University of Zagreb
Faculty of Education and Rehabilitation Sciences

Master's thesis

Visual impairment due to a dyskinetic eye movement disorder in children with dyskinetic cerebral palsy

Mihaela Ivošević

Zagreb, September 2019

University of Zagreb
Faculty of Education and Rehabilitation Sciences

Master's thesis

Visual impairment due to a dyskinetic eye movement disorder in children with dyskinetic cerebral palsy

Mihaela Ivošević

doc.dr.sc. Sonja Alimović

Els Ortibus, MD, PhD

Zagreb, September 2019

Statement of authorship of the work

I confirm that I have personally written the work ***(Visual impairment due to a dyskinetic eye movement disorder in children with dyskinetic cerebral palsy)*** and that I am its author.

All parts of the work, findings or ideas that are cited in the paper or based on other sources are clearly marked as such and are adequately listed in the references.

Name: Mihaela Ivošević

Place and date: Zagreb, September, 2019

Izjava o autorstvu rada

Potvrđujem da sam osobno napisala rad ***(Oštećenja vida uslijed diskinetskih poremećaja pokreta očiju u djece s diskinetskom cerebralnom paralizom)*** i da sam njegova autorica.

Svi dijelovi rada, nalazi ili ideje koje su u radu citirane ili se temelje na drugim izvorima jasno su označeni kao takvi te su adekvatno navedeni u popisu literature.

Ime i prezime: Mihaela Ivošević

Mjesto i datum: Zagreb, rujan, 2019.

Title of the paper: Visual impairment due to a dyskinetic eye movement disorder in children with dyskinetic cerebral palsy

Name of the student: Mihaela Ivošević

Name of the mentor: doc.dr.sc. Sonja Alimović

Name of the co-mentor: Els Ortibus, MD, PhD

The program in which the final exam is taken: Educational Rehabilitation/Rehabilitation, Sophrology, Creative and Art/Expressive Therapies

Abstract: Despite the fact that ocular and cerebral visual abnormalities are shown to be very frequent in cerebral palsy (CP), children with CP are underreferred to rehabilitation services for visual impairments. Visual component is, together with the motor disorder, an integral part of the clinical picture of CP and not an associated symptom. Therefore, an accurate detection of visual disorders and visual function not only lead to a complete clinical diagnosis but also to an appropriate intervention plan. Hence, the need for a study aiming specifically to describe all the aspects of visual involvement in the dyskinetic CP. Research goals were aimed at gaining insights into the nature of visual impairments and functional vision of children with dyskinetic CP, determining the nature of connection between visual functions and functional vision, with an emphasis on searching for dyskinetic eye movement disorder for understanding the difficulties in performing visual activities of two children with dyskinetic CP from the Zagreb's county register of CP, which is part of national C28 RCP-HR-Register of cerebral palsy of Croatia included in Surveillance Cerebral Palsy Europe (SCPE). The data were collected using standardized and non-standardized tests for visual function assessment. Dyskinetic eye movement disorder was tested comparing the tested results of visual functions. Functional vision was tested through the observation of the children's behavior and through open structured questions addressed to parents. Cerebral visual impairment was examined by a questionnaire for cerebral visual impairment screening. Qualitative research analysis shows which ocular and cerebral visual impairments are present as well as their relation to visual functioning. Moreover, it shows clinical features of dyskinetic eye movement disorder that haven't been shown present among tested children. Since this is the second research up to this date, testing an eye movement disorder that specifically occurs in dyskinetic CP, its characteristics are further discussed and defined. In contrary to previous study, in a child where highly inefficient visual functioning was shown, the cause goes wider from the abnormal eye motility. It consists of combination of several motor and sensory problems (lacking binocular visual acuity, contrast sensitivity, fixation, voluntary eye movements and oculomotricity). Due to the small sample and findings that are, on account of heterogeneity of dyskinetic CP hard to compare, future research is needed to expand overall knowledge of functional vision and visual functions needed for planning rehabilitation and education management for children with dyskinetic CP.

Key words: *dyskinetic cerebral palsy, dyskinetic eye movement disorder, visual impairments, visual functions, functional vision*

Naslov rada: Oštećenja vida uslijed diskinetskih poremećaja pokreta očiju u djece s diskinetskom cerebralnom paralizom

Ime i prezime studentice: Mihaela Ivošević

Ime i prezime mentorice: doc.dr.sc. Sonja Alimović

Ime I prezime komentorice: prof.dr.sc. Els Ortibus

Program/modul na kojem se polaže diplomski ispit: Edukacijska rehabilitacija/Rehabilitacija, sofrologija, kreativne i art/ekspresivne terapije

Sažetak: Unatoč činjenici da su okularne i cerebralne vizualne abnormalnosti česte u cerebralnoj paralizi (CP), djeca s CP nedostatno podliježu uslugama rehabilitacije oštećenja vida. Vizualna komponenta je, zajedno s motoričkim poremećajem, sastavni dio kliničke slike CP, a ne samo pridruženi simptom. Točno otkrivanje vizualnog profila djece s CP dovodi do sveobuhvatne kliničke dijagnoze i do odgovarajućeg plana intervencije. Stoga je potrebna studija čiji je cilj opisati sve aspekte vizualne uključenosti u diskinetskom tipu CP. Ciljevi istraživanja bili su usmjereni na stjecanje uvida u prirodu vidnih funkcjia i funkcionalnog vida djece s diskinetskom CP, utvrđivanje prirode povezanosti vizualnih funkcija i funkcionalnog vida, s naglaskom na otkrivanje diskinetskog poremećaja pokreta očiju, kako bi se poboljšalo razumijevanje poteškoća u izvođenju vizualnih aktivnosti dvoje djece s diskinetskom CP iz zagrebačkog županijskog registra CP-a, koji je dio nacionalnog registra C28 RCP-HR cerebralne paralize Hrvatske uključen u Surveillance Cerebral Palsy Europe (SCPE). Podaci su prikupljeni korištenjem standardiziranih i nestandardiziranih testova za procjenu vidnih funkcija. Diskinetski poremećaj pokreta očiju testiran je uspoređujući testirane rezultate vidnih funkcija. Funkcionalni vid testiran je promatranjem dječjeg ponašanja u vizualnim zadacima i kroz otvorena strukturirana pitanja upućena roditeljima. Cerebralno oštećenje vida ispitano je upitnikom za screening na cerebralno oštećenje vida. Kvalitativna analiza podataka pokazuje koja su okularna i cerebralna oštećenja vida prisutna kao i njihov odnos prema funkcionalnom vidu. Budući da je ovo drugo do sada istraživanje ispitivanja diskinetskog poremećaja pokreta očiju koji je karakterističan za diskinetsku CP, opisana su njegova klinička obilježja. Suprotno već objavljenoj studiji, kod djeteta kod kojeg je prikazano vrlo neučinkovito vizualno funkcioniranje, uzrok je širi od abnormalne pokretljivosti oka. Sastoji se od kombinacije nekoliko motoričkih i senzoričkih problema (nedostatna binokularna oštrina vida, kontrastna osjetljivost, fiksacija, voljni pokreti očiju i okulomotorika). Zbog malog uzorka i nalaza koji su zbog heterogenosti diskinetičke CP teško usporedivi, potrebna su buduća istraživanja kako bi se proširilo sveukupno znanje o diskinetskom poremećaju pokreta očiju, funkcionalnom vidu i vizualnim funkcijama potrebnim za planiranje edukacije i rehabilitacije djece s diskinetskom CP.

Ključne riječi: *diskinetska cerebralna paraliza, diskinetski poremećaj pokreta očiju, oštećenja vida, vidne funkcije, funkcionalni vid*

Contents:

1. Introduction

Cerebral palsy (CP) is a blanket term for several conditions resulting in lifelong motor disability. Together they make the most common cause of physical impairment in children, and are responsible for permanent (but not unchangeable) activity limitation and lifetime participation restriction (Pakula, Van Naarden Braun, & Yeargin-Allsopp, 2009; Panteliadis, 2018; Rosenbaum et al., 2007). The definition of CP is still not precise enough to secure agreement as to which people to include under this label, and additional inclusion criteria required are not yet internationally standardised. The latest definition (Rosenbaum et al., 2007) describes CP as "a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to nonprogressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of CP are often accompanied by disturbances of sensation, perception, cognition, communication, and behaviour, by epilepsy, and by secondary musculoskeltal problems" (Rosenbaum et al., 2007). This definition redefined CP from previously used Surveillance of Cerebral Palsy Europe (SCPE) definition, highlighting activity limitation instead of motor function and expressing the possibility that additional impairments coexist. Examples of those impairments include seizures, hearing and visual problems, cognitive and attentional deficits, emotional and behavioural issues, and later-developing musculoskeletal problems. These impairments should be classified as present or absent, and if present, the extent to which they interfere with the individual's ability to function or participate in everyday activities should be described (McIntyre, Morgan, Walker, & Novak, 2011; Novak et al., 2017; Rosenbaum et al., 2007).

Well respected CP classification system based on neurological signs and topography of motor impairment was proposed by SCPE, with agreement on the clinical findings required for each following CP type: spastic (bilateral and unilateral spastic), dyskinetic (dystonic and choreo-athetotic) and ataxic. In the SCPE database, half of children with CP have a bilateral spastic type, nearly a third unilateral spastic type, and the others have either a dyskinetic or an ataxic type. When more than one type is present, the child should be classified according to the dominant clinical feature (Cans et al. 2007 according to Panteliadis, 2018).

1.1. Dyskinetic Cerebral Palsy

Dyskinetic CP cases present involuntary movements, distorted voluntary movements, and abnormal postures due to sustained muscle contractions (Rosenbaum et al., 2007; Krägeloh-Mann et al., 2005 according to Panteliadis, 2018; Monbaliu et al., 2016). Muscle tone is variable, but more commonly on the hypotonic side during infancy. Involuntary postures and movements are induced or exacerbated by emotional factors or movement. SCPE uses dystonic and choreo-athetotic CP subtypes for subgrouping. Choreiform movements are fast, irregular, pathological and involuntary contractions of individual muscles, or small muscle groups; these most often involve the face and bulbar muscles, proximal limb muscles resulting in chorea, as well as toes and fingers.

Athetosis refers to slow, constantly changing writhing or contoring movements mainly of the distal muscles that result in the inability of the child to maintain a position. Dystonia, chorea and athetosis frequently coexist in the child with dyskinetic CP (Monbaliu et al., 2016). Dystonia needs to be differentiated from spasticity. In some cases, however, it may be difficult to delineate chorea and athetosis when features are present from both and then the term dyskinetic CP should be used (Rosenbaum et al., 2007). Dystonia is not velocity-dependent and is not mediated by hyperactive proprioceptive stretch reflexes. Sanger defined it (Sanger et al. 2010 according to Panteliadis, 2018) as a movement disorder in which involuntary sustained or intermittent muscle contraction cause twisting and repetitive movements, abnormal postures or both. Dyskinetic CP is classified into dystonic and choreo-athetotic subtypes based on the dominant neurological sign; the dystonic group constitutes the majority of the cases. Overall, dyskinetic CP accounts for about 6–15% of all cases of CP, and its prevalence appears to be stable in children with a normal birth weight in Europe (Smithers-Sheedy et al., 2014). Hagberg and von Wendt (1989) according to Jan et al. (2001) found that incidence of the dyskinetic form was 0.21 per 1000 live births, while the overall incidence of CP in Western countries is thought to be between 1.5 and 2.5 per 1000. Dyskinetic CP is related to lesions in the basal ganglia and in the thalamus. Around 70% have lesions in the basal ganglia and/or thalamus on MRI, but other brain lesions also occur and a few have normal scans. In a Swedish study, it was found that children with dyskinetic CP were mainly full-term babies with a history of perinatal problems and particularly asphyxia (Himmelmann, Hagberg, Wiklund, Eek, & Uvebrant, 2007).

Panteliadis (2018) emphasizes the importance of early detection of an increased risk of dyskinetic CP at an early developmental stage due to the possibility of intervention long before the emergence of pathological features. Among the most reliable early markers for CP are MRI, neurological assessment and General Movement (GM) assessment. The methodological breakthrough of the GM assessment lies in its predictive value of the development of neurological deficits, in particular of CP, at a very early age (Einspieler, 2001 according to Panteliadis, 2018). In addition to GM advantages, the assessment of GMs is non-intrusive, easy to acquire and cost-effective. Einspieler, Peharz, & Marschik (2016) and Novak et al. (2017) brought out GM markers for dyskinesia, described as poor repertoire of GMs followed by lack of fidgety movements with circular arm movements and spreading the fingers. Characteristically, these abnormal circular arm movements are present at least until the age of 5 months postterm. Poor repertoire of GMs refers to monotonous sequence of movements and less complex movements of the head, trunk and extremities than seen in normal GMs (Ferrari et al., 1990 according to Panteliadis, 2018, Novak et. al, 2017). They are unilateral or bilateral, monotonous, slow forward rotations originating in the shoulder. The monotony in speed and amplitude is the most characteristic quality of such abnormal arm movements. From the age of 3 months onwards, absent fidgety movements and a lack of movements towards the midline are another marker for future dyskinetic CP (Einspieler et al., 2002 according to Panteliadis, 2018). Novak et al. (2017) stresses that on motor tests dyskinesia can be suspected when on voluntary movement twisting arm or neck postures emerge. Furthermore, these children can find midline play complicated to perform, they can prefer toys positioned at shoulder width, switch hands while reaching tasks and they can take a lot more time to initiate movements. In motor behaviour they will show reduced variation.

1.2. Visual Impairments in Cerebral Palsy

Visual impairments are a major and growing health and socioeconomic problem worldwide and the existence of visual impairment limits childhood development to an substantial mode (Alimović, 2012; Fazzi et al., 2012, Guzzeta et al., 2001; Jan et al., 2001; Panteliadis, 2018; Reynell, 1978). The limitations relate to the integration and interpretation of perception from other senses, the development of emotional connectivity, personality and self-confidence, social skills, fine and gross motor skills, language development and others

cognitive terms (Chokron & Dutton, 2016; McIntyre et al., 2011; Panteliadis, 2018). An early ocular, visual, oculomotor, and visuo-perceptual assessment is thus very important. Visual assessment requires a multidisciplinary approach, planned according to the child's age and the severity of CP. Assessment of visual function is also essential. Due to the plasticity of the visual system in CP children, proper visual training programs from an early stage are required to enhance visual function. Higher percentage of visual impairments in children with developmental disabilities is due to various causes of impairment with pre, peri and postnatal factors. Causes of CP, including preterm birth, term hypoxic ischaemic encephalopathy, early brain malformations and congenital or postnatal infections, will contribute to visual outcomes in CP (Salt & Sargent, 2014, Sonksen & Dale, 2002). Therefore, visual impairments are specially increased in children with CP. Dutton & Bax (2010) explain that the spectrum of visual problems in children with CP is very broad and includes peripheral problems related to the anterior part of the visual system (strabismus, reflection disorders and fundoscopic abnormalities) and visual problems of central origin (amblyopia, slower visual maturation and cerebral visual impairment; CVI). Nowdays, some of the disorders, such as cataracts and premature retinopathy, can be prevented or treated, and have become less common cause of vision impairment in children (Guzzetta, Mercuri, & Cioni, 2001; Panteliadis, 2018). After year 2000, a reduction in severe visual problems in CP was reported, but CVI has emerged as a major problem (Aylward, 2002 according to Ortibus, De Cock & Lagae, 2011). From the epidemiological studies available, the prevalence of more severe visual defects in children with CP can be estimated to be around 8% of children with CP (Dufresne, Dagenais, & Shevell, 2014; Guzzetta et al., 2001), but this value is probably an underestimate as the data are often obtained from a review of medical records, rather than a prospective and systematic assessment of all children with CP.

1.2.1. Cerebral Visual Impairment

A condition of visual difficulties caused by the damage in the brain, regularly labelled cortical or CVI, is well accepted but has no internationally confirmed definition (Sakki, Dale, Sargent, Perez-Roche, & Bowman, 2018). In their systematic review, Sakki et al. (2018) found commonalities and diversities in the terminologies and definitions of childhood CVI, indicating partial agreement. In the light of their review, they proposed to

describe CVI as “a verifiable visual dysfunction which cannot be attributed to disorders of the anterior visual pathways or any potentially co-occurring ocular impairment” (Sakki et al., 2018). Components of this definition have to be measurable and quantified using currently available clinical tools. Chokron & Dutton (2016); Fazzi et al., (2012); Guzzetta et al., (2001) and Lehman (2018) describe CVI as the leading cause of visual impairment in children in developed countries and may include poor visual acuity, reduction in visual fields, disorders of eye-movement, strabismus and complex visual-perceptual defects. It relates to damage or malfunction of retrochiasmatic visual pathway and other cerebral areas involved in perception and processing of visual stimuli (Chokron & Dutton, 2016; Guzzetta et al., 2001). Ocular abnormalities are not present or the ocular disease is not sufficient to explain the vision loss and children with display specific behaviours that are characteristic of CVI. Children with CVI are interfered in the way they access, interpret and respond to visual information (Lehman, 2012). CP and CVI share a common origin: 60 to 70% of children with CP also have CVI (Fazzi et al., 2012; Panteliadis, 2018). This is more common in children with periventricular leukomalacia (PVL), who experience difficulties in visual object recognition, visuospatial skills, and visual memory (Boot et al. 2010 according to Panteliadis, 2018). There is some controversy in terminology regarding the terms *cerebral* versus *cortical* visual impairment. Some use the term *cortical* to mean higher level deficits involving visual processing while others use the term *cerebral* as a category under which *cortical* visual impairment would be a subset (Chokron & Dutton, 2016). Lehman (2018) stresses that *cortical* visual impairment is bilateral vision loss associated with damage to areas of the brain associated with visual function. Because subcortical damage to the posterior visual pathways is a standard and accepted cause of the CVI, Sakki et al. (2018) argue that the term *cerebral* is more fitting than the term *cortical* to describe CVI. Dufresne et al. (2014) found that in her study prevalence of cortical blindness in non-ambulatory patients is 22.4% and most of these CP children were spastic quadriplegic and Gross Motor Function Classification System (GMFCS) IV-V. Hoyt (2003) found that children with cortical visual impairment are uncommon to have strabismus (12%), nystagmus (4,8%) and optical atrophy (14%). Sakki et al. (2018) argue that future CVI definitions and diagnostic criteria need to develop further to encompass the wide spectrum of children presenting in clinical ophthalmology services with suspicion on CVI, together with children with higher visual processing dysfunctions but partially intact visual acuity.

1.2.2. Ocular and Visual Abnormalities

Ocular and visual abnormalities are very frequent in CP (50-60%; Dufresne et al., 2014; Panteliadis, 2018) being 10–70 times more common than in general infant and children population. The severity of visual impairment is related to the area and to the extent of the brain damage (Guzzetta et al. 2010 according to Panteliadis, 2018). Most children with CP experience difficulties with their visual acuity; visual fields; contrast sensitivity; binocular vision; ocular alignment; ocular motility (uncoordinated saccades and pursuits, paroxysmal ocular deviations, fixation movement disorder, instability, oculo-motor eye apraxia); visual-guided movements; visual searching; recognition of faces, objects, and routes, visual attention and in maintaining eye contact (Fazzi et al., 2012). Those with very severe attentional problems are at risk of being misdiagnosed as blind (Philp & Dutto 2014 according to Panteliadis, 2018). The level of visual loss is strongly related to the delay in psychomotor development. Ghasia et al. (2008 according to Panteliadis, 2018) showed that the severity of the visual impairment is associated with the degree of motor impairment. Only 4–9% of children with mild CP (GMFCS I) have visual deficits, compared to 58%–60% of children with severe CP (GMFCS V). 70% of the children with severe CP (GMFCS V) have more than one visual deficit. They are also at greater risk for high myopia, dyskinetic strabismus, severe gaze dysfunction, absence of fusion, optic neuropathy, and CVI.

1.2.2.1. Refractive errors

Refractive errors are common in CP children, probably due to the lack of development of a normal optical system. *Hypermetropia* is the most common refractive error in mild to moderate CP (7% in overall CP population; Dufresne et al., 2014), while myopia is commoner in severe CP (2,3% in overall CP population; Dufresne et al., 2014). *Anisometropia* may also be detected in 10–20% of all CP children. Correction of the refractive errors is important to improve the focusing and to improve communication and guidance of movement. Correction of even a small degree of hypermetropia can magnify the text and help those with reading difficulties (Fazzi et al., 2012). More than 70% of children with CP have been found to have low *visual acuity* (Pakula et al., 2009).The Best Corrected

Visual Acuity (BCVA) may be low in many CP children (35–65%), despite normal pupillary responses and normal ophthalmic examination findings, but total blindness is rare. CP children with reduced BCVA can be helped by increasing the size and the proximity of text and images (enlarging the print, double-spacing text, and presenting written material in small sections). Visual acuity may also improve by limiting distractions. According to Panteliadis (2018), *amblyopia* is common in strabismic CP children (70%), but rare in overall CP population (1.4%) according to Dufresne et al. (2014). *Fundus abnormalities* are also not uncommon in CP (30%; Fazzi et al., 2012). A pseudoglaucomatic cupped optic disc (due to subcortical insult of the immature visual system in premature babies) and a temporally pale optic disc (due to cortical insult to the mature visual system in full term babies) are common findings in CP and may be associated with some visual dysfunction. Kozeis et al. (2007) stress that *colour vision* appears to be grossly normal (less than 6% of CP children had abnormal colour vision). However, acquired damage to the temporal lobes in older children can lead to abnormalities of colour perception and interpretation. *Contrast sensitivity* threshold can be significantly reduced in CP children (25–90%). Fazzi et al. (2012). found that 57% of diplegic CP children and 90% of tetraplegic CPP children showed reduced contrast sensitivity. These children require their toys and educational material to be bright and clear, as well as distinct colour boundaries. *Visual field* is usually asymmetrical in CP children (20% according to Panteliadis, 2018 and 25% according to Vasanth, Jacob, & Viswanathan, 2014) with the types of visual field defect depending on the area of brain affected. Damage to the occipital lobes leads to homonymous lack of visual field on the contralateral side. Damage to the posterior parietal region on one side causes lack of attention on the opposite side, imitating homonymous hemianopia, but with a few differences. Children with homonymous hemianopia (1,5% of CP children; Dufresne et al., 2014) have a conjugate gaze deviation toward the hemianopic field; however, children with visual inattention compensate by rotation of the body toward the problematic area. Bilateral superior posterior parietal damage, affecting periventricular white matter, commonly causes lower field impairment, and the affected children look down when they walk as they probe floor boundaries with a foot to check for height change. Bilateral posterior parietal damage also causes inability to see multiple targets simultaneously and impaired guidance of motion (optic ataxia) (Deramore Denver et al. 2016 according to Panteliadis, 2018). *Stereopsis and binocular vision* are very commonly affected in CP children (50–85%), either due to

retrogeniculate damage, amblyopia, or oculomotor dysfunction (Kozeis et al. 2006; Katoch et al. 2007 according to Panteliadis, 2018).

Marked abnormalities of *eye movements* have been found in all types of CP children. Gaze disorders, saccade, pursuit and nystagmus are evident in severe CP children. Those with severe oculomotor disorders, but good head and neck control, use horizontal or vertical head thrusts to facilitate gaze shifts. Oculomotor impairment is very common (28%; Dufresne et al., 2014). A significant proportion of CP children display altered *fixation* (25–80%) that may be either absent or eccentric and is associated with injury to periventricular white matter. As in those with very severe attentional problems, these children are also at risk of being misdiagnosed as blind (Jan et al., 2001). *Smooth pursuits* (tracking movements to follow a slowly moving target) are significantly impaired in CP children (60–98%). Affected children appear with difficulty in seeing multiple targets at once. They also have problems following moving targets, like watching cartoons on the TV. *Saccades* (fast eye movements to alter fixation quickly) are often affected in CP children (60–100%; Kozeis et al., 2007), being either absent, hypermetric, or hypometric. These children adopt compensatory strategies, such as brusque head movements, frequent blinking, or both; they also present with hyperfixation and conjugate gaze spasms (upward and lateral). Kozeis et al. (2006 according to Panteliadis, 2018) found that the microsaccades, being used in reading, are also markedly affected. Convergence and divergence are disconjugate movements, used to maintain fixation on a target as it moves toward or away from the child's face. These movements are impaired in many CP children (30–50%) interfering with the visual focus. A markedly increased incidence of *strabismus* is observed (40–80%) in these children (Collins 2015 according to Panteliadis, 2018). Strabismus patterns in patients with CP are similar to those encountered in the general population but more common (Buckley & Seaber, 1981). *Dyskinetic strabismus* is a specific type of strabismus typical just for CP and is described within specific visual impairments in dyskinetic CP. *Nystagmus* is also common in CP children (30%; Dufresne et al., 2014; Panteliadis, 2018). Fazzi et al. (2012) conducted a similar number (29.4%) of CP children with nystagmus.

Panteliadis (2018) stresses that a high percentage of CP children appear to have altered *optokinetic reflex* (55–87%). Optokinetic reflex causes eye movement in response to objects moving in the periphery while the head is stationary. It combines saccades and smooth pursuit eye movements and maintains the balance of the body in a moving visual environment. It interacts with the vestibular optic reflex when the head is rotating in

stationary visual environment. This reflex plays an important role in everyday experience for most people in the context of driving, as objects tend to move rapidly past the driver in the periphery, which requires a rapid ocular response, while maintaining primary fixation on the road. Furthermore, in CP children, *vestibulo-ocular reflex* is also impaired (50–80%). Vestibulo-ocular reflex stabilizes the image on the retinas during head movements, by producing eye movements in a direction opposite to the head movement, preserving the image on the center of the visual field. It thus maintains the balance of the body by keeping the world steady when the head moves. *Accommodation* (a reflex, which is elicited in response to focusing on a near object) is also impaired in CP children (55–65% according to Vasanth et al., 2014). It comprises coordinated changes in vergence, lens shape, and pupil size which explains why some CP children find it difficult to see near objects and could be misdiagnosed as inattentive. This can affect everyday living and reading skills (Pansell, Hellgren, Jacobson, Brautaset, & Tedroff, 2014). *The pupillary reflex* (the quantity control of light entering the eye) may be slow, especially in tetraplegic CP children (5–75%), rendering them photosensitive (Fazzi et al., 2012). Many CP children (more than 60%) appear to have *visuoperceptual and visuocognitive problems* due to damage to the associated cortical areas (inferotemporal cortex, parietal lobes, frontal eye fields, occipito-parieto-temporal junction) or the ventral and dorsal streams connecting the primary visual cortex with these areas. These children may present with delay in the learning process, as well as the development of perceptual experiences (Kozeis et al. 2006 according to Panteliadis 2018).

The dorsal and ventral stream (two-streams) hypothesis is a model of the neural processing of vision (Hebart & Hesselmann, 2012) which argues that humans possess two distinct visual systems. As visual information exits the occipital lobe, it follows two main pathways (streams); the *ventral stream* (known as the *what pathway*) is involved with object and visual identification and recognition. The *dorsal stream* (*where pathway*) is involved with processing the object's spatial location relative to the viewer (Hebart & Hesselmann, 2012). *The ventral stream* also allows recognition and appreciation of shape, color, texture, recognition of people, and orientation within the surroundings. Ortibus, DeCock & Lagae (2011) stress that *damage to the temporal lobes* can lead to impaired visual recognition of faces and interpretation of facial expressions despite adequate vision (prosopagnosia) and impaired recognition of objects, shapes, letters, and route finding, particularly in new places (topographic agnosia). The dorsal stream helps the motor cortex to organize accurate body and vision-guided movements. It also connects the occipital lobes to the frontal eye fields to

initiate rapid, accurate head and eye movements to assist fixation at chosen targets. *Dorsal stream damage* can lead to impaired visual guidance of movement or optic ataxia, limited capacity to simultaneously see many items at once, and inability to move the eyes to a target despite an intact oculomotor system (gaze apraxia). Visual search is also impaired, making CP children unable to handle complex visual scenes by which they have difficulties in locating an object on a patterned carpet or in a basket of toys, reading crowded print, and recognizing people in a crowd or in the distance. Route finding in crowded environments tends to be difficult, and they tend to watch television from very close. Inaccurate visual guidance of movement or optic ataxia of upper and lower limbs may be misinterpreted as clumsiness, due to accompanied motor disabilities (Chokron & Dutton, 2016; Dutton 2009; Huurneman, Boonstra & Verezen 2013). Damage to periventricular white matter in the parieto-occipital region can cause impairment of movement perception (dyskinetopsia). Children with intact *occipito-parieto-temporal area* (center for movement perception) but with severe occipital lobe damage may exhibit perception of movement as the only visual function (Weinstein 2012 according to Panteliadis, 2018). Although it is not easy to diagnose visuoperceptual and visuocognitive problems, a careful observation of the child's visual behaviour helps distinguish between CP children with visual, oculomotor, visual cognitive, and perceptual difficulties and those without (Jacobson & Dutton 2000 according to Panteliadis, 2018).

1.2.2.2. Visual Impairment in Spastic, Ataxic and Dyskinetic subtypes of Cerebral Palsy

Bilateral spastic CP children are the most severe visually impaired CP subtype (Downie, Frisk & Jakobson 2005 according to Panteliadis, 2018). A high percentage of them (20–47%) have markedly reduced or not assessable best corrected visual acuity and reduced contrast sensitivity. They also show high percentage (30%) of optic nerve disc abnormalities (sectoral pale optic disc, cupped optic disc, optic nerve hypoplasia). 77% of them have gaze dysfunction and more than 75% of the children have a significant refractive error (hyperopia 32%, myopia 67%, and anisometropia 12%). Anisometropic amblyopia (20%), esotropia (40%), and vertical and dyskinetic strabismus are also very common (30%). The majority of the children (80%) have no binocular vision and stereopsis, gaze and movement disorders

(40%), CVI (30%), and visual field defects (11%). Manifestations of CVI usually include ocular deficits such as strabismus or refractive errors, but also fundus abnormalities and optic atrophy (Dutton & Jacobson, 2001). There is a strong correlation between spastic diplegia and prematurity, due to the high possibility of hemodynamic and respiratory instability of the premature newborn resulting in PVL (Tang-Wai, Webster & Shevell 2006 according to Panteliadis, 2018). The visual profile of spastic diplegia is characterized mainly by moderately reduced visual acuity (20%), significant refractive errors (70%) of which hyperopia is the most common 50–73%), absence of stereopsis, reduced contrast sensitivity (57%), amblyopia (60%), and strabismus (60%), dyskinetic strabismus (6%). More than 65% of the children have no binocular vision and stereopsis and impaired ocular motility (especially saccades). Optic disc abnormalities are present in 12% of the cases. *Spastic unilateral CP* is characterized by a slight reduction in best corrected visual acuity (47%); significant refractive errors (70%); reduced contrast sensitivity (25%); amblyopia (60%), that is, mainly strabismic, strabismus (35%) (equal incidence of exotropia, esotropia, and vertical strabismus); reduced visual fields (12%); no binocular vision and stereopsis (50%); and oculomotor impairment (35%) particularly in smooth pursuit and saccades and CVI (12%). Optic disc abnormalities are as often as in spastic diplegia (Fazzi et al., 2012).

Ataxic CP charactarizes irregular movements leading to balance and coordination problems, due to cerebellar dysfunction. Slow eye movements and inability to precisely target what are ataxic CP individuals looking at, lead to problems with depth perception. Sometimes damage to the developing brain is not confined to one location, leading to a variety of symptoms (mixed CP) and entails both spastic and non-spastic characteristics (Fazzi et al. 2009 according to Panteliadis, 2018).

Hoyt (2003) found that the overall distribution of initial acuity levels of children with PVL and those with cortical damage was remarkably similar. After the initial examination, nearly 80% of children with cortical damage showed at least one level of visual improvement, whereas only 40% of those with PVL showed at least one level of visual improvement. Moreover, in the children with cortex damage, 34% improved more than one level. These results suggest that children with PVL are less likely to show visual improvement and if they do so, it is unlikely to be more than one level on functional scale. He also found that the incidence of strabismus (54%), nystagmus (50%), and optic atrophy (38%) in the PVL group is significantly higher than in the group of cortical damage. Oculo-

motor apraxia is the only factor found to be less affected in PVL group (8%) than in cortical damaged children (20%).

In *dyskinetic CP* myopia is a common finding (56%). Guzzetta et al. (2001) observed that normal visual acuity was more often found in dyskinesis. According to Panteliadis (2018), almost 65% of the children have no binocular vision, none of them have stereopsis, many children are amblyopic (44%) (mainly anisometropic), and 22% of them present with optic disc abnormalities, 22% with CVI, and 11% with visual field defects. Esotropia and exotropia are the most common types of strabismus. Ocular and gaze movement dysfunctions are also present in almost one third of the cases. The fixation pattern however appears to be normal. Since basal ganglia regulate the voluntary motor function and eye movements, and cerebellum controls balance and coordination, the ocular movements can be highly affected in these children. Dufresne et al. (2014) compared presence of strabismus, refractive errors and field defects among types of CP. One child (0.06%) with dyskinetic CP had refractive errors (the smallest percentage among other CP subgroups). None of the children with dyskinetic CP had field defect (as well as ataxic children). One child with dyskinetic CP was diagnosed with cortical blindness. 20% of dyskinetic children had strabismus (the percentage was the same for hemiplegic CP and they had the lowest percentage among other CP subgroups). Pansell et al. (2014) explain dyskinetic strabismus as a distinct type of strabismus characterized by variability of the direction and magnitude of the strabismus, unique to severe cases of CP. It is unrelated to accommodative effort or attention and is seen exclusively in these patients where the ocular deviation fluctuates from an esotropia to an exotropia under the same accommodative conditions (Pansell et al., 2014). Constant esotropia and constant exotropia in dyskinetic CP have found to be present in less than 10%, and dyskinetic CP in more than 80% of CP cases (Buckley & Seaber, 1981 and Deramore Denver et al. 2016 according to Panteliadis, 2018). Many of CP patients with dyskinetic strabismus have dyskinetic CP. The association of dyskinetic strabismus with dyskinetic CP suggests that the basal ganglia may be the reason of the malfunction. Buckley & Seaber (1981) report that when a child with dyskinetic strabismus is asked to fix on a given accommodative target, the observer may see one or both eyes slowly converge. Then, with no alteration in attention or accommodation, the eyes may straighten or diverge. Dyskinetic strabismus can even be the first clinical sign of CP. The earliest indication is an esodeviation that is usually noticed by the parents shortly after birth.

1.2.3. Dyskinetic Eye Movement Disorder

Jan et al. (2001) are, up to this date, the only authors describing a movement disorder that specifically occurs in dyskinetic CP. In few other studies (Ghasia, Brunstrom, Gordon, & Tychsen, 2008; Guzzetta et al., 2001; Panteliadis, 2018), dyskinetic eye movement disorder is mentioned, together with other visual impairments occuring in CP, without any further description. In their report, Jan et al. (2001) have described a group of 14 children whose CP is associated with severe dyskinetic eye movement disorders that masks the presence of good visual acuity and results in poor visual function. In the review, dyskinetic eye movement disorder is described as difficulties with fixation, no visual pursuit in any direction, and no field loss. All participants exhibited fluctuating difficulties with the initiation and direction of voluntary saccadic movements, more so with longer saccades. Involuntary saccades did not appear to be affected. The binocular acuity was shown to be close to normal. In all cases of children funduscopic examination was normal, pupils were equal and reacted normally to light. There was no nystagmus. All children were constantly struggling with their eye movements, just as with their trunk and limb movements. CVI was ruled out, and a diagnosis of a severe dyskinetic eye movement disorder was made. This study highlights often misdiagnosing dyskinetic eye movement disorder as CVI. CVI should be diagnosed only when there is loss of acuity due to bilateral disturbance of optic radiation and striate cortex, and when it is confirmed by appropriate testing). Jan et al. (2001) suggested that dyskinetic eye movement disorder should be diagnosed when, similarly to their motor disorder, the patients have an impaired ability to plan and execute voluntary eye movements to the intended pattern and location, and to maintain fixation. Thus, the movement disorder of voluntary saccades and pursuit in patients with dyskinetic CP appears to behave similarly to their general motor difficulties. They believe that most patients with dyskinetic CP also have a dyskinetic eye movement disorder. What is surprising, however, is that their eye movement disorder can be severe enough to mimic blindness and therefore is not diagnosed appropriately sooner. This is because it is not widely understood that children with CP and normal ocular findings can behave as blind (showing a highly inefficient visual function) as a result of eye movement disorder.

A year after Jan et. al.'s report on dyskinetic CP, it was argued that (Matsuba, C., Jan, J., Lyons, C., & Heaven, 2002) it remained unclear how CVI was ruled out and asked whether dyskinetic eye movement disorder and CVI can coexist and if this, in fact, accounted for the

children's visual problems. It was insisted that, in contrary to Jan et. al., control of voluntary eye movements in children with dyskinetic CP is often remarkably preserved, and that the described dyskinetic eye movement disorder is just a particular subgroup of patients who had mainly an oculomotor pathology. Some of the concerns were answered (Matsuba, C., Jan, J., Lyons, C., & Heaven, 2002), explaining the important difference between dyskinetic eye movement disorder and patients with CVI; patients with dyskinetic eye movement disorder had near normal or normal visual acuity, based on forced choice acuity cards, normal peripheral field testing, and EEG results that were inconsistent with CVI (Matsuba, C., Jan, J., Lyons, C., & Heaven, 2002). Furthermore, these children's clinical presentations were different; they could identify targets more easily than CVI children, but dyskinetic eye movements, which arose while attempting to reach or maintain fixation, resulted in a functional inability to use their vision. Also, they said there are patients with a combination of CVI and dyskinetic eye movement disorder. As CVI clinical features increasingly appear, such as poor visual attention, it becomes more difficult to tease these apart from the neurological features of dyskinetic eye movement disorder. Furthermore, patients with multiple impairments often have other severe neurological problems which make the assessment more difficult. The authors advised caution when describing dyskinetic eye movements with other forms of eye movements. For instance, in congenital motor nystagmus, there is a minor reduction in visual acuity, without specific neuroanatomic anomalies and a normal electrophysiologic testing. Patients with dyskinetic eye movements have different features leading to functional visual impairment which is due to erratic fixation and visual pursuit behaviour.

2. Research problem

SCPE classification divines CP according to predominant motor disorder into spastic, dyskinetic or ataxic CP. Dyskinetic CP cases are classified as choreoathetotic or dystonic CP and present involuntary, uncontrolled, recurring, and occasionally stereotyped movements. The primitive reflex patterns predominate, and the muscle tone is varying. Characteristics are involuntary movements, distorted voluntary movements, and abnormal postures due to sustained muscle contractions. Choreo-athetotic CP is dominated by hyperkinesia and hypotonia. Chorea means rapid involuntary, jerky, often fragmented movements, and athetosis means slower, constantly changing, writhing, or contorting movements.

Despite the fact that ocular and visual abnormalities are shown to be very frequent in CP, there is still insufficient number of studies in the literature exploring the visual dysfunction associated with CP, especially with dyskinetic CP. Furthermore, no study has explored all the aspects of visual function in dyskinetic CP. Children with CP are at risk of disturbance in multiple components of the visual system, including the primary visual pathway (eye, optic nerves, thalami, optic radiations, and primary visual cortices), visual association areas, and the oculomotor system. There seems to be a correlation between severity of the motor impairment, as measured by the GMFCS, and both the presence and severity of visual impairment (Panteliadis, 2018). Most CP children experience difficulties with their visual acuity; visual fields; contrast sensitivity; binocular vision; ocular alignment; ocular motility, visual-guided movements; visual searching; recognition of faces, objects, and/or routes, visual attention, and in maintaining eye contact. SCPE classification offers inadequate information on visual impairments in CP (For visual impairment they only determine the presence or absence of such impairment, and then classify the impairment as severe or not, according to the visual acuity (<0,1 in both eyes after correction; Rosenbaum et al., 2007). More important, despite Jan et al.'s clear illustration of the dyskinetic eye movement disorder diagnosis, no further scientific research of that movement disorder present in dyskinetic CP can yet be found.

Hence, the need for a study aiming specifically to describe all the aspects of visual involvement in the dyskinetic CP.

3. The aim of the research and research questions

In accordance with the research problem, research goals were set, aimed at gaining insights into the dyskinetic eye movement disorder, level of visual impairments and functional vision of children with dyskinetic CP. Moreover, the goal was to discover the nature of the connection between visual functions and functional vision, in order to comprehensively determine the difficulties in visual activities of two children with dyskinetic CP from the Zagreb's Registry for Cerebral Palsy.

3.1. Research questions

In accordance with the set objectives of the research, the following research questions have been designed:

1) Is dyskinetic eye movement disorder present in children with dyskinetic CP?

2) How are oculomotor skills developed in children with dyskinetic CP?

3) How are visual function problems related to functional vision?

4. Research methods

The study was approved by the Ethics Committee of the Faculty of Education and Rehabilitation Sciences, University of Zagreb and the Director of the Special Hospital for the Protection of Children with Neurodevelopmental and Motor Disorders, Zagreb, Croatia gave consent to conducting the research.

4.1. Participants

Research participants (L.M. and P.R.) were selected by non-probabilistic and intentional sampling as samples of typical cases of a representative group of children with dyskinetic CP (Miles and Haberman, 1994). The criteria that the children met to enter the study were presence of visual impairment in both eyes without the absence of light perception, absence of retrolental fibroplasia, residency in the City of Zagreb and dyskinetic CP (regardless if choreoathetotic or dystonia subtype) diagnosed according to the SCPE classification. L.M. (2007) was born in Zagreb, in a family of five with an average income. The boy is currently completing the 4th grade of elementary school, which he attends with a customized individualized program for 4 hours a day at the Goljak Center for Education. Twice a week he takes part in the programs of physiotherapy, occupational therapy and speech and language therapy. The boy is diagnosed with a dyskinetic type of CP, GMFCS V (a significantly impaired gross motor functioning). He moves with the aid of a wheelchair that needs to be pushed by another person and needs a great degree of support. L.M. was

born as the third triplet and the only child with developmental disability among them. He was born prematurely as a high-risk infant, at 35 weeks gestational age: weight 1710 g, APGAR 3/8. The birth was complicated, the boy had asphyxia and IVH III. Currently, there is a marked lag in psychomotor development. In early developmental stages he did not adopt the milestones of crawling, holding, walking and speaking. Dysarthria is present; verbal contact is not established and he is not expressing words with meaning, he answers yes/no questions with head movements or smile (yes), but in state of fatigue becomes insecure (lowers his head). He does not use optical aids. Psychological findings indicate the presence of mild intellectual disabilities with influential disabilities. In the classroom he uses Tobii Communicator.

Girl P. R. (2000) was born in a family of three with an average monthly household income. She was born with 3920g at term, in a normal pregnancy without complications, APGAR 10/10. The girl is diagnosed with a dyskinetic type of CP, GMFCS III (she may climb stairs holding onto a railing with supervision or assistance). In early psychomotor development, she adopted crawling at the age of 4, seating and talking at the age of 5. Speech is still not articulated. She has not adopted toilet training in self-care, she feeds on her own, chews and accepts foods of various textures. The girl is currently completing the second grade of elementary school at the Goljak Center for Education, where she attends a customized individualized program. Before coming to Goljak, the girl attended physiotherapy and was enrolled in a speech and language therapy as well as in educational and rehabilitation program. Currently she attends physiotherapy 1 time per month. Ophthalmic findings show neatly evoked potentials and retinography. According to the findings of psychologists, the girl is within normal range of intellectual abilities.

Participants (with the support of their parents) were invited to the study through an official letter to the coordinator of the Zagreb's Registry for Cerebral Palsy. The children and their parents were presented with a telephone introduction of the research, its purpose and the importance of making a scientific contribution, and they were explained why their children were selected for the research participants.

4.2. Variables and assessment tools

Data were collected through:

(1) Visual function assessment

Standardized visual function assessment tests (Lea symbols recognition test with single symbol player cards, Keller grating acuity test cards, Heiding Heidi contrast sensitivity test, Heading Heidi facial expression test and Farnsworth D15 standard color vision test). The tests were used in accordance with the prescribed procedures.

Visual acuity was tested at near (reading distance, approximately 30 - 40 cm) with Lea symbols playing cards with individual symbols (Gräf, Becker & Kaufmann, 2000). This recognition type of test implies that the child recognizes very similar symbols (that is, the smallest distance between two points under which he sees the two points as separate). The results of visual acuity were obtained by recording the value of the M unit (the distance expressed in meters at which the individual optotype was seen at a 5' viewing angle) and calculated and presented in decimal form according to the formula VA = m/M (visual acuity equals distance from which the person looks at testing divided by the distance from which the average eye recognizes an object of a given size). Preferential visual acuity was tested with the Keller grating acuity test cards. Visual acuity at near was examined by Lea symbols playing cards at close range. Distance visual acuity was tested with Lea Symbols.

Contrast sensitivity was examined binocularly using a preference-based test, the Heiding Heidi Low Contrast Face Test (Almoqbel, Irving, & Leat, 2017), where the lowest contrast value was recorded (25%, 10%, 5%, 2.5%, and 1,25%) that a child may notice at a distance of 3 meters. The strongest contrast (lowest score) is 25% and the lowest contrast (the best result) is 1.5%. This is how contrast sensitivity is expressed in the percentage of contrast strength that a child responds to through preferential viewing. The child looks toward the black and white face, relative to the pure white card. The black and white face turns lighter gray on each subsequent card.

The Farnsworth D15 standard color vision test (Bowman, 1982) is based on a method of comparing the colors offered - multicolored tiles that are different in saturation. According to the pilot color (pattern), the task was to sort colors by numbers indicating colors and shades. The examinee had to isolate one scroll of a sample of a particular color, and the examiner extracts the remaining tiles of the same or a similar color from the remaining tile, that is, the

subjects are expected to arrange the tiles different in saturation in a row in order of similarity of color.

Non-standardized tests for assessment of visual functions

The *direct and indirect pupil reaction* was evaluated by the method of alternating illumination of the eyes. Both pupils should narrow when only one eye is lit. Swing lamp test was used, in which a direct pupil reaction was first observed in the right eye only. We lit it several times so that the pupil had time to expand. We repeated the testing on the left eye as well. The pupil's indirect reaction was then examined to determine whether the illuminated pupil accompanied the reaction of the other eye to light. One eye was illuminated (the other was obscured by light), while watching the reaction on the other (obscured) eye. The finding is neat if both pupils narrow at the same time regardless of which eye is lit. In indirect illumination, this result may also indicate the absence of damage to the afferent or efferent pathways involved in the pupil response. In addition to the neurological damage, asymmetry could also indicate brain trauma, eye disease, but it can also occur in a healthy population (Alimović, 2013).

The position and stability of the *monocular corneal reflex* was examined by a narrow beam of light and was used to indicate the position and stability of the retinal fixation. As the subjects looked at the light source with one eye (straight in front of the eye, with the other eye closed) and the position of the light reflex on the pupil was observed. We then slowly shifted the light source in a horizontal direction (within an amplitude of about 1 cm) and monitored whether or not the children kept their gaze on the light, i.e. whether the reflex remained in the same place regardless of the eye and light movement. If the corneal reflex was central and stable, it could have been assumed that the fixation is positioned at or near the macula, which is responsible for the clearest vision in daylight.

To determine with certainty whether the binocular position of the subjects' eyes is symmetrical, we examined the *binocular corneal reflex.* We also evaluated it with a narrow beam of light presented in the middle in front of both eyes at a distance of about 40 cm. The reflex in both eyes was observed and recorded whether they were central and symmetrical (Vinuela-Navarro, Erichsen, Williams, & Woodhouse, 2017).

The *eye motility* was examined by moving the object of interest to the extreme horizontal and vertical limits of the visual field. We wondered if a person could move the eyes in all

directions to the extreme limits or whether the eye remains in a certain position, which could have been a sign of strabismus, paralysis of individual ocular muscles and neurological problems.

Slow tracking movements of object in space were evaluated by tracking objects in motion in different directions within the visual field. The subjects were required to follow the object with their eyes moving in different directions within the visual field at a distance of about 40 cm. It was recorded whether the persons were following the object in all directions, whether the movements were smooth, whether they were compensated for the movements with the head.

The *saccades* (rapid simultaneous movements of both eyes in the same direction) were examined using two objects at a distance of 40 cm from the person, so that the person switches gaze from one to the other object on a verbal stimulus and the result were recorded on a five-point scale depending on the speed fixation shifting and precision movements that establish fixation: 1 - absence of gaze shifting, 2 - slow and inaccurate, 3 - slow and generally accurate, 4 - fast and mostly accurate, 5 - fast and accurate.

Convergence (simultaneous movement of both eyes towards each other) was examined using visually attractive toys that were brought to the nearest point of convergence (meaningful stimuli prevented the underestimation of the quality of eye movement). For functional purposes, the closest convergence point was examined. The examinees were shown visually stimulating objects at a distance of 30 cm. The closest point at which eye convergence was still observed, was noted.

Visual field was examined binocularly by the method of confrontation, for the purpose of functional vision assessment. The respondent was sitting opposite the examiner one meter apart and the sides from which the objects were introduced were selected at random. The examiner moved the stimulus (a visually interesting object for the boy and for the girl) from the periphery of the visual field toward the center.

(2) Screening questionnaire for cerebral visual impairment (Ortibus et al., 2011)

The presence of CVI was assessed through six areas: visual posture (components of fixation, visual field, visual attention, influence of familiar environment), ventral pathway (recognition of facial expressions, objects, persons, familiar environments), dorsal pathway

(finding and recognition of two-dimensional and three-dimensional objects and persons), complex problems (impaired visual attention, non-motor awkwardness in performing motor tasks), other senses (dominance of the use of individual senses when exploring the environment) and related features (rapid satiety with visual information, visual fatigue, general concern, vision compensation to other senses). Described are only those areas that are in connection with the features associated with CVI.

(3) Functional vision assessments and parent-oriented structured questions

Functional vision through the behavior of children (how the child uses his / her vision) in visual tasks (sustained near vision tasks, visual communication, keeping visual information to visual stimuli, orientation and movement). The presence of specific visual behaviors, such as gazing or holding unnatural position of the head when viewing, was observed parallel with the assessments.

Functional vision through observation of children's behavior was assessed through the following areas: use of vision in sustained near vision tasks, which was observed only in the girl; visual search of the table surface, differentiating color of the object from the surface color, estimating position, distance, shape and size of the object. Visual communication assessment included assessment of the focus on visual cues during communication with an adult when binocular viewing, establishing and retaining eye contact, noticing visual cues in communication, and reactions to altered facial expression and signals in communication.

Data on the focus on visual information in everyday skills was collected by examining parents on the focus on visual information while performing their child's activities of daily life (feeding, dressing, personal hygiene...)

Keeping visual information to visual stimuli was recorded by observing the child's focus on visual stimuli during binocular seeing, the speed of directing visual attention on stimuli, and the time of visual attention on stimuli. Whether there were disruptive factors that would have interrupted the maintainance of attention on visual stimuli was also observed.

Orientation and movement were observed when leaving and arriving at the room (based on the child's behavior by assessing binocular orientation and awareness of objects at different places and distances relative to one's body and other objects in the environment) and based on the statements of the parents.

The distance at which the child best uses vision was recorded by measuring the distance at which the child was most interested in seeing visual stimuli. The position of the body when viewing was monitored by looking at whether and how much the position of the body affects visual function. During the assessment, children were properly positioned to control the impact of difficult body position control on visual abilities. Assessment of all areas of functional vision included a behavioral component to reduce the impact of cognitive and communication disabilities on the children's performance.

Open structured parent-oriented questions were used for completing functional vision information (according to Milas, 2019; predefined form and order of questions). Examples of questions for parents: “How does the child look at toys? Does it tilt its head when holding a toy? Where does the child look while eating?")

4.3. Way of conducting research

The surveys were conducted on two occasions; on 5. Juli 2019 at 10AM and on 25. Juli 2019 at 10AM at the Goljak Cabinet for the Assessment of Visual Functions. Boy L. M. was brought from the educational group to the Cabinet, where two other educational rehabilitators attended the assessment with the researcher. Girl P.R. came to the Cabinet accompanied by her mother and the assessment was attended by a researcher and the educational rehabilitator. During the boy's assessment, one educational rehabilitator assisted to maintain proper posture, the second one examined the functions, and the third one recorded the results. When evaluating the girl, one rehabilitator was doing the assessments and the other was recording the results. There was no need for help in maintaining the posture with the girl. In the Cabinet where the observations were conducted, it was important to provide sensory non-irritating conditions: the white sheet was used to cover the desk with materials, the boy was turned in the stroller in the opposite direction from the mirror so that his own reflection would not interfere with his attention.

The window of the assessment room was open, which was a hindrance to the boy because of the sounds coming in and distracting him. On the other hand, visual orientation toward sound outcome has proven to be useful for observation. For the girl, such an unexpected benefit was the entry of the fly into the assessment room. The boy showed signs of excitement about interacting with the examiner and researcher. The girl also showed great

interest and positive affection for the assessment. The total duration of the individual assessments was 60 minutes and 15 minutes of conversation with the parents. In the boy's case, each stage of the assessment lasted longer than planned due to the severity of the motor difficulties. After the observation and return of the children to the educational groups, mothers were discussed with the general information about the usage of the vision in everyday situations.

4.4. Methods of data analysis

This qualitative research was performed via the observation method. It was used to develop a holistic understanding of the phenomena under study to be as objective and accurate as possible given the limitations of the method (DeMunck I Sobo, 1998). In data processing, thematic analysis was used to emphasize the identification, testing, and writing of topics within the data, and to emphasize the organization and rich description of the data set (Guest, MacQueen, and Namey, 2012). Thematic analysis was chosen because it goes beyond simply counting phrases or words in the text and moves on to identifying implicit and explicit ideas within the data, which finally dispels meanings within the dataset (Saldana, 2009). Coding developed themes within raw data by recognizing important moments in the data and structuring it before interpretation (Guest, MacQueen, & Namey, 2012). The following sections present research tables with thematized observational elements (Creswell, 1994), according to modified principles of thematic analysis.

5. Results

Based on a qualitative thematic analysis, the results show that dyskinetic disturbances of eye movement are not present among the respondents. Comparing the observation results with the results of Jan et al. (2001), the boy L. M, as well as the children described in the study of Jan and colleagues have difficulties with voluntary saccades and fixation. L. M. however, exhibits much less difficulty in voluntary saccades (he shows saccadic movements in excessive extent), while children in the study of Jan et al. (2001) have difficulty initiating and directing voluntary saccadic movements, to a much greater extent with longer saccades. P.R. shows nystagmic-like twitches of the eye in the left, right, and upward movements; they

have no significant effect on fixation but on the peripheral visual field. The difficulties of fixation of boy were manifested to a much lesser extent than Jan's study (fluctuating instability of fixation). Neither the boy nor the girl showed great difficulty with fixation; the boy's tracking movements were preserved in all directions, the girl's movements were also preserved, followed by jerkiness in 2 extreme horizontal and 1 vertical movement, while the examined children of Jan et al. could not perform tracking movements in any direction. Also, their participants showed results of binocular visual acuity within normal limits, while L. M. had extremely poor results in assessment of visual acuity. In a girl, visual acuity was at the level of milder visual acuity impairment. As dyskinetic CP is most often the result of injury to the basal ganglia, so are the control of voluntary vision, as well as eye movements partially affected, although dyskinetic eye movement disorder is not evident.

In Table 1., oculomotor visual impairments in examinees are shown. *The direct and indirect pupil reactions* are of importance to accommodation and convergence. The pupil's reaction is one of the basic prerequisites for the use of vision in everyday situations and its orderly functioning indicates that the child can use vision without being disturbed by the hypersensitivity of the eye to light or other paralysis of the optic nerve, efferent or afferent visual pathways. Although the pupil reactions were neat, the boy has a complete absence of *convergence* which has effect on long-term use of vision in visual communication. Since the use of vision in close-range tasks in boy could not be tested, the far-reaching consequences of lack of convergence could not be examined. According to the parents, the boy was able to converge in everyday situations (following the spoon that reaches his mouth). In the girl, the pupil's reactions are neat, as is the convergence. When converting, her left eye was strabbing. *The position and stability of the monocular corneal reflex* gave us important information about the fixation because in the boy it is not laid on or near the macula that is responsible for the clearest vision in daylight and thus the inconsistency of the boy in perceiving visual cues in communication is understandable, and potentially one of the reasons why he doesn't explore the environment in which he moves to a greater extent. By evaluating the monocular corneal reflex, we were able to approximately determine the degree of fixation shift. The shift of the fixation from the macula to the periphery leads worse visual acuity in the boy. Since the degree of shift in the boy is large (25^0), one of the factors that cause worse visual acuity is explained. It is important to consider the distances at which the boy and girl will best use their residual vision. The boy's corneal reflex proved to be fluctuating. In the first assessment it is central and stable, then in the left eye it remains

central, and in the right eye partially nasal (25^0 shift), then again in the right eye it becomes central and thus fluctuates through several assessments. At times when the boy's corneal reflex is observed nasally, the fixation is temporal at that moment. Also, fixation gets momentarily lost, due to the jerky movements that mimic nystagmic eye movements, which improve with finger pointing and are worse in verbal instruction than in physical guidance. In a girl, monocular corneal reflex was also central and stable, with nystagmic-like jerkiness in far left, far right and far up, but it had no significant effect on fixation, only on peripheral visual field. *Binocular corneal reflexes* showed that the girl has strabismus. In functional vision tasks, the boy was be able to maintain the central position of the eyes when looking with both eyes, while the girl was tilting her head when performing close-range tasks. The girl has central corneal reflex in the right eye, and in the left eye it is displaced, which means that the eye does not stand in the central position, that is, convergent strabismus in present in the left eye, which is why the girl is tilting the head when performing some of the tasks. Although the *eye motility* proved to be neat, the boy found it difficult to maintain voluntary eye control in visual communication. In certain directions of movement both girl and boy had jerky eye movements. Neat *slow tracking movements* allowed the girl and boy to retain objects of attention during visual communication and to track movement trajectories. Head stabilization plays a big role in the boy. Boy's *slow tracking movements* have been shown to be neat in vertical-horizontal directions, reaching to the maximal limits. During the slow tracking movements, eye jerkiness were observed which gave the appearance of nystagmus. They occured occasionally and the more the head was positioned, the more noticeable and longer they were. Girl's eye tracking movements have proven to be neat in vertical-horizontal directions, reaching to the maximal limits. Twitches were observed in extreme left-right-up horizontal movements, which was not evident in the downward slow tracking movements. The *fast-tracking (saccadic) movements* allowed the boy to focus his eyes on the object of attention when visual communication and tracking the movement trajectory. Due to the fact that the boy's saccadic movements were wide (he had no narrow saccades) and he had difficulties with the motor control and the speed and fluency of monitoring in visual communication proved to be difficult. Such saccades lead him to great difficulty in the speed and fluency of tracking visual material and persons. His narrow saccades had value "1" which means there is no gaze switching, and wide saccades had value "2" which means they are slow and imprecise. For a girl, fast saccades allowed her to gaze at eyes in communication, form a three-dimensional environment map, plan activities, and read. In the

girl both narrow and wide voluntary saccades are present. Twitching movements were observed during her visual following. Her narrow saccades had value “5” which means they are fast and precise and her wide saccades had value “4” which means they are fast and mostly accurate. Given the fact that the girl understood all of the verbal commands, she was not tested for preferential visual acuity. Results of boy’s *visual acuity* are poor. His result of 0.3 goes under the category of blindness. Recognition visual acuity measured with Lea symbols players cards with individual symbols at 30cm distance show 10M (0.03), at 45cm distance 16M (0.03) and at 20cm he managed to see 6.3M (0.03). Preferential visual acuity measured with Keller grating acuity test carts shows that on this visual acuity test, the boy scored better than on the previous one. Nevertheless, at 40cm he has result of 0.69 cpd, which is significantly worse than expected for his age. On Lea symbols players cards at close range at 40cm distance the girl managed to see up to 1M (0.4) which means 40% residual visual acuity. Measuring distance visual acuity via Lea Symbols, at 3m distance the girl showed a result of 9.5M (0.32; 31.58% rest of visual acuity). The girl followed the letters with a piece of paper placed under the line to be followed. At some moments, she would lean closer to see better, and then the examiner moved away to keep the distance she needed for testing. Also, the girl had three mistakes, which she noticed on her own initiative and corrected it, which was at the end accounted as correct answer. As each shift of fixation from the midpoint results in a decrease in visual acuity, and an increased shift is shown in the boy, a correlation with a lower visual acuity is also expected. Because of the great difficulties in visual acuity, the inability of the boy to understand small details such as changes in facial expressions is understandable. Oculomotor unactivity also affects his visual acuity, as the ability to maintain a stable fixation requires that all eye muscles work together. Also, environmental illumination is important for the boy to have good visual acuity, and one of the assumptions of worse visual acuity unsatisfactory illumination in the room in which he was assessed. The girl showed an optimum level of visual acuity that allowed her to continue focusing on visual information and visual communication Measuring *contrast sensitivity* the boy could see the contrast of 25% at 30cm distance on Heiding Heidi test of contrast sensitivity. Since contrast sensitivity is important for noticing and distinguishing two images of blurred transitions and outlines, identifying low contrast is especially important in visual communication, orientation and movement, and daily sustained near vision tasks. The reduced contrast sensitivity in the boy explains the problems in recognizing small changes in the face of the interlocutor during visual communication. The girl could have seen the

contrast of 1.25% at 1m, which means good contrast sensitivity. It is very important for her to adjust the distance at which her contrast sensitivity is best for orientation and mobility, communication, the landmarks and the distance of the communication signs. The results of *visual field* show that the width of the boy's visual field is within the normal range ($70\text{-}90^0$). Because the boy was non-verbal, the reflexive eye movements towards the stimulus were observed. Girl's visual field is also within the normal range ($70\text{-}90^0$) except in the left eye, where noting of the object has succeeded only to 30^0. The boy did not exploit the full potential that his neatly visual field allows him. He did not estimate the size and relationships of the objects in the rooms through which he moved, which is needed for adequate orientation and movement. *Color vision* has not been tested in the boy due to his intellectual disabilities. On the Farnsworth D15 standard color vision test girl managed to match even the most demanding shades. In her everyday life it means that she can match any color based only on the visible information. The following findings about the boy coincide with some of the literature described in the introduction, which state that children with CP at the GMFCS V have significant difficulties with visual acuity and contrast sensitivity (Panteliadis, 2018). Also, the results of this study coincide with usual visual impairments in CP: uncoordinated saccades, instability of fixation, difficulty of visually guided movements, recognition of changes in the face; grimace and visual attention (Pantliadis, 2018). On the other hand, L.M. showed preserved visual field, binocular vision, and eye motility.

Table 1. Visual functions of children

Elements of opservation	Results L.M.	Results P.R.
Direct and indirect pupil reaction	Direct: nomal Indirect: normal	Direct: normal Indirect: normal

Alignment and stability of monocular corneal reflex	Fluctuating: 1. central and stable, 2. LE: central, RE: nasal (25^0 shift), 3. RE: central Nystagmic-like movement Effect on fixation	Nystagmic-like movememts in bottom right and upwards No effect on fixation
Binocular corneal reflex	Stable and symetric	RE: symetric, LE: convergent strabism
Eye motility	Normal but voluntary movements control hard to control	normal with jerkiness
Slow eye tracking movements	Normal in all H-V lines *Head stabilisation	Normal in all H-V lines *Jerkiness in final L-R H-U
Saccades	No narrow saccades (1) Wide saccades are too extented and unprecise (2)	Narrow saccades fast and precise (5) Wide saccades fast and mostly precise (4)
Convergention	No convergence	LE-convergent strabismus RE- normal adduction
Visual acuity	Recognition VA- 0.03 Preferential VA: 0.69cpd	VA near: 0.4 VA distance: 0.3
Contrast sensitivty	30cm-25% ~~contrast~~	1m-1.25%
Visual field	Normal value (70^0-90^0)	RE: Normal value (70^0-90^0) LE: 30^0 perifferal loss
Color vision	Not tested	Normal

In Table 2. The results show how functional vision is manifested in terms of areas of visual functioning. The boy could not solve *sustained near vision tasks* because of the inability to perform oculomotor activities. While sitting at the table, the girl was visually searching the surface, noticed the objects with the same color as the background (i.e. a brown cone on a brown desk) and between the multiple items found the seeking one (i.e. cell phone), relying solely on visual information. When grasping the objects from the surface, it was observed that she estimates position and distance of the object, as well as the size and shape of the object. When using vision in sustained near vision tasks, the eyes must converge to focus a clear and single image with both eyes. Convergence (along with the accommodation and narrowing the pupil when viewing at close range) is one of the important factors that allow long-term vision to be used in near vision tasks. *Visual communication* is highly dependent on contrast sensitivity, which is responsible for noticing the shadows caused by changing facial expressions. A boy's difficulty in noticing and distinguishing between two images of ambiguous transitions influences the recognition of small changes in the face of the interlocutor, which through development can lead to communication difficulties. The boy also fails to show a change in facial expressions when evaluating functional vision, which can be explained by the difficulty in contrast sensitivity and its maximum utilization at a very short distance within which interaction is rarely established. Despite this, the boy directs his gaze to the face and eyes of the interviewee. However, since he does not have a reduced contrast sensitivity to the extent that he does not notice the basic parts of the face in communication (eyes, mouth), which is observed by the frequent establishment of eye contact when assessing functional vision, it is assumed that he will not develop major problems in establishing an emotional connection with a close person and further problems in socio-emotional development. Also, despite the difficulties in control and excessive extent of saccadic movements, he achieves them, which is important for maintaining eye contact. Long-term maintenance of eye contact was also difficult due to loss of fixation, which means that although managing to maintain visual contact, he had difficulties to fixate it. Despite the lack of speed and fluidity of eye tracking, he achieves it. Regardless of the generally poor performance on the tested visual functions, the boy uses visual information for communication purposes satisfactorily. He will be more successful in nearby visual communication. There is a possibility that the girl will have difficulties in visual communication and recognizing facial expressions over longer distances. In accordance with the findings of the assessment, parental statements confirm that the boy is most focused on

eye contact when communicating. Despite the lack of contrast sensitivity and visual acuity, the boy does use vision very well in communication situations. In the girl, recognition of different facial expressions and gestures during interaction and communication is fully present. Her reaction to the examiner's facial expressions and gestures is appropriate and adapted, and the frequency of gestures and changes in facial expressions accompany the verbal content of the interactions. For both children, parents provide information about spotting familiar people in a group of people. The boy is quicker to *focus his attention on the visual stimulus* than his head instability allows. As soon as his head is physically supported, his attention is longer and more frequent. Although his attention is time-consuming, fixation on the subject is short. Most likely, due to the motor fatigue of the neck area, he cannot hold his head longer to be able to visually explore the environment. The girl maintains her attention long on the moving pictures. The boy is extremely focused on visual exploration of the environment despite the small residual visual acuity and low contrast sensitivity. He does not respond affectively to non-verbal facial expressions or to cards containing images of human faces but responds to sounds (following non-visual cues). Consciously and deliberately directs gaze toward the stimulus which eventually ends up on a visual target. The distance at which the boy *used the best visual acuity at close range* was within 40cm and at the girl it was 30cm (the best handling for communication purposes was within those distances). At a distance of 1m, the girl can best assess the contrast in communication - people should approach her within 1m of communication (when orienting and moving to orient herself within that distance). In the girl, the best use of visual acuity was by far the best within 3m. This distance information is of particular importance as it means that both communication and all rehabilitation tasks should be performed at the indicated distances in order for the children to show themselves as best they can. L.M. showed that, because of his curiosity and openness in interaction with people, better uses vision than the development of visual functions allow him (extroverted people are generally more focused on events around him and will use vision better than introverts). If the boy was more passive and more focused on his own body, he would use the rest of his vision worse. In the boy, the presence of intellectual disabilities is also a limiting factor in directing visual attention and performing visual tasks. On the other hand, the severity of a motor disorder will limit him to better use his vision, due to the inability to move independently. Children with better motor skills will generally use vision better because of the greater need for movement. Thus, these children develop vision better because they use it more often, and then the objects they see at

different distances encourage them to move. In orientation and movement, the girl shows inadequate awareness of her position in space, notices objects in the environment at smaller and longer distances, notices spatial relationships, does not use visual information during movement, does not detect obstacles, and does not adequately assess depth.

Table 2. Functional vision of children according to children's behavior

Elements of opservation	Results L.M.	Results P.R.
Sustained near vision tasks	Not tested	Estimates position, distance, size and shape of objects
Visual communication	Mantaining eye contact, fixation problems, worse after 1m	Continous following, worse after 3m
Visual focus and attention	Excellent focus on visual stimuli (short fixation), non-affectionate reaction to non-verbal facial expressions, focus on auditive signals *motor fatigue, body positioning	Long attention on moving pictures and statical 2D images
Orientation and movement	No visual exploring, pathway following, no room size estimating or interest in learning about the visual stimuli surrounding it	No pathway following, no purposeful looking while O&M
Best distance for looking	Sustained near vision tasks: not tested Long distance: 20-40cm	Sustained near vision tasks: 20-30cm, long distance: under 3m
Influence of position	Big influence, proper head and hips positioning needed for engagement in visual	Tilting head for dominate eye usage (RE)

	tasks	

In Table 3. structured parental statements regarding children's functional vision are shown. It is evident that the boy prefers vibrant colors, although he finds objects equally interesting, regardless of color. It was observed that he prefers visual observation of the toys regardless of the color. The child's excitement for the subject grew with the tone of voice that motivated him to watch the toy. If the child was verbally encouraged to look at the toy, he was more pleased than when the toy was not presented to him verbally. As have the parents noticed, the observation also showed that the boy would respond more to the loud sound of the person than to the bright color of the object. The boy needs physical support to maintain stability of the head, which will greatly influence the direction of attention. Directing attention will also aggravate the instability of fixation on the visual stimulus. In feeding situations, both girls and the boy are visually involved in the interaction. The boy uses the full potential of his eye motility; he follows the feeder while feeding, then visually monitors the flow of food received and after swallowing redirects his gaze to the feeder. Given the complete absence of convergence, it is unusual for parents to report that the boy monitors food delivering to mouth. There is a possibility that he success this activity by compensating with slow tracking movements that he reaches to the limits in all directions. Also, the width of the boy's peripheral visual field is within the normal range, which allows him to accompany both the person sitting next to / across from him and the food coming to his mouth. When brushing his teeth, the boy maintains eye contact with the person that brushes his teeth. In game situations, the boy's visual attention will depend on the visual curiosity of the object being offered (shows more attention towards simpler; visually unsaturated toys). The parents of the boy do not notice any deviation in the way he views the toys, although it is to be expected that the child prefers the toy close to the eyes (20-40cm apart). Although, due to motor difficulties, this is hard to expect. According to her parents, the girl has excellent visual functioning in dealing with daily tasks at sustained near vision tasks in feeding, hygiene and dressing situations. During the assessment in the Cabinet, the use of vision in orientation and movement with respect to the size of the room could not be assessed. When returning the boy to the educational group, he was observed to follow the pathway by which he was moving, mostly directing his gaze to the floor, showing no interest

in scanning the environment; there was no visual exploration (due to the excessive and unstructured visual stimuli in the environment). Given that the boy has a perfectly preserved visual field, it is unusual for him not to use it for environmental exploration or orientation when moving. The parental statements of the boy's orientation and movement overlaps with the assessment of the same; when moving, the boy does not estimate the size of the room and shows no interest in learning about the visual stimuli that surround him. Considering that the assessment was based on a single observation of the situation of orientation and movement, it is not sufficient to draw bigger and firmer conclusions on this aspect. The girl does not walk by her own but with the physical guidance of the caregiver. Her gaze is not purposefully directed while orientation and movement. When moving, she is unaware of her position in the space, because she does not scan the size of the room or obstacles that lie in her way as she walks. Upon entering the assessment room, she scans the contents of the room. Upon leaving the room and upon entering the waiting area, a visual search for the basic spatial relationships (chairs in relation to the cabinet door, entrance door in relation to the chairs) was observed. According to the mother's statement, she does not consistently identify obstacles during movement and does not assess well the depths (visible when walking up the stairs). In a static position, she explores the position of persons in a room in relation to her position and draws further attention to interactions between persons rather than to the contents of the room.

Table 3. Functional vision of children according to open structured parent-oriented questions

Structured questions for parents	Results L.M.	Results P.R.

How does a child look at toys? Does it bring the object closer to their eyes? Does it tilt its head when holding a toy?	Looks at toys in the typical way, it does not seem important to him to move his eyes to a distance that would give an indication of deviation.	Most of the time looks at the toys typically, sometimes tilts her head; when manipulating with one object for a longer time
Where does the child look at while brushing teeth? Does the child look at the person? Does the child look around the room?	When brushing teeth, the boy looks at the person who brushes his teeth in the eyes. Sometimes his head falls to his left side toward his chest, but when he lifts it, he restores eye contact with the person who brushes his teeth. When he is tired, looks at the floor.	She can visually locate washbasin, soap and hand wiping paper In the presence of another person in the toilet room establishes frequent and continuous eye contact
How does the child look while eating? Does the child look at the cutlery, the food, the participants at the table? Does he follow the food that comes to his mouth? Does he look at the person who feeds him?	The boy looks at the person across the table and to the person next to him When feeding, the boy most often looks at the person who feeds him, then visually monitors the course of receiving food by mouth and after swallowing the food again turns his gaze to the person who feeds him	When feeding, the girl looks at the participants at the table, in the cutleries and in the food Visually orienting herself in the kitchen, finding the desired items on the shelves among other items, visually noticing changes in the food created by cooking
How your child uses vision when moving in environment: Is it looking in the	The boy moves in a wheelchair in the space, most often looking in front of him when he is moving, his gaze	She does not look where she walks when moving; moves her legs while walking but does not look purposefully in the direction of

direction of movement? Does the child estimate the size of the room when moving or before starting to move?	is most often directed towards the floor. When moving, there was no appreciation of the size of the room or interest in learning about the visual stimuli surrounding it.	movement but visually explores the elements of the room and the orientation of people in relation to it
Does the child react differently to specific items? Does it prefer certain colors more than other?	The boy prefers vibrant colors, but will respond more emotionally to simpler toys regardless of color than to more complex toys of certain colors He will respond more to a person's loud sound than to the bright color of the subject	There is no problem with matching the color of the garment based on the visible information, she can find the desired garment in the closet

As the results obtained from the *questionnaire for screening for cerebral visual impairment* shown in Table 4. predominantly indicate potential *dorsal pathway damage* in both girl and the boy. They had a reduced interest in complex images and preferred simpler toys. For the girl, it was difficult to look when there were many details on the picture, that is, they fatigued her. She only looked at the details of the picture, not the whole picture. Both subjects experienced difficulties in perceiving more complex components of images or objects. Particular difficulties in orientation and movement were observed in the girl; assessing distances and noticing differences in levels (i.e. while climbing the staircase). It was difficult to see if she can estimate the distances because she was partially carried when moving; when she saw the end of the curb, she stopped. A moving person attracts her attention more than a stationary person. She looked more at people than at objects. Both of the research participants reacted quicker to sound than to visual stimuli and stopped with the activity when there was too much visual stimuli to look at. They rather recognized persons by listening to their voice and looking at their posture than by looking at their faces. They understand facial expressions, even though the boy did not react affective to their changes. The components of the visual attitude (fixation, visual field, visual attention, and influence of familiar surroundings) seem to be mostly neat, both in girl and in the boy, and do not

cause difficulties in visual functioning. They were more visually attracted to people than objects and to moving objects rather than to stationary objects. They both responded more quickly to sound than to visual stimuli, but they used the sense of sight as dominant when performing actions. The boy often looked out of the open window, but the assumption was that it was due to the fixation on the sound sources, not the light sources. He had eye contact present and he could focus on persons or objects, but shorter than the girl. The concepts of functioning with regard to the visual field was difficult to evaluate due to motor impairment. He needs encouragement in the form of physical guidance to look at the object and explore the environment around him. Few objects distracted his visual attention (toys of different visual qualities). He watches objects from a short distance, it is difficult to estimate how many he would view them from a greater distance because he is unable to hold objects independently. Given that parents of the boy were not present during the assessment, the impact of the positive environment (focus on and adherence to parents in different situations) could not have been examined. The girl tilted her head to look at individual objects, but this is probably compensation for the fixation in the strabbing eye. She could focus on persons or objects for a long time. She often stumbled upon clearly visible objects and it was easy for her to fall into something. When the object was harder to spot (e.g. shapes that are similar), it took her longer than expected to be noticed. There are items that interfered with her visual attention, such as a fly flying around the room. She looked at objects from a short distance for fixing the body in a certain position (hands clenched; fixes the position of the body and to calm it). Heiding Heidi test with facial expressions (happy, sad, serious, angry) was used to see the existence of potential CVI components; inability to recognize facial expression (if there is a neat contrast sensitivity and concurrently non-recognition of facial expression, which would be a result in favour of CVI). The girl could recognize all of the shown facial expressions.

Table 4. Cerebral visual impairment

Elements of opservation	Results L.M.	Results P.R.

Visual attitude	Eye contact, focus on people and objects, toys with different visual qualities interfere with visual attention, needs physical guidance, influence of familiar environment not tested	Eye contact present, focus on people and objects, often trips over visible objects, easiliy crashes into obstacles. When objects look alike, needs more time for perception
Ventral pathway	Understands face expressions, rather choses auditive recognition of people	Understands face expressions, rather choses auditive recognition of people
Dorsal pathway	Low interest for complex pictures, visual simplified toys preference. More visual interest for people than for objects	Problems with depth perception (stairs), low interest for detailed pictures, doesn't look at the whole picture, attention on person in movement, on people rather than on objects
Complex problems	Clumsiness and ineffectiveness in motor activities	Clumsiness and ineffectiveness in motor activities
Other senses	First reaction is to auditive signals, second to visual	First reaction is to auditive signals, second to visual
Connected features	Stops with activity after visual fatigue	Stops with activity after visual fatigue

6. Conclusion

The main objective of this study was to see if the subjects had a dyskinetic eye movement disorder. Although the observation of the boy by L. M. and girl P. R. resulted in some overlaps with Jan et al. research, however, our observation did not reveal key things that could potentially be associated with dyskinetic eye movements; children were able to plan and execute tracking movements, and were able to establish and maintain fixation. Even though in certain directions of movement both girl and boy had jerky eye movements, insufficient visual functioning in a boy wasn't due to abnormal eye movements but to

absence of good visual acuity and contrast sensitivity. Girl has shown sufficient visual functioning, with optimally preserved visual acuity and contrast sensitivity. Results from the obtained observations, and findings reported in the study by Jan et al. (2001) do not overlap and to conclude is that the respondents do not have a dyskinetic eye movement disorder.

The second objective was to show the oculomotor skills of the children. Both children had shown neat indirect and direct pupil responses, the girl could converge and the boy had absence of convergence. Boy's monocular corneal refley was fluctuating and girl's was consistent, but in both jerky movements were present. Their binocular corneal reflexes were stable, except in the LE of the girl, because of the strabismus. Their eye motilities were neat, with jerky movements present. Those movements were also occasionally present during their performance of slow tracking movements and wide saccadic movements even though they could have performed them. The girl showed fast and precise, and the boy had shown no narrow saccades. Children have shown normal slow tracking movements in all horizontal and vertical lines. Both of children's visual fields are within normal values. Girl' has shown the full potential of color vision. Contrast sensitivity and visual acuity are described within the first research goal.

The third objective was to relate visual function problems to functional vision. Observation results show that both boys and girls have difficulty functioning visually in different situations. Visual functioning of the boy is difficult in all variables of functional vision, and in the girl, it is most evident in orientation and movement. Given the good visual acuity, convergence and slow tracking movements, the girl successfully and actively manipulated the objects. She used vision in communication excellent which is supported by good contrast sensitivity and saccadic eye movements. The girl recognized different facial expressions and gestures during interaction and communication.The boy A boy's lower results on contrast sensitivity, influence was shown on problems with recognition of small changes in the face of the interlocutor as well as on showing changes in facial expressions. Due to the possibility of establishing and maintaining saccadic movements and slow tracking movemens, he could have maintained eye contact and focus their attention on the visual stimulus.

Given that the available literature on the vision of children with CP deals exclusively with visual functions, this study provides deeper insights into the relationship between visual function and functional vision of children with dyskinetic cerebral palsy. It is only by examining the relationship between visual functions and functional vision that their

influence on the level of participation of the child in the activities of daily living is determined. The primary goal of any rehabilitation program is to develop and improve the child's daily skills. Therefore, knowledge of the association between impairment of specific functions and limitations in activities of daily living is crucial for education and rehabilitation practice.

Following the assessments, it is important to use the results obtained to plan the individualised plan for the vision rehabilitation. Surter and Harvey (2011) have suggested important determinants for vision rehabilitation programming. The first determinant is that vision rehabilitation should be purposeful. Education and rehabilitation expert should have a thorough understanding of each particular procedure, firstly its goals and purpose. Furthermore, vision rehabilitation should be an active process, providing the person with new stimulation. A person should not remain in a passive and familiar state. Vision rehabilitation should encourage decision making based on visual information. Feedback on the results achieved is important to modify the performance for achieving the desired effect. It is also important to emphasize that rehabilitation should be adapted to the person's current abilities. The procedures need to be both manageable and demanding. Vision rehabilitation should occupy and maintain visual attention and focus on the tasks that is to be solved. Procedures should be practiced and repeated until new visual skills are mastered and internalized, so that they are fast, automated and stable over a long period of time. Perhaps most important, vision rehabilitation should integrate multiple senses. The results of the assessment of visual functions and functional vision will narrow us down guidelines in the development of a functional visual profile of a person and give us guidance in further vision rehabilitation planning and adaptation of the environment. When considering the use of vision comprehensively, it is also important to take into account environment-dependent visual factors (clarity of visual stimuli, size, color, contrast, lighting, distance, etc.), personality-related factors (interest, motivation, character, involvement in the community; the number of different situations and activities the person is involved in), as well as economic and family factors. The socio-economic status of the family depends on whether the visually impaired person can afford one of the sophisticated optical aids available, whether he or she will be able to adapt the environment in order to achieve the best possible conditions for the use of vision (change the lighting, colors of walls and furniture). Therefore, since the ability to use vision is not only influenced by visual functions but also by other factors, the assessment of visual functions and functional vision is ultimately not

sufficient for the overarching needs of rehabilitation and education.

7. References

1. Alimović, S. (2012). Visual impairments in children with cerebral palsy. *Hrvatska Revija Za Rehabilitacjiska Istraživanja*, *48*(1), 96–103.
2. Alimović, S. (2013). Development of functional vision in children with perinatal brain damage (*Doctoral dissertation*, University of Zagreb)
3. Almoqbel, F. M., Irving, E. L., & Leat, S. J. (2017). Visual Acuity and Contrast Sensitivity Development in Children. *Optometry and Vision Science*, *94*(8), 830–837. https://doi.org/10.1097/opx.0000000000001101
4. Bowman, K.J. (1982). A method for quantitative scoring of the Farnsworth Panel D-15. Acta Ophthalmologica, 60 (6), 907-916.
5. Buckley, E., & Seaber, J. H. (1981). Dyskinetic strabismus as a sign of cerebral palsy. *American Journal of Ophthalmology*, *91*(5), 652–657. https://doi.org/10.1016/0002-9394(81)90069-6
6. Chokron, S., & Dutton, G. N. (2016). Impact of cerebral visual impairments on motor skills: Implications for developmental coordination disorders. *Frontiers in Psychology*, *7*(OCT), 1–15. https://doi.org/10.3389/fpsyg.2016.01471
7. DeMunck, V. C. & Sobo, E. J. (1998). *Using methods in the field: a practical introduction and casebook*. Walnut Creek, California: AltaMira Pres
8. Dufresne, D., Dagenais, L., & Shevell, M. I. (2014). Spectrum of visual disorders in a population-based cerebral palsy cohort. *Pediatric Neurology*, *50*(4), 324–328. https://doi.org/10.1016/j.pediatrneurol.2013.11.022
9. Dutton, G. N., & Jacobson, L. K. (2001). Cerebral visual impairment in children. *Seminars in Neonatology*, *6*(6), 477–485. https://doi.org/10.1053/siny.2001.0078
10. Dutton, G., & Bax, M. (Eds.). (2010). *Visual impairment in children due to damage to the brain* (Vol. 186). John Wiley & Sons.
11. Einspieler, C., Peharz, R., & Marschik, P. B. (2016). Fidgety movements - tiny in appearance, but huge in impact. *Jornal de Pediatria*, *92*(3 Suppl 1), S64-70. https://doi.org/10.1016/j.jped.2015.12.003
12. Fazzi, E., Signorini, S. G., La Piana, R., Bertone, C., Misefari, W., Galli, J., … Bianchi, P. E. (2012). Neuro-ophthalmological disorders in cerebral palsy: Ophthalmological, oculomotor, and visual aspects. *Developmental Medicine and Child Neurology*, *54*(8), 730–736. https://doi.org/10.1111/j.1469-8749.2012.04324.x
13. Ghasia, F., Brunstrom, J., Gordon, M., & Tychsen, L. (2008). Frequency and severity

of visual sensory and motor Deficits in children with cerebral palsy: Gross motor function classification scale. *Investigative Ophthalmology and Visual Science*, *49*(2), 572–580. https://doi.org/10.1167/iovs.07-0525

14. Gräf, MH, Becker, R., & Kaufmann, H. (2000). Lea symbols: visual acuity assessment and detection of amblyopia. Graefe's archive for clinical and experimental ophthalmology, 238 (1), 53-58.
15. Guest, G., MacQueen, K. M., & Namey, E. E. (2012). Introduction to applied thematic analysis. *Applied thematic analysis*, *3*, 20.
16. Guzzetta, A., Mercuri, E., & Cioni, G. (2001). Visual disorders in children with brain lesions: 2. Visual impairment associated with cerebral palsy. *European Journal of Paediatric Neurology*, *5*(3), 115–119. https://doi.org/10.1053/ejpn.2001.0481
17. Hebart, M. N., & Hesselmann, G. (2012). What Visual Information Is Processed in the Human Dorsal Stream? *Journal of Neuroscience*, *32*(24), 8107–8109. https://doi.org/10.1523/jneurosci.1462-12.2012
18. Himmelmann, K., Hagberg, G., Wiklund, L. M., Eek, M. N., & Uvebrant, P. (2007). Dyskinetic cerebral palsy: A population-based study of children born between 1991 and 1998. *Developmental Medicine and Child Neurology*, *49*(4), 246–251. https://doi.org/10.1111/j.1469-8749.2007.00246.x
19. Hoyt, C. S. (2003). Visual function in the brain-damaged child. *Eye*, *17*(3), 369–384. https://doi.org/10.1038/sj.eye.6700364
20. Jan, J. E., Lyons, C. J., Heaven, R. K. B., Matsuba, C., & Frcp, M. (2001). Visual impairment due to a dyskinetic eye movement disorder in children with dyskinetic cerebral palsy. *Developmental Medicine and Child Neurology*, *43*(2), 108–112. https://doi.org/10.1017/S0012162201000184
21. Kozeis, N., Anogeianaki, A., Mitova, D. T., Anogianakis, G., Mitov, T., & Klisarova, A. (2007). Visual function and visual perception in cerebral palsied children. *Ophthalmic and Physiological Optics*, *27*(1), 44–53. https://doi.org/10.1111/j.1475-1313.2006.00413.x
22. Lehman, S. S. (2012). Cortical visual impairment in children: Identification, evaluation and diagnosis. *Current Opinion in Ophthalmology*, *23*(5), 384–387. https://doi.org/10.1097/ICU.0b013e3283566b4b
23. Lehman, S. S. (2018). Cortical Visual Impairment in the Child with Cerebral Palsy. *Cerebral Palsy*, 1–7.
24. Matsuba, C., Jan, J., Lyons, C., & Heaven, R. (2002). Jan et al. reply. *Developmental*

Medicine & Child Neurology, *44*(5), 356–358. https://doi.org/10.1002/asi.5090180111

25. McIntyre, S., Morgan, C., Walker, K., & Novak, I. (2011). Cerebral Palsy-Don't Delay. *Developmental Disabilities Research Reviews*, *17*(2), 114–129. https://doi.org/10.1002/ddrr.1106
26. Miles, M. B. & Huberman, A. M. (1994). Qualitative data analysis: An expanded sourcebook. Thousand Oaks : Sage Publications
27. Monbaliu, E., de Cock, P., Ortibus, E., Heyrman, L., Klingels, K., & Feys, H. (2016). Clinical patterns of dystonia and choreoathetosis in participants with dyskinetic cerebral palsy. *Developmental Medicine and Child Neurology*, *58*(2), 138–144. https://doi.org/10.1111/dmcn.12846
28. Novak, I., Morgan, C., Adde, L., Blackman, J., Boyd, R. N., Brunstrom-Hernandez, J., … Badawi, N. (2017). Early, Accurate Diagnosis and Early Intervention in Cerebral Palsy: Advances in Diagnosis and Treatment. *JAMA Pediatrics*, *171*(9), 897–907. https://doi.org/10.1001/jamapediatrics.2017.1689
29. Pakula, A. T., Van Naarden Braun, K., & Yeargin-Allsopp, M. (2009). Cerebral Palsy: Classification and Epidemiology. *Physical Medicine and Rehabilitation Clinics of North America*, *20*(3), 425–452. https://doi.org/10.1016/j.pmr.2009.06.001
30. Pansell, T., Hellgren, K., Jacobson, L., Brautaset, R., & Tedroff, K. (2014). The accommodative process in children with cerebral palsy: Different strategies to obtain clear vision at short distance. *Developmental Medicine and Child Neurology*, *56*(2), 171–177. https://doi.org/10.1111/dmcn.12266
31. Panteliadis, C. P. (2018). *Cerebral palsy: A multidisciplinary approach, third edition*. *Cerebral Palsy: A Multidisciplinary Approach, Third Edition. Springer*. https://doi.org/10.1007/978-3-319-67858-0
32. Reynell, J. (1978). Child: care, health and development 1978,4,291-303. *Child: Care, Health and Development*, *4*(5), 291–303.
33. Rosenbaum, P., Paneth, N., Leviton, A., Goldstein, M., Bax, M., Damiano, D., … Jacobsson, B. (2007). A report: the definition and classification of cerebral palsy April 2006. *Developmental Medicine and Child Neurology. Supplement*, *109*(SUPPL.109), 8–14. https://doi.org/10.1111/j.1469-8749.2007.tb12610.x
34. Sakki, H. E. A., Dale, N. J., Sargent, J., Perez-Roche, T., & Bowman, R. (2018). Is there consensus in defining childhood cerebral visual impairment? A systematic

review of terminology and definitions. *British Journal of Ophthalmology*, *102*(4), 424–432. https://doi.org/10.1136/bjophthalmol-2017-310694

35. Saldana, J. (2009). *The Coding Manual for Qualitative Researchers*. Thousand Oaks, California: Sage.
36. Salt, A., & Sargent, J. (2014). Common visual problems in children with disability. Archives of disease in childhood, 99(12), 1163-1168.
37. Smithers-Sheedy, H., Badawi, N., Blair, E., Cans, C., Himmelmann, K., Krägeloh-Mann, I., … Wilson, M. (2014). What constitutes cerebral palsy in the twenty-first century? *Developmental Medicine & Child Neurology*, *56*(4), 323–328. https://doi.org/10.1111/dmcn.12262
38. Sonksen, P. M., & Dale, N. (2002). Visual impairment in infancy: impact on neurodevelopmental and neurobiological processes. *Developmental medicine and child neurology*, *44*(11), 782-791.
39. Suter, P.S. i Harvey, L.H. (2011): Vision rehabilitation: Multidisciplinary Care of the patient following brain injury. SAD, Boca Raton: CRC Press
40. Vasanth, J., Jacob, N., & Viswanathan, S. (2014). Visual function status in children with cerebral palsy. *Optometry & Visual Performance*, *2*(5), 251–254. Retrieved from http://ovidsp.ovid.com/ovidweb.cgi?T=JS&PAGE=reference&D=psyc11&NEWS=N&AN=2014-49302-007
41. Vinuela-Navarro, V., Erichsen, J. T., Williams, C., & Woodhouse, J. M. (2017). Effect of stimulus type and motion on smooth pursuit in adults and children. *Optometry and Vision Science*, *94*(7), 760–769. https://doi.org/10.1097/OPX.0000000000001090

ATTACHMENTS

PRILOZI

Informirani pristanak za sudjelovanje u istraživanju

U svrhu izrade diplomskog rada pod nazivom „Oštećenja vida zbog diskinetskog poremećaja pokreta očiju kod djece s diskinetskom cerebralnom paralizom" provodimo procjenu funkcionalnog vida i vizualnog funkcioniranja, povezanosti između vidnih funkcija i vizualnog funkcioniranja. Procjena se provodi kroz promatranje ponašanja djeteta u zadacima u kojima koristi vid.

Ovim putem Vas i Vaše dijete **molimo da nam pomognete u prikupljanju podataka na način da nam dopustite provođenje procjene vizualnog funkcioniranja s Vašim djetetom.**

Sudjelovanje u istraživanju je *dobrovoljno te dijete može odustati u bilo kojem trenutku* procjene. Nadalje, sukladno Zakonu o edukacijsko-rehabilitacijskoj djelatnosti i Etičkom kodeksu procjena je *anonimna*, nećemo navoditi Vaše osobne podatke niti osobne podatke Vašeg djeteta, kao ni Vaše osobne podatke. Prikupljeni podaci bit će strogo povjerljivi i čuvani, poznati istraživaču i mentoru istraživanja, a u pisanim i usmenim izlaganjima će biti prikazani anonimno.

Rezultati ovog istrživanja dat će uvid u učestalost oštećenja vida i problematiku vizualnog funkcioniranja kod djece s diskinetskom cerebralnom paralizom što će pomoći pri osiguravanju pravovremene dijagnoze i primjerene rehabilitacije djece s diskinetskom cerebralnom paralizom.

Zahvaljujemo Vam na strpljenju i sudjelovanju.

Za dodatne informacije prije, za vrijeme ili poslije ispunjavanja upitnika možete se obratiti istraživačici: Mihaela Ivošević, mob. 099/ 8327 559; e-mail: mihaela.ivosevic@stud.erf.hr i/ili mentorici: doc.dr.sc. Sonja Alimović, e-mail: sonja.alimovic@erf.hr.

Svojim potpisom, ja _____ dajem pristanak za korištenje rezultata dobivenih procjenom funkcionalnog vida mog djeteta.

_____

Potpis

Sveučilište u Zagrebu
Edukacijsko-rehabilitacijski fakultet

POZIV ZA SUDJELOVANJEM U ISTRAŽIVANJU:

„Oštećenja vida uslijed diskinetskog poremećaja pokreta očiju kod djece s diskinetskom cerebralnom paralizom"

Glavni istraživač i kontakt osoba:

Mihaela Ivošević (studentica diplomskog studija Edukacijska rehabilitacija na Edukacijsko-rehabilitacijskom fakultetu Sveučilišta u Zagrebu, E-mail: mihaela.ivosevic@stud.erf.hr)

Članovi istraživačkog tima:

- Doc. dr. sc. Sonja Alimović (Edukacijsko-rehabilitacijski fakultet Sveučilišta u Zagrebu)
- Prof.dr.sc. Els Ortibus (Department of Pediatric Neurology; University Hospitals Leuven, Belgium)
- Mihaela Ivošević (Edukacijsko-rehabilitacijski fakultet Sveučilišta u Zagrebu)

Čast nam je pozvati Vaše dijete i Vas da doprinesete našem istraživanju!

Cilj istraživanja je dobiti uvid u u razinu funkcionalnog vida, utvrđivanje prirode povezanosti između vidnih funkcija i vizualnog funkcioniranja te opservaciju ponašanja u specifičnim vidnim zadatcima za utvrđivanje teškoća u vizualnim aktivnostima djece s diskinetskom cerebralnom paralizom

Koja su okularna i cerebralna oštećenja vida te vidnih funkcija djece s diskinetskom cerebralnom paralizom?

Kakvo je vizuomotoričko i vizuopereceptivno funkcioniranje djece s diskinetskom cerebralnom paralizom ?

Ukoliko je diskinetski poremećaj pokreta očiju prisutan, na koji način se manifestira?

Kakav je odnos refrakcijskih grešaka i poteškoća u okulomotorici te postojanja nedostatno razvijenih vidnih funkcija?

Rezultati ovog istraživanja dat će uvid u učestalost oštećenja vida i problematiku vizualnog funkcioniranja kod djece s diskinetskom cerebralnom paralizom

→ to će pomoći pri osiguravanju pravovremene dijagnoze i primjerene re(habilitacije) djece s diskinetskom cerebralnom paralizom

Kako se funkcionalni vid manifestira s obzirom na područja vizualnog funkcioniranja?

Bit ćemo slobodni kontaktirati Vas putem telefonskog kontakta da provjerimo Vašu spremnost za sudjelovanje u istraživanju