Original Paper

Cerebral Palsy. Considerations Upon 249 Consecutive Patients and Review of Literature

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ABSTRACT: Aim: to determine the prevalence of cerebral palsy (C.P.) among children and to describe its main characteristics (clinical forms, treatment plan, and results). Material and Methods: 249 C.P were studied (120 boys and 129 girls, aged between 0 and 12 years) during 2,321 consecutive clinic visits (incidence 10.7%) to a private pediatric orthopedic clinic in Jeddah, Saudi Arabia, between 2011 and 2016. Spastic type was the main clinical form (231=92.8%), spastic diplegia being the most frequent (166=71.9%). The treatment was complex: conservative only in 42.2%; surgery indicated in 149 (59.8%) cases was performed only in 81 cases, by means of muscle and/or bone procedures, depending on the lesion balance. Results. The postoperative results were excellent in 2.4% of cases, acceptable to some extent in 93.8% and poor in 3.8%. We registered a recurrence rate of 14.8%, a postoperative morbidity rate of 6.17% with a postoperative mortality rate of 0. Treatment results could not be assessed in 129 (51.9%) cases due to lack of follow up. Conclusion: 1. C.P. represents the third most common diagnosis in pediatric orthopedic private practice. 2. The treatment results were acceptable in most cases, but not optimal. 3. Gait analysis using in the preoperative planning could improve significantly the outcome, especially in complicated cases.

KEYWORDS: Cerebral palsy (C.P.), treatment

Introduction

Cerebral palsy is the most common disability at the beginning of childhood in most countries [1]. The incidence varies from one country to another, being accepted around 1-2 cases/1000 live births [2,3]; the prevalence rate in Saudi Arabia is of 2.3% [4]. Among the other studies [5-9], those performed by Al-falahi and Al-turaiki [10] in Riyadh reported that 75% of the severely handicapped patients were children and, 64% of them had cerebral conditions.

Etiology remains unknown in up to 50% of cases, prematurity being prevailing [11]. In most countries the orthopedic burden secondary to cerebral palsy is stationary or increasing, representing the most common diagnosis after a traumatic event [12]. Orthopedic surgery has much to offer in terms of the management of children patients with cerebral palsy, including the cases with spastic hemiplegia, spastic diplegia and spastic quadriplegia [13,14,15].

The newborn baby with cerebral palsy usually has no deformities or musculoskeletal abnormalities at birth, however scoliosis, hip dislocation and fixed contractures develop during the rapid growth of childhood [12].

Material and Method

The objectives of the study were to establish the prevalence of cerebral palsy among the clinic consults performed in Pediatric Orthopedic Clinic Jeddah (Saudi Arabia) in children≤12 years of age, and to established their main etiologic, clinic and therapeutic characteristics according to the literature data 249 cases of cerebral palsy, selected from 2,321 clinic visits, performed in a private pediatric orthopedic clinic in Jeddah (Saudi Arabia), between 2011-2016 were analyzed.

Inclusion criteria: 1. age ≤ 12 years and; 2. musculoskeletal evaluation requested by the family, the patient or requested by another physician.

Children who did not have a clear diagnosis or treatment plan in their files were not included.

Data were collected from and recorded according to the standard protocol and using a data record form. For statistical processing of the data the Statistical Package for Social Sciences (SPSS), version 25 was used; frequency and percentage were used to describe categorical variables, whereas range, arithmetic mean and standard deviation were used to describe continuous variables. A written informed consent was obtained from the patients' caretakers, and the study was approved by the Ethics Comity of "The First Clinic" Pediatric Orthopedic Clinic.

Results

Incidence

The incidence of C.P., according to the literature, is about 1-2 cases/1,000 live births, with a prevalence rate of 2.3% for Saudi. We registered a prevalence rate of 10.7%, calculated on the total number of clinic visits (2,321 visits (confidence interval 9.6-11.8%)).

Demographic data

- *Gender:* A slight prevalence of girls was noticed: 120 boys (48.19%) vs. 129 girls (51.81%) with a sex rate /G) of 0.93.

- Age. The age of the onset is not similar with the age of the first visit, as the diagnosis was previously established by the pediatrician or pediatric neurologist who referred the child to the pediatric surgeon. The mean age at the first visit was at 5.9 ± 3.2 years, most cases (68.2%) belonging to the group of those between 2 and 8 years, while only 7.2% of the cases came in the first year of life (Table 1). We also noticed that about 25% of the cases were seen by a doctor when they were older than 9 years, and that is why most likely the results are not very accurate.

Table 1. Cerebral palsy: age distribution, pediatric orthopedic clinic, Jeddah (2011-2016).

AGE	number	percentage
0-1 year	18 (14M-4F)	7.2%
2-4 years	80 (36M-44F)	32.1%
5-8 years	90 (45M-45F)	36.1%
9-12 years	61 (26M-35F)	24.5%

Etiological factors

The prematurity, the most common etiological factor was registered in 81 (32.5%) cases. Other pathological findings which could be considered as etiological factors were registered in 31 (12.4%) cases as following: brain asphyxia 16, multiple pregnancies 6 (4 cases triplets and 2 cases quadruplets), post head injury 4, septicemia in early childhood due to chest infection, meningitis, gastrointestinal infection and fever of unknown origin 4 cases and brain hydrocephaly 2 cases.

Only one file included the results of the genetic study showed q15 overgrowth syndrome!?

We registered malformative associations in 15 cases (6%): congenital heart disease 5,

seizures 2, inguinal hernia 2, autism and muscular dystrophy one case each and one case for each following associated syndromes: Down, Larsen, windswept syndrome, and Sturge-Weber syndrome.

Diagnosis

All patients included in study, had the diagnosis confirmed by the pediatrician/pediatric neurologist, prior to their presentation to the pediatric orthopedic clinic. We noticed clearly that, any patient who came first to the pediatric orthopedic clinic, without a prior pediatric neurologist consultation, were sent back to them to receive the definitive cerebral palsy diagnosis before starting the treatment plan.

Imaging tests

At the first visit, all patients were prior fully investigated by the pediatrician or pediatric neurologist. The imaging tests were requested by the pediatric orthopedic surgeon, most likely in order to provide the additional data necessary for the establishment of the further therapeutic plan.

Brain MRI performed in 15 cases (6%) found out periventricular leukomalacia (PVL) in 7 cases, brain atrophy in 2 cases, hypoplastic posterior corpus callosum body and splenium in 1 case, absence of corpus callosum body in 1 case, intracranial hemorrhage in 3 cases and ischemia in 1 case.

The routine pelvis X-ray examination performed in all cases, in order to recognize the hip subluxation or dislocation identified hip displacement in 22 (8.8%) cases (14 subluxations and 8 dislocations).

Anatomo-clinical forms

That the most cases (231=92.8%) belonged to the hypertonic-spastic type, diplegia being the most common (67.1%), the rest of spastic hypertonic type being represented by quadriplegia, hemiplegia, and triplegia. Only five (2%) cases were hypotonic forms, while 13 (5.2%) cases did not have a clear subtype in their records (Table 2).

Table 2. Cerebral palsy types.

Anatomo-clinical forms	No. of cases	Percentage
Hypertonic spastic type	231	92.8
- Diplegia	167	67.1
- Quadriplegia	22	8.8
- Triplegia	12	4.8
- Hemiplegia	30	12.9
Hypotonic type	5	2
No clear type recorded	13	5.2

Treatment

The treatment of C.P. was complex, using conservative and/or surgical methods, with the therapeutic plan established by a thorough physical examination, completed by laboratory and imaging tests, if necessary. The main goals of the physical examination were to determine the degree of muscle strength, to evaluate the muscle tone (normal, hypotonic, spastic, or mixed), to assess the reflexes and the sensory function, to evaluate the degree of deformity or muscle contracture at each of the major joints, to assess linear, angular, and torsional deformation of the spine and long bones, and fixed hand or foot deformities, and to appraise balance, equilibrium, and standing or walking postures. The routine pelvis X-ray examination was

performed in all cases in order to recognize hip subluxation or dislocation.

The preoperative gait analysis was introduced in our current practice since 2016 as part of the preoperative planning to help and improve the surgical indication. It was performed in 14 (32.5%) cases with surgical indication, registered during 2016 (43 cases).

In patients with surgical indication, the preoperative preparation consisted of hydroelectrolytic, hematological and vital function balancing.

Depending on the complete lesion balance, the treatment of the patients with C.P. was performed according to one of the following three protocols (Table 3).

Table 3. Cerebral palsy: therapeutic protocol.

Therapeutic protocol	No. of cases	Operated	Non operated
Conservative treatment (Physiotherapy and/or brackets)	100	-	100
Conservative treatment (physiotherapy and/or Botulinum toxin A intramuscular)			
followed by surgery	29	29	-
Surgery indicated at the 1 st visit	120	52	68
Recurrence and revision surgery	12	6	6

1. Conservative treatment only (physiotherapy and/or braces), without requiring late surgery 100 (40.2%) cases, indicated in mild cases or for patients registered before walking age, at which the decision was to start physical therapy and follow up after starting walking.

2. Conservative treatment (physiotherapy and/or intramuscular injections with botulinum toxin A) as a first step, followed by surgery (29 cases=11.6%), the lack of response at conservative therapy (physiotherapy+ botulinum A toxin i.m injections) in 10 cases and recurrence in 19 cases being the main indications for surgery.

3. Surgery was planned since the first visit in 120 cases (48.2%). The indication for surgery were: crouch gait with severe hips adduction,

fixed flexion contracture in the hip or knee joint due to hip flexors muscles or hamstrings muscles, severe gastrocnemius muscles contracture or Achilles tendon shortening with tip toe gait, severe pes planus deformity causing gait disturbance, hip subluxation or dislocation and bony deformity. Surgical treatment consists of single-event multilevel surgery, known as SEMLS.

81 (32.5%) cases were operated on, 52 belonging to this protocol and 29 to the protocol no. 2. Another 12 patients with recurrences needed surgery; 6 of them were operated on and 6 refused surgery. Surgical correction used soft tissue or bone procedure or both, chosen according to the particularities of each case (Figure 1 and Table 4).



Figure 1. Cerebral palsy-surgical procedures.

All surgical procedures were followed by physical therapy, essential to maximize the benefits of most surgical procedures, its goals being to maintain or improve the motion at the joints' level, to regain muscle strength, to maximize ambulation, and improve function, if possible.

Outcomes and post-therapeutic follow up

In the conservative patient's group (100 cases), a significant improvement was registered in 33 patients, who have continued to benefit from intermittent physical therapy treatment, the remaining 67 patients missed follow up. In the operated patients' group, the postoperative anatomic and functional results

were excellent in 2 cases (2.4%), while in 76 cases (93.8%) we noticed an improvement to some extent; 9 cases (3.8%) had poor results (no improvement or even worsening). The recurrence rate was of 14.8% (12 cases); the postoperative morbidity was 6.17% (2 superficial wound infections and 3 heel ulcers) and the postoperative mortality was.

The post-therapeutic evolution of the patients was monitored by follow up visits in 120 (48.1%) patients. The follow up period ranged between 6 months and over 4 years, and the number of visits, between 1 and more than 5 (Table 5).

In 129 (51.9 %) cases we could not assess the treatment's results due to the lack of follow up.

Table 4. Surgical procedure accord	ling to the lesion to be corrected.
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Lesion type	Surgical procedure types	No. cases
Hip Subluxation	Adductor longus muscle and gracilis tendon release+semitendinosus and gracilis tendon release+semimembranosus transfer to the head of gastrocnemius	14
Hip dislocation	Open reduction hip joint	4
	Open reduction hip joint+pelvic osteotomy	2
	Open reduction hip joint+pelvic osteotomy+femural derotation osteotomy	1
	Femoral head resection	1
Crouch gait with fixed knee flexion	Technique used in hip subluxation+gastrocnemius recession or Achilles tendon	30
deformity with tip toe gait	lengthening	
Equines deformity	Gastrocnemius recession or Achilles tendon lengthening	20
Severe pes planus	Tibialis posterior tendon lengthening+subtalar arthrodesis screw	5
	Calcaneus osteotomy	4

Table 5. Cerebral palsy-patients' follow up.

Follow up period (months)	Cases	Number of visits	Cases
6	33	1	39
12	14	2-5	75
12-24	24	>5	6
24-48	38		
>48	11		
No follow up	129		

Discussion

The main causes of disabilities in the Kingdom of Saudi Arabia (K.S.A) are cerebral palsy and developmental delay, followed by road traffic accidents [4]. To our knowledge, previously there was not another previous study in the K.S.A regarding cerebral palsy from pediatric orthopedic perspective.

Demographic characteristics showed a slight prevalence of girls in our study group (sex rate of 0.93), close to the literature that stated there is an insignificant distribution of C.P., depending on sex [16,17].

With respect to the age, only 7.2% were referred to the first visit in the first year of life; this could be explained by the fact that the neurological consequences and the onset of the clinic signs of C.P. were probably delayed for several months due to the immaturity of the nervous system. [18]. Early warning signs of C.P. were developmental delay, toe walking, persistent fisting, and scissoring of the lower limbs [19]. Unfortunately, about 25% of the cases were seen after the age of 9 years, which most likely not led to a good result. Staheli stated that in the cerebral palsies, the most important priority in the first decade is the function, in the second decade the appearance, and afterwards the avoidance of pain. Most functional gains are made in the first decade [12].

There are several C.P. classification systems based on the physiopathology, etiology, and the distribution of motor deficits. Insults resulting in neuronal loss can be 1) cortical (pyramidal), resulting in spasticity, 2) basal ganglia (extra pyramidal), leading to abnormal movements, such as choreoathetosis, 3) cerebellar, resulting in hypotonic, or 4) mixed.

Spastic C.P. is the most common type, which included 92.8% of cases in our study, which is higher (75-88%) than the data reported by others [20,21]. It could be explained by the fact that the

most cases are addressed to the pediatric orthopedic clinics for surgical treatment, which is not applicable in hypotonic cerebral palsy, which is usually very rare (2% in our study).

Classification according to the topographic distribution of motor involvement (motor includes monoplegia, diplegia, deficits) hemiplegia, triplegia, and quadriplegia. Diplegia is present when the lower extremities are first affected, while the upper extremities are not completely spared. Spastic diplegia is the most common type of C.P (71.9% in our study) and is associated with prematurity. The periventricular germinal matrix insult in preterm infants impair the lower limbs more than the upper limbs, resulting in spastic diplegia, according to our MRI findings: periventricular leukomalacia (PVL) reported in seven cases. Hemiplegia (7.4% in our study) is characterized by the involvement of one side of the body, the arm being typically more affected than the leg, due to the larger cortical representation (motor homunculus) of the hand and arm compared to a smaller area for the leg. Hemiparesis is usually the result of very mild hemiplegia. When all four limbs are involved, quadriplegia is the appropriate descriptive term. This is the most debilitating form (25%), the respective children requiring total care. Triplegia is rare and usually is milder and very asymmetric [11,22]. These sub-types can be difficult to separate clinically in some of the children, especially that the degrees of disability can vary widely within these subtypes [21].

The treatment of C.P. is complex, including conservative and/or surgical means.

Non-surgical (conservative) treatment addresses to the management of the spasticity, to prevent the development of fixed contractures. If these occur, before the onset of decompensation is required the correction of the fixed musculoskeletal deformities, because after the complex decompensated joint pathology has already occurred, the surgical options are already limited, the rate of complications increases and the outcome of salvage surgery is frequently not а better one. Specific conservative options for children with C.P. include physical and occupational therapy, orthopedic means (braces and casting), drug treatments of the spasticity (local, intrathecal, systemic), and orthopedic surgical interventions. Most patients require a combination of these methods, but physical therapy is always essential [12]. Conservative treatment is temporary or permanent, focal or generalized

[23,24]. Systemic treatments for spasticity include baclofen, diazepam, dantrolene, and tizanidine, alone or in combinations, commonly used by neurologists, while orthopedic surgeons usually avoid them.

Intramuscular injections are temporary and focal, the most frequently used agent being BTX-A, a potent neurotoxin produced by clostridium botulinum in anaerobic conditions. It binds to cholinergic nerve endings and inhibits the release of acetylcholine, thus blocking the binding of acetylcholine vesicles at the level of the plasma membrane of the motor endplate; the sprouting of new nerve endings then restores neurotransmission. The effect of BTX-A is pharmacologically completely reversible, the onset of action occurring in 3-10 days, with an average therapeutic duration between 3 and 6 months: it can be used before any surgical considerations [25,26]. We used it in 19 cases, all of them ended by surgery.

Surgical management and preoperative workup. Most children with spastic diplegic develop progressive cerebral palsy musculoskeletal deformities as they grow, which need surgical correction [27]. The surgical indications are well standardized including: crouch gait with severe hips adduction; fixed flexion contracture in the hip or knee joint due to hip flexors muscles or hamstrings muscles; severe gastrocnemius muscles contracture or Achilles tendon shortening with tip toe gait, severe pes planus deformity causing gait disturbance, hip subluxation or dislocation and bony deformity.

In the recent years, the gait analysis imposed as one of the most valuable methods for preoperative assessment of children with cerebral palsy, providing data that allow a thorough study of the various components of pathologic gait. Gait analysis is used to identify which muscles require surgery and which gait abnormalities are compensatory and therefore can be spared, and to assess the rotation of the limbs during gait [13]. One study compared the experienced physician's surgical decisionmaking according to the clinical evaluation, with and without using preoperative gait analysis; it indicates that when experienced physicians added gait analysis data to their own clinical evaluations. they changed their surgical recommendations in about half of patients, with more decreases than increases in the number of procedures [28]. Although preoperative gait analysis is theoretically desirable, it is neither possible nor necessary for every child who

undergoes surgery [13]. Since 2016 gait, analysis was included in our standard preoperative work up protocol (14 cases) and we noticed that without using it the preoperative planning was sometimes not fully decided until reexamining the patient under anesthesia in the operating room.

The best surgical procedures for cerebral palsy remain controversial, the evidence of the efficiency of most orthopedic operations is lacking, and the surgical management is not supported by a high level of evidence [29]. In our study the objective of surgery was to correct gait problems, approaching simultaneously all deformities in a single-event multilevel surgery (SEMLS), this being the standard attitude in many studies [12,13,14,24,27,29], since the correction of all orthopedic deformities in one session requires only one hospital admission and one period of rehabilitation.

Correction of bony torsion required rotational osteotomies, and of pes valgus required an extraarticular fusion of the subtalar joint. Types of the muscle procedures performed by us were lengthening of the psoas at the pelvic brim, fractional lengthening of the medial hamstrings, transfer of semimembranosus to medial head of gastrocnemius for a stiff-knee gait and recession of the distal aponeurosis of gastrocnemius. Open lengthening of tendon Achilles is rarely required since it may result in overcorrection, which is impossible to salvage [30].

It is well known that hip subluxation or even dislocation (22 cases in our study) because of cerebral palsy during the rapid growth of the childhood could be a hidden finding, which should not be missed by the pediatric orthopedic surgeon, if it was not explored, by pediatrician or pediatric neurologist. Diagnosis of hip dislocation is very important; it will change the treatment plan as well as the early detection of abnormality will allow preventing the dislocation by soft-tissue surgery only. It was a routine practice in our study to have pelvis X-ray in all C.P. patients. When the dislocation was explored late, we have done a one-stage open reduction, combined with a varus derotation and/or shortening osteotomy of the proximal femur and a pelvic osteotomy.

If severe hip dislocations in the spastic quadriplegia are diagnosed late, management and therapeutic options are considerably limited. In our study, we found one case of severe hip dislocation with huge femoral head displacement in quadriplegic non-ambulating patient, the dislocated hip was affecting the sitting position and hygiene; we did a femoral head resection for improving the positioning and hygiene. The reason for delayed diagnosis in quadriplegic cerebral palsy is simple because the children with hip displacement in spastic hemiplegia or spastic diplegia have such an important gait disorder that they are very quickly referred to the orthopedic surgeon, while hip displacement in the spastic quadriplegia child is silent in the early stages and the parents and pediatricians are focused on much more obvious problems, such as feeding difficulties and seizure management.

The rehabilitation program have to be considered as important as the surgery, similar to other studies [27,31].

The outcome of the surgery and rehabilitation could be determined by a follow-up analysis made 12 to 24 months after surgery [27]. In our study only 73 cases (30%) had been followed for more than 12 months, 129 cases (51.9%) came only once, for consultation. As easily can be noticed, we registered a high rate of missed follow up, that could be due to either financial issues or to the possibility that some of the children that were diagnosed and advised for surgery to seek for another opinion. Also the families of the children with C.P. and chronic disabilities needing a full cooperation of those, couldn't afford the optimal and special care needed in some cases.

Limitations

The present study is a clinic based and captured only the cases of children with cerebral palsy attending private orthopedic surgery in the outpatient clinic of the designated clinic where the study was conducted.

In view of this noted shortcoming, the profile described here should be interpreted with caution.

Conclusion

1. Cerebral palsy represents the third most common diagnosis in pediatric orthopedic private practice, after normal variations and trauma.

2. Although the clinical signs are specific and obvious in most of cases, the diagnosis was relatively late, the average age of the first visit being 5.9 years.

3. The use of gait analysis in the preoperative planning brings a significant decision-making improvement, especially in complicated cases.

4. The treatment is a complex one, using conservative and/or surgical methods, the therapy being established according to the type

and severity of the lesions, the age and general status of the patient and the acceptance of the parents.

5. The treatment results were acceptable in most cases, but not optimal.

Conflict of interests

None to declare.

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