Cerebral palsy of the child in rehabilitation environment: Epidemiologic and clinical profile and therapeutic modalities

La paralysie cérébrale en milieu de rééducation: Profil épidémiologique et clinique et modalités thérapeutiques.

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Abstract

Introduction: Cerebral palsy (CP) is a group of permanent disorders of the development of movement and posture causing activity limitation.
Aim: To evaluate the epidemiological, clinical, and radiological profile of children with CP and to study the therapeutic modalities in daily clinical practice.
Method: This was a retrospective, descriptive study, carried out in a physical medicine and rehabilitation department, including all the patients referred with the diagnosis of cerebral palsy between January 2000 and December 2016.
We used pre-established records to collect data.
The missing data were collected from the files of the pediatric department.
To classify cerebral palsy, we used a motor impairment classification, topographic classification according to the affected limb and the GMFCS – ER.
Results: Fifty patients were included with a gender ratio of 1.23. The mean age of patients was 7.8 years old. The risk factors for CP were mainly represented by perinatal asphyxia(55%) and prematurity(37%). The spastic form was predominant(74%). Quadriplegia was the most severe clinical form(61%). Neuro-orthopedic deformations were found in 78% of cases. Half of the children had GMFCS score between 1 and 2. Brain MRI lesions were dominated by anoxic-ischemic sequelae(34%). Physical therapy was prescribed for all patients. The most prescribed devices were the dynamic ankle foot orthosis(60%). Baclofen was prescribed in 5% of casess and botulinum toxin injection was performed in 30% of children.
Conclusion: The cerebral palsy defines the clinical and functional disorders caused by non-progressive developmental damage to the brain of the infant which requires a multidisciplinary management.

Key words: Cerebral palsy, GMFCS-ER, rehabilitation, epidemiology, clinical forms, risk factors, spasticity

Résumé

Introduction: La paralysie cérébrale (PC) est un ensemble de troubles permanents du mouvement et/ou de la posture responsables de limitation d'activités.
Objectif: Evaluer le profil épidémiologique, clinique et radiologique des enfants atteints de paralysie cérébrale et d'étudier les modalités thérapeutiques.
Méthodes: Il s'agit d'une étude retrospective et descriptive, réalisée dans un service de médecine physique et de readaptation fonctionnelle, incluant tous les patients adressés avec un diagnostic de paralysie cérébrale entre Janvier 2000 et Décembre 2016.
La collecte des données a été réalisée à partir des dossiers médicaux des patients.
Les données manquantes ont été complétées à partir des dossiers du service de pédiatrie.
Pour classer la paralysie cérébrale, nous avons utilisé une classification des déficiences motrices, une classification topographique selon le membre atteint et le GMFCS - ER.
Résultats: Cinquante patients ont été inclus avec un genre ratio de 1,23. L'âge moyen des patients était de 7,8 ans. Les facteurs de risque de la PC étaient principalement représentés par l'asphyxie périnatale (55%) et la prématurité (37%). La forme spastique était prédominante (74%). La quadriplégie était la forme clinique la plus sévère (61%). Des déformations neuro-orthopédiques ont été constatées dans 78% des cas. La moitié des enfants avait un score GMFCS entre 1 et 2. Les lésions cérébrales à l'IRM étaient dominées par des séquelles anoxo-ischémiques (34%). La rééducation fonctionnelle était prescrite chez tous les patients. L'appareillage le plus prescrit était l'attelle tibio-pédieuse de jour (60%). Le baclofène a été prescrit dans 5% des cas et une injection de toxine botulique a été réalisée chez 30% des enfants.
Conclusion: La paralysie cérébrale regroupe les troubles moteurs et limitations fonctionnelles imputables à une atteinte non progressive du cerveau en voie de développement du foetus ou du nourrisson qui nécessitent une prise en charge multidisciplinaire.

Mots clés: Infirmité motrice cérébrale, GMFCS-ER, rééducation, épidémiologie, formes cliniques, facteurs de risque, spasticité.

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INTRODUCTION

Cerebral palsy (CP) is a group of permanent disorders of the development of movement and posture causing activity limitation[1] due to non-progressive disturbances that happened in the developing fœtal, newborn or rarely in infant brain, which occurs in two to three out of 1,000 live births[2]. In addition to motor impairments, the definition includes associated neurobehavioral disorders. These are almost constantly observed in different degrees. Sensory impairments, intellectual and cognitive disabilities, epilepsy and behavioural disorders are often described[1]. Patients with cerebral palsy have clinical presentations that evolve with age combining spasticity, retractions and abnormal movements. The functional and social impact depends on these different impairments and their association with intellectual and cognitive deficiencies. Long-term and multidisciplinary follow-up of these patients is therefore essential, from early childhood through adulthood, to ensure a sufficient degree of autonomy, a good quality of life and a social integration. A good knowledge of the different clinical presentations and the specificities of management are essential to ensure the best treatment. Few studies in the world and even less in Tunisia, have treated this pathology which represents a frequent cause of motor and cognitive difficulties in children. Against this backdrop, we thought it would be interesting to describe the clinical and radiological profile of cerebral palsy, and to outline the different therapeutic alternatives in daily clinical practice in a Physical Medicine and Rehabilitation (PMR) department.
The aim of our study was to evaluate the clinical and radiological profile of children with CP and to study the therapeutic modalities in daily clinical practice.

METHODS

Study design:
This study was carried out at the physical medicine and rehabilitation (PMR) department, including all the patients referred with the diagnosis of cerebral palsy between January 2000 and December 2016. The missing data were collected from the files of the pediatric department.
This was a transverse, single-center, observational, retrospective and descriptive study. The selection of patients was exhaustive. Patients with progressive encephalopathy were not included and those who did not consult in two years or lost to follow-up were excluded. We used pre-established records to collect socio-demographic data, risk factors, clinical and therapeutic data.
To classify cerebral palsy, we used a motor impairment classification (Spastic, ataxic, athetoid/dyskinetic) and topographic classification according to the affected limb (diplegic, hemiplegic, quadriplegic).
We also used the GMFCS – ER[3] that describes the gross motor function of children and youth with cerebral palsy on the basis of their self-initiated movement with particular emphasis on sitting, walking, and wheeled mobility. Distinction between levels is based on functional abilities, the need for assistive technology, including hand-held mobility devices (walkers, crutches, or canes) or wheeled mobility, and to a much lesser extent, quality of movement. The general headings of each level are represented in Figure 1. We also noted the different types of treatments received and the data from magnetic resonance imaging (MRI) if it was done.

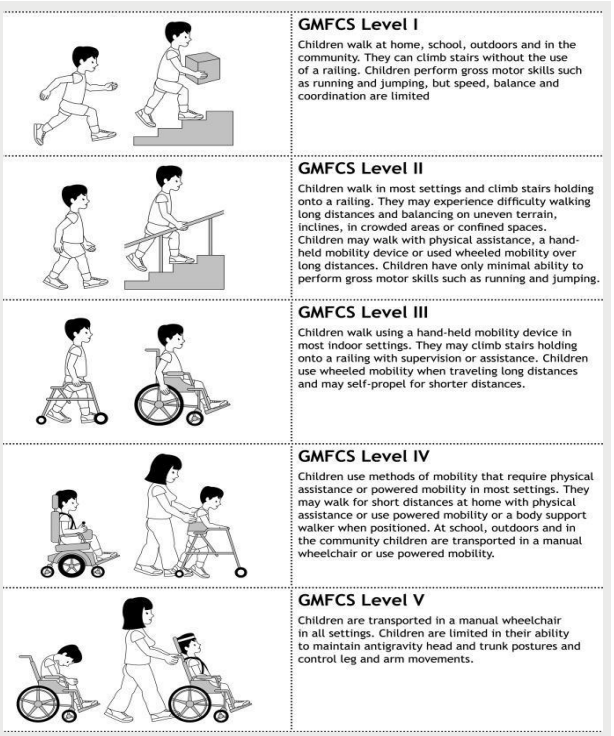

Figure 1. Gross motor function classification system expanded and revised (GMFCS – ER) [4]

Statistical analysis
Data was collected and analyzed using SPSS version 20 software. In the descriptive part we determined the mean, the median and the extreme for quantitative variables and the relative percentages of each category for qualitative variables. In the analytical part, the comparison of two means was done using of Student's t test. The comparison of two percentages was carried out by the Pearson's chi-square test, and in case the conditions of application of this test was not satisfying, the comparison was made by Fisher's two-tailed exact test. The study of the link between two quantitative variables was made by the correlation of Pearson. For all statistical tests, the significance level was set at 0.05.

RESULTS

Fifty patients were included with a gender ratio of 1.23. The mean age at the moment this study was conducted was 7.8±4.7 years old. Thirty percent of patients were born to consanguineous marriages. The risk factors for cerebral palsy are detailed in Table 1. No etiological agent was found in 7% of the patients.

Table 1. Risk factors for cerebral palsy.

Risk factors	Values (%)
Antenatal:	**42**
-Cerebral malformation (hydrocephaly, microcephaly, arachnoid cyst, tumor…)	18
-Intrauterine growth restriction	10
-Multiple pregnancies	8
-Gestational diabetes	6
Perinatal:	**67**
-Premature birth	37
-Perinatal asphyxia	55
-Maternal-fetal infection	9
Post-natal:	**16**
-Infection (meningitis, encephalitis)	12
-Hypothyroidia	3
-Ischemic stroke	2

The spastic form was predominant either bilateral (74%) or unilateral(16%). Dyskinesia and ataxic forms were present in 6% and 4.5% of our population respectively and they were mostly observed in full-term born children (86% of cases). Quadriplegia was the most severe clinical form, and it was found in 61% of spastic forms. Spastic hemiplegia was present in 18% of times, followed by diplegia (16%).

In our study, perinatal asphyxia was present in 55% of children with hemiplegia, in 82% of children with diplegia and in 49% of those with quadriplegia. Most of children with quadriplegia (67%) and hemiplegia(64%) were full-term born. Diplegia was mainly observed in preterm infants (55%).

Neuro-orthopedic deformations were found in 78% of cases. In the lower limbs, the main joint limitations were ankle equinus(45%) and knee flessum(31%). In the upper limbs, wrist rollover and finger wrap were present in 7.5%. A scoliosis was present in 12% of the cases. In 73.5% of the time, the upper limb was functional. Nineteen children (28.4%) were unable to walk. Half of the children had GMFCS score between 1 and 2.

Associated conditions included epilepsy(52.2%), mental disability(42%), strabismus(16%), autism(3%) and an association of multiple disabilities in 28% of the cases.

Among the 37 patients who had reached school age (6years), 16 were enrolled in a normal school and 7 had good grades. Thirteen patients were enrolled in a specialized center and 13 did not go to school.

MRI was performed in 75% of cases. Brain MRI lesions were dominated by anoxic-ischemic sequelae(34%) followed by periventricular leukomalacia lesions(14%) and brain malformations(12%).

Physical therapy was prescribed for all patients. Speech therapy and occupational therapy were prescribed in 22% and 29% of patients respectively. The most prescribed devices were the dynamic ankle foot orthosis(60%) followed by the dynamic knee ankle foot orthosis(51%).

Baclofen was prescribed in 5% of cases.

Botulinum toxin injection was performed in 30% of children. The average time between sessions was 12.2+/-8.5 months. The injection was performed in the lower limb in 76.6 % of cases. Triceps sural, hip adductors and hamstrings were the most injected muscles. For the upper limb, the most injected muscle were the superficial finger flexor muscle. Only one child received scoliosis correction and six children underwent multi-site lower limb surgery.

There was a statistically significant association between schooling and GMFCS(p=0.019) on the one hand and between schooling and topographic clinical forms on the other hand(p=0,046).

Concerning risk factors, we did not find a significant association between the term of birth and clinical presentation(p=0.426) neither between perinatal asphyxia and clinical presentation(p = 0.144).

In our study, these impairments were more frequent in children with quadriplegia(p=0.035). A statistically significant association was noted between quadriplegia and mental retardation (p=0.015).

Concerning the imaging data, there was a significant link between the clinical presentation and anoxo-ischemic lesions on MRI (p=0.025), between prematurity and periventricular leucomalacia lesions on MRI (p=0.005) but also between perinatal asphyxia and anoxo-ischemic lesions on MRI (p=0.036).

DISCUSSION

According to the Surveillance of Cerebral Palsy in Europe (SCPE), Cerebral palsy (CP) is an umbrella term for a group of disorders of movement and posture, caused by a non-progressive interference in the developing brain [5].

Risk factors for CP span the periods before and around the time of conception, during pregnancy, the perinatal period, and up to 2 years-old.

This study allowed us not only to study the clinical and functional characteristics of cerebral palsy, but also to study risk factors, imaging, and therapeutic modalities in daily clinical practice. Our survey also allowed us to look for significant statistical links between CP risk factors, the clinical presentation, GMGCS scores and lesions observed with brain MRI.

CP is the major cause of motor impairment in children, the data of recent studies show that the prevalence of cerebral palsy in Europe, United States and Australia is between 2 and 2.5 per 1000[6,7] . We did not find any valid data on developing countries, this can be explained by the fact that obtaining data on the prevalence of PC is a difficult process which requires the use of national registers or rather complex epidemiological investigations.

The results confirm the evidence found in literature that the prevalence of CP is relatively higher in male patients[8,9]. A study conducted by Reiss et Al in the United States[10] showed that the volume of white matter is reduced in boys born preterm compared to those born at term. Whereas for girls, this volume is comparable whether they are born prematurely or at term, this could be explained by genetic factors, hormonal and by neurobiological differences between the neurons of both sexes[11].

Concerning risk factors, obstetrical and neonatal factors that influence the occurrence of cerebral palsy are well described in the literature, but its pathophysiology still includes obscure points and the causes of the onset of the disorders remain unclear known. This is due to the complexity of the anatomical lesions and the development of brain throughout fetal life and during the first years of extrauterine life[12,13].

These CP risk factors can be antenatal, perinatal, neonatal or postnatal and they can act synergistically (multifactorial pathology) in the disease process[14–16]. Prematurity[17] and perinatal asphyxia are the most studied factors[18,19]. In view of the frequency of CP and its family and socio-professional repercussions, improving morbidity for premature babies is a priority. This requires close collaboration between the Neonatal Medicine and neonatal resuscitation team and the obstetrics team.

According to the literature, spastic forms are the most frequent[20,21]. The topography of the motor disorders in spastic forms was quadriplegia in 61% of cases and it was the most severe clinical form[22].

The most frequently observed neuro-orthopaedic disorder in our series was the ankle equine (45%) caused by the retraction of the sural triceps. The knee flessum is due to the retraction of the hamstrings. It can lead to a decrease in pitch length or a compensatory dorso-lumbar cyphosis. A scoliosis was present in 12% of cases. It is distinguished from idiopathic scoliosis by lower limb deformations and muscular weakness which cause imbalace of the pelvis and spine[23,24].

In fact, it is the muscular imbalance caused by spasticity and/or weakness that causes a vicious attitude. When permanent, this attitude will lead to a disparity in growth between the agonist and antagonist muscles and between the muscles and the bones structure. This modifies the pressures applied on the growth plate.

This results in an irreducible deformity of musculo-tendinous origin[14,25].

According to the SCPE[5], 29% of children with CP had a severe intellectual impairment, 7% had a severe visual impairment and 20% had epilepsy.

In our study, we found that these impairments were more frequent in children with quadriplegia(p=0.035). A statistically significant association was noted between

quadriplegia and mental retardation (p=0.015).
In our study, one in three children could not walk and had to use a wheelchair pushed by a third party. These children were at risk of orthopaedic deformations, back pains and difficulties in obtaining a comfortable sitting position. Add to that, this reduced autonomy can make them non-active members of society, and even a burden on parents who have to be on hand.
The severity of the lesions were correlated to their topography. There was a significant link between the GMFCS score and clinical forms(p=0.009). A GMFCS between 1 and 2 (slight motor disability) was associated to diplegia and hemiplegia and between 3 and 5 (severe motor disability) was associated to quadriplegia. The GMFCS is also predictive of the cognitive development of children, their autonomy and their social interactions[26–28] .
According to the SCPE, there is a tendency to enroll these children in ordinary schools. In certain situations, however, the use of specialized institutions is necessary[29].
In our study, 67% of the children with diplegia and 71% of those with hemiplegia, went to an ordinary school. However, 87% of children with quadriplegia did not attend school.
There was a statistically significant association between education and GMFCS (p = 0.019) on the one hand and between schooling and topographic clinical forms on the other (p=0,046). A GMFCS between 1 and 2 was associated with integration into ordinary schooling system.
In Tunisia, Law 83-2005 stipulates that «The State guarantees the right to education, education, rehabilitation and training in the mainstream system for children with disabilities and provide them with equal opportunities for the enjoyment of this right»[30]. In practice, school integration remains difficult because of psychological barriers (reluctance of teachers but also of parents) but above all material barriers (accessibility, very high health costs)[31].
Morphological exploration of the brain is a major challenge in neonatology. It is achieved thanks to the progress of magnetic resonance imaging. MRI is the examination of choice when there is a suspicion of anoxic-ischaemic pathology[32] but in the premature baby, its place is always in second intention after transfontanellar ultrasound (TFUS)[33].
The brain lesions observed are classified into categories that differ from one author to another[34,35].
In our study, brain MRI was performed in 50 children. It showed abnormalities in 27 cases(54%), the most common of which were anoxic-ischaemic sequelae (34%), periventricular leukomalacia lesions (14%) and cerebral malformations (11%).
However, the development of cerebral palsy can occur in children without neonatal brain injury[36–38].
So we think that the recommendation of early detection through the use of new neuroimaging techniques seems to be an essential objective to make progress in management.
The management of CP must be early, multidisciplinary (pediatricians, neuropediatricians, physical physician, orthopedic surgeons, physiotherapists, occupational therapists...)[39]. It must focus on the child and his environment (parents, siblings and sisters, school environment...). The objective of the care is to make the child acquire the best autonomy and to prevent deformities. This treatment requires a systemic and/or local drug treatment accompanied by regular rehabilitation, adapted equipment and sometimes surgery.
To treat spasticity, oral medications can be used, such as Baclofen® (gamma aminobyturic acid agonist)[40]. It is effecient mainly on the spinal cord by increasing the inhibitory effect of the alpha motor neuron. However, this oral treatment has been abandoned because of its side effects (sedation, drowsiness, nausea, hallucinations…).
When spasticity is localised, botulinum toxin is used in local injections. It is a purified neurotoxin complex derived from the anaerobic bacterium Clostridium Botulinium[41]. When injected in a spastic muscle[42], it improves joint amplitude and delays the occurrence of fixed deformations and it improves the position of the joints and segments of the limb, allowing a better mobility. According to studies, early use of botulinum toxin (BT) provides a maximum response with a longer effect and delays definitive surgery. The ideal age of BT injection is between 2 and 5 years for the lower limb and above 4 years for the upper limb. The time between two injections should be at least 3 months and can vary from 6 to 12 months[42]. So why not create a systematic BT injection protocols for patients with CP according to GMFCS or to clinical presentation ?
Rehabilitation or physiotherapy is the cornerstone of CP management. The main objectives are to improve postural control, joint range of motion, motor control, muscle strength and mobility[43,44]. Several rehabilitation techniques have been shown to be effective, but there is no clear consensus[45]. This should lead to the development of pre-established protocols combining rehabilitation and toxin injections according to the GMFCS.
The medical equipment and devices correct and prevent deformations and stimulate standing or sitting[43–45]. Braces are available for the upper or lower limb and the spine. They allow new aquisition and enhance psycho-motor development and social interactions.
The othopaedic surgery includes muscle elongation or transfer (single tenotomy, tendon elongation, proximal muscle disintegration, tendon transfer), correction of osteoarticular deformities (osteotomy, arthrodesis, arthroplasty), and even neurosurgery (peripheral selective neurotomy)[46,47].
Nowadays, multi-site surgery aims to correct all defects (musculotendinous or osteoarticular) at once[46,48]. The efficacy of multi-site surgery has been demonstrated by numerous studies showing a decrease in the number and total duration of hospitalizations during childhood, improved walking, increased joint range of motion and improved functional abilities[49–52].
Ideally, surgery should be performed in adolescence, otherwise growth-related retractions may lead to repeated interventions[50,53].
In our series, the average age at surgery was 6 years, which can be explained by the the fact that the indications are limited to severe or complicated forms. Won't early surgery in the most severe cases bring better results?
Retrospective data collection exposes this study to selection bias. The number of patients was limited and the average of patients lost to follow-up was high (34%). This may be explained by limited access to the hospital, relocation or transfer of follow-up to the private sector.
In addition, this was a mono-centric study concerning a pathology whose follow-up is multidisciplinary (PMR, pediatrics, neurology and orthopedics).
All this could reduce the fiability of our results.

CONCLUSION

It is important to assess the clinical features of cerebral palsy and to be aware of the factors influencing its progression, so that multidisciplinary care can be provided to ensure social integration, a minimum of autonomy and a good quality of life. On a national stage, multidisciplinary collaboration is needed to research and to create a national registry for this disease and for the development of care centres for children with CP. This will help us to act on risk factors and target therapeutic practices for improvement, ensuring a minimum of autonomy, a better quality of life

for children and their families, and integration into school and society.

We also propose to create a CP research unit at the Military Hospital of Tunisia, including all of the above-mentioned specialists, and to continue collecting data prospectively over the next few years.

ABBREVIATIONS LIST

BT: Botulinum toxin

CP: Cerebral palsy

GMFCS-ER: Gross Motor Function Classification System Expanded and Revised

LB: Life birth

MRI: Magnetic resonance imaging

PMR: Physical Medicine and Rehabilitation

SCPE: Surveillance of Cerebral Palsy in Europe

TFUS: Transfontanellar ultrasound

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