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Our experience regarding rehabilitative, orthopedic integrative interdisciplinary approach in patients with disabling neurological posttraumatic sequelae. Case series and some related literature pointing OF SCIENCE

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Abstract

Introduction Traumatic brain injury (TBI) and / or spinal cord injury (SCI) usually occur in a polytraumatic context, and may produce catastrophic central nervous system (CNS) damages and secondarily extensive dysfunctional biomechanical alterations. This study aims to illustrate the interdisciplinary collaboration between neurorehabilitation and orthopedic clinics in our hospital, focusing on the results of surgical interventions intended to correct the fixedflexion deformity of knees, in patients with disabling sequelae after CNS severe lesions.

Material and methods Between 2005-2018, in the Neuromuscular Rehabilitation Clinic of Teaching Emergency Hospital "Bagdasar-Arseni", 13 young patients (mean age 37.4 +/- 12.6; median 31; limits 26-43) with multiple articular stiffness and joint deposturing sequelae after severe CNS trauma have been transferred from other medical units. Twelve had bilateral knee flexion contractures, two associated additional elbow stiffness, and in three patients ectopic ossifications of the hips, with ankylosis in extension or painful flexion were found. Patients were subsequently transferred for iterative orthopedic interventions: hamstring lengthening (pes anserinus and femoral biceps tendon transpositions) in 12 cases, associated with posterior knee capsulotomy, traction and/or resection of neurogenic heterotopic ossification around the knee or hip joints and casting in 8 of them.

All orthopedic interventions were followed by progressive rehabilitation programs. Spasticity was assessed with modified Ashworth scale (mAS). In pre-/ and post orthopedic surgery, all patients were assessed using an adaptation for adults of the Gross Motor Function Classification Scale, Expanded and Revised (GMFCS – E&R).

Results Twelve patients had knee joint stiffness and chronic flexion contracture: 77% were severely limited in their walking ability, depending on wheelchair (GMFCS – E&R level IV), respectively 23% were bedridden, non-ambulate and totally dependent in all aspects of care (GMFCS – E&R level V).

Knee orthopedic serial interventions were followed by iterative, individualized rehabilitation treatments, and 50% subjects have regain their capacity to walk independently (GMFCS – E&R level II), respectively 50% succeeded to walked with assistive devices (GMFCS – E&R level III).

Discussion Both neuro-muscular system deficits and joint disorders can produce locomotor system abnormalities, joint complications and limb dysfunctional problems. These disturbances represent targets and therapeutic objectives for rehabilitation. Chronic knee flexion contracture, stiff elbows and/or hips, periarticular neurogenic heterotopic ossification: all represents major challenges in the complex management of patients with sequelae after CNS severe

Posterior capsulotomy addressed to a stiffed, distorted knee joint, corrects the limb axis and expands the range of motion (through the angle gained by the eliminated flexion contracture), and sometimes restores the patient's ability to walk. Serial orthopedic interventions, followed by sustained postoperative rehabilitation, had a decisive influence on obtaining good functional results.

Conclusions Comprehensive, multiprofessional approach and collaboration between neurorehabilitation and orthopedic teams are essential for the therapeutic management of patients with severe contractures post neuraxial

Proper evaluation and goal setting are mandatory for rehabilitative management, pre-/ and post orthopedic corrective surgery. Harmonized timing for iterative interventions, followed by postoperative structured, sustained (often for lifetime) rehabilitation are essential for obtaining functional results. Adequate prophylaxis of complications represents a main therapeutic objective, as well.

Key words: traumatic brain injury (TBI), spinal cord injury (SCI), vegetative status, spasticity, contracture, capsulotomy, orthopaedic surgery, neurorehabilitation

Introduction

Traumatic brain injury (TBI) and / or spinal cord injury (SCI) usually occur in a polytraumatic context, and may produce catastrophic central nervous system (CNS) damages, with secondarily extensive dysfunctional biomechanical alterations (1-3).

This study aims to illustrate the interdisciplinary collaboration between neurorehabilitation and orthopedic clinics in our hospital, focusing on the results of surgical interventions aimed to correct the fixed-flexion deformity of knees, in patients with disabling sequels after CNS severe lesions, and respectively, the consequent facilitated progress reached in their complex rehabilitative approach.

Orthopedic surgeons are treating the secondary effects of neurologic lesions, as expressed in articular and periarticular tissues dysfunctions. Orthopedic surgery and neurorehabilitation interventions have common goals: relief of pain, prevent and adjust deformities and joint dislocations, preservation and recovery of function. Maintenance of multilevel joint balance is essential for efficient standing and sitting.

Knee flexion contracture is one of the most common complications in CNS lesions, irrespective its etiology: vascular, traumatic, infectious, neoplastic, etc. Causes of knee contracture lie in hamstring spasticity, hip flexion contracture, gastrocnemius and soleus muscle tightness, and posterior knee capsule stiffness.

Orthopedic management should be assessed to the correction of rotational deformities, lengthening of shortened muscles, or shortening the elongated ones, correction of joint contractures, followed by sustained rehabilitation – orthoses and physical-kinesiological therapy. Hamstring lengthening might be sufficient for the correction of knee flexed contracture; if associated with posterior capsulotomy it prevents recurrences (4).

Treatment should reduce (as possible) disability and facilitate mobility toward patient's independence.

Material and methods Between 2005-2018, in the Neuromuscular Rehabilitation Clinic of Teaching Emergency Hospital "Bagdasar-Arseni", 13 young patients (mean age 37.4 +/- 12.6; median 31; limits 26-43) with multiple articular stiffness and joint deposturing sequels after severe CNS trauma have been transferred, from other medical units.

Demographics, medical history, neurologic and orthopedic diagnosis were recorded. Patients and/ or their kin expressed their agreement to be photographed for academic purpose. Data anonymization and careful removing personally identifiable information were performed. Ethics approval of the study was obtained from our hospital's Bioethics Commission.

Twelve subjects had severe knee joint stiffness and bilateral chronic flexion contracture; average preoperative knee flexion contracture was 95.5° (range 60° to 120°).

Besides knee joint stiffness, ectopic ossifications of the hip with ankylosis in extension or painful flexion was found in three of them, and in two subjects additional elbow stiffness (90° flexion).

Previously to admission in our department, all patients had inefficient preventive procedures, unsuccessful physical therapy and bracing. All were nonambulatory because of the catastrophic CNS sequelae lesions and /or joint contractures.

Patients were carefully evaluated by a multidisciplinary team, who assessed the general health status, foregoing to decide a specific orthopedic intervention. All patients had a stabile clinical condition, neither pulmonary nor urinary infections, and no pressure sores.

Prior to the goniometric articular evaluation, modified Ashworth scale – mAS (5) was used to evaluate resistance during passive soft-tissue stretching, applied throughout the entire (possible) range of motion (ROM). All patients were quoted 4 on mAS (segmental rigidity, in flexion/ extension, abduction/ adduction), passive movement being not possible. Preoperative total ROM was measured in all patients. All patients were evaluated and submitted a daily program of passive extension-flexion movements, aimed to improve ROM amplitude, prior to the orthopedic intervention (-s).

Pre-/ and post orthopedic surgery global functional status of the patients was assessed using a corresponding for adults adaptation (we have made) of the GMFCS – E&R (6), because it was useful and easier to apply than other evaluation tools (Tegner Lysholm knee score (7) and/or other scales (8). Table I summarizes the general criteria for sample selection, description and case stratification with GMFCS – E&R scale.

Surgical Techniques Different orthopedic techniques were used to correct the severe dys-

posturings: teno-muscular transpositions, capsulotomies, arthrolysis, heterotopic ossification excision surgery of the hip or knee joints.

Table I. Adapted GMFCS (- E&R) general criteria for cases stratification

	General headings for each level				
Level I	Walks without limitation	Near normal gross motor function, walks independently			
Level II	Walks with limitations	Walks independently, but has difficulty with uneven surfaces; minimal ability to jump			
Level III	Walks using hand-held mobility device(s)	Walks using assistive devices: canes, crutches, anterior or posterior walkers; "may require a seat belt for pelvic alignment and balance. Sit-to-stand and floor-to-stand transfers require physical assistance from a person or support surface" [6]			
Level IV	Severely limited self- mobility. The mobility is based on the wheelchair. Subject can use powered mobility	Might use powered mobility (a scooter or other type of powered mobility device controlled with the joystick or electrical switch, that enables independence); "require adaptive seating for pelvic and trunk control" [6]			
Level V	Transported in a manual wheelchair	Non-ambulate, totally dependent in all aspects of daily living, activities and participation			

Postural correction of the knee flexed contraction/ stiffness and deformity was achieved by the following surgical interventions: hamstring lengthening (hamstring/ pes anserinus lengthening and femoral biceps tendon transpositions) in 12 cases, combined with posterior knee capsulotomy in 8 cases, followed by traction and/ or casting, respectively resection of the neurogenic heterotopic ossification.

In the case of irreducible knee stiffness (flexum above 100°) or in those in which physical therapy hasn't been able to obtain less than 80° of the extension deficit, a 2nd step surgical approach was performed: the first surgical intervention, consisted of tendons transposition, was followed by 3 months of physical therapy, leading up to a 30°-40° of

extension deficit, then a second orthopedic intervention, consisting of posterior capsular disinsertion (fig.1 and 2).

Results Pre-/ and postoperative outcomes and global functional status of patients with chronic knee flexion contracture are presented in Table II.

Table II. Pre-/ and post-surgical global functional status (on the adapted GMFCS-E&R scale)

GMFCS (- E&R)	Level I	Level II	Level III	Level IV	Level V
Pre operative	0	0	0	77%	23%
Post operative	0	50%	50%	0	0

Pre-surgical assessment: all patients had severe knee flexion contracture and stiffness. About 77% subjects were severely limited in their walking ability, being assisted in wheelchair (adapted GMFCS-E&R: level IV), respectively 23% were non-ambulate, being totally dependent in all aspects of care (adapted GMFCS-E&R: level V).

After knee orthopedic serial interventions, coupled with iterative, subsequent, individualized neurorehabilitation treatment, all subjects regained an almost complete knee extension (corrected to less than a critical 15° angle of flexion). Half of them were able to ambulate using a knee-ankle-foot orthosis, with or without crutch(es) or waking frame (adapted GMFCS-E&R: level III) support. The other half regained the capacity to walk independently without help, on flat surfaces (level II).

For a better illustration, we present patient DF (meta-initials), male 27 years-old. Medical history and neurological evolution: he submitted a severe TBI (in Oct. 2014), followed by vegetative state (in Feb. 2015), and minimally conscious state (in Aug. 2015).

He was admitted in the Neurorehabilitation Clinic in different evolutionary moments after iterative, corrective surgical interventions for bilateral knee flexion contractures (fig.1, 2, and table III), presenting gradual improvement of his neurological status and on the adapted GMFCS-E&R scale.

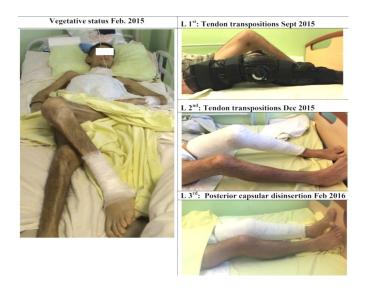


Fig.1. Patient DF (meta-initials), male 27 years-old. Flexion contraction in both knees (more severe in the left lower limb). Postsurgical evolution of the left knee after successive orthopedic corrective interventions (L1st,L2nd,L3rd), presented in Table III. Postoperative improvements in the right knee: noncritical flexion contracture, fewer than 15°, subsequently addressed with kinetotherapy and knee orthosis.

Table III. Schematic synopsis of iterative orthopedic interventions (patient DF (meta-initials), fig.1 and 2

Right Knee	Left Knee		
	16/09/2015 hamstring release (tendon transposition)		
07/10/2015 hamstring release, orthotic device			
04/11/2015 excision of heterotypic calcification around the knee + medial collateral ligament transposition			
	07/12/2015 tendon transposition + excision of heterotypic calcification		
	14/02/2016 posterior capsulotomy, ossification excision, manipulation under anesthesia (followed by casting)		



Fig.2. Patient DF (meta-initials), male 27 years-old (Oct 2015). Good recovery after the first couple of bilateral successive orthopedic interventions: partial correction of knees flexion contractures. Postoperative rehabilitation outcomes: **Level III** on the adapted GMFCS-E&R scale, possibility to exercise (indoor) walking in household perimeter for limited distances, with long mobile orthotic devices and human assistance.

Discussion The hip and knee joints have essential biomechanical role for gait, by flexing and rotating, providing stability during the activities of daily life. Human normal posture and motility devolve upon extensive cinematic mechanisms and subtle sensorial tuning loops for balance recovery and control (**fig.3**).

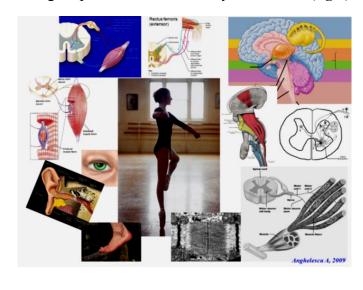


Fig.3. Functional interrelation, subtle sensorial tuning loops, cinematic mechanisms of balance and motility and interdependence in the Neuro-Myo-Artro-Kinetic (NMAK) apparatus.

These implied inseparable functional interdependence of at least two main systems (the

nervous and muscular ones) integrated in neuro-muscle-skeletal complex interrelations. The synergic collaboration of the afore mentioned systems is reflected by the modern concept of "Neuro-Myo-Artro-Kinetic (NMAK) apparatus, who's functional unit (named the "kinetic unit") is composed at segmental level by the morph-functional triad: nerve – muscle – joint. Disruptions at any level of the NMAK apparatus may induce extensive dysfunctional biomechanical alterations (1-3, 9).

Spasticity is a disabling problem for many adults and children with a variety of neurological disorders such as multiple sclerosis, stroke, cerebral palsy, traumatic brain or spinal cord injuries. When the appropriate stretching is not carried out systematically in these patients, muscle shortening and contractures can occur. Spasticity causes pain, contractures, and impaired function. Ultimately, patients experience loss of function and mobility, hygiene difficulties, and orthopedic deformation (1-3, 9-17).

Severe central nervous system (CNS) lesions, usually (mainly) affect descending pathways, including the cortico-spinal tract. Lesions of the upper motor neuron cause imbalance between the excitatory vs. inhibitory stimuli, which converge to the peripheral neuronal pool (alfa and gamma lower motor neurons) in the spinal cord. Besides paralysis with specific topography, the affected muscles will become immobilized in a shortened position (10), and postural imbalances between agonists / antagonists / synergists / stabilizers occurs. In immobilized muscle occurs an increase quantity of connective tissue that reduces muscle compliance (11) and alters the rheological properties of the muscle, its plasticity and viscous-elasticity.

Disturbed rheological intrinsic properties of the muscle and the pathological disruption of the regulatory mechanisms interact with the limb control, position and movement. Resistance in recent-onset spasticity is reflex-induced, whilst resistance in chronically spastic muscle involves rheological changes, contracture, fibrosis, stiffness and atrophy (6, 9-22). Spasticity aggravates contracture and vice-versa, in a vicious cycle, resulting dysposturing, secondary severe myo-tendoligamental and joint-capsulo retractions, joint complications and limb dysfunctional problems. These disturbances usually represent targets/therapeutic objectives for (Neuro-)Rehabilitation,

are addressed in multi-/ interdisciplinary corrective interventions, and therefore represent the subject of this communication.

Pending on the two main path-physiological clinical models of spasticity – the spinal (tetra-/ paraplegia) or cerebral model (hemiplegia) – presence of comorbidities, acquired complications, economic and technical endowment, different therapeutic procedures are indicated.

Sometimes neurosurgical interventions may be indicated, when spasms and spasticity cannot be satisfactorily controlled by medication and physical therapy (11, 19): neurotomy, rhizotomy, myelotomy, cordectomy, spinal cord stimulation, intrathecal baclofen or/ and morphine pumps.

The indications for orthopedic surgery are:

- 1.deterioration or uncontrollable spastic posture;
- 2.fixed deformity that interferes with function

3.secondary complications such as bony deformities, dislocation of the hip and/or joint instability (19).

Severe flexion contractures involving the knee or the hip represent major impediment to functional weight-bearing and ambulation, and the management of such deformities is challenging and problematic (4, 18-22). Significant knee contractures ($\geq 30^{\circ}$) are difficult to brace and severely limit standing and functional efficient ambulation.

Chronic (fixed) knee flexion contracture, elbows and/or hip ankylosis, or periarticular heterotopic ossification represent major challenges in the therapeutic management of patients with sequelae after CNS catastrophic traumatic events. Posterior capsulotomy addressed to a distorted, flexed knee joint, corrects the limb axis and expands the ROM (by the angle of the eliminated flexion contracture), and even may restore the patient's ability to walk. This orthopedic technique, followed by sustained postoperative rehabilitation, has a decisive influence on obtaining good functional results (4, 19, 22, 23). Provided the deformity is controlled by other measures, there is no urgency about operations. In some cases, it may be better to delay until patients

stabilise and then correct them in 1–2 steps (19). Comprehensive preoperative assessment and goal setting in patient-centred multidisciplinary team, carefully evaluation of the general status, prophylactic and curative management are essential objectives of the physical and occupational therapy, oriented to secondary and tertiary prevention and

limitation of the complications. Respecting careful selection criteria, almost all of our subjects succeeded to provide good outcomes (Table II).

Braces and serial casting were used to maintain a spastic limb in a reflex-inhibiting posture and prevent relapsing contractures.

One of the main conditions for a possible orthopedic corrective intervention is represented by a stabile biological, clinic and functional somatic condition. Most of our cases have resulted from polytrauma events, and were admitted to neurorehabilitation with "poor" or "depleted" biological reserves, malnutrition or even "complex cashexia" (24), so these are patients at risk. In this context of frail biological general status, orthopedic interventions are not risk-free (especially procedures necessitating extended articular approaches).

In afore presented small group of patients, no post-acute orthopedic complications occurred. Any pathological situation (thrombophlebitis, respiratory or digestive infections, including sepsis with multiresistant drugs germs, or pressure sores) represent contraindications – at least temporary – for orthopedic surgery

Even in cases where conservative therapy is apparently overcome, general and focal myorelaxant treatment for the spastic somatic muscles might be effective. The case reported below is illustrative (fig. 4, 5).



Fig.4. Patient BRC (meta-initials), male 23 years-old (Apr 2018). Medical history and neurological evolution: in Oct 2007 he submitted a severe TBI (diffuse axonal injury – DAI) after hetero-aggression. He was hospitalized for 6 months in Spain and then transferred in our clinic in Apr. 2008, in a chronic

stage, with severe neuropsychological impairments: spastic tetraparesis (right hemiplegia), posttraumatic encephalopathy, mixed non-fluent aphasia, neurogenic bladder.

Important somatic impairments: flexion contractures involving

the knees, hips and elbows. Apparently irreducible contractures: right knee fixed contracture (125°, mAS 4), flexion of the 4th and 5th fingers of the right hand ("pseudocubital paresis"), and irreducible varus equin of the left foot. Cerebral MRI: diffuse cortical atrophy, bilateral hippocampus atrophy, lacunae in the mesencephalon. EMG: normal bioelectric parameters of the right cubital nerve, excluded a peripheral lesion.



Fig.5. (A,B) Patient BRC (meta-initials), male 23 years-old (Dec. 2008). Systemic miorelaxant treatment (dantrolene 50 mg/day) and focal chemodenervation with botulinum toxin therapy 1500 UM Dysport, addressed to the right upper limb and both lower limbs), followed by sustained kinetotherapy and deposturing ortosis (maintained for 6-8 hours daily). At discharge (after 8 weeks): unipodal walking with the roller frame. At 16 weeks: bipodal walking, using the roller frame. Fig.5 (C,D). Patient BRC, male 24 years-old (Apl 2009), after 12 months. Independent for basic ADL, substantial neuropsichological improvement. No motor deficit, good orthostatic posture, able to maintain monopodal balance (D), and rise on tip toes (C). Total dissipation of the contracture in his right hand.

Essential medical information and educational endeavors linked to the individual pathology are provided for both the patient and caregivers, who are taught to avoid pathological postures, and prevent complications.

Sometimes (especially in patients with vegetative status), the last / unique therapeutic solution for these joint contractures remained the palliative orthopedic approach, for severe dysposturing

contractures that interfere with shoe wearing, wheelchair positioning and/or personal hygiene.

Conclusions Chronic (fixed) knee flexion contracture, elbows and/or hip ankylosis, or periarticular heterotopic ossification represent major therapeutic challenges in the management of patients with sequelae after CNS severe traumatic events.

Comprehensive, inter-/ multidisciplinary collaboration between the orthopedic and neurorehabilitation doctors and connected multiprofesional teams are essential for complex therapeutic approach of patients with severe limb contractures. Orthopedic intervention is only a part of the ensemble of the necessary corrective therapeutic procedures.

Proper evaluation and goal setting are mandatory for the rehabilitative management, pre-/ and post orthopedic corrective surgery. Harmonized timing for iterative interventions, followed by postoperative structured and sustained (often for life-time) rehabilitation programs are essential for obtaining best possible functional results. Adequate prophylaxis of complications represent a main therapeutic objective.

Abbreviations:

mAS, modified Ashworth scale
CNS, central nervous system
DAI, diffuse axonal injury
GMFCS (-E&R), Gross Motor Function
Classification System (Expanded and Revised)
NMAK, Neuro-Myo-Artro-Kinetic
ROM, range of motion
SCI, spinal cord injury
TBI, traumatic brain injury

All authors have made substantial contribution to the work, and approved it for publication.

Conflicts of interest

The authors do not have conflicts of interest to declare.

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