabilizing weight only
2	Fair passive assist	Can hold on to object placed in hand
3	Good passive assist	Can hold on to object and stabilize it for use by the other hand
4	Poor active assist	Can actively grasp object and hold it weakly
5	Fair active assist	Can actively grasp object and stabilize it well
6	Good active assist	Can actively grasp object and then manipulate it against other hand
7	Spontaneous use, partial	Can perform bimanual activities easily and occasionally uses the hand spontaneously
8	Spontaneous use, complete	Uses hand completely independently without reference to the other hand

Tab. 3.1. 6 Ferrari manipulation pattern

	Integrated	Semi-functional	Synergic	Imprisoned	Excluded
Main core	Subterminal/ terminal	Subterminal lateral	Stereotypically	Indirect grasping	Functionally

	pinch, Possible selective finger movements, Mastery of intrinsic motricity for manual exploration,	pinch with basically adduced thumb, Scarce/absent intrinsic motricity	expressed grasping and release within flexion and extension synergies. Active loading of object.	(passive loading) Imprisoned thumb or positioned underneath palm.	ineffective or negligible grasping
Physiotherapy	Useful in early age	Useful through perceptive exercises	Useful for both affected and unaffected hand	Limited effectiveness	useless
Botulinum toxin	Rarely required	Occasionally required	Required to reduced forearm pronation and wrist ulnar deviation	Useful to inhibit spasticity hand	Rarely required
Orthoses	Useless	Occasionally nocturnal splint	Occasionally thumb and wrist dynamic splint are needed	Occasionally nocturnal splints are useful	Wrist splint are sometimes required
Multilevel upper limb surgery	Contraindicated	No indication	Useful for muscle retraction	Useful for muscle retraction and bone deformity	Aesthetic indication

Tab. 4. 4 Sample features

PZ ID	Age at time of study	Age at time of surgery	House	Ferrari manipulation pattern
1.1	17	13	3	IMPRISONED
1.2	17	15	3	IMPRISONED
2	19	16	0	EXCLUDED
3	16	13	0	EXCLUDED
4	15	12	4	SYNERGIC
5	21	18	3	IMPRISONED
6	16	14	4	SYNERGIC
7	14	13	5	SYNERGIC
8	15	13	3	IMPRISONED
9.1	24	19	4	SYNERGIC
9.2	24	21	4	SYNERGIC
10	11	10	3	IMPRISONED
11	12	11	5	SYNERGIC

Tab. 4.2 Data "group by group"

ID	T0		T1						T2					
PZ	MUUL	AHA	GASP	GASF	GASA	GASTOT	MUUL	AHA	GASP	GASF	GASA	GASTOT	MUUL	AHA
SYNERGIC HAND GROUP (HOUSE 4, 5)														
4		42	53,67	43,59	50,00	47,84		53	50,00	62,40	43,80	52.1		
6	31,34%		62,35	50,00	50,00	55,83	63,87%							
7	73,64%		68,45	60,87	31,64	64,31	82,35%		76,52	50,00	61,64	63,27		
9.1			50,00	50,00	50,00	50,00			50,00	60,00	50,00	53,33		
9.2			70,00	60,08	50,00	57,55								
11		70	62,27	73,27	50,00	64,91		79	63,57	57,63	61,74	62.27		73
MEAN			61.12	56.30	46.94	56.74			60.02	57.50	54.29	57.74		
IMPRISONED HAND GROUP (HOUSE 3)														

1.1	41.8%	55.10	63.39	43.20	53.44	48.36%	36.40	50.00	50.00	49.72	46.72%
1.2	46.72%	64.41	68.70	62.40	65.04	53.28%	69.39	68.70	62.40	66.83	
5	60.75%	54.56	28.67	50.70	43.93		50.00	42.24	51.41	47.65	68.91%
8		54.91	57.36	54.56	55.55		74.54	47.55	54.56	58.61	
10		58.26	56.20	62.40	58.94		38.06	31.39	37.60	35.75	
	MEAN	57.45	61.99	54.65	55.83		53.68	47.97	51.19	51.71	
EXCLUDED HAND GROUP (HOUSE 0)											
2	38,52%	52,10	43,80	39,43	47,77		48,80	48,30	51,75	61,17	
3		50,75	70,93	58,64	59,79		44,78	43,96	70,17	49,56	
	MEAN	51.43	57.37	49.04	53.78		46.79	46.13	60.96	55.37	

Tab. 3.1. 1 ***House Functional Classification***

House Functional Classification		
Grade	Designation	Activity Level
0	Does not use	Does not use
1	Poor passive assist	Uses as stabilizing weight only
2	Fair passive assist	Can hold on to object placed in hand
3	Good passive assist	Can hold on to object and stabilize it for use by the other hand
4	Poor active assist	Can actively grasp object and hold it weakly
5	Fair active assist	Can actively grasp object and stabilize it well
6	Good active assist	Can actively grasp object and then manipulate it against other hand
7	Spontaneous use, partial	Can perform bimanual activities easily and occasionally uses the hand spontaneously
8	Spontaneous use, complete	Uses hand completely independently without reference to the other hand

Tab. 3.1. 1 Ferrari manipulation pattern

	Integrated	Semi-functional	Synergic	Imprisoned	Excluded
Main core	Subterminal/ terminal pinch, Possible selective finger movements, Mastery of intrinsic motricity for manual exploration,	Subterminal lateral pinch with basically adduced thumb, Scarce/absent intrinsic motricity	Stereotypically expressed grasping and release within flexion and extension synergies. Active loading of object.	Indirect grasping (passive loading) Imprisoned thumb or positioned underneath palm.	Functionally ineffective or negligible grasping
Physiotherapy	Useful in early age	Useful through perceptive exercises	Useful for both affected and unaffected hand	Limited effectiveness	useless
Botulinum toxin	Rarely required	Occasionally required	Required to reduced forearm pronation and wrist ulnar	Useful to inhibit spasticity hand	Rarely required

			deviation		
Orthoses	Useless	Occasionally nocturnal splint	Occasionally thumb and wrist dynamic splint are needed	Occasionally nocturnal splints are useful	Wrist splint are sometimes required
Multilevel upper limb surgery	Contraindicated	No indication	Useful for muscle retraction	Useful for muscle retraction and bone deformity	Aesthetic indication

Tab. 3.1. 1 correspondence between Ferrari manipulation pattern classification and House classification

HOUSE CLASSIFICATION	FERRARI MANIPULATION PATTERN CLASSIFICATION
House 0	Excluded hand
House 1,2,3	Imprisoned hand
House 4,5,6	Synergic hand
House 7	Semifunctional hand
House 8	Integrated hand

Tab. 4. 1 Sample features

PZ ID	Age at time of study	Age at time of surgery	House	Ferrari manipulation pattern
1.1	17	13	3	IMPRISONED
1.2	17	15	3	IMPRISONED
2	19	16	0	EXCLUDED
3	16	13	0	EXCLUDED
4	15	12	4	SYNERGIC
5	21	18	3	IMPRISONED
6	16	14	4	SYNERGIC
7	14	13	5	SYNERGIC
8	15	13	3	IMPRISONED
9.1	24	19	4	SYNERGIC
9.2	24	21	4	SYNERGIC
10	11	10	3	IMPRISONED
11	12	11	5	SYNERGIC

Tab. 4. 1 Multilevel surgery description

PZ ID	House	Ferrari manipulation pattern	Multilevel surgery description
1.1	3	IMPRISONED	FPB, UFC, RFC, B release. ECRB retention.
1.2	3	IMPRISONED	Adhesion release. AP tenotomy Transfer UFC and RFC to EBCR
2	0	EXCLUDED	BB fasciotomy. Release PT and SFF. Transfer UFC to EBCR. EUCB tenotomy.
3	0	EXCLUDED	I MCFJ arthrodesis. BB fasciotomy. UFC, FBP, PT release. PG tenotomy.
4	4	SYNERGIC	PT release.
5	3	IMPRISONED	PG tenotomy.
6	4	SYNERGIC	BB and PM fasciotomy. Epicondylar muscle tenotomy. UFC release.
7	5	SYNERGIC	Transfer UFC to EBCR
8	3	IMPRISONED	UFC, RFC, PT, FFDP Fasciotomy. PGtenotomy.
9.1	4	SYNERGIC	Carpus arthrodesis, UFC tenotomy
9.2	4	SYNERGIC	Plate removal. FBP myotomy

10	3	IMPRISONED	BB fasciotomy.
11	5	SYNERGIC	Transfer UFC to ECRB.

Tab. 4.1. 1 Synergic hand group data

ID	T0		T1						T2						T3					
PZ	MUUL	AHA	P	F	A	TOT	MUUL	AHA	P	F	A	TOT	MUUL	AHA	P	F	A	TOT	MUUL	AHA
4		42	53,67	43,59	50,00	47,84		53%							50,00	62,40	43,80	34,26		
6	31,34		62,35	50,00	50,00	55,83	63,87													
7	73,64		68,45	60,87	31,64	64,31	82,35		79,98	58,19	61,64	72,21	92,44%		76,52	50,00	61,64	63,27		
9.1			50,00	50,00	50,00	50,00			50,00	50,00	50,00	50,00			50,00	60,00	50,00	53,33		
9.2			70,00	60,08	50,00	57,55														
11		70	62,27	73,27	50,00	64,91		79%						62.27	63,57	57,63	61,74		73%	

Tab. 4.2.1 Imprisoned hand group data

ID PZ	T0		T1						T2						T3					
	MUUL	AHA	P	F	A	TOT	MUUL	AHA	P	F	A	TOT	MUUL	AHA	P	F	A	TOT	MUUL	AHA
1.1	41,8%		55,10	63,39	43,20	53,44	48,36%								36,40	50,00	50,00	49,72	46,72%	
1.2	46,72%		64,41	68,70	62,40	65,04	53,28%								69,39	68,70	62,40	61,42		
5	60,75%		54,56	28,67	50,70	43,93			50,00	44,18	54,23	49,28	76,47%		50,00	42,24	51,41	47,65	68,91%	
8			54,91	57,36	54,56	55,55			50,00	57,36	54,56	54,01			74,54	47,55	54,56	58,61		
10			58,26	56,20	62,40	58,94			38,06	31,39	37,60	35,75			38,06	31,39	37,60	35,75		

Tab. 4.3.1 Excluded hand group data.

ID PZ	T0		T1						T2						T3					
	MUUL	AHA	P	F	A	TOT	MUUL	AHA	P	F	A	TOT	MUUL	AHA	P	F	A	TOT	MUUL	AHA
2	38,52%		52,10	43,80	39,43	47,77			53,30	53,40	50,00	64,52	47,9%		48,80	48,30	51,75	61,17	40,34	
3			50,75	70,93	58,64	59,79			50,75	70,93	58,64	59,79			44,78	43,96	70,17	49,56		