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Systematic Review
Revisão Sistemática

Effectiveness and Determinant Variables of Augmentative and Alternative Communication Interventions in Cerebral Palsy Patients with Communication Deficit: a Systematic Review

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Keywords

Cerebral Palsy
Communication Skills
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Determinant Variables
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ABSTRACT

Purpose: Assess the effectiveness of augmentative and alternative communication (AAC) interventions in patients with CP and to reveal determinant variables of main intervention outcomes: receptive and expressive language. **Research strategies:** The search was performed in following databases: MEDLINE (Ovid); PubMed (NLM); Embase (Ovid); Cochrane Database of Systematic Reviews; Cumulative Index to Nursing and Allied Health Literature; Database of Abstracts of Reviews of Effects; Cochrane Central Register of Controlled Trials; Health Technology Assessment database and PEDro. **Selection criteria:** Full-text and peer-reviewed studies in English studying the effectiveness of AAC in patients with cerebral palsy were included. Studies with patients (<18 years) diagnosed with CP were included. **Data analysis:** A narrative analysis was conducted to evaluate the efficacy of AAC methods. A random-effects model meta-analysis was used to assess determinants of AAC intervention outcomes. **Results:** The online database and manual reference search revealed 445 records. Nine studies investigating a total of 294 subjects with CP met predefined eligibility criteria: 4 studies with single subject, multiple baseline research designs, 3 longitudinal cohort studies, 1 case control study and 1 case series. Results revealed moderate-quality evidence that AAC interventions improve the receptive and expressive communication skills in patients with CP. The random-effects model meta-analysis revealed the power of identified determinant variables affecting the AAC intervention outcomes. **Conclusion:** Diversity of CP patients requires proper analysis of determinant variables to ensure the efficacy of AAC assessment and intervention. More studies of high methodological and practical quality assessing the efficacy of AAC interventions are needed to clarify the evidence.

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INTRODUCTION

Cerebral palsy is one of the most common neurological conditions leading to physical disability in children. The prevalence of cerebral palsy (CP) has been estimated 2.1 in 1000[1]. CP is traditionally classified based on the motor manifestations of the patient, though variety of conditions are associated with the motor impairments. Among these conditions, the rate of speech, language, and hearing problems is rather high, resulting in a life-long communication deficit[2]. Previous studies had estimated that 30% to 80% of children with CP have communication impairment[3-5]. Communication disorders can limit not only social interaction and community participation but also the educational opportunities of individuals with CP[6-10]. Application of augmentative and alternative communication (AAC) methods in children with cerebral palsy is part of the integrated interventional approach supporting and developing the communication skills in this category of children[11,12]. Limited evidence is accumulated analyzing the efficacy of AAC services for children with CP. Majority of CP patients did not receive intervention at an early age due to misdiagnosis of communication disorders[13-15]. The preliminary findings suggested that AAC application usually targeted the expressive skills of the children with CP. Hustad and Miles[16] studied the speech and language services among pre-school children with CP and revealed that over 50% of the children who needed AAC had AAC-related tasks in their educational process. This evidence requires special attention, as majority of children with CP are likely to have speech and/or language disturbances at a level requiring AAC support[17,18]. At around age 2 when delay in speech development becomes clearly obvious, the opportunities for early intervention to reduce the speech deficit may be missed. Adopting a "wait and see" strategy may prevent the CP patients from accessing to alternative tools of communication[19]. Speech therapists report that introducing AAC to CP patients at an early age is necessary to improve communication and language skills[20]. There are some reports where parents mentioned that they wished the AAC methods had been introduced to their children earlier[21]. The cited studies revealed very limited information on communication variables essential for the proper assessment and intervention in patients with CP[5]. Identification of variables assists the professionals in the process of therapy planning and differentiated intervention. The predicting and moderating variables facilitate assessment and the selection process of intervention tools[22]. These variables do not represent the appropriate outcome measures, but instead provide the therapist with levels of assessed functions, environmental influences and possible factors that can mediate the outcome[22]. Early prediction and identification of variables required for the assessment process are vital to manage communication issues effectively in children with CP[18]. However, little is known about the early predictors and moderators of post-interventional communication performance in children with CP[14,23]. Closing the existing gap between early predictors, moderators, mediators and outcomes of communicational intervention can potentially result in earlier intervention for children with cerebral palsy[24]. This is an essential issue that should be involved in the assessment process of CP patients as communication disorders may persist lifelong.

To go deeper in the mechanism of how a pediatric patient with CP responds to different categories of AAC interventions, we need to look at a scope of interventions and outcomes associated with them. In a review presented by Lund and Light[25] the patient's age, level of intellectual disability and intervention setting were analyzed as potential moderating factors influencing the efficacy of AAC interventions including picture-based systems like PECS and speech generating devices (SGDs). Both categories of AAC methods (PECS and SGDs) proved to be effective in improving the speech outcomes, but the data analysis revealed higher efficacy of SGDs in pediatric patients with CP without a co-manifestation of intellectual disability[25]. The evidence collected by Lund SK and Light focused on potential variety of predictive factors influencing the AAC intervention outcomes. The authors evaluated the predictive role of cognition and looked into this domain trying to reveal all social-cognitive factors involved. The work scrupulously analyzes all pre-intervention patient characteristics that possessed a potential to predict communication outcomes for patients with CP. The evidence suggests that the identified factors: age, comorbid conditions as well as social-cognitive factors are critical variables affecting the outcomes. The evidence accumulated is only for limited number of AAC methods such as PECS, SGDs and new studies are needed to evaluate the predicting or moderating role of all the influencing factors for a large variety of low or high-tech AAC devices. Identification of the possible predictors, moderators, and mediators that may influence the outcomes of AAC intervention is of major importance, as they should be discussed as potential targets included in AAC assessment questionnaires, making the possible outcome more effectively measureable.

PURPOSE

The objective of the presented review was to evaluate the effectiveness of AAC interventions in patients with CP, pointing on those determinant variables of main intervention outcomes that have to be considered by the specialists in the assessment and intervention process.

RESEARCH STRATEGY

The systematic review was conducted based on the Cochrane Collaboration guidelines[26] and reported using the statement of PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses)[27]. A search strategy was developed by the team of the review authors (T.R.P., H.H.M., AVA; F.A.S., J.M.A.). The search strategy was based on the inclusion of indexed keywords, title and/or abstract text terms from database records. An author specialized in information management (J.M.A.) completed the search strategy, identified the keywords according to the features of each database, and conducted the computer-aided search. The search encompassed the following databases: MEDLINE (Ovid); MEDLINE In-Process Citations & Daily Update (Ovid); PubMed (NLM) (Internet)[28]; Embase (Ovid); Cochrane Database of Systematic Reviews (CDSR;

Cochrane Library: Wiley); Cumulative Index to Nursing and Allied Health Literature (CINAHL; EBSCO); Database of Abstracts of Reviews of Effects (DARE; Cochrane Library: Wiley); Cochrane Central Register of Controlled Trials (CENTRAL; Cochrane Library: Wiley); Health Technology Assessment (HTA) database (Cochrane Library: Wiley) and PEDro (Internet)[29]. Studies included in review articles and reference lists of the reviews were checked to reveal relevant studies. EndNote bibliographic software (Clarivate Analytics, Philadelphia, PA, U.S.A.) was used to identify and download the references.

SELECTION CRITERIA

Full-text and peer-reviewed studies in English studying the effectiveness of augmentative and alternative communication in patients with cerebral palsy were included. No publication date or publication status restrictions were imposed (including dissertations and theses to address publication bias). Two review authors (T.R.P. and H.H.M.) performed the screening of results independently and selected the articles by title and abstract according to initially elaborated eligibility criteria. Any disagreement between the two authors was resolved by a third author (F.A.S.) of the review. All selected items were independently checked as full text by the same review authors. In case of disagreement the full text was assessed by the third author (F.A.S.) to gain consensus. Studies with pediatric patients (<18 years) diagnosed with CP were included (Table 1). CP diagnosis should include the description of motor dysfunction (spastic, atetoid, ataxic), as well as define all other co-manifestations (sensory deficit, speech disorder, mental disorder, etc.).

TYPES OF INTERVENTIONS

AAC is defined as a multimodal intervention including at least (1) an access mode (e.g., upper limb function, eye gaze, etc.) and AAC method (e.g., unaided AAC (body movements, gestures, facial expressions, signs); aided AAC (objects, object symbols, photographs, picture symbol boards and books; communication devices). The formulation of AAC was based on the recently conducted studies and systematic reviews[30,31]. AAC interventions had to be provided individually or in a group and conducted by a speech therapist (or an aligned specialist) working in specialized centers and departments (special education center, a local primary care facilities, schools or preschool centers, office of a speech therapist, a hospital speech therapy department). For a more detailed analysis of variables contributing to outcomes, data reported by other partners (e.g., peers, family members) were also included in the review. Studies evaluating multidisciplinary interventions provided by different professionals were not included. The control interventions (if applicable) included primary care treatments without an AAC method (e.g., usual speech therapy, language therapy, swallowing and communication therapy), no treatment or the waiting list. Not all the studies included in this review were with a comparator (comparing strategy of intervention), yet we were interested in estimation of variables not only between the interventions, but also within the selected AAC intervention.

Types of outcome measures

The primary outcomes were determined based on the recently conducted reviews on communication therapy interventions[32,33] and included the following: receptive and expressive language skills. The primary outcomes were derived from assessment tools applied in included studies (Vocabulary Acquisition (Mean proportions of errors); MLU; PLS-4; FOCUS©; PPVT-R; TACL-R; GSRT; Analysis of conversational samples).

Secondary Outcomes were also derived from assessment tools applied in the pooled studies (Improvement in child's literacy skills; Reduction in child's frustration; Battelle age equivalence scores; Increase in CACE vocabulary; YC-PEM; CP- Quality of Life Questionnaire; DMQ; GAS; RAACS; Arc's Self-Determination Scale; The Quality of Life Profile for People with Physical and Sensory Disabilities). Outcomes were classified as short term (up to 3 months), medium term (>3 to 12 months), and long term (>12 months). Measurement categories and derived outcomes are presented in Table 2.

DATA ANALYSIS

Data extraction from the selected studies was performed by one of the review authors (T.R.P.) and then screened by a second review author (A.V.A) to increase the number of relevant studies identified for use in this systematic review. Extracted data contained relevant information about the study participants (age, gender, type and duration of intervention); intervention type (unaided or aided AAC methods); type of comparators (if applicable); type of analysis methodology; outcomes at baseline and at follow-up. The review of the extracted data from all selected studies revealed that the studies were heterogeneous

Table 1. Criteria for inclusion and exclusion of the reviewed studies

Inclusion criteria	Exclusion criteria
1. Studies on individuals with the diagnosis cerebral palsy and report of the presence of communication deficit.	1. Studies where the case definitions are not clearly stated as cerebral palsy or if it is inconsistently applied
2. Studies which reported data on any outcome of interest for CP patients with communication deficit who use AAC tools.	2. No communication skills related data were reported on any of the outcomes
3. Studies in individuals below 18 years	3. Reported data for individuals with cerebral palsy are not necessitating application of AAC methods
4. Studies with known designs including observational and interventional studies (single subject, multiple baseline research design, case series, cross-sectional, case control, cohort and randomized control trials).	4. Methods of data collection and documentation are not reported

Table 2. Measurement categories and derived outcomes

Measurement category	Abbreviation	Outcome
Vocabulary Acquisition (Mean proportions of errors)	-	
Mean Length of Utterance in morphemes	MLUm	
Preschool Language Scale-4-th edition	PLS-4	Receptive and Expressive
Focus on the Outcomes of Communication Under Six	FOCUS©	Language Skills
Peabody Picture Vocabulary Test - Revised	PPVT-R	
Test for Auditory Comprehension of Language (Receptive)	TACL-R	
Gray Silent Reading Test	GSRT	
Analysis of conversational samples	-	
Functional Assessment of Communication Skills for Adults	ASHA-FACS	Communication and cognitive skills
Improvement in child's literacy skills	-	Literacy skills
Reduction in child's frustration	-	Level of frustration
Battelle age equivalence scores	-	Developmental delay
Increase in CACE vocabulary	-	
Young Children's Participation and Environment Measure	YC-PEM	Participation motivation
CP- Quality of Life Questionnaire		Quality of life
Dimensions of Mastery Questionnaire	DMQ	Mastery motivation
Goal Attainment Scaling	GAS	Goal attaining
The Responsive Augmentative and Alternative Communication Style Scale	RAACS	Responsiveness
Arc's Self-Determination Scale;	-	Self-Determination
The Quality of Life Profile for People with Physical and Sensory Disabilities	-	Quality of life
Canadian Occupational Performance Measure	COPM	Activities of daily living

to initiate a meta-analysis. Therefore, a narrative analysis was conducted to evaluate the efficacy of AAC methods. A random-effects model meta-analysis was used to assess determinants of AAC intervention outcomes.

The Grades of Recommendation, Assessment, Development, and Evaluation (GRADE) approach was used to assess the overall quality of the evidence and the strength of recommendations that could be applicable across a wide range of interventions and studies. The GEADE provides the best estimate of the effect size for each outcome in the review. The overall quality rating in GRADE is addressed to the body of evidence across outcomes, and selects the lowest quality of evidence from all of the outcomes that are influential to decision making. Evidence was categorized as high, moderate, low or very low-quality evidence[34]. A random-effects model meta-analysis was conducted for each predictor. For this purpose comprehensive Meta-Analysis software (V. 2; Biostat, Englewood, NJ) was used. The significance was acknowledged at $p < 0.05$. We have used Cohen's[35] conventions for the interpretation of effects sizes as small (Cohen's d = 0.2, Pearson's r = 0.1), medium (Cohen's d = 0.5, Pearson's r = 0.3), or large (Cohen's d = 0.8, Pearson's r = 0.5). The main effects of each meta-analysis were processed using one of two statistics: Pearson's r to show the relationships between continuous predictors and outcomes; and Cohen's d. to show the relationships between nominal predictors and outcomes. The heterogeneity was assessed using a chi-square test of the Q value and estimated the magnitude of heterogeneity using the I^2 value[36]. The value of I^2 was classified as low (< 0.25), moderate (0.50), or high (0.75). Potential moderators of the relationship between predictors and outcomes were analyzed with meta-regression for the group of continuous variables (method-of-moments model) and subgroup analysis for nominal variables. The moderation analyses are applied in cases when moderate or high heterogeneity ($I^2 > 0.25$) was revealed (a minimum of eight studies are required for the meta-regression)[37].

Risk for bias assessment

Risk of bias assessment aimed to provide transparency of results and findings presented in the review. It was performed for all included studies to eliminate bias in their findings, reported in results or conclusions. Risk of bias assessment tried to reveal the validity of findings and objectively estimate the intervention effect. The risk for bias assessment was performed independently by two review authors (T.R.P. and H.H.M.), and in case of disagreement a third review author (J.M.A.) was involved in the assessment process. The authors of the review were not blinded for the name of the journal, author names or institutions. Corresponding authors were contacted, if there was a need for additional data. To perform a quality analysis of included studies the EPHPP (Effective Public Health Practice Project) tool was used, as it ensures assessment of multiple studies with different study designs[38,39]. According to guidelines of EPHPP each study was assessed regarding its adequacy with respect to (a) addressing selection bias, (b) research design, (c) limiting confounders, (d) blinding, (e) data collection methods, and (f) accounting for withdrawals and dropouts from the study. In EPHPP assessment, the studies are rated as strong, moderate or weak on each component and the overall quality rating is derived from the combination of all scores. Studies with less than four strong component ratings and no more than one weak rating were considered to have an overall assessment of moderate. The authors individually evaluated studies against the EPHPP

criteria and reached 85% on individual rating components and 100% on the overall rating score. Inconsistencies in the ratings were discussed until a 100% agreement was reached.

RESULTS

The online database search revealed 442 records (Figure 1). Three additional records were identified through manual reference checking. All the duplicates were removed and 257 records remained and were checked on title and abstract. The screening identified 19 items to be evaluated as full text for eligibility. After the screening 9 studies met predefined eligibility criteria and were included in the systematic review[40-48]. From 9 studies included in the systematic review 4 were studies with single-subject multiple-baseline research designs, 3 were longitudinal cohort studies, 1 case control study and 1 case series. All of the selected items were written in English. Studies were conducted in the USA[40-44], Malaysia[45], Spain[46], UK[47] and Australia[48]. Among the included studies seven were interventional and two observational studies. All studies provided data at baseline, and post-intervention assessment.

Participants

Participants of the studies were recruited via speech therapist (or an aligned professional) at special education centers, local primary care facilities, schools or preschool centers, offices of a speech therapist, hospital speech therapy departments. The retrospective searches of patient records were also included. The sample sizes of the selected studies ranged from 4 to 215 participants and the overall sample size of included CP patients was 309 (Table 3). Mean levels of expressive and receptive communication skills were assessed by different standardized measurement tools, with moderate to severe scores at baseline (Table 3).

Interventions

The studies have used different types of unaided (eye gaze, facial expressions, gesturing, and/or pointing, eye codes; manual sign, facial expression, eye gaze, Words for Life; Vantage Lite with Unity3; Nova Chat; Eye tracker systems: Tobii PCEye Go and myGaze; Child–parent interactive play with unaided AAC such as manual signs and gestures; pictogram-based AAC) and aided (communication boards, books, and/or pictures, VOCAs or SGDs, computerized AAC devices, iPad with LAMP, communication book & makaton; communication board and typing; mobile AAC apps; communication book; communication book and writing; low-technology boards, switches, high technology devices) AAC devices.

Two studies compared an AAC intervention method with no speech and no AAC therapy[40,47]. One study used another AAC method (Computerized AAC devices for children without disability) as a comparator[41]. One other study[46] used the same AAC strategy in the control group with other category of patients (Table 3).

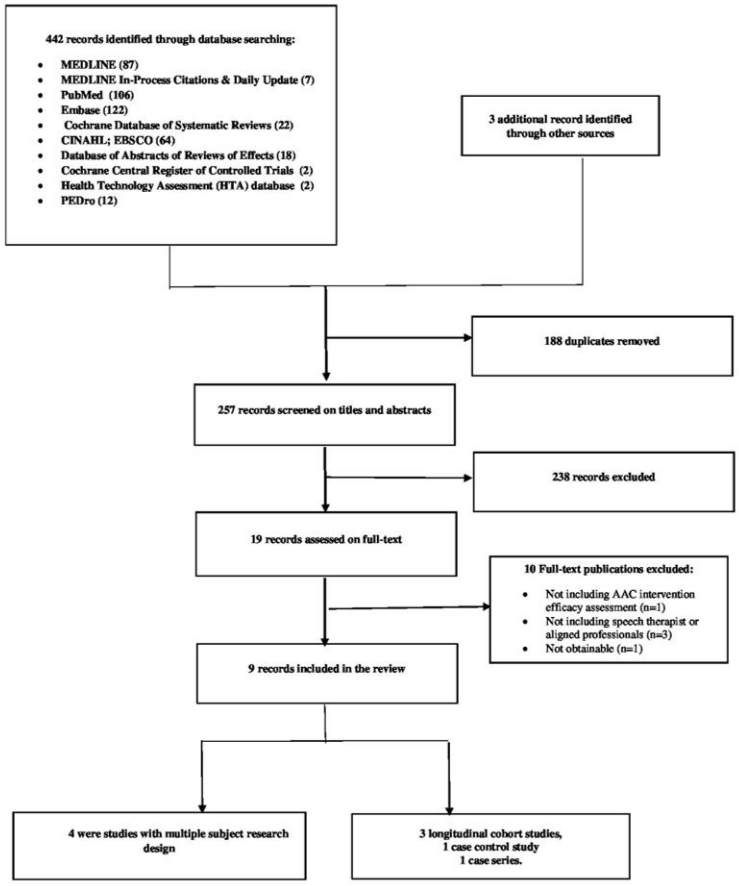

Figure 1. Study flow diagram

Table 3. Characteristics of studies included in the review. N/A = Not applicable.

Author	Country	Design	Participants	Intervention	Duration of intervention	Comparison	Outcome measurement tool	time point of measures	Results/ significance
Hidecker et al.[40]	USA	case series	215 children with CP (from 2 to 17 years)	Unaided AAC (Eye gaze, facial expressions, gesturing, and/or pointing; manual signs); Aided AAC (Communication boards, books, and/ or pictures, VOCAs or SGDs)	12 months***	No speech and no AAC therapy; Speech therapy	Communication skills (receptive and expressive);	Baseline: 3; 6; 12 months after baseline	P=0.0006
Hochstein et al.[41]	USA	case control study	16 children with CP and motor disability (3.2-8.1 year, months)	Computerized AAC devices	2 weeks*	Computerized AAC devices (for children without motor disability)	Communication skills (receptive and expressive); Vocabulary Acquisition (Mean proportions of errors)	Baseline: 2 weeks	P< 0.01
Joginder Singh et al.[45]	Malaysia	Single-subject multiple-baseline research design	8 children with CP (3–12 years)	Communication book & Makaton; Communication board and typing; Mobile AAC apps; Communication board and mobile AAC apps; Communication book; Communication book and writing	1.1-2.3*** years/ months	Not applicable (N/A) (Qualitative research design)	Communication skills (receptive and expressive); Improvement in child's literacy skills; Reduction in child's Frustration.	Baseline: 1.01; 1.09; 2.01; 2.02 years	N/A
Medeiros et al.[42]	USA	Longitudinal cohort study	16 children with CP (9–27 months),	Child–parent interactive play with unaided AAC such as manual signs and gestures.	18 months***	N/A	Communication skills (receptive and expressive); Battelle age equivalence scores (Communication and Motor)	Baseline: 9 months; 18 months	Motor skills: $p < 0.001$; Language skills: $p < 0.01$
Pahisa-Solé and Herrera-Joancomartí[46]	Spain	Single-subject multiple-baseline research design	4 patients with CP (13-17 years)	Pictogram-based AAC	30 years***	The same communication device without the compansion system	Communication skills (receptive and expressive); Increase in CACE vocabulary*	Baseline: 30 years	Increase for 41.59%; P<0.05
Smith and Hustad[43]	USA	Longitudinal cohort study	26 children with CP (two-year-old)	Aided AAC (low-technology boards, switches, high-technology devices); Unaided AAC (facial expression, sign, eye gaze, and gestures)	6 months**	N/A	Communication skills (receptive and expressive); Preschool Language Scale-Fourth Edition) and expressive (Mean length of utterance in morphemes) language skills	Baseline: 6 months	PLS raw: P<0.05; MLUm: P<0..01
Soto and Clarke[47]	UK	Single-subject multiple-baseline research design	4 teenage girls (14 - 18 years)	iPad with LAMP Words for Life; Vantage Lite with Unity3; Nova Chat	12 weeks**	Conversation-based intervention	Communication skills (receptive and expressive): Verbs; Pronouns; Bound morphemes; Spontaneous clauses	Baseline: 12 weeks	Communication skills: Verbs (p= 0.004; 0.0005; 0.07; 0.06); Pronouns (P= 0.005; 0.002; 0.09; 0.59); Bound morphemes (p= 0.006; 0.39; 0.92; 0.03); Spontaneous clauses (p= 0.06; 0.002; 0.003; 0.20)
Karlsson et al.[48]	Australia	Single-subject multiple-baseline research design	5 children with CP (aged 3–5 years)	Eye tracker systems: Tobii PCEye Go; myGaze (Software: (Look to Learn and Grid 3)	6 weeks*	N/A	Communication skills (receptive and expressive); PLS-4); FOCUS©; YC-PEM; CP- Quality of Life Questionnaire; DMQ; COPM; GAS; RAACS	Baseline: 6 weeks	significant results for: COPM; GAS
Lund and Light[44]	USA	Longitudinal cohort study	15 patients with CP (age: 4-8 years)	Vocalizations; Head shake/nod; Communication board with alphabet; Eye pointing; Vocalizations; Eye codes;	15 years***	N/A	Communication skills (receptive and expressive); PPVT-R; TACL-R; GSRT; Analysis of conversational samples; ASHA-FACS; Arc's Self-Determination Scale; The Quality of Life Profile for People with Physical and Sensory Disabilities	Baseline: 15 years	Significant results for: PPVT-R; TACL-R

Outcomes were classified as short term *(up to 3 months), medium term; **(>3 to 12 months), and long term; ***(>12 months)

Speech therapist

The number of speech therapists (or aligned professionals) providing the AAC interventions was different within the selected studies (from an individual therapist to professional group intervention). Speech therapists mostly conducted training program with a duration ranging from 2 weeks to a maximum of 27 weeks (Table 3). Exception were two studies with single-subject multiple-baseline research design that have evaluated the AAC efficacy after a longer application period[45,46]. Majority of therapists received special training and instructions on the applied AAC method.

Analysis of determinant variables that influence the AAC outcome in CP patients

The basic design of the review includes the assessment of AAC intervention efficacy in pediatric patients with CP discussing the effectiveness of different aided or unaided alternative communication strategies and pointing to factors that may potentially influence the efficacy of intervention. These determinant factors are potentially associated with intervention outcomes and in majority of studies are not properly considered or interpreted. In almost all studies where complex intervention methods are used the component factors involved are not analyzed. Nine studies, involving 309 participants, contributed to the analysis of the relationship between the selected predictors and expressive and receptive language outcomes (Figure 2).

According to the random effects meta-analysis results, there was a statistically significant main effect of the correlation between dysarthria severity and language skills ($r = 0.369$, $p=0.029$). Heterogeneity was moderate ($I^2 =42.315$, $Q = 22.326$, $p < 0.05$). Statistically significant correlation existed also between the cognitive ability and language skills ($r=0.340$, $Q=12.717$, $P=0.042$ and $I^2=29.228$). However, none of the hypothesized moderators— playing skills of the child, play interaction, object exploration, speech imitation, joint attention explained a significant amount of heterogeneity. Examination of a funnel plot revealed asymmetry (Figure 3), and so we applied a Duval–Tweedie trim-and fill correction. This correction reduced the main effect but did not change the finding ($r = 0.126$, $p < 0.01$). Sensitivity analysis indicated that the finding is robust to the removal of any individual sample. Results of the meta-analyses can be categorized as medium significant effects, small significant effects, and nonsignificant effects. Visual impairments, hearing impairments and limb paresis demonstrated a small significant effect on expressive and receptive language outcomes.

Dysarthria severity and cognitive ability showed medium significant effects. The nominal variables such as the age

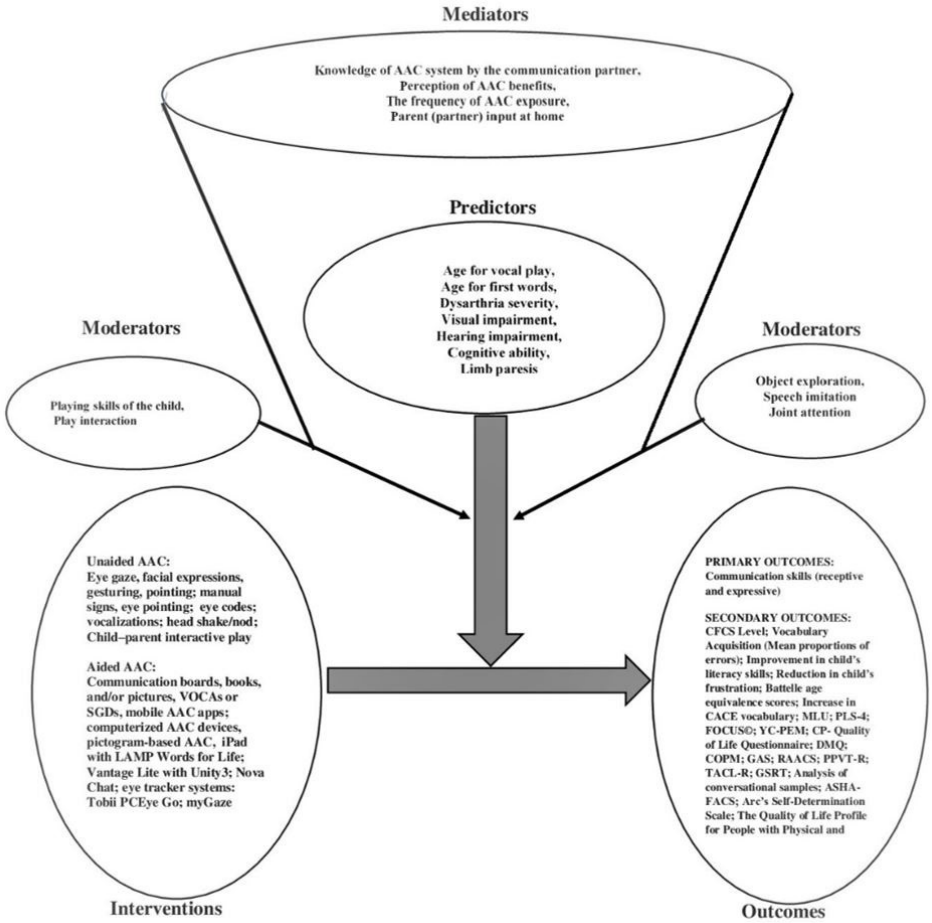

Figure 2. Interaction of predicting, moderating and mediating factors. Influence of determinant factors on intervention outcomes

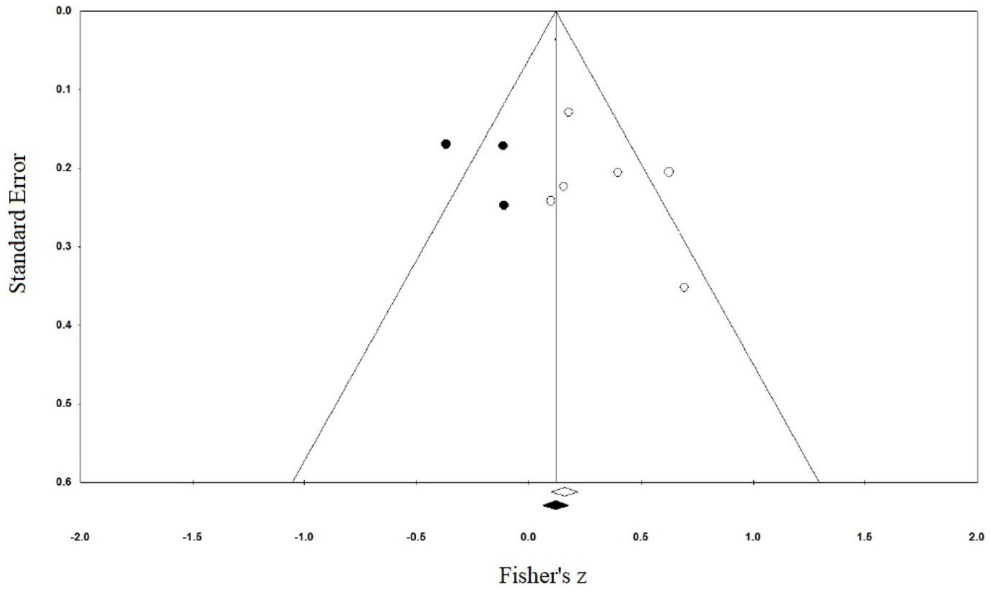

Figure 3. Funnel plot of Fisher's *z* on standard error for dysarthria severity, showing asymmetry. Empty shapes represent included studies and filled shapes represent the Duvall Tweedie Trim and Fill correction.

Table 4. GRADE evidence profile: AAC interventions for pediatric patients with CP

Quality assessment						
No of studies	Risk of Bias	Inconsistency	Indirectness	Imprecision	Publication bias	Overall quality of evidence
9	Serious	Not serious	Not serious	Not serious	None	Moderate

for vocal play (AVP) and age for first words (AFW) showed nonsignificant effects. Our results indicate that dysarthria severity is a better predictor than hearing or visual impairments. The findings show that children with better cognitive ability have a better prognosis for language development. The results support the concept insisting that issues in comprehension are correlated with the most severe language disorders[49,50]. From the list of continuous variables the hearing or visual impairments showed small significant effect, possibly due to different aiding methods used in therapy process. The nonsignificant correlation between AFW (and/or AVP) and language outcomes shows that early word acquisition could be qualitatively different from the types of language/grammatical skills that are difficult for children with CP. The determinant factors can serve not only as predicting variables, but also may represent a group of variables that influence the strength of relationship between the predicting variable and the outcome. The described variables are moderators and in the studies included in this review they are identified as child characteristics: speech imitation, joint attention, play interaction, object exploration. These factors are not directly predicting the outcome but may enhance the influence of predictors (positively or negatively) upon the outcome variable. In addition, there is a need to emphasize the mediator variables that explain the mechanism by which the independent predicting variable affects the dependent outcome variable. Usually the mediators are analyzed after the study completion and are helpful to explain why and how the intervention was effective. Among the mediating factors identified in this review are the influences of the communication partners on different components of intervention. Mediators responsible for the reinforcement of predicting influences on outcomes are the frequency of therapy, communication partner knowledge, perception of AAC benefits, and parent (partner) input at home.

Risk for bias assessment

The number of studies included in the review was small and the risk for overestimation of AAC intervention efficacy was very high. Possible high risk is explained by variety of factors: research favoring positive results, different study design and large variety of outcomes. Strength evaluation for included studies and applied design was based on the Grading of Recommendations, Assessment, Development and Evaluation guidelines. The scores of GRADE (Table 4) represent different required components: level of evidence, quality of evidence, relevance of evidence; statistics precision and size of effect[51]. Our review does not contain any randomized control trials that are known to have less risks of bias. On the other hand selection of study design does not in itself increase or decrease the research quality. Effective Public Health Practice Project (EPHPP) was used to perform quality assessment of included multimodal studies: The Table 5 shows results of EPHPP assessment for individual studies and components, as well as the overall rating of quality assessment.

Outcomes

All studies included in the review reported improvement in the receptive and expressive language skills of patients with

Table 5. EPHPP quality assessment of included studies

Studies included in the review	Selection bias	Design	Confounders	Blinding	Data collection	Withdrawals/ dropouts	Overall rating
Hidecker et al. (2018)[40]	Moderate	Moderate	Strong	Weak	Strong	Strong	Moderate
Hochstein et al. (2003)[41]	Moderate	Moderate	Strong	Moderate	Strong	Strong	Moderate
Joginder Singh et al. (2017)[45]	Moderate	Moderate	Strong	Weak	Strong	Strong	Moderate
Medeiros et al. (2016)[42]	Moderate	Strong	Strong	Moderate	Strong	Moderate	Moderate
Pahisa-Solé and Herrera-Joancomartí (2017)[46]	Moderate	Strong	Strong	Weak	Strong	Strong	Moderate
Smith and Hustad (2015)[43]	Weak	Moderate	Strong	Weak	Strong	Strong	Moderate
Soto and Clarke (2018)[47]	Moderate	Moderate	Strong	Weak	Strong	Strong	Moderate
Karlsson et al. (2018) [48]	Moderate	Moderate	Strong	Weak	Strong	Strong	Moderate
Lund and Light (2006)[44]	Moderate	Moderate	Strong	Moderate	Strong	Moderate	Moderate

CP. These primary outcomes were derived from corresponding assessment tools (Vocabulary Acquisition (Mean proportions of errors); MLU; PLS-4; FOCUS©; PPVT-R; TACL-R; GSRT; Analysis of conversational samples) applied in studies included in the review. Seven studies out of nine showed quantified evidence of improvement in communication skills. Secondary outcomes were also derived from assessment tools applied in the pooled studies (Improvement in child's literacy skills; Reduction in child's frustration; Battelle age equivalence scores; Increase in CACE vocabulary; YC-PEM; CP- Quality of Life Questionnaire; DMQ; GAS; RAACS; Arc's Self-Determination Scale; The Quality of Life Profile for People with Physical and Sensory Disabilities). They were distributed with certain level of overlap in included studies, though the design of the studies was very heterogeneous. All secondary outcomes were scores of standardized measurements (for participation motivation, quality of life, activities of daily living, goal attaining, responsiveness, improvement in child's literacy, level of frustration, developmental delay, cognitive deficits, self-determination) and in all studies deploying the specific tools significant improvement was registered. In five studies this improvement was statistically significant which exposes the instruments that are more reliable and flexible for use with various AAC strategies (Table 3).

CONCLUSIONS

In general, this systematic review produced moderate-quality evidence indicating that AAC interventions have proven medium and long-term efficacy in improving communication of CP patients. In studies where AAC intervention is compared to speech therapy (or no therapy), high-quality evidence was revealed that AAC is improving communication at medium and long-term interventions[40,47]. This systematic review does not include studies with RCT design, which usually produces the highest level of evidence and lowest level of the risk for bias, compared to other interventional or observational designs. Hence, certain level of bias was included in all studies where the patients, therapists (professionals), and outcome assessors were not blinded. This systematic review included studies with diverse strategies of patient selection, including primary sources such as neurologists, speech therapist, special educator, as well as secondary care specialists. Possible categories of applied assessment tools, many of which are standardized measurement methods, have been integrated and presented in review results. The review tried to focus on all possible outcome measures, both primary and secondary that have been used by the authors of included studies. However, heterogeneity existed among the included studies and part of the included studies were with methodological and practical limitations. It is essential also to select therapists or other professionals based on their AAC skills or to properly train and support them in providing AAC services. Trained therapist might be capable of identifying patients with AAC need (for temporary therapeutic or permanent use). The practical implication of the determinant variables is essential for intervention and assessment planning. Any assessment tool containing more of the determinant variables may ensure the accuracy of the AAC need evaluation or AAC intervention efficacy. In general, the majority of determinant factors identified in this review were rather large categories (language skills, cognition, language comprehension, etc.) and there is a need for new studies of high methodological and practical quality focusing on detailed, clusterized analysis of determinant variables to clarify the evidence.

REFERENCES

1. Jonsson U, Eek MN, Sunnerhagen KS, Himmelmann K. Cerebral palsy prevalence, subtypes, and associated impairments: a population-based comparison study of adults and children. Dev Med Child Neurol. 2019;61(10):1162-7. http://dx.doi.org/10.1111/dmcn.14229. PMid:30950519.
2. Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy. Dev Med Child Neurol Suppl. 2007;109:8-14. PMid:17370477.
3. Pellegrino L. Cerebral palsy. In: Batshaw ML, editor. Children with disabilities. 5th ed. Baltimore, MD: Brookes; 2002. p. 443-66.
4. Odding E, Roebroeck ME, Stam HJ. The epidemiology of cerebral palsy: incidence, impairments and risk factors. Disabil Rehabil. 2006;28(4):183-91. http://dx.doi.org/10.1080/09638280500158422. PMid:16467053.
5. Zhang JY, Oskoui M, Shevell M. A population-based study of communication impairment in cerebral palsy. J Child Neurol. 2015;30(3):277-84. http://dx.doi.org/10.1177/0883073814538497. PMid:25051968.
6. Frisch D, Msall ME. Health, functioning, and participation of adolescents and adults with cerebral palsy: a review of outcomes research. Dev Disabil Res Rev. 2013;18(1):84-94. http://dx.doi.org/10.1002/ddrr.1131. PMid:23949832.
7. Balandin S, Berg N, Waller A. Assessing the loneliness of older people with cerebral palsy. Disabil Rehabil. 2006;28(8):469-79. http://dx.doi.org/10.1080/09638280500211759. PMid:16513580.
8. Cooper L, Balandin S, Trembath D. The loneliness experiences of young adults with cerebral palsy who use alternative and augmentative communication. Augment Altern Commun. 2009;25(3):154-64. http://dx.doi.org/10.1080/07434610903036785. PMid:19591004.
9. Hemsley B, Balandin S, Togher L. "I've got something to say": interaction in a focus group of adults with cerebral palsy and complex communication needs. Augment Altern Commun. 2008;24(2):110-22. http://dx.doi.org/10.1080/07434610701830579. PMid:18465365.
10. Hemsley B, Sigafoos J, Balandin S, Forbes R, Taylor C, Green VA, et al. Nursing the patient with severe communication impairment. J Adv Nurs. 2001;35(6):827-35. http://dx.doi.org/10.1046/j.1365-2648.2001.01920.x. PMid:11555030.
11. Clarke M, Price K. Augmentative and alternative communication for children with cerebral palsy. Paediatr Child Health. 2012;22(9):367-71. http://dx.doi.org/10.1016/j.paed.2012.03.002.
12. Pennington L. Cerebral palsy and communication. Paediatr Child Health. 2008;18(9):405-9. http://dx.doi.org/10.1016/j.paed.2008.05.013.
13. Andersen G, Mjøen TR, Vik T. Prevalence of speech problems and the use of augmentative and alternative communication in children with cerebral palsy: A registry-based study in Norway. Perspect Augment Altern Commun. 2010;19(1):12-20. http://dx.doi.org/10.1044/aac19.1.12.
14. Cockerill H, Elbourne D, Allen E, Scrutton D, Will E, McNee A, et al. Speech, communication and use of augmentative communication in young people with cerebral palsy: the SH&PE population study. Child Care Health Dev. 2014;40(2):149-57. http://dx.doi.org/10.1111/cch.12066. PMid:23656274.
15. Sigurdardottir S, Vik T. Speech, expressive language, and verbal cognition of preschool children with cerebral palsy in Iceland. Dev Med Child Neurol. 2011;53(1):74-80. http://dx.doi.org/10.1111/j.1469-8749.2010.03790.x. PMid:21039439.
16. Hustad KC, Miles LK. Alignment between augmentative and alternative communication needs and school-based speech-language services provided to young children with cerebral palsy. Early Child Serv (San Diego). 2010;4(3):129-40. PMid:22194705.
17. Bax M, Tydeman C, Flodmark O. Clinical and MRI correlates of cerebral palsy: the European cerebral palsy study. JAMA. 2006;296(13):1602-8. http://dx.doi.org/10.1001/jama.296.13.1602. PubMed: 17018805.
18. Hustad KC, Allison K, McFadd E, Riehle K. Speech and language development in 2-year-old children with cerebral palsy. Dev Neurorehabil. 2014;17(3):167-75. http://dx.doi.org/10.3109/17518423.2012.747009. PMid:23627373.
19. Cress CJ, Marvin CA. Common questions about AAC services in early intervention. Augment Altern Commun. 2003;19(4):254-72. http://dx.doi.org/10.1080/07434610310001598242.
20. Geytenbeek J. Prevalence of speech and communication disorders in children with CP. Dev Med Child Neurol. 2011;53(1):10-1. http://dx.doi.org/10.1111/j.1469-8749.2010.03803.x. PMid:21171218.
21. Marshall J, Goldbart J. "Communication is everything I think." Parenting a child who needs augmentative and alternative communication (AAC). Int J Lang Commun Disord. 2008;43(1):77-98. http://dx.doi.org/10.1080/13682820701267444. PMid:17852533.
22. Rosenbaum P, Eliasson AC, Hidecker MJ, Palisano RJ. Classification in childhood disability: focusing on function in the 21st century. J Child Neurol. 2014;29(8):1036-45. http://dx.doi.org/10.1177/0883073814533008. PMid:24810083.
23. Coleman A, Weir KA, Ware RS, Boyd RN. Relationship between communication skills and gross motor function in preschool-aged children with cerebral palsy. Arch Phys Med Rehabil. 2013;94(11):2210-7. http://dx.doi.org/10.1016/j.apmr.2013.03.025. PMid:23583864.
24. Mei C, Reilly S, Reddihough D, Mensah F, Pennington L, Morgan A. Language outcomes of children with cerebral palsy aged 5 years and 6 years: a population-based study. Dev Med Child Neurol. 2016;58(6):605-11. http://dx.doi.org/10.1111/dmcn.12957. PMid:26566585.
25. Lund SK, Light J. Long-term outcomes for individuals who use augmentative and alternative communication: part II--communicative interaction. Augment Altern Commun. 2007;23(1):1-15. http://dx.doi.org/10.1080/07434610600720442. PMid:17364484.
26. Higgins JPT, Green S. Cochrane Handbook for Systematic Reviews of Interventions. Chichester, UK: John Wiley & Sons Ltd; 2011
27. Liberati A, Altman DG, Tetzlaff J, Mulrow C, Gøtzsche PC, Ioannidis JP, et al. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate healthcare interventions: explanation and elaboration. BMJ. 2009;339:b2700. http://dx.doi.org/10.1136/bmj.b2700. PMid:19622552.
28. NIH: National Library of Medicine. [Internet]. Bethesda, MD: NIH; 2020 [cited 2020 Aug 13]. Available from: http://www.ncbi.nlm.nih.gov/pubmed
29. PEDro: Physiotherapy Evidence Database. [Internet]. Australia: PEDro; 2020 [cited 2020 Aug 13]. Available from: http://www.pedro.org.au/
30. Broomfield K, Harrop D, Judge S, Jones G, Sage K. Appraising the quality of tools used to record patient-reported outcomes in users of augmentative and alternative communication (AAC): a systematic review. Qual Life Res. 2019;28(10):2669-83. http://dx.doi.org/10.1007/s11136-019-02228-3. PMid:31214931.
31. Judge S, Randall N, Goldbart J, Lynch Y, Moulam L, Meredith S, et al. The language and communication attributes of graphic symbol communication aids - a systematic review and narrative synthesis. Disabil Rehabil Assist Technol. 2020;15(6):652-62. http://dx.doi.org/10.1080/17483107.2019.1604828. PMid:31012746.
32. Damiano DL, Gilgannon MD, Abel MF. Responsiveness and uniqueness of the Pediatric Outcomes Data Collection Instrument compared to the Gross Motor Function Measure for measuring orthopedic and neurosurgical outcomes in cerebral palsy. J Pediatr Orthop. 2005;25(5):641-5. http://dx.doi.org/10.1097/01.bpo.0000167079.83835.22. PMid:16199947.
33. Mandl L, Schindel D, Deutschbein J, Frick J, Schenk L. [Quality of German-Language Guidelines for Post-Stroke Rehabilitation of Aphasia and Dysarthria - Results of a Systematic Review and of an International Comparison]. [Rehabilitation (Stuttg). 2019;58(5):331-8. [German]. http://dx.doi.org/10.1055/a-0651-9838. PMid:30273948.
34. Guyatt GH, Oxman AD, Vist GE, Kunz R, Falck-Ytter Y, Alonso-Coello P, et al. GRADE: an emerging consensus on rating quality of evidence and strength of recommendations. BMJ. 2008;336(7650):924-6. http://dx.doi.org/10.1136/bmj.39489.470347.AD. PMid:18436948.
35. Cohen J. Statistical power analysis for the behavioral sciences. 2nd ed. Hillsdale, NJ: Erlbaum; 1988
36. Borenstein M, Hedges LV, Higgins JP, Rothstein HR. A basic introduction to fixed-effect and random-effects models for meta-analysis. Res Synth Methods. 2010;1(2):97-111. http://dx.doi.org/10.1002/jrsm.12. PMid:26061376.

37. Armijo-Olivo, S., Ospina, M., Bruno, R.D.C., Egger, M., Saltaji, H., Fuentes, J, et al. Poor reliability between Cochrane reviewers and blinded external reviewers when applying the Cochrane risk of bias tool in physical therapy trials. PLoS One. 2014;9(5):e96920. http://dx.doi.org/10.1371/journal.pone.0096920.

38. Evans N, Lasen M, Tsey K. Appendix A: effective public health practice project (EPHPP) quality assessment tool for quantitative studies. In: Evans N, Lasen M, Tsey K, editors. A systematic review of rural development research: characteristics, design quality and engagement with sustainability. USA: Springer; 2015. p. 45-63. SpringerBriefs in Public Health. http://dx.doi.org/10.1007/978-3-319-17284-2.

39. Johnson RE, Jones GT, Wiles NJ, Chaddock C, Potter RG, Roberts C, et al. Active exercise, education, and cognitive behavioral therapy for persistent disabling low back pain: a randomized controlled trial. Spine. 2007;32(15):1578-85. http://dx.doi.org/10.1097/BRS.0b013e318074f890. PMid:17621203.

40. Hidecker MJC, Slaughter J, Abeysekara P, Ho NT, Dodge N, Hurvitz EA, et al. Early predictors and correlates of communication function in children with cerebral palsy. J Child Neurol. 2018;33(4):275-85. http://dx.doi.org/10.1177/0883073817754006. PMid:29366365.

41. Hochstein DD, McDaniel MA, Nettleton S, Neufeld KH. The fruitfulness of a nomothetic approach to investigating AAC: comparing two speech encoding schemes across cerebral palsied and nondisabled children. Am J Speech Lang Pathol. 2003;12(1):110-20. http://dx.doi.org/10.1044/1058-0360(2003/057). PMid:12680818.

42. Medeiros KF, Cress CJ, Lambert MC. Mastery motivation in children with complex communication needs: longitudinal data analysis. Augment Altern Commun. 2016;32(3):208-18. http://dx.doi.org/10.1080/07434618.2016.1179789. PMid:27184193.

43. Smith AL, Hustad KC. AAC and early intervention for children with cerebral palsy: parent perceptions and child risk factors. Augment Altern Commun. 2015;31(4):336-50. http://dx.doi.org/10.3109/07434618.2015.1084373. PMid:26401966.

44. Lund SK, Light J. Long-term outcomes for individuals who use augmentative and alternative communication: part I--what is a "good" outcome? Augment Altern Commun. 2006;22(4):284-99. http://dx.doi.org/10.1080/07434610600718693. PMid:17127616.

45. Joginder Singh S, Hussein NH, Mustaffa Kamal R, Hassan FH. Reflections of Malaysian parents of children with developmental disabilities on their experiences with AAC. Augment Altern Commun. 2017;33(2):110-20. http://dx.doi.org/10.1080/07434618.2017.1309457. PMid:28387140.

46. Pahisa-Solé J, Herrera-Joancomartí J. Testing an AAC system that transforms pictograms into natural language with persons with cerebral palsy. Assist Technol. 2019;31(3):117-25. http://dx.doi.org/10.1080/10400435.2017.1393844. PMid:29045194.

47. Soto G, Clarke MT. Conversation-based intervention for adolescents using augmentative and alternative communication. Augment Altern Commun. 2018;34(3):180-93. http://dx.doi.org/10.1080/07434618.2018.1490926. PMid:30043650.

48. Karlsson P, Bech A, Stone H, Vale C, Griffin S, Monbaliu E, et al. Eyes on communication: trialling eye-gaze control technology in young children with dyskinetic cerebral palsy. Dev Neurorehabil. 2019;22(2):134-40. http://dx.doi.org/10.1080/17518423.2018.1519609.

49. Lambon Ralph MA, Snell C, Fillingham JK, Conroy P, Sage K. Predicting the outcome of anomia therapy for people with aphasia post CVA: both language and cognitive status are key predictors. Neuropsychol Rehabil. 2010;20(2):289-305. http://dx.doi.org/10.1080/09602010903237875. PMid:20077315.

50. Moulton E, Magno S, Valabregue R, Amor-Sahli M, Pires C, Lehéricy S, et al. Acute diffusivity biomarkers for prediction of motor and language outcome in mild-to-severe stroke patients. Stroke. 2019;50(8):2050-6. http://dx.doi.org/10.1161/STROKEAHA.119.024946. PMid:31272324.

51. NHMRC: National Health and Medical Research Council. NHMRC additional levels of evidence and grades for recommendations for developers of guidelines. Canberra: Australian Government; 2016.

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AVA: Data collection, writing of the manuscript; HM: Data collection, data analysis; FAS and JAM: Data selection, Data analysis; TP: Study design, writing of the manuscript