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Clinical characteristics and functional status of children with different subtypes of dyskinetic cerebral palsy

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Abstract

Dyskinetic cerebral palsy (CP) is the second major subtype of CP. Dyskinetic CP can be classified into different subtypes, but the exact clinical characteristics of these subtypes have been poorly studied. To investigate the clinical characteristics and functional classification of dyskinetic CP from the perspective of neurologic subtypes in a hospital-based follow-up study.

This was an observational study of consecutive children with dyskinetic CP treated at The Affiliated Women & Children Hospital of Qingdao University (China) from October 2005 to February 2015. The children were stratified according to their neurologic subtype and assessed with the Gross Motor Function Classification System (GMFCS), Manual Ability Classification System (MACS), and Communication Function Classification System (CFCS). MRI scanning was conducted at 1 year of age for most children.

Twenty-six participants (28.0%) had dystonic CP, 26 (28.0%) had choreoathetotic CP, and 41 (44.1%) had mixed CP. Auditory impairment and basal ganglion lesions occurred more frequently in the dystonia group (n = 8, 31%; and n = 16, 67%), while seizures, microcephaly, white matter lesions, and mixed lesions were more frequent in the mixed type (n = 14, 34%; n = 10, 24%; n = 15, 41%; n = 12, 32%). Functional classification levels were distributed unequally among the 3 subgroups ($P < .01$). No significant difference between GMFCS and MACS was found among the 3 subgroups ($P > .05$).

Different subtypes of dyskinetic CP have specific comorbidities, radiological characteristics, and functional attributes according to their etiological factors and brain lesions. Children with dystonic CP have more limited functional status than children with choreoathetotic CP.

Abbreviations: AAC = augmentative and alternative communication, ABR = auditory brainstem response, CFCS = Communication Function Classification System, CP = cerebral palsy, DIS = Dyskinesia Impairment Scale, GMFCS = Gross Motor Function Classification System, HIE = hypoxic-ischemic encephalopathy, ICF = International Classification of Functioning, Disability, and Health, LD = laryngeal dystonia, MACS = Manual Ability Classification System, SCPE = Surveillance of Cerebral Palsy in Europe, SD = spasmodic dysphonia.

Keywords: clinical subtype, Communication Function Classification System, comorbidities, dyskinetic cerebral palsy, Gross Motor Function Classification System, Manual Ability Classification System

1. Introduction

Dyskinetic cerebral palsy (CP) is the second most important type of CP after spastic CP.[1,2] It is also one of the most disabling type of CP.[1,2] It has been reported that the prevalence of dyskinetic CP is increasing.[3,4] Dystonia and choreoathetosis are 2 common clinical manifestations of dyskinetic CP. They are present simultaneously in most cases, although with some differences.[5,6] According to the Surveillance of Cerebral Palsy in Europe (SCPE), dyskinetic CP can be classified into the dystonic or choreoathetotic subtypes. Many children with dyskinetic CP have additional movement abnormalities presenting as spasticity, and they have traditionally been grouped within a "mixed" category, but some recent studies classified these cases as dyskinetic CP.[4,7] The clinical characteristics of children with dyskinetic and spastic CP are different from those with pure dyskinesia. Therefore, it could be more adequate to classify dyskinetic CP into the dystonic, choreoathetotic, and mixed subtypes.[8] A more comprehensive recognition of different CP subtypes could be clinically significant and have etiological usefulness.

The various clinical presentations of dyskinetic CP depend on the affected brain structures.[9–11] Previous studies have shown that lesions in the basal ganglia and thalamus are common in dyskinetic CP, and that white matter lesions are also common.[12,13] So far, no study has provided a description of the underlying brain lesions from the point of view of the clinical subtypes.

A number of assessment tools are available for CP. The International Classification of Functioning, Disability, and Health (ICF) put emphasis on function and participation. The

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Gross Motor Function Classification System (GMFCS), the Manual Ability Classification System (MACS), and the Communication Function Classification System (CFCS) have been widely used to assess gross motor, fine motor, and communication ability in children with CP, and they have been proved to have good reliability and validity.[14–16] The combination of these 3 classification systems provides a profile of the overall functional status of a child with CP. Nevertheless, it remains unclear whether various dyskinetic CP subtypes differ in functional status.

Therefore, the aim of this hospital-based study was to investigate the clinical characteristics and functional status of each dyskinetic CP subtype, including etiological factors, accompanying impairments, and neuroimaging findings, as well as gross motor, fine motor, and communication disabilities.

2. Methods

2.1. Study design and patients

This was an observational study of consecutive children with dyskinetic CP treated at the Department of Neurology & Rehabilitation of The Affiliated Women & Children Hospital of Qingdao University (China) from October 2005 to February 2015. The functional assessment database was implemented in 2009 to 2012. Therefore, children already >4 years of age during this period were retrospectively identified and their data were retrospectively compiled. Children <4 years old seen in 2009 to 2012 were prospectively compiled into the database when they reached 4 years of age.

The inclusion criteria were: diagnosed and clinically classified as dyskinetic CP; >4 years of age; and without progressive or degenerative diseases. The exclusion criterion was: the child had already underwent orthopedic or neurological treatment.

This study was approved by the ethics committee of The Affiliated Women & Children Hospital of Qingdao University (China). Informed consent was provided by the parents.

2.2. Diagnosis

The diagnosis and classification of dyskinetic CP were based on the SCPE definitions.[1] The same 2 physicians (>30 years of experience) diagnosed the disease and performed the subtype classification. The major evidence for clinical classification was the abnormal clinical signs and abnormal movement patterns.

Children with CP and a mixed profile of dyskinetic and spastic features were classified as mixed subtype when the dyskinetic type dominated or when no specific type dominated. Signs of spasticity include exaggerated tendon reflexes, increased velocity-dependent muscle tone, and positive Babinski sign.

For children with dyskinetic CP only, the subgroup classification of dystonia vs. choreoathetosis was mainly performed in accordance to the Taskforce on Childhood Movement Disorders,[5] which reached a consensus on the definition and classification of childhood movement disorders in 2008, and the SCPE that recommended to classify the subgroups from the clinical aspect.[1] For the hard-to-determine subgroups, the classification was mainly conducted according to the Dyskinesia Impairment Scale (DIS).[17] The clinical subtypes were differentiated by 2 associate chief physicians or associate chief physician.

2.3. Peri/neonatal adverse factors

The peri/neonatal period ranged from the onset of labor to the 28th day of life of the newborn. Cases of hypoxic-ischemic encephalopathy (HIE), hyperbilirubinemia, asphyxia, verified intracranial hemorrhage/stroke, cerebral infection (viral or bacterial meningitis/meningoencephalitis), and premature delivery were considered as peri/neonatal adverse events. In addition, congenital brain malformations that were found on later cranial MRI were considered to be caused by abnormal gestational events.

2.4. Brain MRI

Brain MRI was performed using a 1.5 or 3.0 T Siemens Verio scanner (Siemens Healthcare, Erlangen, Germany) to obtain axial T1- and T2-weighted images and coronal and sagittal T2-weighted images. Images were analyzed by a radiologist who was blinded to all clinical information. MRI was performed at admission or when the patient reached 1 year of age, whichever came first.

2.5. Accompanying impairments

The following comorbidities were examined: cortical visual dysfunction, substantial auditory impairment, learning disability, coexisting seizures, microcephaly, and inspiratory laryngeal stridor. Data about those comorbidities were obtained from the medical records and from parental interviews conducted during follow-up. Visual dysfunction was diagnosed by an ophthalmologist. Substantial auditory impairment was defined as a 70-dB or greater hearing loss (bilateral). Coexisting seizure was defined as the occurrence of at least 2 events of unprovoked seizures or 1 event of unprovoked seizure occurring in the setting of a predisposing cause. Learning disability was defined as an IQ less than 70, measured by the Wechsler scales or estimated from clinical observation. Microcephaly was diagnosed in the presence of a head circumference at least 2 standard deviations lower than the head circumference for the same age and gender. The diagnosis of inspiratory laryngeal stridor was made according to the clinical manifestations.

2.6. Functional assessment

The GMFCS, MACS, and CFCS[15,16,18] were used to evaluate the functional status for gross motor function, manual abilities, and communication ability. They all grade the severity from level I (the most able) to level V (the least able). The functional assessments were made by the doctors and mothers when the child was 4 years old or older (Table 1).

2.7. Statistical analysis

The distribution of the continuous data was tested with the Kolmogorov–Smirnov test. Normally distributed continuous data were presented as mean ± standard deviation and analyzed using ANOVA and the Tukey post hoc test. Categorical data were presented as frequencies and tested using the Chi-square test. The Kruskal–Wallis *H* test was used for comparisons between dystonia, choreoathetosis, and mixed type of the different functional levels of the GMFCS, MACS, and CFCS. Similarly, the distributions of the 5 levels of the GMFCS, MACS, and CFCS in dyskinetic CP subgroups were compared using the Kruskal–Wallis *H* tests. The distribution of each functional level was compared dichotomously between 2 different subtypes. The distribution of levels between 2 different functional classification systems in individual subtype was compared using the Wilcoxon rank sum tests. Statistical analysis was conducted with SPSS 18.0

Table 1

The 5 levels of the GMFCS, MACS, and CFCS.

Level	GMFCS	MACS	CFCS
I	Walks without limitations	Handles objects easily and successfully	Sends and receives information with familiar and unfamiliar partners effectively and efficiently
II	Walks with limitations	Handles most objects but with somewhat reduced quality and/or speed of achievement	Sends and receives information with familiar and unfamiliar partners but may need extra time
III	Walks using a hand-held mobility device	Handles objects with difficulty; needs help to prepare and/or modify activities	Sends and receives information with familiar partners effectively, but not with unfamiliar partners
IV	Self-mobility with limitations; may use powered mobility	Handles a limited selection of easily managed objects in adapted situations	Inconsistently sends and/or receives information even with familiar partners
V	Transported in a manual wheelchair	Does not handle objects and has severely limited ability to perform even simple actions	Seldom effectively sends and receives information even with familiar partners

CFCS = Communication Function Classification System, GMFCS = Gross Motor Function Classification System, MACS = Manual Ability Classification System.

(IBM, Armonk, NY). Two-sided *P* values <.05 were considered statistically significant.

3. Results

3.1. Characteristics of the patients

Ninety-three children (61 (65.6%) males and 32 (34.4%) females) with a diagnosis of dyskinetic CP were included in this study (Table 2). The children were 4 to 13 years of age (mean, 5.5 ±1.8 years) at the last follow-up. Twenty-six children belonged to the dystonic group, 26 to the choreoathetosic group, and 41 to the mixed type group (dyskinetic with spastic features).

3.2. Maternal and birth characteristics

Of all 93 children, 33 (35.5%) were born before the end of the 37th week of gestation (which is defined as preterm), 59 (63.4%) were born at term, and 1 (1.1%) was born at 42 weeks. Seven (7.5%) children were one of twins (Table 2). The occurrence of neonatal adverse events across dyskinetic CP subtypes are presented in Table 3. Peri/neonatal adverse events were present in 79 (85.0%) children. None of the 93 children had postneonatally acquired CP.

3.3. Neuroimaging

MRI findings were available for 87 participants. The brain MRI findings were normal in 10 participants; 28 participants showed lesions in the basal ganglia or/and thalamus; 22 showed pure white matter lesions; and 19 had lesions involving thalamus or/and basal ganglia, combined with cortico-subcortical and white matter lesions. Cortico-subcortical lesions were found in 3 children. Representative images demonstrating the differences in lesion distribution in dyskinetic CP children are shown in Fig. 1. Congenital brain malformation was found in 5 patients out of 14 (15.03%) children without obvious peri/neonatal adverse. Those malformations included 3 macrogyria, 1 diencephalon fusion, and 1 gray matter heterotopia. The brain MRI findings according to the neurologic subtypes are shown in Table 4.

Table 2

Characteristics of the patients.

	Choreoathetosis, n = 26	Dystonia, n = 26	Mixed type, n = 41
Gender, n (%)			
Male	18 (69.2)	15 (57.7)	28 (68.3)
Female	8 (30.8)	11 (42.3)	13 (31.7)
Birth weight			
<2500 g	5 (19.2)	4 (15.4)	17 (41.5)
2500–3999 g	19 (73.1)	20 (76.9)	21 (51.2)
≥4000 g	2 (7.7)	2 (7.7)	3 (7.3)
Twins or multiplets	2 (7.7)	2 (7.7)	3 (7.3)

Table 3

Neonatal adverse events according to the subtypes of dyskinetic CP.

	Choreoathetosis, n = 26		Dystonia, n = 26		Mixed type, n = 41	
Neonatal adverse events	Premature, n = 6	Term, n = 20	Premature, n = 11	Term, n = 15	Premature, n = 16	Term, n = 25
Hyperbilirubinemia + HIE	1 (17)	—	—	2 (13)	4 (25)	1 (4)
Hyperbilirubinemia + asphyxia	—	—	2 (18)	2 (13)	3 (19)	2 (8)
Hyperbilirubinemia	1 (17)	5 (25)	8 (73)	7 (47)	3 (19)	6 (24)
Asphyxia	3 (50)	1 (5)	—	—	3 (19)	—
HIE	—	4 (20)	1 (9)	2 (13)	—	9 (36)
Total	5	10	11	13	13	18

HIE = hypoxic-ischemic encephalopathy.

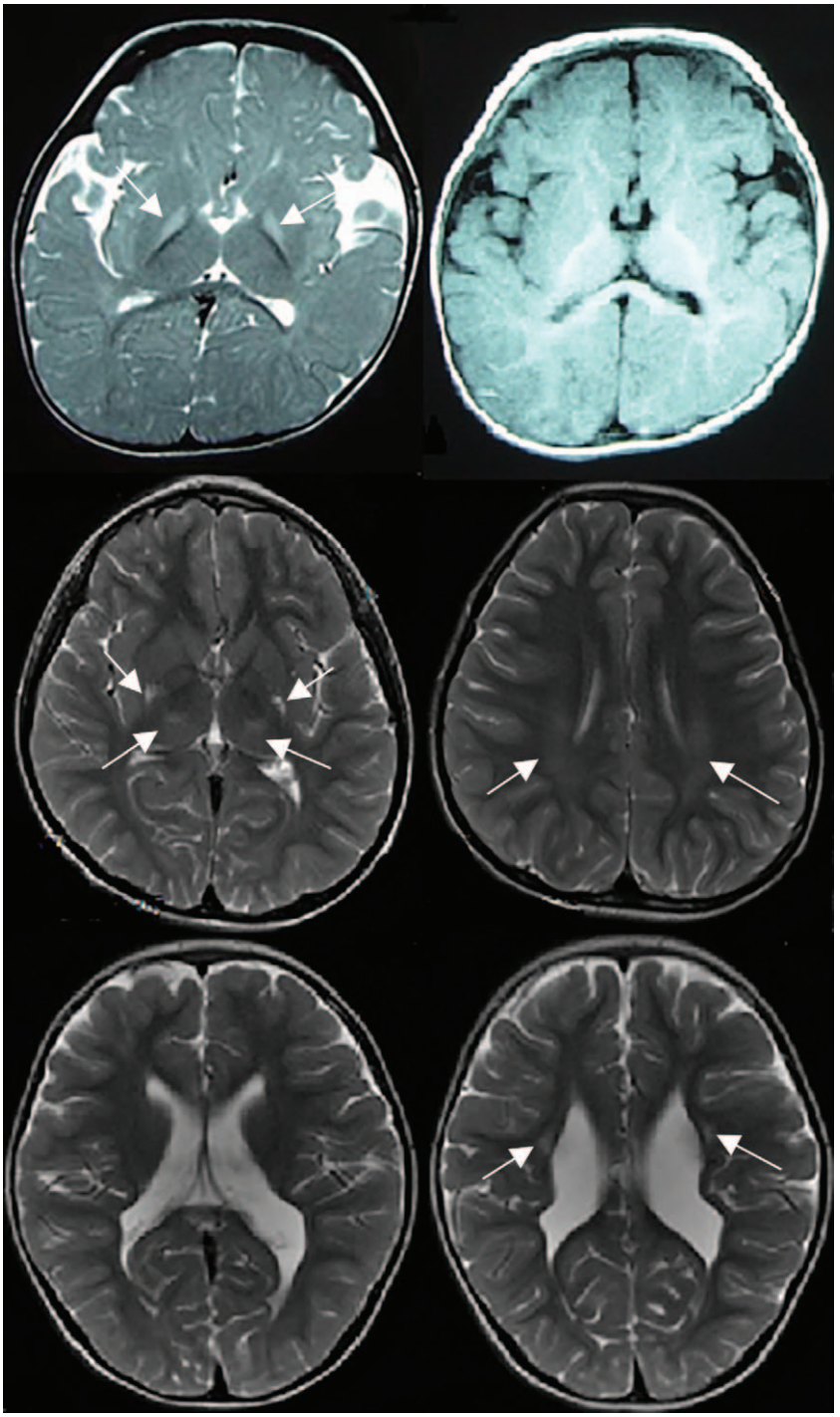

Figure 1. Upper: Axial MRI of bilateral basal ganglia lesions in a 19-month-old female with dystonic CP and a history of hyperbilirubinemia. Left: Axial T2-weighted image demonstrates bilateral, symmetric hyperintensity in the globus pallidus (arrows). Right: Axial T1-weighted image showed no signal-intensity abnormality in the globus pallidus. Middle: Axial MRI of basal ganglia/thalamus lesion combined with white matter lesion in a 8-year-old male with choreoathetosic CP and a history of profound HIE. Axial T2-weighted images showed putament, thalamus, and paracentral white matter lesions (arrows). Lower: Axial MRI of white matter lesions in a 34-month-old female with mixed CP, 29^{+4} week gestation and a history of mild asphyxia. Axial T2-weighted image demonstrates periventricular leukomalacia, arrow indicates the periventricular gliosis.

3.4. Accompanying impairments

Learning disability was present in 45 children, especially in the dystonia and mixed subtypes. Children suspected of auditory impairment through clinical observation had auditory brainstem response (ABR) testing, which showed sensorineural deafness in 10 children, 7 of them having a >95-dB hearing loss. Epilepsy was found in 21 children: 6 with infantile spasms and 15 with partial epilepsy (with 5 out of those 15 with partial seizure secondarily generalized). The frequencies of different comorbidities according to the neurologic subtype are shown in Table 5. For all comorbidities except for inspiratory laryngeal stridor, the

Table 4

Neuroimaging findings according to the subtypes of dyskinetic CP.

Neuroimaging finding	Choreoathetosia, n (%)	Dystonia, n (%)	With spasticity, n (%)
Basal ganglia lesion	6 (23)	16 (67)	6 (16)
White matter lesion only	6 (23)	1 (4)	15 (41)
Basal ganglia/thalamus/cortico-subcortical lesion combined with white matter lesion	5 (19)	5 (21)	9 (24)
Basal ganglia/thalamus lesion/cortico-subcortical lesion	—	—	3 (8)
Maldevelopment	2 (8)	1 (4)	2 (5)
Normal findings	7 (27)	1 (4)	2 (5)
Not done or lost	—	2	4
Total	26	26	41
% of abnormalities	19/26 (73)	23/24 (96)	35/37 (95)

Table 5

Frequency of comorbidities according to the neurological subtypes.

Comorbidities, n (%)	Choreoathetosis (n = 26)	Dystonia (n = 26)	Mixed type (n = 41)	P_1	P_2
Cognition impairment	5 (19)	19 (70)	21 (51)	.627	<.001
Microcephaly	1 (4)	1 (4)	10 (24)	.003	1.000
Epilepsy	3 (12)	4 (15)	14 (34)	.018	1.000
Cortical visual dysfunction	1 (4)	2 (8)	9 (22)	.021	1.000
Severe auditory impairment	2 (8)	8 (31)	0	.008	.035
Inspiratory laryngeal stridor	2 (8)	5 (19)	4 (10)	.821	.416

P_1: choreoathetosis + dystonia vs mixed type.
P_2: choreoathetosis vs dystonia.

differences in the distribution among dyskinetic CP subtypes were significant.

3.5. Functional status

The functional performance from levels I to V for each scale of the 3 subgroups is provided in Table 6. The distributions of GMFCS, MACS, and CFCS levels among different dyskinetic CP subtypes are shown in Fig. 2. GMFCS, MACS, and CFCS were distributed disproportionately in the dystonia ($P < .001$) and mixed subgroups ($P = .001$), except for the choreoathetosis group ($P = .39$). The functional level distribution of each classification system in the 3 subgroups was statistically significant ($P < .001$).

There were no significant difference between GMFCS and MACS levels in the 3 subgroups ($P > .05$). In the dystonia and mixed groups, a significant difference was found between GMFCS and CFCS, as well as between MACS and CFCS ($P < .01$). Statistical comparison between 2 different subtypes for functional levels revealed that gross motor and fine motor functional levels were distributed disproportionately between the dystonia, choreoathetosia, and mixed type groups ($P < .01$).

4. Discussion

Dyskinetic CP is the second major subtype of CP. Dyskinetic CP can be classified into different subtypes, but the exact clinical characteristics of these subtypes are poorly defined. Therefore, this study aimed to investigate the clinical characteristics and functional classifications of dyskinetic CP from the perspective of neurologic subtype in a hospital-based follow-up study. The results showed that in dyskinetic CP, different subtypes have their own specific comorbidities, radiological characteristics, and functional status attributes according to their etiological factors and brain lesions. Children with dystonia have more limited functional status than children with choreoathetosia.

Dystonia and choreoathetosis are often simultaneously present, and dystonia often dominates in dyskinetic CP. In addition, spasticity is present in about 70% of the children with dyskinetic CP. Despite the fact that dystonia and choreoathetosis

Table 6

Distribution of GMFCS, MACS, and CFCS levels by subtypes of dyskinetic CP functional level.

	Dystonia (n = 26)			Choreoathetosia (n = 26)			Mixed with spasticity (n = 41)		
	GMFCS, n (%)	MACS, n (%)	CFCS, n (%)	GMFCS, n (%)	MACS, n (%)	CFCS, n (%)	GMFCS, n (%)	MACS, n (%)	CFCS, n (%)
I	0	0	1 (4)	9 (35)	6 (23)	7 (27)	4 (10)	1 (2)	11 (27)
II	0	1 (4)	5 (19)	3 (12)	12 (46)	14 (54)	6 (15)	16 (39)	15 (37)
III	1 (4)	2 (8)	7 (27)	9 (35)	5 (19)	4 (15)	12 (29)	5 (12)	6 (15)
IV	6 (23)	5 (19)	8 (31)	5 (19)	2 (8)	1 (4)	12 (29)	12 (29)	8 (20)
V	19 (73)	18 (69)	5 (19)	0	1 (4)	0	7 (17)	7 (17)	1 (2)

In the dystonia group: $P < .001$ for GMFCS vs CFCS, and $P < .001$ for MACS vs CFCS.
In the mixed type group, $P = .001$ for GMFCS vs CFCS, and $P = .002$ for MACS vs CFCS.
CFCS = Communication Function Classification System, GMFCS = Gross Motor Function Classification System, MACS = Manual Ability Classification System.

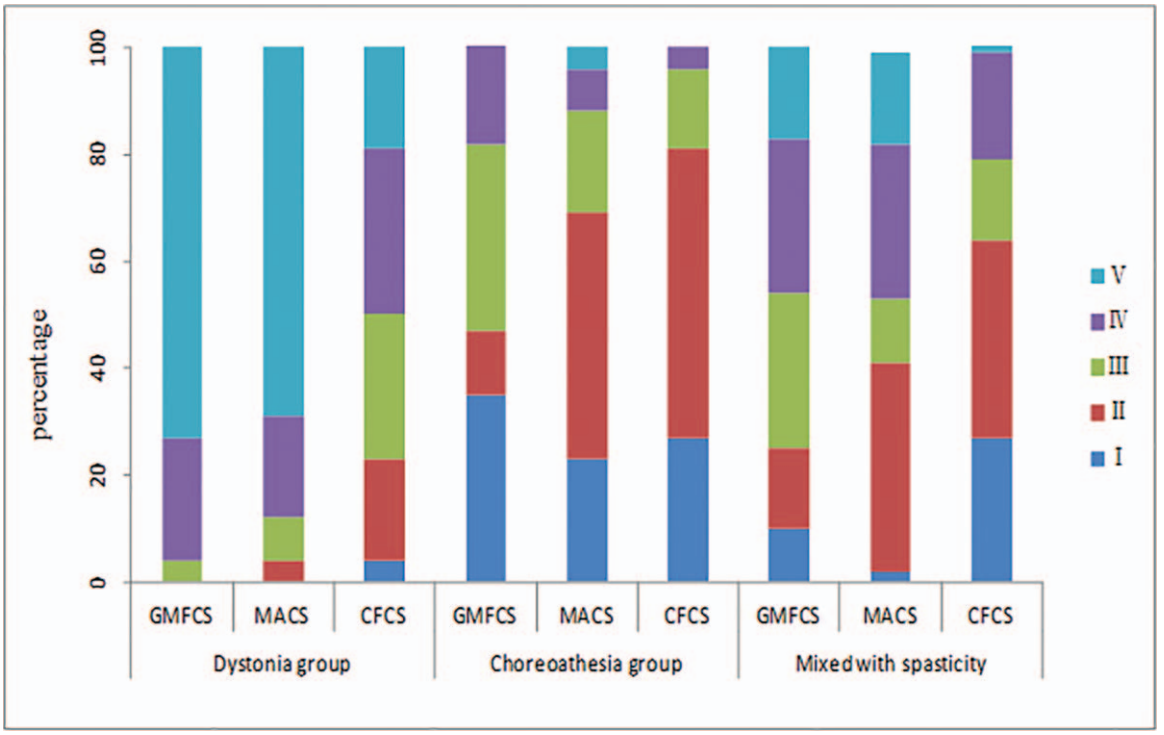

Figure 2. Distributions of GMFCS, MACS, and CFCS levels by subtypes of dyskinetic CP in 93 children. For choreoathetosis vs dystonia, all $P < .001$ for all 3 scales. For choreoathetosis vs mixed, $P = .005$ for GMFCS, $P = .002$ for MACS, and $P = .26$ for CFCS. For dystonia vs mixed, $P < .001$ for GMFCS, $P < .001$ for MACS, and $P = .001$ for CFCS (Kruskal–Wallis H test).

are often present simultaneously, those 2 features are independent, leading to 2 independent categories. In addition, involuntary spasms are also found in some children with dyskinetic CP, leading to the mixed category. This classification is supported by recent guidelines.[1,5] In the present study, all patients but 2 could be easily classified. Nevertheless, this classification remains controversial. Indeed, dyskinetic CP is very common in the clinical practice. Some previous studies have classified children with CP and involuntary spasms as spastic CP, while children with spasms with dominant dyskinesia were classified as dyskinetic CP of the mixed subtype. The spasms of these children were characterized as velocity-dependent hypermyotonia, stretch hyperreflexia, and with positive pathological signs and ankle clonus. Although spasms and dystonia are relatively hard to distinguish in some cases (2 cases in the present study), the abnormal signs in the physical examination could help the classification. In addition, the children with dyskinetic CP only could be classified as the dystonic and choreoathetotic subtypes.[8] Actually, the proportion of children with CP and both of these 2 manifestations is higher than the proportion of children with only one of the 2 manifestations. The clinical presentations of these 2 subtypes are somewhat different, mainly regarding the positions, velocity, and predictability of the involuntary movements, as well as the patterns of the abnormal movements. The present study was performed according to the 2008 consensus of the Taskforce on Childhood Movement Disorder[5] and our clinical experience, and most of the children could be classified into subtypes according to the clinical presentations. For the 2 children that the subtypes could not be easily classified, we referred to the DIS[17] for subtype classification. Finally, 1 child was classified as dystonic, and the other 1 was classified as choreoathetotic.

In the present hospital-based study, the mixed type accounted for a larger proportion than the dystonic and choreoathetosic types, while the latter 2 types accounted for an equal proportion. Hyperbilirubinemia was present in 47 cases and significantly more common in the dystonic subtype (21/26, 80.8%), as supported by recent studies.[4,19] Hyperbilirubinemia occurred more frequently in the mixed subgroup (21/41, 51.2%). Two primary injury patterns, the "central" model and the "peripheral" model, are caused by hypoxic-ischemic brain damage (HIBD) with different clinical sequelae.[20,21] Both models have been observed in the present study. If the typical lesion site (thalamus and putamen) is involved, dyskinesia may be predominant with relatively well-preserved intellect; when the cortex structure or/and optic radiations are also involved, profound intellectual impairment or vision disorder is present; and when the periventricular white matter is affected, spastic symptoms are present. Therefore, the different clinical manifestations lie in the affected anatomic location. The specific patterns of brain injury help distinguish HIBD from injury following kernicterus, in which the globus pallidus, subthalamic nuclei, brainstem auditory, and oculomotor nuclei are more vulnerable, and lead to diverse clinical manifestations as auditory or/and movement disorders.[22]

In the present study, 88.5% (77/87) of the MRI examinations were abnormal, and the different lesions were not equally distributed among the subtypes of dyskinetic CP. The most frequent lesions were in the basal ganglia, which were observed more commonly in the dystonia group (87.5% of the cases, 21/24), compared with 48% in the mixed and 42% in the choreoathetosia groups, respectively. Pure white matter or combined white matter lesions were less frequent, and mainly occurred in the mixed type, accounting for 64.9% of the cases,

followed by the choreoathetosia subtype (11/26, 42.31%). Cortico-subcortical lesions were present in 32% of children with the mixed type. Congenital brain malformation was found in 5 children, which were suspected to be associated with the congenital dysplasia caused by adverse factors during the pregnancy. The distribution of the 3 subtypes in these 5 children was not significantly different (n=2, n=1, and n=2). Ten children showed normal brain MRI imaging; they were mainly in the choreoathetosis group (7/26, 26.9%).

Comorbidities were different among the different subtypes. For mixed type with spasticity, microcephaly, epilepsy, and cortical visual dysfunction occurred especially frequently. Cognition impairment and auditory impairment was more common among children with the dystonic subtype, while there was no difference for laryngeal stridor.

In the present study, 10 children had auditory dysfunction, with eight in the dystonia dominant group and 2 in the choreoathetosia group. They all had experienced perinatal adverse events and their neuroimaging findings all showed abnormal signals in the globus pallidus, which supported hyperbilirubinemia as a perinatal etiology.

In children with smaller head circumference, hypoxic-ischemic brain damage was present in 5 children, intracranial hemorrhage in 2, prematurity in 3, and 2 showed a congenital cerebral malformation (macrogyria). Except for the 2 cases of macrogyria, the MRI findings all showed lesions in the pericentral white matter. Therefore, we presume that microcephaly is most probably secondary to the paracentral white matter volume loss.

Epilepsy occurred more frequently in the mixed subtype with spasticity (14/41, 34.15%) than in the dyskinetic subtype (7/52, 13.46%). In most cases, lesions in the white matter and deep gray matter unlikely gave rise to epilepsy unless the cortex is involved. In the present study, the majority of children with epilepsy had adverse events of hypoxic-ischemic brain damage with lesions in the rolandic areas or other cortico-subcortical areas. Furthermore, congenital brain malformation (macrogyria) and gray matter heterotopia were observed in the 3 children with epilepsy and could also be a strong predictive factor for epilepsy.

Inspiratory stridor was common in dyskinetic CP children. It is caused by laryngeal dystonia (LD),[23] which is related to lesions in the basal ganglia. In some cases, LD also manifests as spasmodic dysphonia (SD), and it may be more common than inspiratory stridor as the manifestation of LD.[24] In the present study, cognition impairment occurred more frequently in the dystonia and mixed subtypes. In the dystonia group, lesions in the basal ganglia were common and had less adverse effects on cognition than lesions in the cortex, which could be seen commonly in the mixed group. The presence of poor results in IQ tests is associated with the severity of motor disability and communication impairment.

In dyskinetic CP, GMFCS and MACS levels among the 3 subgroups showed no significant difference. Thus, it illustrated that GMFCS levels correlated well with MACS levels.[25] Dyskinesia movements can be seen in the upper and lower limb regions in pure dyskinetic CP. On the other hand, spasticity was more predominant in the lower extremities while dyskinesia was more pronounced in the upper limbs in the mixed type. The combination had a negative effect on mobility and handling object performance. In the dyskinetic and mixed groups, gross motor and manual ability were more limited than the communication function, as the advent of augmentative and alternative communication (AAC) can benefit to communicate with others. In this study, the proportions of children with grades IV and V GMFCS, MACS, and CFCS scores were significantly higher in the dyskinetic group than in the choreoathetosis group. Dystonia-predominant children showed more severe functional classification levels than the choreoathetosis subgroup, in particular for GMFCS and the MACS. The presence of dystonia indicated more negative effect on motor functions and on communication than choreoathetosis due to dystonia patterns hampering posture maintenance, coordination, and trigger of voluntary movement.

The present study is not without limitations. It was a hospital-based study and the patients were from a clinical case series rather than population-based; so, their characteristics do not represent those of a population-based sample. Furthermore, the distinction of subtypes was determined according to the clinical manifestations and physical signs, which were based on the definitions of the SCPE[1] and the Taskforce on Childhood Movement Disorders,[5] while no discrimination scales were used to evaluate the presence and severity of dystonia, choreoathetosis, and spasticity. To improve the reliability of subtype differentiation, 2 experienced doctors collaborated to determine the classification of CP types according to their clinical observations. In addition, this study was a single center study, and the sample size was relatively small. We hope to further establish a multicenter network for the management of CP, and increase the sample size in our future studies.

5. Conclusion

In dyskinetic CP, different subtypes have their own specific comorbidities, radiological characteristics, and functional attributes according to their etiological factors and brain lesions. Children with dystonia have more limited functional status than children with choreoathetosia.

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Validation: Dianrong Sun, Mei Hou, Yutang Li, Rong Yu.
Visualization: Jianhui Zhao.
Writing—original draft: Dianrong Sun.
Writing—review & editing: Dianrong Sun.

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