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CEREBRAL PALSY AND LANGUAGE: A SYSTEMATIC REVIEW

Dissertação apresentada à Universidade Católica Portuguesa para obtenção do grau de mestre em

Neuropsicologia

Por

Filipa Pais Gonçalves

Lisboa, 2021

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A PARALISIA CEREBRAL INFANTIL E A LINGUAGEM: UMA REVISÃO SISTEMÁTICA

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Filipa Pais Gonçalves

Sob a orientação de Professora Doutora

Maria Vânia Silva Nunes

Lisboa, 2021

Abstract

Aim: The aim of this systematic review was to describe the language's development in children with cerebral palsy and its association with other variables, such as the Cerebral Palsy (CP) subtype and the level of Gross Motor Function Classification System (GMFCS).

Method: A systematic review was carried out, following the prisma check-list. The databases consulted were *PUBMED*, *SCOPUS* and *Web of Science*. Articles published in other languages, than English, and articles that used only functional measures to assess expressive language, were excluded. The Systematic Review Protocol was registered in the PROSPERO Register.

Results: A total of 723 publications were found in the PUBMED, SCOPUS and WEB of SCIENCE databases. Of these articles, 374 duplicates were excluded. Most of the included studies were developed in European countries and in the last 10 years. There were only 2 case-control studies, which compared tipically developped (TD) children with children with CP, as all the others being observational cohort studies, organized either by CP subtype or by other functional classifications. Regarding the evaluated language subdomains, all studies assessed receptive language and most all of those also assessed expressive language. Within these two main areas, there was great variability in the specific domains evaluated, from semantics, phonological awareness, articulation, lexical development.

Conclusion: The data found in this systematic review reinforces the idea proposed by previous researchers that the motor disorders are often accompanied by disturbances in cognition and, in particular, in language. In the studies where the CP children were compared with TD children, although there was significant variability among subdomains, the performance was, in general, poorer. In what concerns the association with the GMFCS level and the CP subtype, all articles showed that as GMFCS level increases, the language impairments also increase and the dyskinetic subtype seemed to be the one with poorer performance in language tasks, results that are in line with previous research.

Key words: Cerebral palsy; Language; Gross Motor Function Classification System

Glossary

BGN: BASAL GANGLIA NECROSIS

CAS: CHILDHOOD APRAXIA OF SPEECH

C-BILLT: COMPUTER BASED INSTRUMENT FOR LOW MOTOR LANGUAGE TESTING

CFCS: COMMUNICATION FUNCTION CLASSIFICATION SYSTEM

CG: CONTROL GROUP

CP: CEREBRAL PALSY

DDST II: DENVER DEVELOPMENTAL SCREENING TEST II

EG: EXPERIMENTAL GROUP

GMFCS E&R: GROSS MOTOR FUNCTION CLASSIFICATION SYSTEM EXTENDED AND REVISED

GMFCS: GROSS MOTOR FUNCTION CLASSIFICATION SYSTEM

IQ: INTELLIGENCE QUOTIENT

MACS: MANUAL ABIILITY CLASSIFICATION SYSTEM

PNVPC: PROGRAMA NACIONAL DE VIGILÂNCIA DA PARALISIA CEREBRAL

PVL: PERIVENTRICULAR LEUKOMALACIA

SLPG: SPEECH LANGUAGE PROFILE GROUP

SR: SYSTEMATIC REVIEW

SSPI: SEVERE SPEECH IMPAIRMENTS

TD: TIPICALLY DEVELOPED

VSS: VIKING SPEECH SCALE

VTPP: VOCABULARY TEST BY PEABODY PICTURE

WPPSI: WECHSLER PRESCHOOL AND PRIMARY SCALE OF INTELLIGENCE

Index

Index: Figures and Tables

1. Introduction

Cerebral palsy is a set of movement and posture disorders that occurs during fetal development or in childhood, and it is not progressive in nature (Levitt & Addison, 2019). It presents a great variability, existing from cases of children with mild motor disorders, to children with bilateral motor impairment, absence of language and totally dependent on their caregivers (Fairhurst, 2012a).

Although it is considered that the primary deficit of these children occurs in the motor domain, it is extremely common, and transversal to the different types of cerebral palsy, the presence of sensory and cognitive impairments , with executive, visuospatial, attention and language functions as the most impoverished areas (J. Fluss & Lidzba, 2020). Although there is not an agreement regarding the methods used in language assessment among children with CP, it is known that language difficulties is one of the most common impairments, affecting their quality of life and participation (Barty et al., 2016; Fluss & Lidzba, 2020; Vendrell & Narberhaus, 2009).

This project proposes the development of a Systematic Review on language in cerebral palsy, in order to understand, on one hand, how and in what specific domains, is this cognitive function altered in children with cerebral palsy and, on the other hand, if there is evidence that points to a relationship between its degree of functionality, with regard to gross motor skills (Palisano et al., 2008) the cerebral palsy subtype and language development, at the level of different domains involved in both expressive and receptive language.

2. Theoretical Framework

2.1. Cerebral palsy

Cerebral palsy is a set of permanent disorders of motor and postural development, with an incidence of approximately 2 in every 1000 live births, a pathology that, despite major improvements, in terms of maternal and child health, especially in developed countries, maintains a constant number of cases and constitutes the most common cause of physical disability in pediatric age (Eunson, 2016; Rosenbaum, 2014).

These disorders are explained by disturbances or lesions in the brain of the fetus or in the post-natal period (Rosenbaum, 2014). In addition, there are risk factors repeatedly associated with the diagnosis of CP such as prematurity (Fairhurst, 2012), male babies, multiple gestation, intrauterine viral infections, mothers with diseases of the thyroid functioning (Jacobsson & Hagberg, 2004). In a systematic review on this topic, the following risk factors were found, consistently, throughout the various studies analyzed: placental problems; low birth weight; meconium aspiration; caesarean delivery; prenatal asphyxia; neonatal seizures, hypoglycemia, neonatal infections and respiratory distress syndrome (McIntyre et al., 2013).

More recently, the research regarding the genetic role in cerebral palsy has been growing and evidence shows, consistently, that a potential genetic etiology can be identified in, approximately, 20% of individuals with cerebral palsy, mostly, monogenetic rare diseases (Maclennan et al., 2019). Studies suggest that DNA variants could modify an individual's susceptibility to insults such as thrombosis or haemorrhage, which impact motor development (Eyk et al., 2018; Maclennan et al., 2017). However, it is not clear yet that there are specific cerebral palsy genes and it's argued that, in most cases, it's sustained by an epigenetic etiology (Eyk et al., 2018; Maclennan et al., 2019). Currently, the International Cerebral Palsy Genomic Consortium is developing an international database of genetic variants involved in cerebral palsy. Since there is such a wide intradiagnostic varibility, the deeper understanding of the genomics and other development mechanisms may help to classify the individuals, more accurately (Maclennan et al., 2019).

Regarding the diagnostic criteria, and according to the definition proposed by Rosenbaum (2014), the designation of CP must be limited to changes in motor development, even if deficits of another nature arise, and it must concern etiologies prior to the end of the neonatal period (Love et al., 2007).

Although there is a huge intradiagnosis variability, some authors point to the fact that lesions in the basal ganglia, associated with lesions in cortical structures are responsible for the worst prognosis (Himmelman & Uvebrant, 2011).

In Portugal, according to the Report of the National Cerebral Palsy Surveillance Program at 5 years of age, there is a decrease in the incidence rate in children born in Portugal between 2001 and 2007, from 2.01-live births born in 2001 to 1.55 live births born between 2001 and 2007. Despite this, the Report warns of the fact that there is a national trend towards an increase in the birth of twins, as well as an increase in the average age of mothers, which brings to discussion if the incidence rate will continue to decrease (PNVPC, 2017).

In a study carried out in Canada, the data points to a decline in prevalence since the early 1990s, with this reduction being even more significant among groups III-V of GMFCS, those diagnosed with bilateral spastic CP and with severe cognitive impairments (Hadjinicolaou et al., 2019). In another study, carried out in Sweden, which analyzed 186 children born with CP, between 1999 and 2002, the epidemiological data show that 40% of the children had unilateral spastic CP and 39% bilateral; 16% had CP of the dyskinetic type and 5% of the ataxic type (Himmelman & Uvebrant, 2011).

Three main types are known in which children with cerebral palsy are grouped: spastic, ataxic and dyskinetic (Surveillance of Cerebral Palsy in Europe, n.d.).

The first one is characterized by a large increase in muscle tone, which originates, in most cases, from cortical injuries (Eunson, 2016). Furthermore, within the spastic type, hyperreflexia and pyramidal signs are described, such as the babinski reflex. Spastic cerebral palsy is also characterized by an abnormal pattern of posture and movement (Surveillance of Cerebral Palsy IN

Europe, n.d.). According to the European Classification of Cerebral Palsy, the spastic type can be divided in bilateral, which refers to cases of affected limbs in both sides of the body and unilateral, which regards cases of affected limbs in only one side of the body. The classification of diplegia or tetraplegia is no longer used (Fairhurst, 2012) and it's recommended, instead, the use of functionality profiles.

The second one, the ataxic type, is identified by a loss of muscular coordination, a decrease in tone, balance problems, which are shown through difficulties in the use of force and in the sense of rhythm and accuracy and are associated in lesions in the cerebellar pathways (Rosenbaum, 2014). The tremor is present but it is called an intention tremor, because it's notable at the end of a voluntary and intentional movement, by opposition of the rest tremor, seen in other diseases. The ataxia of the trunk and gait is also present and in the intentional movements, it's usual to watch a behaviour of past-pointing (Surveillance of Cerebral Palsy IN Europe, n.d.).

In the third case, the dyskinetic type, we may find slow movements and chorea, which seems to be associated with lesions in the basal ganglia and in the extrapyramidal pathways (Eunson, 2016). The general profile is defined by involuntary, uncontrolled and often stereotyped movements, the muscle tone fluctuates between spasticity and hypotonia and there is a predominance of primitive reflex (Levitt & Addison, 2019).

This cerebral palsy type is divided into two subtypes: the dystonic and the choreo-athetotic. The first one is mainly defined by its abnormal postures and, although fluctuating, there is a predominance of spastic tone. On the other hand, the choreo-athetotic subtype is characterized by hyperkinesia and a predominant hypotonic tone (Surveillance of Cerebral Palsy IN Europe, n.d.).

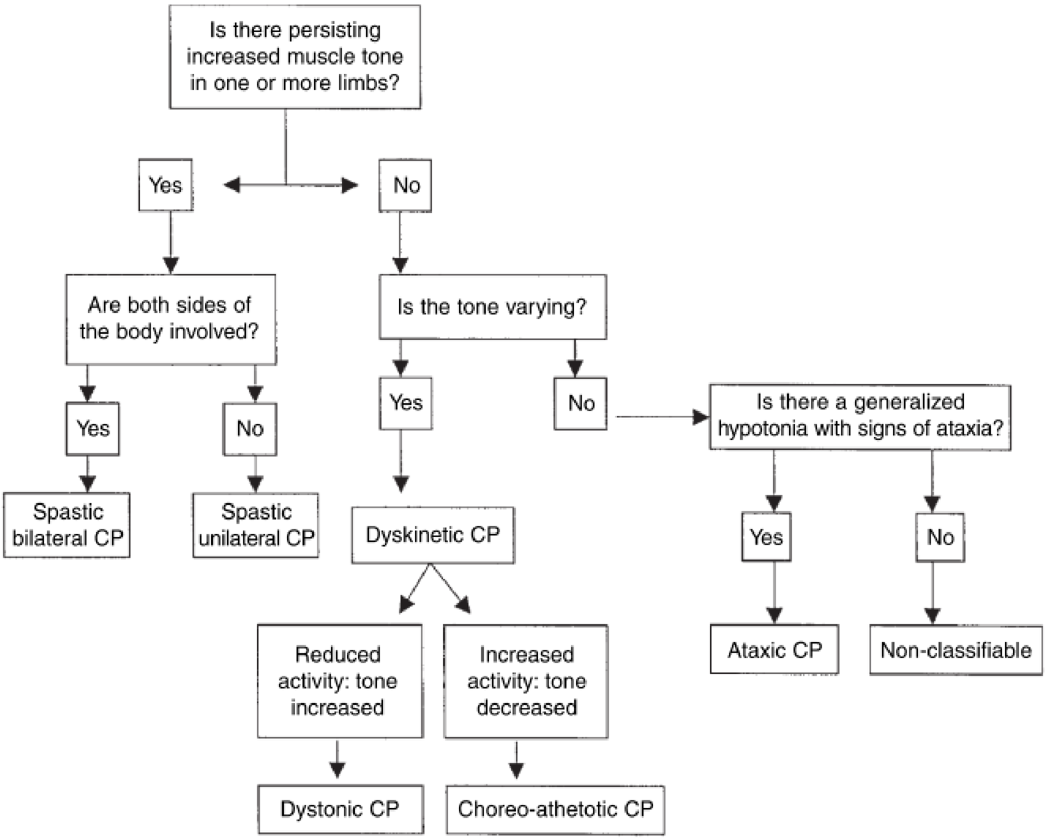

Figure 1: Hierarchical classification tree of cerebral palsy subtypes (Cans, 2000).

Furthermore the division of CP in diferente subtypes, some authors tried to introduce the concept of "severe CP" (Dakovic et al., 2014; Horber et al., 2020), although this isn't consensual.

In this study, conducted from the data from the SCPE, an index for a high level of impairment was defined, which applied to 39% of the children: they did not walk or had a severe intellectual delay (IQ <50), with comorbidities for epilepsy, visual and / or hearing impairment (Horber et al., 2020).

Regarding the CP types, in any of those, the presence of other impairments, namely sensory, cognitive and language, is common (Rosenbaum, 2014). Children with cerebral palsy may have a deficit in sensory integration, as a result of neurological dysfunction, or due to limited access to sensory experiences, as a result of their motor difficulties (Bumin, 2001).

Due to this fact, in the last 15 years, several proposals have emerged for a new framework for the discussion of cerebral palsy, namely its renaming, becoming known as *“brain injury / interference of early development”*, thus removing the centrality of motor deficits (Shusterman, 2014).

More recently, a new definition added clearly the idea of how common are other disturbances, stating that the motor disorders are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behavior, and /or by epilepsy (Rosenbaum, 2014).

There is yet another proposal that argues the renaming to cerebral palsy spectrum disorder, in order to promote a more comprehensive awareness and to enhance the development, evidence based, of more holistic and truly child-oriented diagnostic and intervention approaches (Shevell, 2019).

In a population study, learning difficulties are described in 45% of children; epilepsy in 44%; communication difficulties in 49% and total absence of speech in 30%; 17% of children also had visual impairments and 6% had a neuropsychiatric diagnosis (Himmelman & Uvebrant, 2011).

In the case of cognitive impairments, in general, they appear to be positively correlated with the level of motor deficit and with comorbidity with epilepsy. Most children with cerebral palsy have neuropsychological deficits, especially in the scope of visuospatial, executive and attention functions, and ¼ of these children have language deficits (Aisen et al., 2011; J. Fluss & Lidzba, 2020).

In another study, which included only children with spastic bilateral CP subtype, receptive language was impaired in 85% of participants, visuospatial abilities in 90%, short-term memory in 21-58%, declarative memory in 47-67%, praxis comprehension in 20% and executive functions in 58-74%, which means that visuospatial abilities and receptive language were the most frequent impairments within this group (Vendrell & Narberhaus, 2009).

In short, several studies have pointed to the importance of deepening the knowledge about the impairments associated with cognitive aspects, describing that, in several cases, these can be even more disabling than the motor deficit itself (Himmelman & Uvebrant, 2011). Thus, along with the classification of the cerebral palsy subtype and the use of functional classification systems (Hidecker, 2020; Horber et al., 2020), it seems to be essential, during the evaluation process of these children, to ensure the screening of frequently associated conditions, with particular emphasis on cognitive impairments and epilepsy (Stadskleiv, 2020).

2.2. The Gross Motor Function Classification System

The Gross Motor Function Classification System (GMFCS) is an instrument for the functional classification of gross motor skills of children with CP, allowing a simple look at the child's abilities and limitations (Palisano et. al., 1997; Palisano et al., 2008).

This system is divided into 5 levels and grouped by 4 age groups and, although it was initially designed to classify children between 2 and 12 years of age, its expanded and revised version includes now children between 12 and 18 years old (Palisano et al., 2008, 2018). In 2007, it was also created an adaptation for the portuguese population (Andrada et al., 2007).

GMFCS E&R, as a functionality classification instrument, thus seeks to distinguish between capacity and performance, the first one referring to what an individual can do in his natural environment and the second one to what he does when he is asked to perform a task in a formal context of evaluation (Palisano et al., 2008, 2018).

In a descriptive study, regarding children born between 1990 and 2016, in Europe, 32% had a GMFCS of III – V; Of those, 15% had a GMFCS of IV and V and a severe intelectual impairment, accompanied by epilepsy. The spastic bilateral CP is the one presenting a higher number of impairments. Inside the group of dyskinetic CP, it was not found an association between cognitive development and the GMFCS level.

In the ataxic group, 72% could walk without help and more than a half of those children had a normal IQ (Horber et al., 2020).

In a population study, mentioned above, of the 186 children studied, 51% were classified as GMFCS level I; 14% at level II, 3% at level III; 11% at level IV and 22% at level V (Himmelman & Uvebrant, 2011). Regarding the relationship between the anatomy of the lesion and the level of GMFCS, the study shows that 76% of children, with lesions in white matter, were classified as levels I and

II; on the other hand, 67% of those with lesions in the basal ganglia would be at levels IV and V (Himmelman & Uvebrant, 2011).

Also, several studies point to this association between GMFCS and the existence of language impairments , however other authors argue that it is excessive to consider GMFCS as a predictor, in itself, of language impairments, which may have motivated the creation of the CFCS (Communication Function Classification System), which seems to make a more adjusted contribution to the planning of the intervention and, consequently, to the decision making on the use of alternative and augmentative means of communication (Hidecker, 2020).

2.3. Language's development: the case of Cerebral Palsy

Articulation problems and communication disorders are common results of the diagnosis of cerebral palsy and can affect the child's development in different areas of his life (Barty et al., 2016).

Recent studies, using neuroimaging techniques emphasize that areas such as the cerebellum, the dorsolateral prefrontal cortex and the basal ganglia are activated, both during the execution of motor tasks and during the execution of linguistic tasks (Houwen et al., 2016). In addition, some areas directly and highly related to language, such as Broca's area, are activated by performing motor actions. Some studies also point to motor development as an important predictor of children's performance in reading and arithmetic tasks (Adi-Japha et. al., 2011) and others demonstrate, through functional imaging techniques, the clear involvement of the supplementary motor area in the processing of language (Courson & Tremblay, 2017).

It is known that children with cerebral palsy, at preschool age, have a low language development, having a lexicon lower than expected for their age, as well as significant phonological, syntactic and articulatory problems (Chorna et al., 2017). Despite this, other authors argue that children with cerebral palsy may have language skills appropriate for their age, however, the same does not seem to be the case, in general, for reading skills (Critten et al., 2019). In line with the literacy area, another study reports that the fact of many children with cerebral palsy suffer from articulatory problems hinders their phonological awareness, which is considered one of the main predictors of reading (Peeters et al., 2009).

It is also argued that, in children with cerebral palsy, communication problems, namely poor speech from a lexical point of view, can result directly from their motor deficit and can have a considerable negative impact on their socialization (Chorna et al., 2017).

It was also found, in one of the studies, a linear association between the quality of the articulation and the memory span of children with CP, highlighting the fact that children with bigger motor problems have to face multiple challenges in order to develop language (Peeters et al., 2009).

In another study, among the group of children who did not have significant articulatory difficulties, there were lower performances in complex tasks of fine motor skills, which can be rooted in a problem of motor programming and coordination (Estil et al., 2003).

Another study points to the fact that many of these children often have difficulties in visuospatial skills, it can contribute to a low memory regarding the orientations of the forms of the letters themselves and, consequently, lead to reading and writing problems (Critten et al. , 2019).

Not only that, but also the intelligence factor seems to have a relevant mediation role between the motor and language domains, as shown by the results of a study in which half of children with normal intelligence (IQ> 70) had, for the most part, motor impairments; on the other hand, children with subnormal intelligence (IQ <70) showed impairments, both in the motor domain and in the language domain (Pirila et al., 2007). It is understood that the neurological injury, which is the source of language impairments, may also be responsible for a lower intellectual capacity (Peeters et al., 2009).

Some data suggest that 25% of children with cerebral palsy will not develop language, and those who suffer from spastic bilateral and / or diskynetic type cerebral palsy are the ones most at risk (Novak & Spirit-jones, 2017). These data suggest the proximity between language and motor skills and, according to several authors, their interdependence (Courson & Tremblay, 2017; Ding et al., 2015; Estil et al., 2003; Fonseca, 2001), which, in disorders such as cerebral palsy, highlights the need to consider clinical exercise and rehabilitation programs, from a multidisciplinary and synergistic perspective (Bonnechère, B., Jansen, B., Omelina, L., Degelaen, M., Wermenbol, V., Rooze, M., Sint Jan, 2014).

In a study of the prevalence of speech disorders in Norway, in children with cerebral palsy in which it was possible to assess speech development, 48% had a normal speech; 16% had a slightly imperceptible speech; 9% clearly imperceptible; 6% seriously imperceptible and 19% had no speech, which means that half of the children were impaired in this domain (Andersen & Trust, 2010).

Speech problems were more prevalent in children of the dyskinetic type and in children with a GMFCS level IV and V classification. Among the children whom speech problems were identified, 54% used an augmentative and / or alternative means of communication (Andersen & Trust, 2010).

Geytenbeek (2015), in a study on the understanding of oral language, in children with cerebral palsy, shows that 33% of the variability of the results is explained by the chronological age, its level of GMFCS and cerebral palsy subtype. The same author, in another subsequent study, shows the importance of this last factor, even in the understanding of oral language, since children of the dyskinetic type obtained better results than children of the spastic type (Geytenbeek et al., 2015).

A recent Systematic Review (Vaillant et al., 2020), which sought to present evidence on the factors that influence the oral comprehension of children with cerebral palsy, suggests the need to deepen the understanding of the effect of certain dimensions, such as the subtype, the classification of the GMFCS and even comorbidity for epilepsy.

In terms of assessment, the studies provide very different instruments and, consequently, results (Rosenbaum et al., 2014). The Platform for Surveillance of Cerebral Palsy in Europe recommends the use of the Viking Speech Scale, which consists in a classification system, divided in 4 levels, according only to the expressive language abilities presented for the child (Surveillance of Cerebral Palsy IN Europe, n.d.). This assessment keeps in mind the association with the motor disorder, stating, for example, in Level 1: *“Speech is not affected by motor disorder”.*

Another classification system, the Communication Function Classification System (CFCS), created by the same authors as GMFCS, it can be applied to children from 2 years of age and, like GMFCS, it is divided into 5 levels, however, unlike the latter, it is not divided into several age groups, which means that it’s expected the child progresses through the various levels (Hustad et al., 2015; Palisano et al., 2018). Unlike the Viking Speech Scale (VSS), the CFCS focuses on the assessment of the child's communication, in any modality, that is, it classifies not only the expressive speech, but also the child's ability to use gestures, the eye gaze and, if applicable, alternative and augmentative communication systems (Hidecker et al., 2011; Hustad et al., 2015).

Whether at VSS or CFCS, the assessments seem to leave space for some subjectivity, since both instruments can be used, either by health technicians, rehabilitation teams, but also by families, which can generate inconsistencies, or because of the subjectivity of those assessing, either by the characteristics of the contexts that can enhance, more or less, the child's abilities (Geytenbeek, 2016; Hustad et al., 2015).

Other authors also propose the Speech Language Profile Groups (SLPG) which seeks to classify children, on four levels, based on two assumptions: on one hand, children who have articulatory problems that affect speech and, on the other hand, children who, in addition to articulation problems, may also have cognitive impairments, which also contribute to explain their communication difficulties (Hustad et al., 2015).

All of this evidence reinforces the need for language assessments to be made, as early as possible, and in a coordinated manner with the assessment of other cognitive functions, in order to adjust the planning of interventions to the real needs of children (Geytenbeek, 2016).

Thus, in summary, there are several authors who point to the influence that the subtype of cerebral palsy, the level of GMFCS, and the presence of other deficits, such as epilepsy, have on the severity of language problems (J. Ballester-Plané et al., 2018; Coleman et al., 2013; Estil et al., 2003; Fairhurst, 2012b; Hidecker, 2020; Vitrikas et al., 2020). However, the specificities of the language deficits of these children remain to be clarified, and it is not clear, in several studies, whether it's performed only a communication assessment (expressive language) or if other impairments are present, regarding comprehension, reading, writing and other countless subdomains of language, such as morphosyntax, semantics, phonetics and prosody (J. Geytenbeek, 2016; Vaillant et al., 2020).

Thus, the need and interest arose to carry out a systematic review that allows analyzing the level of evidence with regard to language development in children with cerebral palsy. This review hopes to deepen the knowledge in this area, and, from this, it may pave the way for the discussion on specific domains of language assessment and intervention in cerebral palsy.

3. Methods

A systematic review was carried out, following the *Prisma check-list* (Tricco et. Al., 2018)

3.1 Research Questions:

Our main research question is if there is an influence of cerebral palsy in language skills. The main outcome is language status in children with cerebral palsy and main comparator is language status in children with typical development.

Specific questions:

a) Are there differences between the language skills, receptive and expressive, of children with cerebral palsy and those of children with typical development?

1. If there are differences, which linguistic domains are most affected?

b) Is the GMFCS level related to the development of language skills?

c) Is the cerebral palsy subtype related to the development of language skills?

3.2 Search strategy

A systematic literature was conducted, following the *Prisma-ScR* checklist (Tricco et. Al., 2018). The databases consulted were *PUBMED*, *SCOPUS* and *Web of Science*.

The research was conducted in english, using uncontrolled terms, resorting to the use of *Boolean* operators, such as AND and OR, and truncations, so that possible variations of the term can be captured (Donato, 2019).The syntax used was: *("cerebral palsy" AND child* OR youth AND gmfcs OR "Gross Motor Classification System" AND language OR speech OR communication Or linguistic skills OR linguistic competences".)*

The mesh terms were available for almost all the search terms, as listed below:

Cerebral Palsy [C10.228.140.140.254]

Child [M01.060.406]

Adolescent [M01.060.057]

Language [F01.145.209.399]

Speech [L01.559.423.676]

Communication [L01.143]

Linguistic skills e linguistic competences (*no mesh terms available*)

GMFCS e Gross Motor Function Classification System (*no mesh terms available*)

To ensure that an appropriate research strategy was being used, *PRESS* was used, a tool used to validate information search strategies, thus helping researchers to present the most accurate review possible (McGowan, 2016).

3.3 Inclusion criteria

- Articles, published in peer-reviewed journals, were included
- Articles, in english, portuguese, french and spanish were included
- The study designs of the articles are randomized clinical trials; non-randomized trials; cohort studies; cross-sectional observational studies and case studies.
- Articles were included that, in their keywords and / or abstracts, include the terms “cerebral palsy” combined with “GMFCS” and/or “language” and / or “language skills” and / or “oral language” and / or “reading skills and writing ”and / or“ verbal comprehension ”. and / or oral expression and / or verbal production and/or communication, in the four languages supported in this systematic review.
- Studies were included whose sample includes at least one child, up to 18 years old, diagnosed with cerebral palsy.

- Studies that investigate the association between different aspects of language (oral comprehension, oral expression, written comprehension, written production, reading) and the diagnosis of cerebral palsy were included.
- Studies that investigate the association between different aspects of language (oral comprehension, oral expression, written comprehension, written production, reading) and the subtype of cerebral palsy and / or at the level of motor functionality (GMFCS scale) were included.
- Studies that describe the language assessment instruments used were included.
- Studies that provide specific data regarding language assessment were included.

3.4 Exclusion criteria

- Articles published in other languages, than english, Portuguese, French or spanish were excluded.
- Articles that use only functional measures to assess expressive language (speech) were excluded.

3.5 Systematic Review Protocol

The Systematic Review Protocol was registered in the PROSPERO Register: Prospective Register of Systematic Reviews, thus avoiding duplication of reviews (Donato, 2019):
https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42021229257

3.6 Selection and evaluation of the study's quality

Mendeley, a reference management program, was used. Duplicate articles were deleted through the Rayyan Platform. In a first phase, studies that meet the defined criteria were selected, which resulted from an analysis of the titles and abstracts. This initial screening led to the elimination of many studies, and then the documents were analyzed in full (Cochrane, 2020).

The selection of articles was ensured, using the Rayyan platform, by two researchers.

At the end of the selection, a flowchart was produced that illustrates all the stages of the process. To assess the quality of the studies, a checklist was created, inspired by the Newcastle-Ottawa Scale adapted for cross-sectional studies. This scale evaluates domains related with the representativeness of the sample, the sample size, the data regarding the non-respondents, the ascertainment of the exposure, the comparability of subjects, the way the outcome is assessed, and the statistical tests used (NOS, n.d.).

3.7 Data Extraction and Synthesis

A data extraction form was created, based on the support checklist of the Cochrane Collaboration (Cochrane, 2020).

The synthesis of the data illustrates the strength of the evidence, describing, therefore, whether the associations found are consistent throughout the articles. Finally, the research questions were resumed, concluding whether the evidence found, throughout the systematic review, allows them to be answered.

4. Results

A total of 723 publications were found in the PUBMED, SCOPUS and WEB of SCIENCE databases. Of these articles, 374 duplicates were excluded, through the RAYYAN platform.

After that, the titles and abstracts of the articles were analyzed, and it was possible to exclude 318 articles, as they did not meet all the defined inclusion criteria.

Thirty articles were selected for full reading, of which 21 were excluded due to various reasons: a) wrong outcome, that is, the results of the investigation were not directly linked with the assessment of language domains (5 articles); b) the assessment of language domains was only assessed using indirect measures of inquiry to parents and caregivers (9 articles); c) the assessment was exclusively focused on communication, using only functional classification instruments (7 articles).

The 10 articles included met all the inclusion criteria defined for this systematic review.

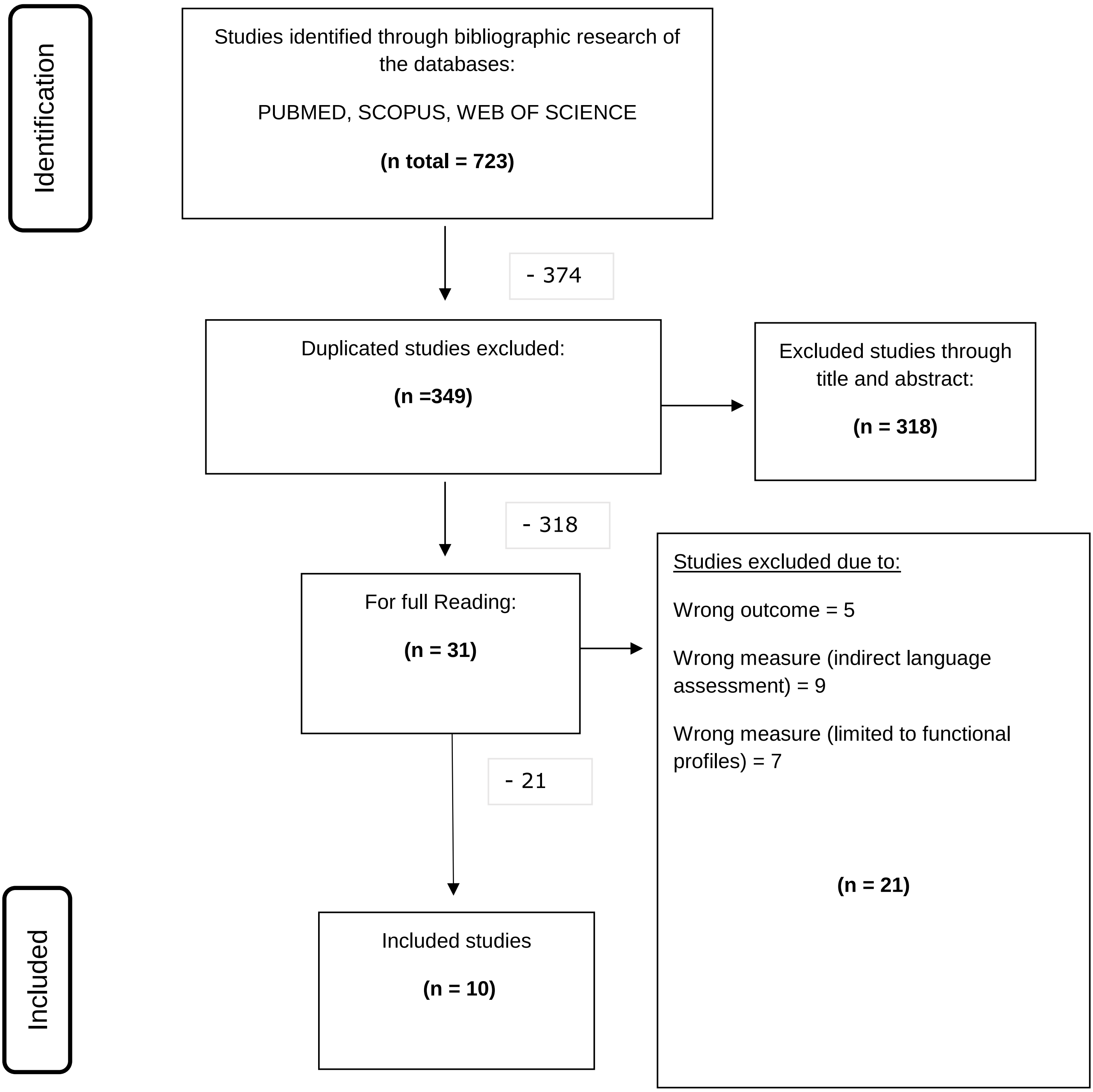

Figure 2. Flow of studies, following PRISMA selection (Moher et al., 2009)

As can be seen in table 1, most of the included studies were developed in European countries and in the last 10 years. There were only 2 case-control studies, which compared typically developed children with children with CP, as all the others being observational cohort studies, organized either by CP subtype or by other functional classifications such as verbal/non-verbal children. The ages of the children ranged between 3 and 12 years old, and in all studies there was the inclusion of a balanced number of male and female participants.

Number	Authors	Year	Country	Journal	(n)	Gender (n)		Age range/Age average	Study Type
						M	F		
1	Ballester-Plané et. al	2017	Spain	European Journal of Paediatric Neurology	104 (52=PCI; 52=TD)	56	48	Mean = 34 months	Descriptive observational study: case-control study
2	Dahlgreen Sandberg, A.	2006	Sweden	Developmental Medicine & Child Neurology	6	1	5	6 > 12 years	Descriptive observational study: cohort study
3	Geytenbeek, J. et. al.	2015	Netherlands	Developmental Medicine & Child Neurology	87	44	43	Mean = 6 years and 8 months	Analytical observational study: cohort study
4	Geytenbeek, J. et. al.	2015	Netherlands & Portugal	European Journal of Paediatric Neurology	80	41	39	7 > 12 years	Analytical observational study: cohort study
5	Lamonica, D. et. al.	2015	Brazil	Journal of Communication Disorders, Audiology and Swallowing.	20 (10=PCI; 10=TD)	4	6	44 > 83 months	Descriptive observational study: case-control study
6	Mei et. al.	2020	Australia & UK	Developmental Medicine & Child Neurology	84	47	37	4 years and 11 months > 6 years and 6 months	Descriptive observational study: cohort study
7	Mei et. al.	2015	Australia & UK	Developmental Medicine & Child Neurology	84	47	37	5 > 6 years	Descriptive observational study: cohort study
8	Sigurdardottir, S.; Vik, T.	2010	Iceland & Norway	Developmental Medicine & Child Neurology	152	74	78	Mean = 5 years and 6 months	Descriptive observational study: cohort study
9	Sigurdardottir, S. et. al	2008	Iceland & Norway	Developmental Medicine & Child Neurology	127	60	67	Mean = 5 years and 6 months	Descriptive observational study: cohort study
10	Soriano, J. & Hustad, K.	2020	USA	Developmental Neurorehabilitation	27	15	12	10>12 years	Analytical observational study: cohort study

Table 1: Included studies in the Systematic Review.

Regarding the quality assessment of the studies, the evaluation grid of the *Newcastle-Ottawa Scale* was used, adapted to cross-sectional studies, with most studies (7/10) varying between Good Study and Very Good Study. None of the included studies were classified as Unsatisfactory.

	SELECTION				COMPARABILITY	OUTCOME		TOTAL SCORE	QUALITATIVE SCORE
	Representativeness of the sample	Sample size	Non respondents	Ascertainement of the exposure (risk factor)		Assessment of outcome	Statistical test		
Ballester-Plané, J., Laporta-Hoyos, O., Macaya, A., Póo, P., Meléndez-Plumed, M., Toro-Tamargo, E., Gimeno, F., Narberhaus, A., Segarra, D., & Pueyo, R. (2018). Cognitive functioning in dyskinetic cerebral palsy: Its relation to motor function, communication and epilepsy. *European Journal of Paediatric Neurology*, *22*(1), 102–112. https://doi.org/10.1016/j.ejpn.2017.10.006	___ c)	___ c)	*	**	**	**	*	8	Good Study
Dahlgren Sandberg, A. (2006). Reading and spelling abilities in children with severe speech impairments and cerebral palsy at 6, 9, and 12 years of age in relation to cognitive development: A longitudinal study. *Developmental Medicine and Child Neurology*, *48*(8), 629–634. https://doi.org/10.1017/S0012162206001344	___ c)	___ c)	____ c)	**	**	___ d)	*	5	Satisfactory study
Geytenbeek, J. J. M., Vermeulen, R. J., Becher, J. G., & Oostrom, K. J. (2015). Comprehension of spoken language in non-speaking children with severe cerebral palsy: An explorative study on associations with motor type and disabilities. *Developmental Medicine and Child Neurology*, *57*(3), 294–300. https://doi.org/10.1111/dmcn.12619	*	___ ©	____ c)	**	**	**	*	8	Good Study

Geytenbeek, J. J., Oostrom, K. J., Harlaar, L., Becher, J. G., Knol, D. L., Barkhof, F., Pinto, P. S., & Vermeulen, R. J. (2015). Language comprehension in nonspeaking children with severe cerebral palsy: Neuroanatomical substrate? *European Journal of Paediatric Neurology*, *19*(5), 510–520. https://doi.org/10.1016/j.ejpn.2015.06.001	__ c)	___ c)	_____ c)	**	N/A	**	*	5	Satisfactory study
Lamônica, D. A. C., Paiva, C. S. T., Abramides, D. V. M., & Biazon, J. L. (2015). Communication skills in individuals with spastic diplegia. *Codas*, *27*(2), 135–141. https://doi.org/10.1590/2317-1782/20152013060	___c)	___c)	_____c)	**	**	**	*	7	Good Study
Mei, C., Reilly, S., Reddihough, D., Mensah, F., Pennington, L., & Morgan, A. (2016). Language outcomes of children with cerebral palsy aged 5 years and 6 years: A population-based study. *Developmental Medicine and Child Neurology*, *58*(6), 605–611. https://doi.org/10.1111/dmcn.12957	*	*	*	**	**	**	*	10	Very good study
Sigurdardottir, S., Eiriksdottir, A., Gunnarsdottir, E., Meintema, M., Arnadottir, U., & Vik, T. (2008). Cognitive profile in young Icelandic children with cerebral palsy. *Developmental Medicine and Child Neurology*, *50*(5), 357–362. https://doi.org/10.1111/j.1469-8749.2008.02046.x	*	*	c)	**	**	**	*	9	Very good study
Sigurdardottir, S., & Vik, T. (2011). Speech, expressive language, and verbal cognition of preschool children with cerebral palsy in Iceland. *Developmental Medicine and Child Neurology*, *53*(1), 74–80. https://doi.org/10.1111/j.1469-8749.2010.03790.x	*	*	*	**	**	**	*	10	Very good study

Soriano, J. U., & Hustad, K. C. (2021). Speech-Language Profile Groups in School Aged Children with Cerebral Palsy: Nonverbal Cognition, Receptive Language, Speech Intelligibility, and Motor Function. *Developmental Neurorehabilitation*, *24*(2), 118–129. https://doi.org/10.1080/17518423.2020.1858360	___ c)	__c)	b)	**	N/A	**	*	5	Satisfactory study
Mei, C., Reilly, S., Bickerton, M., Mensah, F., Turner, S., Kumaranayagam, D., Pennington, L., Reddihough, D., & Morgan, A. T. (2020). Speech in children with cerebral palsy. *Developmental Medicine and Child Neurology*, *62*(12), 1374–1382. https://doi.org/10.1111/dmcn.14592	*	____ b)	*	**	_____ b)	**	*	7	Good study

Table 2: Quality assessment of the included studies, based on the Newcastle Ottawa Scale (NOS)

Regarding the evaluated language subdomains, all studies assessed receptive language and most all of those also assessed expressive language. Within these two main areas, there was great variability in the specific domains evaluated, from semantics, phonological awareness, articulation, lexical development, and specific skills such as reading and spelling.

As for other relevant domains assessed, it is noteworthy that gross motor skills were assessed in most studies (7/10) using the GMFCS and in 4 of the studies the *Leiter International Performance Scale* and the *Columbia Mental Maturity Scale* were used as measures directed for the assessment of non-verbal cognition.

Regarding the measures used to assess language, the variability is immense, and no common assessment protocol was found among the included studies. In 3 of the articles, the *Peabody Picture Vocabulary Test* was used, which assesses the receptive vocabulary. In 2 of the articles, the *Language Subscale of the Bayley Scales of Infant Development* is used, which also assesses the receptive and expressive domains, particularly semantic but also morphosyntactic development. In most studies, different instruments are used to assess language, fact that may be explained by the great variability within the sample: not all children were verbal and, in these cases, the researchers resorted to specific instruments, leaving others that require verbalization to the verbal children (Table 3).

Study	Language Measures	Assessed domains	Other relevant measures	Main outcomes
Ballester-Plané, J., Laporta-Hoyos, O., Macaya, A., Póo, P., Meléndez-Plumed, M., Toro-Tamargo, E., Gimeno, F., Narberhaus, A., Segarra, D., & Pueyo, R. (2018). Cognitive functioning in dyskinetic cerebral palsy: Its relation to motor function, communication and epilepsy. *European Journal of Paediatric Neurology*, *22*(1), 102–112. https://doi.org/10.1016/j.ejpn.2017.10.006	Screening test of Spanish Grammar Peabody Picture Vocabulary Test Rey Auditory Verbal learning Test Pattern and Verbal Recognition Memory	Receptive Language Verbal Learning Verbal memory	Communication: **CFCS** Functional motor development: **GMFCS**	Children with CP (GMFCS II e III) had lower scores in language tests, compared with TD children. Functional communication (CFCS) was negatively correlated with verbal attention, receptive vocabulary, basic grammar comprehension and visual short term memory. In the CP group, as motor severity increases, more cognitive functions are impaired.
Dahlgren Sandberg, A. (2006). Reading and spelling abilities in children with severe speech impairments and cerebral palsy at 6, 9, and 12 years of age in relation to cognitive development: A longitudinal study. *Developmental Medicine and Child Neurology*, *48*(8), 629–634. https://doi.org/10.1017/S0012162206001344	All the tests used were constructed by the author	Reading Spelling Phonological awareness	N/A	Children with CP and severe speech impairment showed improvement in reading and spelling abilities. Children with CP showed slow progress in phonological abilities.

Geytenbeek, J. J. M., Vermeulen, R. J., Becher, J. G., & Oostrom, K. J. (2015). Comprehension of spoken language in non-speaking children with severe cerebral palsy: An explorative study on associations with motor type and disabilities. *Developmental Medicine and Child Neurology*, *57*(3), 294–300. https://doi.org/10.1111/dmcn.12619	C-BILLT	Receptive language (spoken language comprehension)	N/A	The type of CP, the GMFCS level and the chronological age explain 33% of the variance in the eaw scores (C-BILLT). The dyskinetic type had higher scores than the spastic type.
Geytenbeek, J. J., Oostrom, K. J., Harlaar, L., Becher, J. G., Knol, D. L., Barkhof, F., Pinto, P. S., & Vermeulen, R. J. (2015). Language comprehension in nonspeaking children with severe cerebral palsy: Neuroanatomical substrate? *European Journal of Paediatric Neurology*, *19*(5), 510–520. https://doi.org/10.1016/j.ejpn.2015.06.001	C-BILLT	Receptive language (spoken language comprehension)	N/A	Linear association between spoken language comprehension and white mass reduction in frontal and parieto occipital regions and thinning of the corpus callosum.
Lamônica, D. A. C., Paiva, C. S. T., Abramides, D. V. M., & Biazon, J. L. (2015). Communication skills in individuals with spastic diplegia. *Codas*, *27*(2), 135–141. https://doi.org/10.1590/2317-1782/20152013060	Denver Developmental Screening Test (Language Subescale) Peabody Picture Vocabulary Communicative behavior (observation)	Receptive and expressive language.	Mental age establishment: **Stanford Binet Method.** Functional classification of motor development: **GMFCS**	Lower results in the receptive vocabulary domain, in the CP group. In about 50% of the children with CP, it was also found attention deficits, which is proposed by the authors as a negative factor for language development.

Mei, C., Reilly, S., Bickerton, M., Mensah, F., Turner, S., Kumaranayagam, D., Pennington, L., Reddihough, D., & Morgan, A. T. (2020). Speech in children with cerebral palsy. *Developmental Medicine and Child Neurology*, *62*(12), 1374–1382. https://doi.org/10.1111/dmcn.14592	Diagnostic Evaluation of Articulation and Phonology Communication Symbolic Behaviour Scales** Preschool Language Scale	Articulation Phonology Expressive communication Spoken language comprehension	Functional classification of motor development: **GMFCS**	Delayed or disturbed speech in 83% of the children. In 54%, it was identified articulation problems; in 44%, phonological problemes and 78% of the sample completed the criteria for dysarthria.
Mei, C., Reilly, S., Reddihough, D., Mensah, F., Pennington, L., & Morgan, A. (2016). Language outcomes of children with cerebral palsy aged 5 years and 6 years: A population-based study. *Developmental Medicine and Child Neurology*, *58*(6), 605–611. https://doi.org/10.1111/dmcn.12957	Preschool Language Scale (PLS) Peabody Picture Vocabulary Test 4th Edition	Receptive and expressive language	Functional classification of motor development: **GMFCS** Non verbal cognition: **Columbia Mental Maturity Scale.**	45% of the children showed receptive language impairment and 54% expressive language impairment. Co-ocurring language and cognitive impairment was found in 30% of the participants. GMFCS IV and V positively correlated with language impairment. Sintax tasks scored lower than morphologic ones.

Sigurdardottir, S., Eiriksdottir, A., Gunnarsdottir, E., Meintema, M., Arnadottir, U., & Vik, T. (2008). Cognitive profile in young Icelandic children with cerebral palsy. *Developmental Medicine and Child Neurology*, *50*(5), 357–362. https://doi.org/10.1111/j.1469-8749.2008.02046.x	**WPPSI-R:** full scale/verbal scale **Bayley Scales of Infant Development: language subscale. Reynell Zinkin Developmental Scales:** verbal comprehension and expressive language subscales***	Expressive Language Receptive Language Vocabulary Semantics	**Functional classification:** verbal/non verbal & normal/mild dysarthria/severe dysarthria. **Functional classification for motor development:** GMFCS **Columbia Mental Maturity Scale* Leiter International Performance Scale***	The GMFCS level was negatively correlated with the verbal scale of the WPPSI. The dyskinetic type had the lower scores.
Sigurdardottir, S., & Vik, T. (2011). Speech, expressive language, and verbal cognition of preschool children with cerebral palsy in Iceland. *Developmental Medicine and Child Neurology*, *53*(1), 74–80. https://doi.org/10.1111/j.1469-8749.2010.03790.x	**WPPSI-R:** full scale/verbal scale **Bayley Scales of Infant Development: language subscale. Reynell Zinkin Developmental Scales:** verbal comprehension and expressive language subscales***	Expressive Language Receptive Language Vocabulary Semantics	**Functional classification:** verbal/ non verbal & normal/mild dysarthria/severe dysarthria. **Functional classification for motor development:** GMFCS **Columbia**	25% of the non verbal children were from the dyskinetic type and 67% had quadriplegia. 83 % of the non verbal children were at stages IV and V of the GMFCS. 74% of the verbal children were at stage I of the GMFCS.

			Mental Maturity Scale* **Leiter International Performance Scale***	
Soriano, J. U., & Hustad, K. C. (2021). Speech-Language Profile Groups in School Aged Children with Cerebral Palsy: Nonverbal Cognition, Receptive Language, Speech Intelligibility, and Motor Function. *Developmental Neurorehabilitation*, *24*(2), 118–129. https://doi.org/10.1080/17518423.2020.1858360	TACL-4 - Test of Auditory Comprehension of Language. TOCS+ - Test of Children's Speech Plus.	Comprehension of receptive language Expressive Language (repetition)	**Functional classification for motor development:** GMFCS **Leiter International Performance Scale**	More than 76% of children with speech motor involvement and typical language comprehension scored above the cutoff score for cognitive impairment **VS** 80% of children with speech motor involvement and language comprehension impairment scored below the cutoff for cognitive impairment.

Table 3: Summary of the instruments, assessed domains and main results of the included studies.

Below it’s presented a brief summary of each of the articles included , based on the Cochrane domains (Higgins & Thomas, 2021):

Ballester-Plané, J., Laporta-Hoyos, O., Macaya, A., Póo, P., Meléndez-Plumed, M., Toro-Tamargo, E., Gimeno, F., Narberhaus, A., Segarra, D., & Pueyo, R. (2018). Cognitive functioning in dyskinetic cerebral palsy: Its relation to motor function, communication and epilepsy. European Journal of Paediatric Neurology, 22(1), 102–112. https://doi.org/10.1016/j.ejpn.2017.10.006

Participants and inclusion criteria: 52 cases of CP with dyskinetic CP and 52 TD people matched by age and gender were recruited for the control group. Of these 52 cases, only 18 were children, with ages ranging from 7 to 17 years old. The inclusion criteria were a) Clinical diagnosis of CP with mainly dyskinetic features; b) age over six years old; c) presence of an inteligible yes/no response system; d) being able to understand simple instructions as assessed by the Screening Test of Spanish Grammar (receptive language).

Assessment protocol:

a) Motor status: the authors used the GMFCS to assess lower limb function; the BFMF and the MACS for the upper limbs.
b) Communication: the performance on daily communication was assessed through the CFCS.
c) Epilepsy: the authors used the International League Against Epilepsy criteria, classifying it as active epilepsy, resolved epilepsy or non-epilepsy.
d) *Cognitive and Language assessment:* the authors used *Raven’s Colored Progressive Matrices* to assess inteligence; *Spatial and Digit Span subtests on the Wechsler scales* to assess visual and verbal attention; *Benton’s Judgement of Line Orientation Test and Facial Recognition Test* to assess visuospatial and visuoperceptual abilities; *Peabody Picture Vocabulary Test III* to assess receptive vocabular; the *Screening Test of Spanish Grammar* to assess basic gramar comprehension; the

Rey Auditory Verbal Learning Test to assess verbal learning; the *Pattern and Verbal Recognition Memory subtest of the Cambridge Neuropsychological Test Automated Battery* to assess visual and verbal memory and the *Wisconsin Card Sorting Test-64* to assess cognitive flexibility.

Outcomes and results:

- All participants with dyskinetic CP showed a poorer performance than the control group in all cognitive functions.
- Participants with a moderate motor impairment (GMFCS II-III) had lower scores on language-related tests (receptive vocabulary ($p= 0{,}004$); verbal learning ($p= <0{,}001$) and basic gramar comprehension ($p= <0{,}001$).
- Participants with a severe motor impairment (GMFCS IV-V) had lower scores than controls in all cognitive and language tasks, except for verbal memory. ($p= 0{,}062$)
- Communication (CFCS) was negatively correlated with performance in verbal attention ($r_s= -0{,}38$), receptive vocabulary ($r_s = -0{,}4$) , basic grammar comprehension ($r_s = -0{,}41$) and visual short term memory ($r_s = -0{,}29$).
- The presence of epilepsy was associated with poorer performance in intelligence, visuospatial abilities ($r_s= -0{,}51$), basic grammar comprehension ($r_s= -0{,}37$), verbal learning ($r_s=-0{,}35$).

Key conclusions:

- As motor severity increases, more cognitive functions are impaired compared to healthy controls.
- Subjects at any GMFCS level performed significantly worse than healthy controls in attention, visuoperception and visual memory, which can be due to the involvement of basa ganglia and thalamic functional systems.

- The poorer daily communication was associated with lower performance mainly in receptive language-related functions, which can be explained by the fact that motor difficulties that inhibit speech ability in dyskinetic CP affect the frequency and the quality of the communicative acts.

Dahlgren Sandberg, A. (2006). Reading and spelling abilities in children with severe speech impairments and cerebral palsy at 6, 9, and 12 years of age in relation to cognitive development: A longitudinal study. Developmental Medicine and Child Neurology, 48(8), 629–634. https://doi.org/10.1017/S0012162206001344

Participants and inclusion criteria: Six children (one male; five females) diagnosed with CP and severe speech impairments (SSPI) and six TD children, matched by age and IQ with the children of the EG. All the participants used the *Bliss System*[1] as their primary communication mode, since none of them could produce any intelligible speech.

Assessment protocol: all the tests used were constructed by the author, including tasks of phonological awareness, spelling and Reading. The author also used the *Digit Span* test and the *Corsi Blocks*, in order to establish memory indicators.

Outcomes and results: This is a longitudinal study, focused on understanding the development of literacy skills and in the three times where children were assessed, there was an improvement in the EG, in all reading and spelling tasks. In terms of phonological awareness, there was no improvement in the EG, in any of the tasks. On the other hand, in the CG, all the participants improved from time 1 to time 3. At last, the EG didn't improve aswell in the auditory or in visuospatial short term memory tasks.

[1] **The Bliss System** is a semantic graphical language used by individuals with severe speech and physical impairments: https://www.blissymbolics.org/

Key conclusions: The results imply that children with CP and with SSPI have problems developping fluent reading and spelling skills, however it's important to highlight that the children with CP didn't show any reading or spelling skills at preschool age, but they improved consistently from preschool to school aging.

Geytenbeek, J. J. M., Vermeulen, R. J., Becher, J. G., & Oostrom, K. J. (2015). Comprehension of spoken language in non-speaking children with severe cerebral palsy: An explorative study on associations with motor type and disabilities. Developmental Medicine and Child Neurology, 57(3), 294–300. https://doi.org/10.1111/dmcn.12619

Participants and inclusion criteria: 87 children with CP, with a GMFCS level IV or V, with a productive spoken vocabular of fewer than five words; without a diagnosis of auditory problems.

Assessment protocol: the children were assessed with the C-BiLLT, a computer-based instrument for low motor language testing[2], that provides information on the child's ability to understand spoken sentences, which are accompanied by the correspondent images.

Outcomes and results: the authors found a significant positive correlation between age and C-BiLLT raw score, aswell better scores among the children with dyskinetic CP, compared with the ones from the spastic group.

The children from the level IV of GMFCS also had higher scores than the ones from the level V.

Key conclusions: This study confirmed that children with CP, particularly the ones in the most severe levels of GMFCS, show a significantly delayed level of comprehension of spoken language, when compared with tipically developped children[3]. The finding, regarding the diferences between the CP subtype is in line with previous studies, where the language understanding was less affected

[2] https://c-billt.com/about-c-billt/

[3] There are no data available regarding the comparison with tipically developped children.

among the children with dyskinetic CP, in relation to the ones of the spastic subtype (p= 0,035). The previous data supporting this study shows that the underlying brains lesions affect not only the motor impairment level, but also the language abilities and highlight the importance of clearly distinguish the assessment of receptive and expressive language.

Geytenbeek, J. J., Oostrom, K. J., Harlaar, L., Becher, J. G., Knol, D. L., Barkhof, F., Pinto, P. S., & Vermeulen, R. J. (2015). Language comprehension in nonspeaking children with severe cerebral palsy: Neuroanatomical substrate? European Journal of Paediatric Neurology, 19(5), 510–520. https://doi.org/10.1016/j.ejpn.2015.06.001

Participants and inclusion criteria: 80 children with severe CP, aged between 1 and a half and 12 years old; with a GMFCS level IV or V, with a productive spoken vocabular of fewer than five words/severely disarthric; without a diagnosis of auditory, visual, metabolic or any neuromuscular problems. It was mandatory to have brain imaging exams.

Assessment protocol: the children were assessed with the C-BiLLT, a computer-based instrument for low motor language testing[4], that provides information on the child's ability to understand spoken sentences,that are accompanied by the corespondent images.
Regarding the MRI imaging, it was classified according to 4 main patterns: a) Periventricular Leukomalacia (PVL); b) Basal Ganglia Necrosis (BGN); c) Brain malformations due to, for example, viral infections; d) Miscellaneous findings.

Outcomes and results: the C-BiLLT scores were better for children with the pattern of BGN, then the ones from the other 3 patterns *(p=0,013).* Among the group of PVL and brain malformation, the language performances were moderately to severely delayed in 100% of the participants.

[4] https://c-billt.com/about-c-billt/

The authors didn't find association between spoken language comprehension scores and cortical or subcortical grey mass brain lesions.

Lower C-BiLLT scores were found among the children with cerebellum abnormalities, when compared with the ones without *(p= 0,05).*

For all the patterns, the authors found a significant linear association between spoken language comprehension performance and white mass reduction in the frontal area, in the parieto-occipital area and the thinning of the corpus callosum.

Key conclusions: Generally, children with a BGN pattern scored better than children with a PVL one. The findings of this study emphasise the consistent association between white mass lesions and neurodevelopmental outcomes.
Diffuse thining of corpus callosum and diffuse reduction of the frontal and parieto-occipital white mass areas showed a clear correspondance with poor spoken language comprehension, which stresses the value of inter-to intrahemispheric connectivity in what concerns language development.

Lamônica, D. A. C., Paiva, C. S. T., Abramides, D. V. M., & Biazon, J. L. (2015). Communication skills in individuals with spastic diplegia. Codas, 27(2), 135–141. https://doi.org/10.1590/2317-1782/20152013060

Participants and inclusion criteria: 10 children with spastic CP and diplegia and 10 TD children, matched by age and IQ score. The chronological age ranged from 44 to 83 months and children with sensorial, auditory or visual impairments were excluded, aswell children with an IQ below 70 points.

Assessment protocol:
Communication skills (indirect measure): *MacArthur Development Inventory on Communicative Skills* is an instrument applied to the parents in order to assess receptive and expressive vocabular.

Communicative Behavior: it was analyzed through a qualitative method, through 30 to 40 minutes of footage of the participants performing interactive activities. The answers produced by the children were analyzed according to 26 communicative categories.

Receptive Vocabulary: *Vocabulary Test by Peabody Picture* (VTPP).

Language: it was assessed through the *Denver Developmental Screening Test II (DDST-II),* which also evaluates personal-social skills; fine-adaptative motor skill and Gross Motor Skill.

Outcomes and results: in all the instruments used to evaluate language, the authors didn't find significant differences between groups (*receptive vocabulary: p = 0,105; expressive vocabulary: p = 0,295; language: p = 0,854*).

Key conclusions: Children with CP are not an homogeneous group, which makes very challenging the attempt to draw a generic linguistic profile. It is crucial to start considering subgroups according to the GMFCS and the brain lesion.

When assesing language skills in children with CP, it's very important take into account that there is a high prevalence of attention impairments in these individuals, which obviously has an impact in verbal learning.

Mei, C., Reilly, S., Bickerton, M., Mensah, F., Turner, S., Kumaranayagam, D., Pennington, L., Reddihough, D., & Morgan, A. T. (2020). Speech in children with cerebral palsy. Developmental Medicine and Child Neurology, 62(12), 1374–1382. https://doi.org/10.1111/dmcn.14592

Participants and inclusion criteria: 84 children diagnosed with CP, verbal or minimally verbal with ages ranging from 4 years/11 months to 6 years/6 months. More than a half of the recruited children (57%) were at levels I and II of the GMFCS; the dyskinetic and the ataxic subtype represented only 2% of the sample.

Assessment protocol:

Speech classification: a) age appropriate speech development; b) articulation delay or disorder; c) phonological delay or disorder; d) dysarthria; e) childhood apraxia of speech (CAS).

Dysarthria and CAS: through a 10 minutes sample conversation, the authors used the modified version of the *Mayo Clinic dysarthria classification system* and the diagnostic criteria for CAS were adapted from previous research, based on the American Speech Language Hearing Association.

Phonological assessment: the authours made this evaluation through the *Percentage consonants correct* (PCC), a scale that varied from: a) speech is unintelligible; b) only isolated words or phrases are intelligible; c) half of the message is understood; d) speech is intelligible with some exceptions; e) speech is completely intelligible.

Oromotor function: the authors used the *Verbal Motor Production Assessment for Children* and this scale was administered only to the verbal participants. The purpose was to distinguish between dysarthria and CAS.

Gross and fine motor functions: the authors used the GMFCS and the MACS.

Language: the authors used the *Preschool Language Scale*, 4th Edition, which evaluates verbal comprehension and expressive language.

Cognition: the authors used the *Columbia Mental Maturity Scale* as a cognitive development measure.

Outcomes and results: 82% of the participants showed a delayed or disordered speech. In the group of the verbal participants, articulation was age appropriate in 46%. The most frequently absent consonants were fricatives and, in the non verbal group, only half was able to produce some consonants (*Mean = 2.6*).

The phonological development was age appropriate in 57%, being important to highligh that 72% of this sample was in the levels I-III from the GMFCS.

Dysarthria was seen in 78% of the participants, but it had a mild presentation in 53% of the cases. Among verbal participants, 17% met criteria for CAS and the speech delay/disorder was positively associated with language impairment.

Key conclusions: The authors concluded that speech delay/disorder is independente from cognition and that it represented a huge challenge try to distiguish dysarthria from CAS, since there are various common criteria. In line with previous studies, it was concluded that speech disorder/delay was always present in all the participants of the non spastic subtypes and of the GMFCS V.

Mei, C., Reilly, S., Reddihough, D., Mensah, F., Pennington, L., & Morgan, A. (2016). Language outcomes of children with cerebral palsy aged 5 years and 6 years: A population-based study. Developmental Medicine and Child Neurology, 58(6), 605–611. https://doi.org/10.1111/dmcn.12957

Participants and inclusion criteria: 84 children, diagnosed with CP from the Victorian Cerebral Palsy Register, with ages ranging from 5 to 6 years old.

Assessment protocol:

Receptive and Expressive Language: the authors used the Preschool Language Scale. When the participants were not fatigued, the authors also administered the Peabody Picture Vocabulary Test. For the non verbal participants, the authors used the Communication and Symbolic Bejaviour Scales-Developmental Profile Caregiver Questionnaire.

Non verbal cognition: Columbia Mental Maturity Scale.

Motor function: GMFCS.

Outcomes and results: receptive and/or expressive language impairment was identified in 61% of the entire sample (verbal and non verbal children). Mixed receptive-expressive language impairments occurred in 44% of the children. Cognitive impairment was associated with an increased *odds ratio* of language impairment. GMFCS level IV and V were associated with language impairment. Among the verbal participants, the ones with language impairment had receptive semantic deficits. The comprehension of syntactic structures was more often affected than for the morphological aspects.

Key conclusions: the vocabulary and syntax abilities are below average, which is consistent with previous research, and highlights the importance of cognition for language development. It is however important to deepen the understanding of what cognitive components are central for language.

Sigurdardottir, S., Eiriksdottir, A., Gunnarsdottir, E., Meintema, M., Arnadottir, U., & Vik, T. (2008). Cognitive profile in young Icelandic children with cerebral palsy. Developmental Medicine and Child Neurology, 50(5), 357–362. https://doi.org/10.1111/j.1469-8749.2008.02046.x

Participants and inclusion criteria: 127 children from the State Diagnostic and Counselling Centre (SDCC), of Kopavogur in Iceland.

Assessment protocol:

Language: the *Verbal Scale of the Wechsler Preschool and Primary Scale of Intelligence* (WPPSI) was used. When children were not able to complete this scale, there were other alternative instruments presented below.

Cognition: the full scale of *Wechsler Preschool and Primary Scale of Intelligence (WPPSI)* OR the *Bayley Scales of Infant Development* OR the *Columbia Mental Maturity Scale* OR the *Leiter International Performance Scale* OR the *Reynell Zynkin Developmental Scales* OR the *Test of Nonverbal intelligence (TONI-2)*.

Outcomes and results: Children from the ataxic subtype showed lower verbal scores thant the ones from the other subtypes (*Median = 72.5*). On the other hand, children from the spastic type and the ones with quadriplegia had better scores in the verbal scale than in the performance one[5].
Within the groups with a more severe motor impairment, there was found a lower median in all WPPSI subscales.

Key conclusions: Particular attention should be given in what concerns the choice of the assessment protocols. An early and properly conducted evaluation can be decisive for the intervention planning.

Sigurdardottir, S., & Vik, T. (2011). Speech, expressive language, and verbal cognition of preschool children with cerebral palsy in Iceland. Developmental Medicine and Child Neurology, 53(1), 74–80. https://doi.org/10.1111/j.1469-8749.2010.03790.x

Participants and inclusion criteria: 152 children with a diagnosis CP, comprising the total of children born in Iceland between 1989 and 2004.

Assessment protocol:
Language: the *Verbal Scale of the Wechsler Preschool and Primary Scale of Intelligence* (WPPSI) was used. When children were not able to complete this scale, there were other alternative instruments presented below.
Speech: the authors classified through a functional 3 levels system, based on direct observation of children, a) normal speech; b) mild dysarthria; c) severe dysarthria.

Cognition: the full scale of *Wechsler Preschool and Primary Scale of Intelligence* (WPPSI) OR the *Bayley Scales of Infant Development* OR the *Columbia Mental Maturity Scale* OR the *Leiter International Performance Scale* OR the *Reynell Zynkin Developmental Scales* OR the *Test of Nonverbal intelligence* (TONI-2).

[5] There are no p-values available, regarding the comparison among subtype CP groups

Results and Outcomes: 84% were verbal communicators. Among the non verbal communicators, 67% had quadriplegia and 25% had dyskinetic CP. On the other hand, among the verbal group, only 15% had quadriplegia and 10% were from the dyskinetic type. Most of the non verbal children had severe intellectual impairments. Children with severe dysarthria were more likely to be unable to walk and to be of the dyskinetic CP subtype (*p = 0,02; p = <0,01*). The verbal subescale of the WPPSI-R showed better results within the group of hemiplegia and diplegia, when compared with the dyskinetic and ataxyc subtype (*p = 0,018*).

Key conclusions: Both expressive language function and speech status were highly associated with gross motor function, CP subtype and cognitive functioning. Severe dysarthria should be well distinguished from severe cognitive impairment, since 25% of that group revealed normal or borderline level (WPPSI-R). Almost half of the total group had verbal intelligence scores in the normal range, being the vocabulary and similarities subtests the ones with better results.

Soriano, J. U., & Hustad, K. C. (2021). Speech-Language Profile Groups in School Aged Children with Cerebral Palsy: Nonverbal Cognition, Receptive Language, Speech Intelligibility, and Motor Function. Developmental Neurorehabilitation, 24(2), 118–129. https://doi.org/10.1080/17518423.2020.1858360

Participants and inclusion criteria: 1) have a medical diagnosis of CP, 2) have hearing within normal limits according to a distortion product otoacoustic emission screening, and 3) have no co-occurring diagnosis of autism spectrum disorder. In the present study, additional inclusion criteria required that the children 4) be 10–12 years of age. Twenty-seven children met the inclusion criteria.

Assessment protocol:

Receptive language: the authors used the *Test of Auditory Comprehension of Language* (TACL-4), which includes 3 subtests: Vocabulary, Grammatical Morphemes and Elaborated phrases and sentences.

Non verbal cognition: the authors used the *Leiter International Performance Scale* (Leiter-R).

Speech: the authors used the *Test of Children's Speech Plus* (TOCS+), which involves the repetition of individual words and sets of utterance.

Outcomes and results: Better non verbal abilities were strongly correlated with better receptive language ($r = 0.75$, $p = <0{,}001$). The majority of the participants with typical language development, independently of their speech motor involvement *status* (with or without) scored above the cutoff score for cognitive impairment. On the contrary, the majority of children wiith language comprehension impairment and speech impairment scored below the cutoff score for cognitive impairment ($p = <0{,}001$; $p = 0{,}003$).

Key conclusions: In general, the findings of this study contribute to the already existing evidence that the level of GMFCS is associated with language, speech and intellectual abilities in children with CP.

The fact that non verbal abilities worked as a kind of predictor for linguistic subdomais, such as syntax and vocabular was a particularly valuable finding, in what concerns the possibility of using more straightforward measures to assess children with CP, especially those with severe motor impairments.

5. Discussion

The main research question of this Systematic Review is if there is an influence of cerebral palsy in language skills, being the main outcome the language status in children with cerebral palsy and the main comparator the language status in children with typical development.

In general, considering the results of all articles included in this systematic review, the answer to this question seems to be positive, although the enormous variability of the results, the specificities of each case are notorious, as already presented by previous authors (Andersen & Trust, 2010; Critten et al., 2019; Hustad, Katherine; Oakes et al., 2015; Lipscombe et al., 2016).

The idea proposed by the new definition for CP seems to be in line with the results found in this SR, reinforcing that the motor disorders are often accompanied by disturbances of sensation, cognition, communication, perception, and/or behavior, and /or by epilepsy (Rosenbaum, 2014).

Also, in the studies where the CP children were compared with TD children, although there were significant variability among subdomains, the performance was, in general, poorer, which seems to correspond to the data found in population-based studies, where learning difficulties were described in 45% of children; communication difficulties in 49% and total absence of speech in 30%; (Himmelman & Uvebrant, 2011) and another one that highlights the presence of language impairments in ¼ of children with CP (Fluss & Lidzba, 2020).

Specific questions:

a) Are there differences between the language skills, receptive and expressive, of children with cerebral palsy and those of children with typical development?

1. If there are differences, which linguistic domains are most affected?

Among the included studies, there were only two with a case-control design, comparing CP children with TD children (Ballester-Plané et al., 2018; Lamônica et al., 2015), and in both, it is important to highlight that the recruited sample was very specific and it wasn't representative of all the pediatric population living with CP. In the first one, the CP participants were diagnosed with one specific subtype of CP, the dyskinetic one, and the sample also included adults, which demands a careful look to the conclusions (Ballester-Plané et al., 2018). This information is important for several reasons, one of them is related to the neurodevelopmental aspect, it being clear that brain plasticity mechanisms do not act or remain the same way between childhood and adulthood (Novak & Spirit-jones, 2017). On the other hand, the subtype specifically analyzed by these authors has been already associated in previous research with more severe clinical conditions, given the lesions in the basal ganglia and the cortical structures to which they are connected (Himmelman & Uvebrant, 2011).

In this sense, the analysis of the results and their comparison with the TD group participants must be done with special care.

In the second study mentioned above, (Lamônica et al., 2015) the participants are children diagnosed with spastic-type CP, and with diplegia, a definition that no longer seems to be consensual among the authors, remaining the recommendation for the use of the designation hemiplegia, in the sense of identifying whether individuals with CP have both, or just one side of the body affected (Fairhurst, 2012).

Regarding the spastic subtype, a previous study shows impairments in receptive language in 85% of these children, thus calling attention to beware of generalizations to all other CP subtypes and, particularly, their respective levels of functionality (Vendrell & Narberhaus, 2009).

In the study that evaluated dyskinetic subtype participants, the results point to a poorer performance in all tasks related to language, with the exception of verbal memory, when compared to the CG. The number of affected domains increases as the degree of motor functionality also increases, and this study ensured the inclusion of participants from all degrees of motor functionality (GMFCS). This idea of severity of the cognitive impairment associated with the severity of the motor impairment has been widely defended by several authors, although it is discussed whether the degree of GMFCS should be understood as a predictor of cognitive status or just as a mirror of the extension and location of the lesion (Chen et al., 2013; Horber et al., 2020). Also, in another study, it was found that 67% of individuals with lesions in the basal ganglia were at levels IV and V of the GMFCS (Himmelman & Uvebrant, 2011), with this brain area being activated, either in performing motor or linguistic tasks (Houwen et al., 2016).

Despite this, another study showed that in bilateral spastic CP, cognitive impairments increased concomitantly with the degree of GMFCS, an association that has not been described in the dyskinetic subtype group (Horber et al., 2020).

In neuroanatomical terms, it has been consistently shown that white matter lesions are strongly associated with poorer neurodevelopmental prognoses, namely CP (Iwata et al., 2012; Smithers-Sheedy et al., 2014; Vaillant et al., 2020). Interestingly, although the studies mentioned above point to more severe conditions in the group of the dyskinetic subtype, a study that analyzed brain images of children with CP found that, in language tasks, individuals with basal ganglia necrosis, an area usually affected in dyskinetic type CP, presented better results in language tasks when compared to those who had periventricular lesions (white matter) (Geytenbeek et al., 2015).

On the other hand, in the case control study that analyzed children from the spastic subtype, with diplegia, (Lamônica et al., 2015), no significant differences were found between the EG and the CG in any of the *Denver Developmental Screening Test II subscales*, including in Language. Regarding the results in the assessment by the *Vocabulary Test by Peabody Picture*, no statistically significant differences were found either, although the authors highlight a greater variability of the results within the group of children with CP. It is important to note that the sample in this study, in addition to being small (10 CP participants + 10 TD participants), did not include participants from all levels of the GMFCS, with 70% of the sample being level II and the rest being level III.

Through a closer look at the results of this study, it appears that, although the differences between the groups are not statistically significant, there is a considerable disparity between the minimum and maximum values obtained[6], which accentuates the need to reflect on whether the diagnosis of CP is, in itself, sufficient to consider a profile of language skills, or if, on the other hand, it is essential to associate the diagnosis with specific assessment protocols, which characterize each of the cases "caught" by the clinical diversity that this concept seems to encompass.

b) Is the GMFCS level related to the development of language skills?

The discussion around the interaction between motor skills and language has been developed by different authors who highlight anatomical aspects to support this interaction, among which the activation of Broca's area, during the performance of motor tasks, as well as the importance of the suplementar motor area for language processing (Courson & Tremblay, 2017). Studies of a different nature also present motor development as a predictor of children's performance in school-related tasks, such as reading and arithmetic, as well as an association between articulatory skills and working memory of children with CP, calling for attention to the fact that not only linguistic aspects play an important role in language acquisition by these children, but also other cognitive dimensions that need to be associated with it for this process to be successful (Peeters et al., 2009).

[6] On the DDCM Expressive Vocabulary subscale, the minimum value of the CG is 90.2, while that of the EG is only 56.2. In the assessment of Receptive Vocabulary by the VTPP, the minimum value of the CG is 4, which corresponds to the lower high interval, while in the EG it is 2, corresponding to the upper low interval.

Regarding the studies included in this systematic review, half of those clearly analyzed the association of the motor function level (assessed through the GMFCS) and language's development. In all of those, it was confirmed that, as the level of the GMFCS increases, the language impairments also increase (Ballester-Plané et al., 2018; Geytenbeek et al., 2015; Mei et al., 2016; Sigurdardottir et al., 2008; Sigurdardottir & Vik, 2011)), evidence that seems to be in line with previous studies which demonstrated that speech problems were more prevalent in the group of children from GMFCS IV and V (Himmelmann et al., 2013; Himmelmann & Uvebrant, 2011; Virella et al., 2016).
However, it is worth analyzing these data more carefully, as it is particularly challenging to find studies that, on the one hand, focus their assessment on language as a cognitive function, and not just on its expressive modality (speech) and, on the other hand, the great disparity in assessment protocols and sample characteristics also raises, namely with regard to the age range of the included participants, important challenges when it comes to potential generalizations.

One of the included studies focuses the investigation on one of the CP subtypes, which, regardless of the level of GMFCS associated with it, has its own characteristics, namely with regard to the anatomy of the lesion, often associated with the basal ganglia (Ballester- Plané et al., 2018; Surveillance of Cerebral Palsy IN Europe, nd). In two other studies, the sample focuses on children without speech or with severely affected speech and classified in levels IV and V of the GMFCS, which seems to correspond to the idea advocated by other studies, however, leaving out the possibility of analyzing the language situation in children located at other levels of the GMFCS (Dahlgren Sandberg, 2006; Geytenbeek et al., 2015). In each of the studies, very different skills are assessed, with the first proposing the assessment of reading and, particularly, spelling skills, both associated with the acquisition of phonological awareness (Binder et al., 2009; Estil et al., 2003) and, in the case of the second, the assessment of receptive language (comprehension of spoken language), thus promoting the involvement of interconnected but distinct brain areas.

A meta-analysis of fMRI studies (Binder et al., 2009) showed that in the context of language processing, in particular semantics, there is a predominance of the left hemisphere, although this is discussed by several authors, in a complex connection between the parietal, temporal, and frontal lobes, all of which play an essential role in heteromodal and integrative processes.

One of the areas highly involved seems to be the angular gyrus, located in the parietal lobe region, with associations of lesions in this area with dyslexia, dysgraphia, impairments in sentence comprehension, dyscalculia and ideomotor apraxia (Binder et al., 2009; Straube; et al., 2012). In another one of the studies, difficulties in integrative aspects of receptive language, which involve the ability to categorize, organize and interpret, were affected in almost all participants, regardless of their GMFCS level, which denotes the importance of combining a look at the nature, extent and location of the injury and not just the severity of motor functionality (Mei et al., 2016).

On the other hand, the inferior frontal gyrus seems to be more involved in aspects related to phonological awareness, working memory, syntactic processing and motor planning, corresponding to a crucial interaction region for motor behavior and aspects of language production as is the case of Broca's area. Thus, in this case, lesions in these areas, which may be common in individuals with a CP diagnosis, a primarily motor condition, will probably affect the domains of expressive language and specific tasks such as reading (Peeters et al., 2009). This complexity reinforces the importance of ensuring specific assessment protocols that clearly reflect on the constructs being assessed, so that the intervention goals may be effectively adjusted to the needs of each case.

In another study, differences in performance on language tasks (*Verbal Scale: WPPSI-R*) were found between GMFCS level I and all other levels, with the first group showing median performances and all other levels in the lower middle range (Sigurdardottir et al., 2008). Although the II-V levels were all in the lower mean range, it is important to emphasize that the minimum and maximum values for group IV-V were significantly lower than those for group II-III, as well as the distribution of the sample, with approximately 5 times more participants in group II-III than in group IV-V, which can bias the results.

In another study, conducted by the same authors (Sigurdardottir & Vik, 2011), expressive language was shown to be strongly associated with the level of GMFCS, however, the same did not happen with verbal cognition, which was between the medium and medium low range in all GMFCS groups. Similar to what had already been postulated by previous studies, the investigation in the field of CP seems to be strongly biased by the domains of expressive language, limiting the assessment of language to a functional classification that takes little account of the linguistic potential of these children at other levels, stressing the need to clearly distinguish articulatory problems with language impairments, as a cognitive function itself (Sigurdardottir & Vik, 2011).

In the case-control study, which evaluated individuals with dyskinetic CP (Ballester-Plané et al., 2018), the ones with higher levels of GMFCS have shown to be positively associated with increased cognitive impairments, not only in language domains, but also from other cognitive functions such as memory, attention and visuoperceptive skills. Once again, the apparently simple association between the level of motor function and the development of other cognitive functions hides the characteristics of the lesion, which, the more diffuse it is, not only causes a worse motor prognosis, but also a cognitive one (Ballester -Plané et al., 2018; Iwata et al., 2012).

It is noteworthy that, in this same study, regardless of the degree of GMFCS, all participants had a lower performance compared to the control group in cognitive domains fundamental to language development, such as attention, the visual-perceptive domain and memory. In previous studies, in TD individuals, growing evidence had already demonstrated about the important role of the basal ganglia, not only in motor tasks, but also in cognitive ones, namely those that require verbal skills (Leisman et al., 2014) and that demand the activation of the working memory (Arsalidou et al., 2013), which is of unquestionable importance in the language development process.

*“Nothing in biology makes sense except in the light of evolution” (*Theodosius Dobzhansky *cited in* Dilley, 2019)*.*

This quote by Ernst Meyr is particularly relevant in the context of the role of the basal ganglia, since these structures fully recruited for aspects of motor control, in our ancestral past, would have been, throughout evolution, implicated in cognitive skills and, in particular, in language.

In another cohort study, involving preterm infants, a positive association between white matter lesions and increased cognitive impairments, as well as the incidence of CP, was demonstrated (Iwata et al., 2012). White matter, made up mostly of axonal tissue, is responsible for connecting all the cortical areas: the gray matter. According to these studies, a failure in these circuits contributes to a worse cognitive outcome than an injury in gray matter areas (Iwata et al., 2012; Woodward et al., 2012).

In the case of cerebral palsy, this fact is relevant because, on the one hand, it is known that this diagnosis often has underlying PVL conditions, which are predictors of a worse prognosis, namely, in terms of language (Geytenbeek et al., 2015) and, on the other hand, because gray matter is more permeable to external aspects, such as stimulation and also to the mechanisms of neuroplasticity itself (Iwata et al., 2012). Early knowledge of the nature of injuries may contribute to the design of more adjusted and comprehensive interventions, which combine motor development goals with those in the areas of cognition and language.

d) Is the cerebral palsy subtype related to the development of language skills?

None of the studies included in this systematic review carried out a comparative analysis between the CP subtypes, including those that focused only on one of these groups and, moreover, none of the studies makes a particular analysis of the language outcomes of children of the ataxic subtype, even though it is the less prevalent one (Cans, 2000).

Previous studies suggest that children of the dyskinetic subtype, as well as those of the bilateral spastic subtype, have a worse prognosis regarding language acquisition and development, when compared to the others (Novak & Spirit-jones, 2017)

This also appears to be in line with the previous research question, as it is these subtypes that seem to be most associated with the most severe levels of GMFCS.

In one of the included studies (Geytenbeek et al., 2015), consisting only of children of the dyskinetic and spastic (bilateral) subtypes, with GMFCS levels IV-V, those in the first group had better results in the receptive language test (spoken language comprehension) than those of the second, which can be understood in line with the underlying lesion, since children of the bilateral spastic type may have lesions that are more diffuse at the cortical level, namely, PVL conditions that compromise the areas most associated with semantics and verbal comprehension (Binder et al., 2009; Iwata et al., 2012). On the other hand, children in the dyskinetic group, also non-verbal, may have an underlying lesion that is more focal, which compromises them in terms of motor planning and, consequently, of speech, but which has preserved the areas associated with the processing of semantics and comprehension.

With regard to the ataxic subtype, it is not only the least common group, but also the one in which, at age 5, an alteration in the diagnosis is most frequently seen, as it is confused with other ataxias of a genetic and non-progressive nature. In terms of neuroimaging, this subtype is also the most difficult to identify, since it is common not to identify injured areas, but only hypoplasia or slight malformations of the cerebellum (Dan, 2020). The low prevalence of this subtype, together with the little information obtained through neuroimaging exams, has contributed to a lack of knowledge about the specific characteristics of language development, as well as other cognitive dimensions, within this group. Only two of the included studies found information broken down by CP subtype, which demonstrated that children of the ataxic subtype were in the lower range, compared to the average, in terms of verbal cognition, with values similar to those presented by the dyskinetic group and below those presented by those of the spastic subtype (Sigurdardottir et al., 2008; Sigurdardottir & Vik, 2011). Once again, it should be noted that this subtype represented only 4% of the sample in the studies mentioned above, with no presentation of joint data between the subtype and the level of GMFCS, leaving other characteristics of functioning of these individuals to be investigated.

In all the studies in which data are available on the CP subtype, and in particular, in which the spastic subtype is divided into hemiplegia and diplegia, the first subgroup is the one with the best results in language assessment. Usually, hemiplegia results from lesions in the pyramidal pathways, the major motor pathways involved in the control of voluntary movements (Iloeje & Ogoke, 2017; M. I. Shevell et al., 2009). These descending pathways directly involved in motor behavior do not seem to be involved in language functions, or rather, in verbal cognition, and may only have an impact on the articulatory level of the muscles of the mouth and larynx. Furthermore, hemiplegia results from an injury in only one of the hemispheres, which, despite its impact, at an early age, can still be compensated by mechanisms of neuronal plasticity and by extrinsic factors of early intervention (Fairhurst, 2012; Kułak et al., 2011; Masoud et al., 2017).

In this sense, contrary to what seems to be verified in the case of the degree of GMFCS, the subtype of CP does not establish such a clear relationship with the development of language, in line with what several authors have defended. CP, as a neurodevelopmental disorder, implies manifestations that seem to be part of a very broad spectrum, which, even within the different subtypes, presents great variability.

6. Conclusion

The present systematic review clearly points to the need to rethink language assessment protocols, in order to adapt them to the enormous spectrum that cerebral palsy seems to encompass. In fact, with the exception of one of the studies (Mei et al., 2020), they all analyze language, based on functional aspects, especially expressive language and, in circumstances where there is a distinction, this is limited to receptive and expressive modalities, it is never clear whether there are differences, for example, between the domains of semantics, morphosyntax, phonology, etc. Only in Mei's study (2020), within the discursive modality, specific data on aspects such as prosody, loudness, pitch, resonance etc. are presented, which are altered in an important part of the sample of this study, with emphasis on: altered pitch (73%), harsh voice (73%), reduced speech rate (86%), consonants imprecisely articulated (96%) and prolonged phonemes (78%). The same study shows that the comprehension of syntactic structures was more often affected than for the morphological aspects, highlighting the importance of cognition for language development and the need to deepen the understanding of what cognitive components are central for language and how should it be addressed in rehabilitation context.

It should also be noted that the large intragroup variability makes it difficult to carry out a comparative analysis between TD children and CP children, knowing that as the degree of motor functionality increases, as well as in cases of lesions in the white matter (as is the case with periventricular lesions) the amount and severity of associated impairments also seems to increase (Iwata et al., 2012; Smithers-Sheedy et al., 2014; Vaillant et al., 2020).

Nevertheless, the lack of assessment protocols for specific domains of language, among these children, means that most assessments are limited to expressive language, which undoubtedly suffers the consequences of the motor impairment, present in the cases of cerebral palsy (Pirila et al., 2007). Although the GMFCS classification has been established as a simple and useful tool, with regard to anticipating the needs of these children in different areas, it is a matter of great complexity, which also has an underlying brain injury. This means that more diffuse lesions not only cause a more severe motor condition, but also a cognitive one; see, for example, the case of hemiparesis, which generally present lighter motor functioning and more favorable cognitive prognoses (Ballester -Plané et al., 2018; Iwata et al., 2012).

Also from the perspective of the location and extent of the lesion, the studies analyzed still raise some doubts since; on the one hand, bilateral spastic cerebral palsy seems to have associated more severe prognoses, in addition to the dyskinetic subtype. However, in the case of the former, periventricular lesions are frequent, which can affect important portions of white matter, commonly associated with poor neurological prognosis (Geytenbeek et al., 2015), while in cases of dyskinesia, the lesion tends to concentrate in the basal ganglia (grey matter), which are more permeable to external aspects such as stimulation and to the neuroplasticity mechanisms themselves (Iwata et al., 2012). Having said that, with such a distinct origin of lesions being likely, why, in several studies, do these two subtypes appear side by side in terms of underlying severity? A possible explanation may be precisely related to the nature of the assessment protocols, as children in the dyskinetic group, given the nature of their injury, may have impairments in motor planning, which affects their speech, however, it is likely that their areas of understanding and semantic processing are preserved (Binder et al., 2009; Iwata et al., 2012). In this sense, when assessment protocols are aimed almost exclusively at the assessment of expressive language, they may, on the one hand, not be capturing the potential of these children and, on the other hand, it might be a lost opportunity to reflect on more accurate rehabilitation programs, namely with regard to the design of augmentative and alternative communication systems.

From the subtypes point of view, it is difficult to find studies that allow a real comparative analysis, as the distribution between the groups is not uniform: see the case of the ataxic subtype, which always seems to perform poorly (S. Sigurdardottir et al., 2008; Solveig Sigurdardottir & Vik, 2011), however, their representativeness in the sample is also low, being in line with the prevalence data consulted (Bošnjak & Daković, 2013; Surveillance of Cerebral Palsy IN Europe, nd).

In short, this systematic review hopes to contribute to a closer look at the specifics of language assessment in children with cerebral palsy, reaffirming the need to better organize this diverse clinical group and to bring together a joint effort to define neuropsychological assessment protocols, which may be able to respond to the most fragile areas of these children and, consequently, promote an earlier and more effective rehabilitation response.

7. Study Limitations

The main limitations of this systematic review are related, on the one hand, to methodological aspects, namely, the fact that it was not possible to include a larger number of databases, given the constraints of time and resources. And on the other hand, with the constraints regarding the very nature of the research questions. In fact, the study of language in cerebral palsy has traditionally been associated with the study of functionality, which makes it difficult to identify articles that deal with a formal assessment of language as a cognitive function.

Furthermore, it is important to emphasize that the number of articles, which reached the final stage of this systematic review, is quite small so that generalizations can be carried out, especially given the diversity of instruments used to assess language.

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