The Effects of Associated Impairments on Severity of Cerebral Palsy: Insights from a Single-Center

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Abstract

Aim: The aim of the study was to describe the distribution of the impairment index across Cerebral Palsy (CP) subtypes and Gross Motor Function Classification System (GMFCS) levels and investigate the relationship between CP subtypes, birth weight, gestational age, and the impairment index.

Method: This retrospective study was conducted with 423 children with CP aged between 0-18 years. Data were gathered from the medical records of children. Birth weight and week, functional classification levels, intellectual impairment, presence of vision, hearing problems, epilepsy recorded. All children were classified according to the GMFCS, Manual Abilities Classification System (MACS), Communication Function Classification System (CFCS), Eating and Drinking Abilities Classification System (EDACS), and CP subtypes. Participants were categorized according to Impairment Index (II) that consist of gross motor, intellectual, vision and hearing impairments and epilepsy. Multivariate backward modelling linear regression model was used to explain relations between impairment index and functional classification systems, birth weight and birth week.

Results: Amongst the 423 children (mean age 6.38±4.57 years) analyzed, 130 (30.7) of the children had low impairment, 159 (31.7%) of them had moderate impairment, and 134 (31.7%) of them had high impairment according to the II. In unilateral spastic type CP, 61.5% had a low impairment index ($p<0.05$), in bilateral spastic CP, 44.2% of children had a moderate impairment index ($p<0.05$), in dyskinetic CP 67.9% of children had a high impairment index ($p<0.05$). In ataxic type there were not any difference significantly between impairment index levels ($p=0.06$). As a result of regression analysis gross motor function level (Beta=0.85, $p<0.01$) and birth weight (Beta=-0.05, $p=0.04$) were predictors of the Impairment Index, and explained 73% of the variance.

Conclusion: Nearly one third of the children had high II; birth weight and gross motor functional level are the predictors of II. These results can help improve rehabilitation and social services.

Keywords: Cerebral palsy, physiotherapy, impairment, ICF.

Özgün Araştırma Makalesi (Original Research Article)
Geliş / Received: 14.08.2024 & **Kabul / Accepted:** 20.03.2025
DOI: https://doi.org/10.38079/igusabder.1529388

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ETHICAL STATEMENT: Gathered from Hacettepe University, Non-invasive Clinical Researches Ethical Committee (Number: GO 20/356; Date: 17.04.2020).

Serebral Palside Eşlik Eden Bozuklukların Serebral Palsi Şiddetine Etkisi: Tek Merkez Bulguları

Öz

Amaç: Çalışmanın amacı, Serebral Palsi (SP) alt tipleri ve Kaba Motor Fonksiyon Sınıflandırma Sistemi (GMFCS) seviyeleri arasında Bozukluk İndeksi (Bİ) dağılımını tanımlamak ve Bİ, SP alt tipleri, doğum ağırlığı ve doğum haftası arasındaki ilişkiyi incelemektir.

Yöntem: Bu retrospektif çalışma 0-18 yaş aralığında 423 SP'li çocukla yürütüldü. Veriler çocukların tıbbi kayıtlarından toplandı. Doğum ağırlığı ve haftası, fonksiyonel sınıflandırma seviyeleri, bilişsel bozukluk, görme, işitme sorunları, epilepsi varlığı kaydedildi. Tüm çocuklar GMFCS, El Becerileri Sınıflandırma Sistemi (MACS), Yeme ve İçme Becerileri Sınıflandırma Sistemi (EDACS), İletişim Fonksiyonu Sınıflandırma Sistemi (CFCS) ve SP alt tiplerine göre sınıflandırıldı. Katılımcılar, kaba motor, bilişsel, görme ve işitme bozuklukları ile epilepsiden oluşan Bİ'ye göre kategorize edildi. Bİ ile fonksiyonel sınıflandırma sistemleri, doğum ağırlığı ve doğum haftası arasındaki ilişkileri açıklamak multivariat doğrusal regresyon modeli kullanıldı.

Bulgular: Analiz edilen 423 çocuktan (ortalama yaş 6,38±4,57 yıl) Bİ'ye göre 130'unda (%30,7) düşük düzeyde bozukluk, 159'unda (%31,7) orta düzeyde bozukluk ve 134'ünde (%31,7) yüksek düzeyde bozukluk saptandı. Unilateral spastik SP'li çocukların %61,5'inde düşük düzeyde Bİ ($p<0,05$), bilateral spastik SP'li çocukların %44,2'sinde orta düzeyde Bİ ($p<0,05$), diskinetik SP'li çocukların %67,9'unda yüksek düzeyde Bİ $p<0,05$) saptandı. Ataksik tipte ise Bİ düzeyleri arasında anlamlı bir fark yoktu ($p=0,06$). Regresyon analizi sonucunda kaba motor fonksiyon düzeyi (Beta=0,85; $p<0,01$) ve doğum ağırlığının (Beta=-0,05; $p=0,04$) Bİ'nin yordayıcıları olduğu ve varyansın %73'ünü açıkladığı görüldü.

Sonuç: Çocukların yaklaşık üçte biri yüksek Bİ'ye sahipti; doğum ağırlığı ve kaba motor fonksiyonel seviyesi Bİ'nin prediktörleridir. Bu sonuçlar, SP'de rehabilitasyona yönelik yaklaşımların geliştirilmesine ve sosyal hizmetlerin iyileştirilmesine yardımcı olabilir.

Anahtar Sözcükler: Serebral palsi, fizyoterapi, bozukluk, ICF.

Introduction

Cerebral palsy (CP) is the most prevalent cause of physical disability of childhood and defined as a group of lifelong non-progressive disorders that impact the development of posture and movement in the developing brain and resulted by activity limitation[1,2].

CP characterized by motor function, posture and communication[3] impairments; moreover, associated problems like epilepsy and intellectual impairment as well as impairments of hearing and vision are common. The degree of these impairments varies from mild to severe inability[4].

With use of the International Classification of Functioning, Disability and Health (ICF) framework, professionals started to apply the ICF framework both for interventions and research purposes[5] and the influence of impairments of bodily structure and functions on social as well as personal life is understood better in accordance with the ICF framework. In those with CP, beside to CP definition, based on motor function and its neurological frame, the existence or lack of associated impairments significantly affect participation and as well as well-being[6].

Although the prevalence of associated problems in people with CP varies according to the various surveillances, common associated impairments are intellectual disability within the range of 30–50%, epilepsy within 15-55%, vision within 10–100% and hearing impairment 30–40%. The prevalence of associated problems according to various functional levels in CP is unclear. Therefore, investigation among functional state and associated problems in CP can be helpful for planning proper intervention programs[7].

CP has been classified using a combination of the motor characteristics as well as classification systems such as the Gross Motor Function Classification System (GMFCS), the Manual Ability Classification System (MACS), the Communication Function Classification System (CFCS), and the Eating and Drinking Ability Classification System (EDACS). On the other hand, these classification systems explain functional capacity for each single ability, such as gross motor abilities, manual abilities or communication rather than severity level. There is no commonly agreed definition of “severe CP”, definitions may refer to motor function only or to multiple impairments[8]. Therefore, recently, in order to evaluate the level of impact in people with CP together with motor, cognitive, and associated impairments, the "impairment index" has been defined, which evaluates the motor involvement severity, the associated impairments presence, and cognitive inadequacy together and defines the impact of CP on the child as mild, moderate, and severe impairment[6].

This study analyses data hospital based data retrospectively and centered on the definition of CP based on the severity of impairment using the impairment index, which aims to describe impairments combination of as well as severity of these impairments. This study aims to describe the distribution of the impairment index across subtypes of CP and GMFCS levels, and evaluate the association between CP subtypes, birth weight, gestational age, and impairment index.

Material and Methods

This single-center retrospective study was completed at Hacettepe University, Faculty of Physical Therapy and Rehabilitation, between July 2020 - July 2022. Ethical approval for the study was gained from Hacettepe University, Ethical Committee of Non-invasive Clinical Researches (Number: GO 20/356; Date: 17.04.2020).

Study Population, Exclusion Criteria

Data of the study were collected from medical records of children. Inclusion criteria were being age <18 years, diagnosis of CP. Date of birth, birth weight and week, functional classification levels, intellectual impairment, presence of vision, hearing problems and epilepsy recorded. Children with missing any of these relevant records and any neurological, genetic, or metabolic disorders co-existence with CP were excluded.

Classification of Functional Abilities

The Turkish versions of GMFCS[9], MACS[10], CFCS[11], and EDACS[12] were used to classify the functional levels. All Turkish versions were valid and reliable[9-12].

Gross Motor Function Classification System (GMFCS): classifies motor functional abilities in CP. The distinction between levels are based on functional limitations, the necessity for hand-held aids or wheeled devices for mobility[9,13].

Manual Abilities Classification System (MACS): classifies the hand skills of children with CP, like object grasping and releasing in daily life within five levels[10,14].

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Eating and Drinking Abilities Classification System (EDACS): classifies eating and drinking abilities in children with CP into five levels. EDACS aims to differentiate and classify the daily eating and drinking abilities in people with CP[12,15].

Communication Function Classification System (CFCS): classifies the communication function of people with CP in daily life within five levels (Level I–V)[11,16].

Cerebral Palsy Classification

CP subtypes was classified according to the classification of Surveillance of CP in Europe as unilateral spastic (US-CP) bilateral spastic (BS-CP), dyskinetic, and ataxic[17].

Associated Impairments

Intellectual impairment was determined according to the guidance research center of the Ministry of National Education; visual and hearing impairments, and epilepsy were recorded according to medical records of the last testing. In case of lack of medical reports, the patient excluded from the study.

Intellectual impairment was categorized into three intelligence quotient (IQ) categories or estimated IQ: normal/near-normal intellect (IQ>70), mild to moderate intellect (IQ between 50-69), and severe impairment of intellect (IQ<50)[6]. Impairments of vision and hearing were noted as the existence or non-existence. Presence of epilepsy, which is described as having two unprovoked seizures or receiving antiepileptic treatment, determined by a child neurologist according to medical reports.

Impairment Index

Impairment index consists of three levels according to the severity and impairment combinations.

Low impairment was defined as GMFCS Level I–II (able to walk without any support), IQ>70, neither impairment of visual nor hearing, and not having epilepsy.

High impairment was defined as GMFCS Level IV–V (inability to walk) and/or IQ<50 (severe impairment of intellect) with or without at least one of the impairments of visual, hearing, and active epilepsy.

Moderate impairment was defined as GMFCS Level I–II, IQ>70, but with one or more of the impairment of visual, hearing, and active epilepsy; or being GMFCS Level I–II with an IQ between 50-70, with or without at least one of the impairment of visual, hearing, and active epilepsy; or GMFCS Level III (walking with support), with an IQ<50 with or without one or more of the impairment of visual, hearing, and active epilepsy[6].

Statistical Methods

The Statistical Package for Social Sciences (SPSS) software version 21.0 was used. Categorical variables were given as numbers and percentages. Chi-square test was used as appropriate for measurement of relations. Multivariate backward modelling linear regression model was used to explain relations between impairment index and functional classification systems, birth weight and birth week.

Results

423 children with CP who met the inclusion criteria included. Majority of children, 76.6% (n=324) were spastic type (unilateral and bilateral), following dyskinetic (n=78, 18.4%),

and 5% (n=21) were ataxic. The social and demographic properties are presented in Table 1.

Table 1. Social and demographic properties of participants

Social and demographic properties		**Mean±Standard Deviation (min – max)**	
Age (years)		6.38±4.57 (2-18)	
Birth Week		35.40±4.63 (23 – 43)	
Birth Weight (grams)		2541.83±929.36 (660 – 5750)	
Gender	**Female n (%)**	179 (42.3)	
	Male n (%)	244 (57.7)	
Clinical Type		**n**	**%**
Unilateral Spastic		109	25.8
Bilateral Spastic		215	50.8
Dyskinetic		78	18.4
Ataxic		21	5.0

n, number; % percentage

Among classification systems, according to the GMFCS, 23.6% (n=100) were level 1, 27.0% (n=114) were level II, 18.9% (n=80) were level III, 16.3% (n=69) were level IV and 14.2% (n=60) were level V; according to the MACS, 41.8% (n=117) were level 1, 23.9% (n=101) were level II, 13.9% (n=59) were level III, 12.3% (n=52) were level IV and 8.0% (n=34) were level V. According to the CFCS, 57.7% (n=224) were level 1, 14.9% (n=63) were level II, 13.9% (n=59) were level III, 7.8% (n=33) were level IV and 5.7% (n=24) were level V and according to the EDACS, 64.8% (n=274) were level 1, 15.1% (n=64) were level II, 9.9% (n=42) were level III, 4.5% (n=19) were level IV and 5.7% (n=24) were level V.

The distribution of GMFCS, MACS, CFCS and EDACS according to clinical types are presented in Table 2.

Table 2. Distribution of GMFCS, MACS, CFCS and EDACS according to clinical types

Clinical type		**GMFCS n (%)**	**MACS n (%)**	**CFCS n (%)**	**EDACS n (%)**
Unilateral spastic (n=109)	Level I	63 (57.8)	56 (51.4)	91 (83.5)	100 (91.7)
	Level II	43 (39.4)	34 (31.2)	15 (13.8)	5 (4.6)
	Level III	3 (2.8)	17 (15.6)	3 (2.8)	4 (3.7)
	Level IV	-	2 (1.8)	-	-
	Level V	-	-	-	-
Bilateral spastic (n=215)	Level I	30 (14.0)	102 (47.4)	121 (56.3)	140 (65.1)
	Level II	45 (20.9)	42 (19.5)	27 (12.6)	29 (13.5)
	Level III	67 (31.2)	25 (11.6)	32 (14.9)	18 (8.4)
	Level IV	39 (18.1)	25 (11.6)	19 (8.8)	13 (6.0)
	Level V	14 (15.8)	21 (9.8)	16 (7.4)	15 (7.0)
Dyskinetic (n=78)	Level I	3 (3.8)	8 (10.3)	14 (17.9)	18 (23.1)
	Level II	15 (19.2)	17 (21.8)	20 (25.6)	25 (32.1)

	Level III	5 (6.4)	15 (19.2)	22 (28.2)	20 (25.6)
	Level IV	29 (37.2)	25 (32.1)	14 (17.9)	6 (7.7)
	Level V	26 (33.3)	13 (16.7)	8 (10.3)	9 (11.5)
Ataxic (n=21)	Level I	4 (19.0)	11 (52.4)	18 (85.7)	16 (76.2)
	Level II	11 (52.4)	8 (38.1)	1 (4.8)	5 (23.8)
	Level III	5 (23.8)	2 (9.5)	2 (9.5)	-
	Level IV	1 (4.8)	-	-	-
	Level V	-	-	-	-

n, number; % percentage

Associated impairments and gross motor function in overall CP. Among all the participants, 214 (50.6%) were walking without support (GMFCS Level I-II), 80 (18.9%) was walking with support (GMFCS Level III), and 129 (30.5%) of children were GMFCS Level IV-V. Most of the cases (n=231, 54.6%) had an IQ of 70 (normal intellect), following 135 (31.9%) an IQ between 50-69, and 57 (13.5%) an IQ of < 50. Impairment of the vision was in 133 (31.4%) of children, and impairment of hearing was in 23 (5.4%) of children. Overall, 103 (24.3%) had epilepsy. According to the Impairment Index, 130 (30.7) of the children had low impairment, 159 (31.7%) of them had moderate impairment, and 134 (31.7%) of them had high impairment. The frequency of categories according to the Impairment Index and associated impairments according to the CP subtypes were presented in Table 3.

Table 3. Impairment Index and associated impairments in CP subtypes

		Unilateral Spastic n (%)	**Bilateral Spastic n (%)**	**Dyskinetic n (%)**	**Ataxic n (%)**
Impairment Index	**Low**	67 (61.50)	43 (20.0)	10 (12.8)	10 (47.6)
	Moderate	42 (38.50)	95 (44.20)	13 (16.7)	9 (42.9)
	High	-	77 (35.80)	53 (67.9)	2 (9.5)
Gross Motor Function	**Level I-II**	106 (97.2)	75 (34.9)	18 (23.1)	15 (71.4)
	Level III	3 (2.8)	67 (31.2)	5 (6.4)	5 (23.8)
	Level IV-V	-	73 (34.0)	55 (70.5)	1 (4.8)
Intellectual	**IQ <50**	-	36 (16.7)	20 (25.6)	1 (4.8)
	IQ 50-70	20 (18.3)	66 (30.7)	45 (57.7)	4 (19.0)
	IQ >70	89 (81.7)	113 (52.6)	13 (16.7)	16 (76.2)
Visual	**Yes**	84 (22.0)	87 (40.5)	18 (23.1)	4 (19.0)
	No	85 (78.0)	128 (59.5)	60 (76.9)	17 (81.0)
Hearing	**Yes**	2 (1.8)	15 (7.0)	6 (7.7)	-
	No	107 (98.2)	200 (93.0)	72 (92.3)	21 (100.0)
Epilepsy	**Yes**	24 (22.0)	55 (25.6)	22 (28.2)	2 (9.5)
	No	85 (78.0)	160 (74.4)	56 (71.8)	19 (90.5)

n, number; % percentage

The results of the backward modelling linear regression analysis showed that among the independent variables, gross motor function level (Beta=0.85, p<0.01) and birth weight

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(Beta=-0.05, p=0.04) were strong predictors of the Impairment Index, and explained 73% of the variance (Table 4).

Table 4. Results of regression analyses

Model: Multiple linear regression (backward modeling)						
Dependent variable	**Independent variable**	**B**	**Std. Error**	**Beta**	**p**	**R²**
Impairment Index	**Step 1**					
	Constant	0.902	0.183		.000	0.731
	Birth weight	-0.041	0.030	-0.053	0.172	
	Birth week	-0.002	0.007	-0.011	0.784	
	Clinical type	-0.022	0.030	-0.022	0.474	
	GMFCS	0.472	0.028	0.816	.000	
	MACS	-0.007	0.030	-0.012	0.813	
	EDACS	0.030	0.035	0.042	0.386	
	CFCS	0.025	0.030	0.039	0.404	
	Step 2					
	Constant	0.904	0.182		.000	0.732
	Birth weight	-0.042	0.030	-0.053	0.165	
	Birth week	-0.002	0.007	-0.011	0.774	
	Clinical type	-0.021	0.030	-0.021	0.487	
	GMFCS	0.469	0.024	0.811	0.000	
	EDACS	0.028	0.033	0.039	0.403	
	CFCS	0.024	0.030	0.037	0.421	
	Step 3					
	Constant	0.859	0.093		0.000	0.732
	Birth weight	-0.048	0.022	-0.061	0.029	
	Clinical type	-0.022	0.030	-0.022	0.470	
	GMFCS	0.470	0.024	0.812	.000	
	EDACS	0.026	0.033	0.037	0.418	
	CFCS	0.024	0.030	0.036	0.424	
	Step 4					
	Constant	0.836	0.088		.000	0.733
	Birth weight	-0.049	0.022	-0.063	0.023	
	GMFCS	0.464	0.023	0.802	0.000	
	EDACS	0.027	0.033	0.038	0.414	
	CFCS	0.024	0.030	0.037	0.414	
	Step 5					
	Constant	0.830	0.087		.000	0.733
	Birth weight	-0.047	0.021	-0.060	0.029	
	GMFCS	0.470	0.021	0.813	0.000	
	CFCS	0.038	0.024	0.058	0.116	
	Step 6					
	Constant	0.829	0.087		.000	0.732
	Birth weight	-0.044	0.021	-0.056	0.041	
	GMFCS	0.493	0.016	0.852	0.000	

Impairment index among to the CP subtypes varied significantly according to the Chi-Square test. In unilateral spastic CP, 61.50% of children had a low impairment index ($p<0.05$), in bilateral spastic CP, 44.2% of children had a moderate impairment index ($p<0.05$), in dyskinetic CP 67.9% of children had a high impairment index ($p<0.05$). In

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ataxic type there were not a significant difference between impairment index levels (p=0.06) (Table 3).

Discussion

This study was a retrospective with a large population to understand the frequency of associated impairments among children with CP, especially targeting on the relatively numerous of children with CP, which gives the opportunity to investigate the effects of associated problems on all CP subtypes, as well as this study is one of the few studies, and in our knowledge the first study from Türkiye based on a novel classification, the impairment index which is in order to determine which children have relatively moderate or more severe impairment, it is important to characterize the combined effects of the many disabilities present in children with CP. This is because the children's needs and level of participation will be directly impacted by these.

Although the current study was not focused on the participation domain of ICF, according to the several studies, the effects of impairments on participation are well documented, and the influence of associated problems is significant and effects both function and participation of people with CP, and therefore it is ultimate to evaluate, understand, and follow the presence of impairments, such as motor, epilepsy, intellectual impairment as well as vision and hearing[18]. For instance, amongst factors affecting participation, gross motor function is the one of the most important factor in CP[19] and according to the Alghamdi et al[20]. (2017), Dang et al[21]. (2015), McManus et al. (2008)[22], and Colver, et al[23]. (2012) studies, motor skill is the major factor effecting participation of people with CP. Duke et al (2021) indicate that there is a relationship between comorbidities and limitation in school participation[24]. Similarly, according to study of Burgess et al., decrease of daily activity related with presence of an intellectual impairment; and epilepsy was associated with decrease of daily activity as well as self-care in CP[25]. Additionally, according to Crotti et al., (2024) within relation to motor function vision in daily life as manual abilities, and may influence the quality of life by limiting self-esteem, emotional and social well-being of these children[26]. Moreover, van Gorp et al. were well documented that factors in childhood account for 79–90% of the variation in participation of young adult in home life as well as interpersonal relationships of individuals with CP. Children with decreased motor capacity, intellectual disability or epilepsy are at risk for limitation in participation in young adulthood[27]. Therefore, as in current study, understanding impairments altogether is a key element to have opportunity enhancing participation in children with CP. Although classification systems like GMFCS, MACS, EDACS and CFCS are important tools for constitute common language to describe better and communicate about the largely heterogeneous functional skills of people with CP[8] there is not a consensus on definition of “severity of CP”: definitions mostly refer a function as motor function only. Therefore, the Impairment Index provides a framework to describe the severity in a child with CP that closely related with participation.

In our study population sample, nearly one third of children (31.7%) were thus classified high impairment index level that will require comprehensive care and services to support because of this group has one or more associated severe impairment. One important finding of this study is that, when compared with spastic CP subtype, dyskinetic children

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have much more tendency to having severe impairments, with 67.9% of these children had a high impairment index. Dyskinetic CP is the second largest subtype of CP affecting 10–20% of children with CP[28] and consistent with current study severe motor impairment and intellectual impairment are more frequent in dyskinetic CP than in other subtypes[29].

According to the regression analysis, beside the GMFCS, birth weight is one of the predictors of the severity of CP in term of the Impairment Index. Esih et al., in their study found an inverse association between birth weight and CP developing risk[30].

Horber et al. found a weak relationship between severity and birthweight, as well as birth week, in our study, we found birth weight as the predictor but not birth week[6]. These contradictory results indicate more researches need to investigate relationship between impairments, birth week and weight. On the other hand, similar to current study, Delacy et al. analyzed the associated impairments and gross motor function levels and found that with increasing GMFCS level, the amount of people with CP with each associated impairment also increased[31].

The main limitation of the study was lack of changing severity by the time. Additionally, impact of etiological factors on severity should be investigated in future researches. Another limitation was, although the study based on such a large sample size, participants of the study based on a single center, therefore multicenter studies need for covering whole country.

Conclusion

In conclusion, according to the findings, dyskinetic subtype of CP is more severe form with bilateral spastic CP. Since, CP is a lifelong condition characterized by changes in function across the lifespan; the importance of interventions to improve outcomes in motor disorders associated with the condition[32]. Therefore, understanding the effects of associated impairments is important to organize and planning rehabilitation as well as social services. Clinical use of the disability index may guide effective and multidisciplinary rehabilitation practices. In the future studies it is recommended that to evaluate the effects of different disability levels of the disability index on activity and participation according to the ICF perspective.

Conflict of Interest: Authors declare that no conflict of interest.

Ethical Approval: Gathered from Hacettepe University, Non-invasive Clinical Researches Ethical Committee (Number: GO 20/356; Date: 17.04.2020).

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