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Review Article

Speech and Language Therapy for Acquired Central Dysgraphia in Neurological Patients: A Systematic Review to Describe and Identify Trainings for Clinical Practice

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ABSTRACT

Purpose: Acquired central dysgraphia is a heterogeneous neurological disorder that usually co-occurs with other language disorders. Written language training is relevant to improve everyday skills and as a compensatory strategy to support limited oral communication. A systematic evaluation of existing writing treatments is thus needed.

Method: We performed a systematic review of speech and language therapies for acquired dysgraphia in studies of neurological diseases (PROSPERO: CRD42018084221), following the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) checklist with a search on several databases for articles written in English and published until August 31, 2021. Only methodological well-designed studies were included. Further assessment of methodological quality was conducted by means of a modified version of the Downs and Black checklist.

Results: Eleven studies of 43 patients in total were included. For each study, we collected data on type of population, type of impairment, experimental design, type of treatment, and measured outcomes. The studies had a medium level of assessed methodological quality. An informative description of treatments and linkages to deficits is reported.

Conclusions: Although there is a need for further experimental evidence, most treatments showed good applicability and improvement of written skills in patients with dysgraphia. Lexical treatments appear to be more frequently adopted and more flexible in improving dysgraphia and communication, especially when a multimodal approach is used. Finally, the reported description of treatment modalities for dysgraphia in relation to patients' deficits may be important for providing tailored therapies in clinical management.

Acquired central dysgraphia is a disorder in writing letters, syllables, words, or phrases that affects phonology, orthography, and semantic processing (Weekes, 2005). It usually co-occurs with disorders of other language modalities (i.e., reading, speaking, word retrieval, auditory comprehension) as in the case of aphasia (Damasio, 1998) after brain injury, tumor, infection, or stroke (Hallowell & Chapey, 2008). Oral language appears to be predominant in everyday communication (Nickels, 2002) and often prioritized in the treatment. However, written language may be easier to recover (e.g., when the lexical level is fairly preserved whereas articulation deficits are predominant), and it is recommended as a practical strategy to support and complement oral communication rehabilitation (Beeson &

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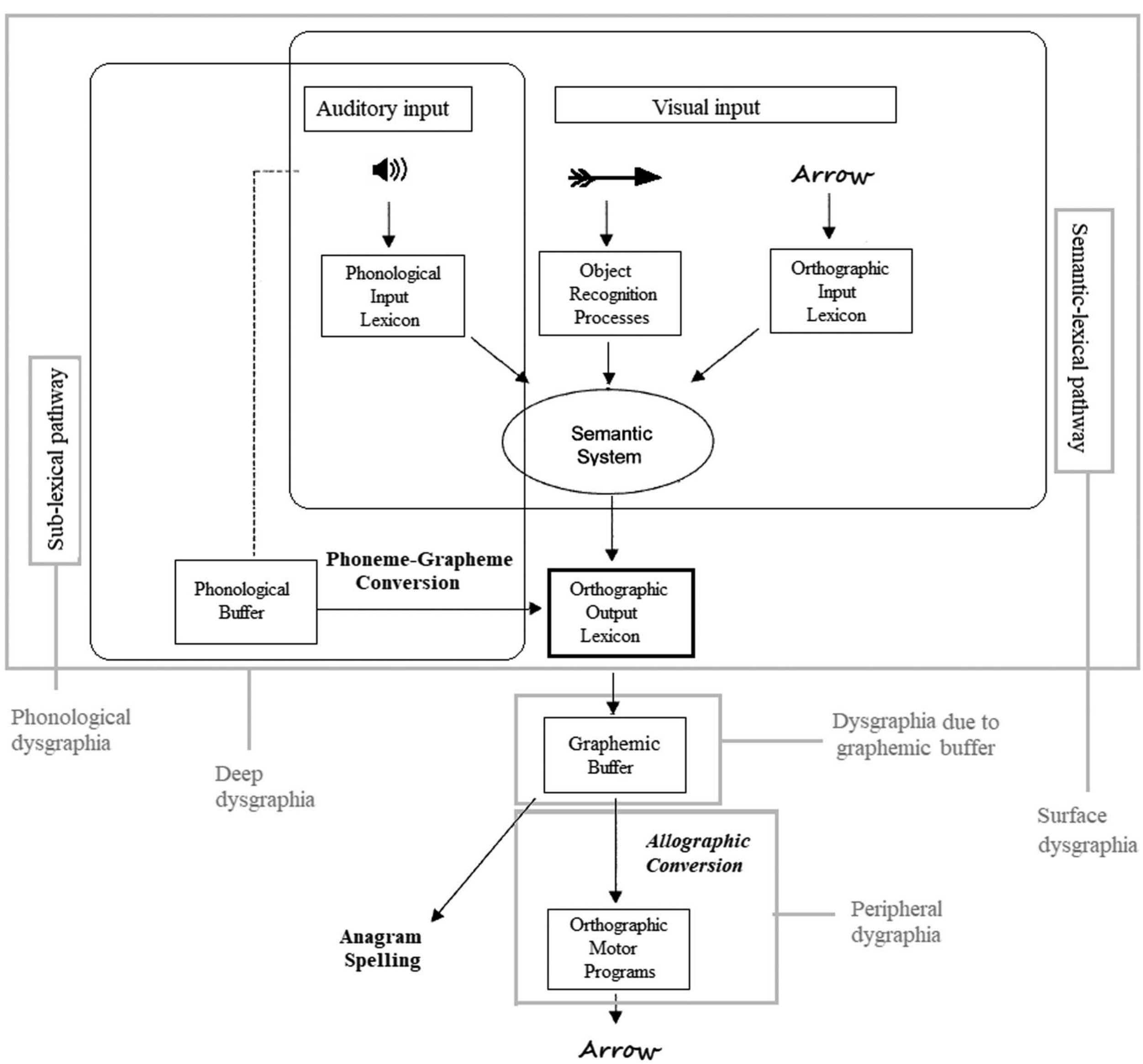
Rapcsak, 2002). In addition, writing plays a relevant role in activities of daily living, especially given the increasing use of mobile phones, tablets, and personal computers in work and social situations.

Writing consists of cognitive, linguistic, and motor components, so it is susceptible to impairment from brain injuries of varying severities and manifestation types (Rapp & Fischer-Baum, 2015) that affect the underlying spelling mechanism (Rapcsak et al., 2009). A cognitive model explains that the convergent input of semantics and phonology enables writing via semantic-lexical or sublexical pathways (see Figure 1; Beeson et al., 2002). This model is useful for identifying writing difficulties and giving guidance on their dysgraphia classification by damaged route: surface, phonological, deep dysgraphia, and dysgraphia due to damage of the graphemic buffer and peripheral dysgraphia (see Figure 1; Beeson et al., 2002).

The different types of dysgraphia exhibit specific manifestations that suggest particular areas of cerebral damage (Weekes, 2005). Regarding surface dysgraphia, which is often associated with left parietal lobe injuries (Rapcsak et al., 2009), the regularization of irregular word spellings is one of the main symptoms, whereas the spelling of regular words and nonwords is more accurate (Rapcsak et al., 2007). Patients with damage to the left perisylvian cortical area (Rapcsak et al., 2009) often show

Figure 1. Cognitive model for writing. The gray squares and labels indicate the types of dysgraphia associated with a specific disruption of the writing routes (this figure is based on the work of Beeson et al., 2002).



phonological dysgraphia, with difficulty in writing nonwords and unfamiliar words. Extensive lesions to the supramarginal gyrus and the insula usually cause deep dysgraphia (Rodrigues et al., 2014), leading to difficulties in writing nonwords and unfamiliar and abstract words, and to the appearance of semantic paragraphias (Jefferies et al., 2007). In the case of graphemic buffer disorders, usually with damage in the left parietal cortex (Sakurai et al., 2007), the deficit involves working memory during writing; hence, a length effect can be seen (Miceli & Capasso, 2006). The last cluster of symptoms belongs to peripheral dysgraphia, often associated with damage to the left temporo-parieto-occipital cortex, and results in problems in selecting the appropriate motor sequences needed to write letters (Beeson & Rapcsak, 2002).

With central dysgraphia, it is useful to classify the type to predict possible symptom clusters; however, adding information about the underlying processes may be significant both for setting and monitoring the rehabilitation program (Beeson & Rapcsak, 2002) and for understanding the possible copresence of oral language disorders.

In the literature, several speech and language therapies have been proposed to improve single-word writing. These involve writing tasks such as copying, delayed copying, and anagram tasks (Ball et al., 2011; Beeson et al., 2002; Miceli & Capasso, 2006) or the written naming of pictures (Kiran, 2005). Other studies have been directed toward promoting sound–letter conversion (Kiran, 2005; Luzzatti et al., 2000) or the production of written sentences (Salis & Edwards, 2010). Finally, some studies have focused on improving functional written communication, such as its use in conversation (Clausen & Besson, 2003). An evidence-based synthesis is of clinical importance in research on the treatment of language disorders (Dollaghan, 2007).

A narrative review (Thiel et al., 2015) attempting to narrowly summarize therapies with functional purposes found various types of treatment but also some with poor methodological paradigms, which affected the synthesis of considerations. While the authors provided a general representative overview of treatment categories, specific clinical guidance on the appropriate rehabilitation of dysgraphia was minimal and remains scarce in the extant literature. The treatment of dysgraphia is a relevant issue because of the limitations to communicative activities of daily living and the crucial role of treatment in supporting oral communication and enhancing its recovery (Beeson & Rapcsak, 2002). For these reasons, a comprehensive overview of evidence-based methods in written language treatments is needed to describe existing rehabilitation protocols. Moreover, an accurate knowledge of reported speech and language treatments and the link to patient deficits may be the key to informing clinicians on tailored training applications.

This review is limited to considering only central dysgraphias (i.e., dysgraphias consisting of specific cognitive impairments that do not overlap with those of a motor or visual–spatial nature). It excludes peripheral dysgraphias, which relate more to motor than linguistic aspects and exceed the aim of this review due to the possible visual and spatial complexity in proceeding letter shapes (D'Imperio et al., 2020; Ingles et al., 2014). Central dysgraphias can be interpretable within a coherent theoretical framework that dictates specific rehabilitation techniques. Therefore, the goals of this systematic review are (a) to identify whether there is methodological well-designed evidence for the treatment of acquired central dysgraphia and its outcomes; (b) to assess the quality of each study for methodological considerations, which comprises an important clinical issue in treatment research; and (c) to provide an informative description of treatments for acquired neurological dysgraphia in order to obtain a comprehensive picture of their clinical application. By providing a systematic review of speech and language treatments of acquired dysgraphia, this study ultimately aims to provide insight into rehabilitation tailored to deficits to guide clinicians in promoting the improvement of writing skills.

Materials and Method

Search Strategy

Since this current review relates to health care, its protocol was registered to the International Prospective Register of Systematic Reviews (PROSPERO: CRD42018084221). We followed the PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) 2020 guidelines for reporting systematic reviews (Page et al., 2021) to ensure an appropriate and transparent method (Caulley et al., 2020; Innocenti et al., 2020). The search was conducted through several databases: Cochrane Central Register of Controlled Trials (CENTRAL), PubMed, Web of Science, Embase, and Google Scholar (gray literature), from the beginning until August 31, 2021. The following keywords were used: *dysgraphia, agraphia*, *writing impairment*, *writing disorder*, *acquired dysgraphia*, *acquired agraphia*, *speech therapy*, *intervention*, *treatment*, *rehabilitation*, and *exercise*. The final intent was to identify all the types of rehabilitative strategies for written language recovery in cases of neurological deficits. (Appendix A shows the details of search terms and strategies.)

Selection Criteria

We included only papers written in English. The studies eligible for inclusion investigated rehabilitation of written language impairments in adult patients with acquired dysgraphia due to neurological diseases. Only patients with acquired focal neurological events were selected. Studies of patients with dementia, brain tumors, and progressive diseases were excluded due to expected differences in clinical evolution and treatment purpose. We only included randomized controlled trials, controlled trials, single-subject designs, and controlled before–after studies in order to focus specifically on methodological well-designed studies. These studies ensured that comparisons within or between treatments were conducted. Thus, case series, single-case reports, and cohort studies (without control subjects or design) were excluded. The considered outcomes were improvements in written lexical access, phonological processing, and the spelling of written words and/or sentences.

Data Collection and Analysis

Initially, two authors of this study (S.N. and S.R., both speech-language pathologists) independently screened titles and abstracts of the searched records. Articles selected by at least one of the reviewers were then independently read by both for final inclusion. Any disagreement was resolved through discussion or, if needed, by a third author (F.B., speech-language pathologist). Three authors (S.N., F.B., and C.B.) independently extracted data from each record, which were then reviewed by one of the others.

First, study characteristics were investigated, including type of speech and language treatment, main results, and study design and characteristics for methodological assessment. Additional data were then extracted to provide useful information for clinical application, such as participant information (sex, age, education, etiology, and time of onset), linguistic manifestations of dysgraphia and type of aphasia (if present), outcomes assessed, and descriptions of the intervention (duration, frequency, and follow-up periods).

Methodological Quality Assessment

A critical appraisal of the methodological quality of included studies was conducted by means of a modified version of the Downs and Black checklist (Downs & Black, 1998) by two authors (F.B. and C.B.). This is a well-established tool that was developed using rigorous psychometric methods and tested for reliability and validity (Saunders et al., 2003). The checklist assesses five main points: reporting, internal validity (bias), internal validity (confounding), external validity, and power. We adapted the checklist by removing some items for an ad hoc modified version to overcome the lack of gold standard tools in the design of the included studies, as in the case of single-subject experimental designs (Wendt & Miller, 2012). Three ratings are available for each item of the checklist: yes (1), no (0), and unable to detect (0). Scores range from 0 to 16, and a higher total indicates a lower risk of bias (see Appendix B).

Results

The database search identified 486 studies, of which 81 were duplicates and removed, leaving 405 titles and abstracts to be screened. Many records were excluded because they did not meet the PICO (patient, intervention, comparison, and outcome) conditions mentioned in the selection criteria (Eriksen & Frandsen, 2018). Only 19 abstracts were thus selected for full-text reading; eight did not meet the inclusion criteria (see Appendix C). Therefore, 11 studies were included in the final review (see Figure 2), as well as a similar number of articles appearing in reviews on specific aspects of medical rehabilitation (Innocenti et al., 2019; Nordio et al., 2018).

The included studies were all single-subject multiple-baseline designs, adopting a prototypical single-case experimental methodology (Tate et al., 2016). Among them, two studies had a cross-over design (Raymer et al., 2010; Thiel et al., 2016). No randomized controlled trials or controlled studies were included.

The studies were conducted in the United Kingdom (Thiel & Conroy, 2014; Thiel et al., 2016) and the United States (Beeson et al., 2003, 2010; Beeson & Egnor, 2006; Clausen & Besson, 2003; Kim et al., 2015; Kiran, 2005; Rapp, 2005; Rapp & Kane, 2002; Raymer et al., 2010) with publication dates between 2002 and 2019.

Study Characteristics

The entire sample consisted of 43 participants. Two participants took part in two different studies (Rapp, 2005; Rapp & Kane, 2002) but with different treatments and experimental designs.

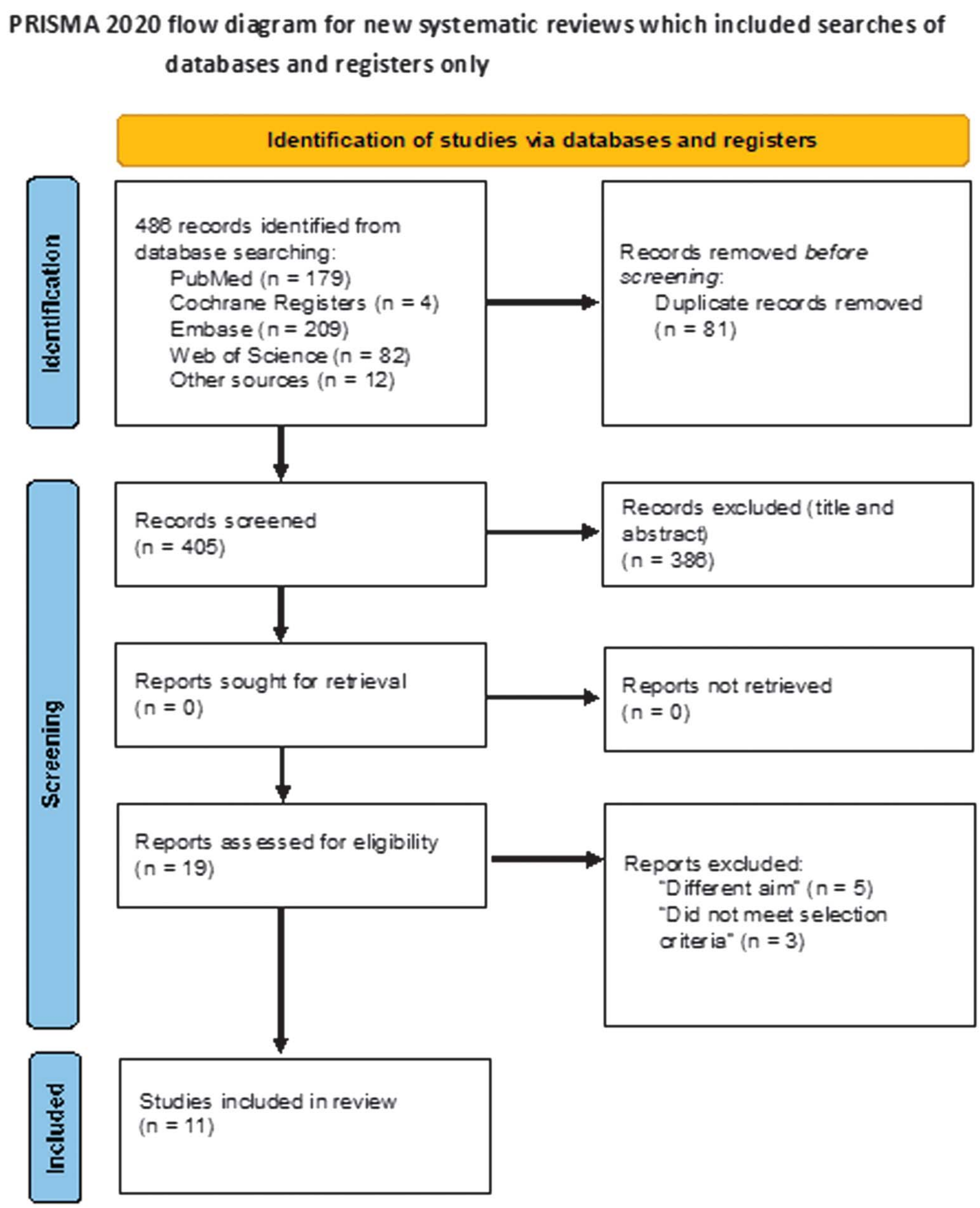
Assessment of Types of Treatment and Main Results

Types of Treatment for Dysgraphia

The types of dysgraphia treatment varied among the studies. As clearly described in Table 2, these were lexical, phonological, and interactive treatments.

The lexical treatments were administered in eight studies (Beeson et al., 2003; Beeson & Egnor, 2006; Clausen & Besson, 2003; Rapp, 2005; Rapp & Kane, 2002; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016), based on word spelling exercises. The treatments involved three different protocols: Copy and Recall Treatment (CART), spell–study–spell treatment, and repetition treatment. CART is a homework protocol that

Figure 2. PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) flow diagram of article selection.



includes repeated copying and recall of target words (Beeson et al., 2003; Clausen & Besson, 2003) with simultaneous oral language treatment in weekly group sessions. In the spell–study–spell treatment protocol, the participant repeats a listed word and attempts to spell it (Rapp, 2005; Rapp & Kane, 2002). The repetition treatment consists of oral repetition of target words in conjunction with CART (Beeson & Egnor, 2006).

Some studies using these lexical treatments also explored intervention approaches, which may be helpful in the application of rehabilitation. Intervention approaches systematically adopted included errorless treatments, designed to minimize errors, and errorful treatments, with progressive orthographic cues, to overcome errors (Raymer et al., 2010; Thiel & Conroy, 2014). Both approaches confirmed significant improvements in spelling accuracy in eight patients, but Raymer et al. (2010) noted a possible advantage of errorful treatments, whereas Thiel and Conroy (2014) found no significant difference between the effects of the two approaches and remarked on a risk of between-therapy interaction. However, these two studies had differences in methodologies, making them hard to compare.

Other approaches adopted were the unimodal approach, which utilizes only one task at a time, and the multimodal approach, which involves multiple concurrent tasks (Thiel et al., 2016). In these approaches, all eight participants achieved significant improvements in trained and untrained words, with no predominant differences in spelling accuracy (with the exception of one patient in a multimodal task); however, in the multimodal therapy, researchers observed a qualitative flexibility in performance training tasks and the proactive use of strategies (Thiel et al., 2016; see Table 2).

Phonological therapy was applied in only one study (Kiran, 2005), where it was not used in combination with other approaches. It focused on sound–letter correspondences: participants had to write some words dictated to them using a series of steps that emphasize phoneme–grapheme conversion rules (see Table 2).

Two studies evaluated the interactive treatments (Beeson et al., 2010; Kim et al., 2015). These studies combined lexical and phonological exercises in the rehabilitation protocol and aimed to practice problem-solving strategies (Thiel et al., 2015). They took advantage of the use of electronic devices that check and correct spelling errors. In the work of Kim et al. (2015), the treatment was associated with the multiple oral reading approach that focuses on reading aloud exercises to improve word recognition and support reading (see Table 2).

Main Results of Treatments

In terms of outcome, all reported treatment types showed some promising positive training results. However, the studies with lexical treatments led to better outcomes for a larger number of patients, namely, 33 out of 35 participants (Beeson et al., 2003; Beeson & Egnor, 2006; Clausen & Besson, 2003; Rapp, 2005; Rapp & Kane, 2002; Thiel & Conroy, 2014; Thiel et al., 2016). In the successful interventions, participants showed significant statistical increasing or large effect sizes on spelling accuracy of trained words. Some of them also reported improvement on untrained words with significant effects (Rapp, 2005; Rapp & Kane, 2002; Thiel & Conroy, 2014; Thiel et al., 2016) or minimal changes that were not supported with statistical analysis (Beeson & Egnor, 2006). In two studies with lexical treatments, the authors directly compared treatment effects in relation to the type of dysgraphia (i.e., orthographic output lexicon or the graphemic buffer). They reported that all subjects had improvements in repeated and delayed copying of words, but only patients with selective graphemic buffer deficit showed generalization effects to untrained words (Rapp, 2005; Rapp & Kane, 2002). Finally, a study with a CART protocol and combined oral repetition task assessed improvement in oral language, showing significant improvements in spoken lexicon in two patients with anomic and conduction aphasias (Beeson & Egnor, 2006).

Regarding phonological treatment, following the intervention, two out of three participants showed improvement in almost all measures for both treated and untreated words in writing to dictation and written naming tasks, and for treated oral spelling words except oral naming and irregular words. No significant improvements were found for one participant (Kiran, 2005).

However, all five participants who underwent interactive treatment showed significant enhancements from the start (Beeson et al., 2010; Kim et al., 2015). Statistical analyses in all the studies showed significant improvements or large effect sizes on the spelling accuracy of trained and untrained words. Beeson et al. (2010) reported a significant improvement in the spelling of untrained regular and irregular words for both of the two participants when using the electronic speller and for one of them also without using the device. Kim et al. (2015) reported an improvement in the reading rate of trained passages and in single-word speed-reading for three patients.

It is noteworthy that in some selected articles, participants previously received varying amounts of traditional language treatments (Beeson et al., 2003, 2010; Beeson & Egnor, 2006; Clausen & Besson, 2003; Kim et al., 2015), with some successful results from other writing treatment experiments focusing on writing (Clausen & Besson, 2003; Thiel et al., 2016) or spoken language skills (Beeson et al., 2010; Clausen & Besson, 2003); these studies showed a general responsiveness of patients to treatment. Finally, some authors pointed to participants' strong motivation to improve their written language skills (Beeson & Egnor, 2006; Beeson et al., 2010; Kim et al., 2015) as a possible factor in treatment responsiveness (Dalemans et al., 2010).

Intervention Characteristics

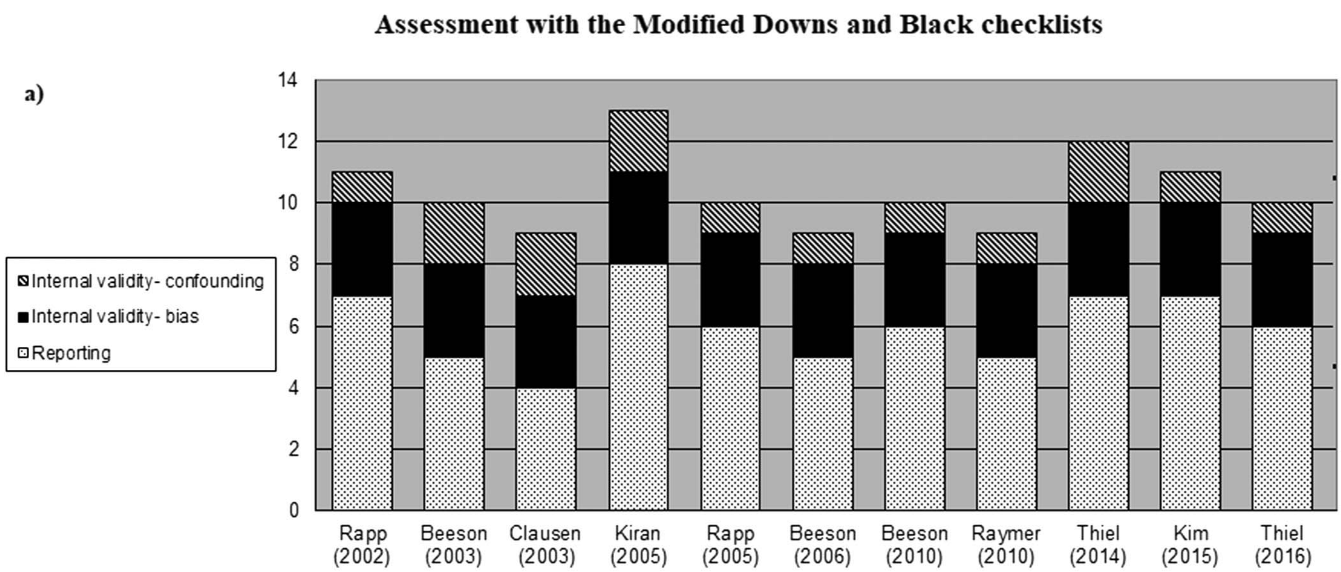
All the studies administered individual sessions of 1 to 2 hr in duration, 1 to 3 times per week. Two studies added weekly group sessions on oral language (Beeson et al., 2003; Clausen & Besson, 2003). In most studies, daily homework was also requested (Beeson et al., 2003, 2010; Beeson & Egnor, 2006; Clausen & Besson, 2003; Kim et al., 2015; Raymer et al., 2010). The treatment duration lasted 4 to 14 weeks; in the majority of cases, the treatments ended when the established criterion for each protocol was achieved (Beeson et al., 2003, 2010; Kiran, 2005; Rapp, 2005; Rapp & Kane, 2002; Raymer et al., 2010; see Table 2). Five studies had a follow-up assessment after 4 to 112 weeks (Rapp, 2005; Rapp & Kane, 2002; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016). Two studies had two different treatments applied in a cross-over design (Raymer et al., 2010; Thiel et al., 2016), and in two other studies, all participants received two different treatment approaches during each daily session, with an alternating order of administration from session to session (Beeson & Egnor, 2006; Thiel & Conroy, 2014).

Assessment of Studies' Quality

With respect to critical appraisal using the modified Downs and Black checklist, the "Reporting" subscale obtained the best score. None of the studies received scores on the "external validity" questions; the "internal validity – bias" subscale obtained a better score than the "internal validity – confounding" subscale (see Figure 3).

Concerning the reporting values, all the studies clearly described aims and interventions in terms of task procedures, characteristics of stimuli, and the duration and frequency of sessions. Moreover, most of the studies (seven of 11) clearly reported the main findings with raw data, except for four studies (Beeson et al., 2010; Rapp, 2005; Raymer et al., 2010; Thiel et al., 2016). However, outcomes were not always adequately detailed in the Method section but only in the Results section (Beeson et al., 2003; Beeson & Egnor, 2006; Clausen & Besson, 2003; Thiel & Conroy, 2014; Thiel et al., 2016). Most of the studies (seven of 11) reported the *p* values or effect sizes for the main outcomes. Only three studies described the chosen inclusion and exclusion criteria of the sample in terms of etiology, type of aphasia and dysgraphia, time postonset, and education (Kiran, 2005; Thiel & Conroy, 2014; Thiel et al., 2016). Particularly noteworthy is that only some studies clearly reported the type of dysgraphia (Beeson et al., 2010; Kim et al., 2015; Kiran, 2005; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016). The clinical representations of dysgraphia were generally well detailed, except in Clausen and Besson's (2003)

Figure 3. Assessment with the modified Downs and Black checklist: (a) graph of results and (b) reported results for each item and article.



b)

	ITEMS	Rapp (2002)	Beeson (2003)	Clausen (2003)	Kiran (2005)	Rapp (2005)	Beeson (2006)	Beeson (2010)	Raymer (2010)	Thiel (2014)	Kim (2015)	Thiel (2016)
Reporting	1	1	1	1	1	1	1	1	1	1	1	1
	2	1	0	0	1	1	0	1	1	0	1	0
	3	0	0	0	1	0	0	0	0	1	0	1
	4	1	1	0	1	1	1	1	1	1	1	1
	6	1	1	1	1	0	1	0	0	1	1	0
	7	1	1	1	1	1	1	1	1	1	1	1
	9	1	1	1	1	1	1	1	1	1	1	1
	10	1	0	0	1	1	0	1	0	1	1	1
External validity	11	0	0	0	0	0	0	0	0	0	0	0
	13	0	0	0	0	0	0	0	0	0	0	0
Internal validity - selection bias	16	1	1	1	1	1	1	1	1	1	1	1
	18	1	1	1	1	1	1	1	1	1	1	1
	20	1	1	1	1	1	1	1	1	1	1	1
Internal validity - confounding (selection bias)	21	0	0	0	1	0	0	0	0	1	0	0
	22	0	1	1	0	0	0	0	0	0	0	0
	26	1	1	1	1	1	1	1	1	1	1	1
	Total	*11*	*10*	*9*	*13*	*10*	*9*	*10*	*9*	*12*	*11*	*10*

study; however, they used different classifications since their assessments reflected different tools and tasks. Thus, some studies specified the types of dysgraphia using cognitive models (Beeson & Egnor, 2006; Beeson et al., 2010; Kim et al., 2015; Kiran, 2005; Thiel & Conroy, 2014; Thiel et al., 2016), whereas others only mentioned the damaged areas (Beeson et al., 2003; Beeson & Egnor, 2006; Clausen & Besson, 2003) or levels (e.g., damage to orthographic output lexicon; Kiran, 2005; Rapp, 2005; Rapp & Kane, 2002). Four studies (Beeson et al., 2003; Kim et al., 2015; Kiran, 2005; Thiel & Conroy, 2014) mentioned that some modifications to the protocol (such as additional treatment sessions, homework practices, additional tools, and repetition of training) had been applied to facilitate some participants to accomplish the exercises. These studies showed how the protocol could be adapted to the patients' clinical needs to optimize adherence and administration of training (see Table 2). Among the five studies that reported follow-up (Rapp, 2005; Rapp & Kane, 2002; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016), no performance loss was reported.

With respect to external validity, only two studies specified the source of population (Kiran, 2005; Thiel & Conroy, 2014), and none reported the entire recruitment procedures or described the inclusion and exclusion criteria. Some studies (Beeson & Egnor, 2006; Clausen & Besson, 2003; Thiel & Conroy, 2014) specified the treatment setting (i.e., a university environment or participant's home), but no studies explained whether these settings were representative among those in use (see Appendix B, Question No. 13). Most of the studies (Beeson & Egnor, 2006; Beeson et al., 2010; Clausen & Besson, 2003; Kim et al., 2015; Kiran, 2005; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016) specified that a clinician, a therapist, or an examiner conducted the individual treatment sessions but provided no other information about the practitioner (e.g., job profile, specialization).

Regarding internal validity, all the studies clearly described appropriate statistical techniques (chi-square test and effect size) to assess any change after rehabilitation. Four studies (Beeson et al., 2003; Clausen & Besson, 2003; Kiran, 2005; Thiel & Conroy, 2014) reported a high scoring of reliability as assessed in questions to control for confounding and selection bias (see Appendix B). Moreover, only in the work of Kiran (2005), a blind independent researcher conducted error analysis of the data to reduce the risk of bias. In all the studies, any other retrospective unplanned subgroup analysis was reported.

Finally, two studies (Rapp, 2005; Rapp & Kane, 2002) recognized the occurrence of possible confounders as reported in previous experiments in an attempt to control for confounding. In particular, they considered the severity of the subjects' writing deficits, level of memory, auditory single-word comprehension, and naming abilities. In all the studies, the confounders that may have had an effect on internal validity were age (Wallentin, 2018), time of onset, severity/extent of the lesion, severity and type of aphasia (Lazar & Antoniello, 2008; Pedersen et al., 2004; Watila & Balarabe, 2015) and dysgraphia at baseline, perceptual and spatial abilities (Rapp & Kane, 2002), any other language treatments or pharmacologic therapies underway at the same time (Plowman et al., 2012), and the presence of any other cognitive deficits (Seniów et al., 2009).

Additional Information on Patients and Treatment Characteristics

Participants' Profiles

From the entire sample of 43 participants, two took part in two different studies (Rapp, 2005; Rapp & Kane, 2002) but with different treatments and experimental designs. The 11 studies under review had a sample size that ranged from two to eight participants.

The age of the participants was between 39 and 80 years. They were all literate: Education ranged from 7 to 22 years, and they all had learned to write before brain damage occurred, as reported in the demographic data. Two studies did not specify the years of education, but mid–high instructional level could be assumed by mentioned work position/qualification (Rapp, 2005; Rapp & Kane, 2002). The etiology of cerebrovascular accident was homogeneous for all patients; most of them had a left hemisphere lesion, but some had no specification about localization. In all the studies, patients were in the chronic phase of disease (i.e., above 6 months), except for two patients in a subacute phase (i.e., at 2 months and 2 weeks postonset; Kim et al., 2015; Raymer et al., 2010). Only two studies specified the source population for recruitment: regional speech pathologists and hospitals of the Austin area in the United States (Kiran, 2005) and two stroke support groups (Thiel & Conroy, 2014).

Clinical Assessments

To assess both aphasia and dysgraphia, experimental tasks and standardized tests are used in the following: Western Aphasia Battery (Kertesz, 1982), Boston Diagnostic Aphasia Examination (Goodglass et al., 2001), Psycholinguistic Assessments of Language Processing in Aphasia (Kay et al., 1996), Johns Hopkins University Dysgraphia Battery (Goodman & Caramazza, 1985), Pyramids and Palm Trees Test (Howard & Patterson, 1992), Boston Naming Test (Kaplan et al., 2001), Arizona Battery for Reading and Spelling (Beeson & Rising, 2010), Gray Oral Reading Test–Third Edition (Wiederholt & Bryant, 1999), and Snodgrass and Vanderwart picture set (Snodgrass & Vanderwart, 1980). Some studies included evaluation of general cognitive functioning using the Wechsler Memory Scale–Revised (Wechsler, 1987) and Coloured Progressive Matrices (Raven et al., 1998).

Types of Deficits

We reported the types of dysgraphia as identified by the authors. Dysgraphias were heterogeneous (see Table 1). In some studies, such as that of Beeson et al. (2002), writing impairments were classified in accordance with cognitive models including phonological dysgraphia (Beeson et al., 2010; Kiran, 2005; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016), deep dysgraphia (Kiran, 2005; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016), graphemic buffer disorder (Rapp, 2005; Rapp & Kane, 2002; Thiel & Conroy, 2014; Thiel et al., 2016), surface dysgraphia (Kim et al., 2015; Thiel et al., 2016), and damage to the orthographic output lexicon (Kim et al., 2015; Rapp, 2005; Rapp & Kane, 2002). One participant had severe dysgraphia due to impairments of sublexical, orthographic, and buffer processes (Raymer et al., 2010). Other studies defined dysgraphia on the basis of impaired writing tasks (e.g., in written naming and writing to dictation tasks; Beeson et al., 2003; Beeson & Egnor, 2006). However, it should be noted that all the authors, except Clausen and Beeson (2003), provided detailed descriptions of the type of writing errors made, which are important for identifying and classifying dysgraphia based on the majority of errors produced but also for setting rehabilitative exercises.

Outcome Measures

Regarding specific intervention outcomes, all the studies except three explored the improvement of spelling accuracy of trained and untrained words. The three studies only focused on trained words, all with lexical treatments (Beeson et al., 2003; Clausen & Besson, 2003; Raymer et al., 2010). Moreover, two studies considered the spelling accuracy of repeated words to test for a possible repetition effect (Rapp, 2005; Rapp & Kane, 2002; see Table 1).

Most of the studies evaluated abilities at writing to dictation tasks (Beeson et al., 2003, 2010; Kim et al., 2015; Kiran, 2005; Rapp, 2005; Rapp & Kane, 2002; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016). Beeson et al. (2003) provided not only dictation but also a line drawing of the target word. In three other studies, a written naming task was used to assess spelling accuracy (Beeson & Egnor, 2006; Clausen & Besson, 2003; Kiran, 2005) and in two other studies (Rapp, 2005; Rapp & Kane, 2002), participants were asked to repeat the word before spelling it. Finally, accuracy of sound–letter and letter–sound correspondences (Beeson et al., 2010) and speed of spelling (Thiel & Conroy, 2014) were examined. In addition to these written language abilities, some studies assessed reading abilities, such as reading rate, accuracy, and reaction time (Kim et al., 2015), whereas others focused on oral language abilities such as oral naming, oral spelling accuracy, or comprehension (Beeson & Egnor, 2006; Kiran, 2005).

Regarding functional outcome, a few studies reported positive changes after lexical treatment by describing patient's increasing use of written language in communication (Beeson et al., 2003; Clausen & Besson, 2003; Thiel et al., 2016). However, only one study (Clausen & Besson, 2003), in which authors focused on the conversational use of written language, emphasized this aspect. This study collected data during conversational group sessions in which patients were encouraged to write the target words. Notwithstanding this study, no studies evaluated functional communication in terms of the actual use of written language in daily activities in a systematic way.

Generalization and Maintenance of Effects

With respect to the generalization of untrained outcomes for the lexical treatments, two studies showed positive results for all the participants (12 patients) with this intervention (Thiel & Conroy, 2014; Thiel et al., 2016). In another study, two patients with graphemic buffer disorders had a significant generalization, but the patient with orthographic output lexicon deficits did not (Rapp, 2005). It is not possible to determine whether the CART protocol had generalized effects, because no studies assessed untrained words using this protocol, and one study reported only qualitative minimal changes (without statistical analysis) under this protocol (Beeson & Egnor, 2006).

The two patients who benefited from phonological treatment had generalization in writing to dictation and written naming tasks for untrained regular words; however, this type of treatment did not prove useful for irregular words (Kiran, 2005). Finally, interactive treatments showed a generalization to untrained words for four patients both with and without the use of the electronic speller, whereas for one patient, the generalization was possible only with the support of the device (Beeson et al., 2010; Kim et al., 2015).

In relation to long-term learning, only the lexical treatment seemed to determine the maintenance of effects at follow-up assessments (Rapp, 2005; Rapp & Kane, 2002; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016). Every study reported some significant long-term benefits, but with differences between participants with similar impairments. Considering differences in patients and treatments, some authors suggested that the long-term therapy effects were likely related to certain skills and approaches that were not evaluated (Rapp, 2005).

Discussion

This systematic review examined 11 studies on treatments for acquired central dysgraphia and a similar number of articles appearing in ad hoc reviews in clinical rehabilitation (Innocenti et al., 2019; Nordio et al., 2018).

Although the studies were few, the total sample of 43 patients is an adequate number for analysis.

The majority of neurological patients showed some improvements in dysgraphia following methodological well-designed speech and language therapies, which is a sign of their applicability and efficacy. The methodological approach of the studies was a single-subject design, which allows for consideration of individual statistical results with some attention to methodological quality assessment.

Dysgraphia Improvement Effects

It is promising that all studies reported some improvements in patients after dysgraphia therapy. Among 43 participants, only three did not respond to treatment; however, these three had other co-occurring deficits that could have interfered with treatment, such as an impairment in auditory comprehension (Kiran, 2005) or difficulties in visual processing and severe impairment of the semantic system (Beeson et al., 2003). The majority of patients benefited from the treatments, despite the variety in the type and severity of deficits, time of onset of impairment, and type of treatment. It is important to consider that most of the patient's conditions were chronic. Thus, we can affirm that improvement in writing skills is possible in the chronic phase of the disease, a finding that is in agreement with the literature on the recovery of people with aphasia (Code & Herrmann, 2003; Meinzer et al., 2004; Moss & Nicholas, 2006).

In our assessment of methodological quality, all selected studies had a good level of reporting as demonstrated by an adequate explanation of aims and interventions. This detailed presentation was recognized as a strength that permitted the writing interventions to be clearly described (see Table 2).

We can summarize that all the reported treatments were based on impairment models that asked participants to "relearn" skills established before the condition (Thiel et al., 2015). Particular attention was given to improvement in single-word writing and the use of spelling procedures at semantic-lexical or sublexical levels (see the cognitive model of writing in Figure 1).

The largest group of studies evaluated lexical treatments (eight studies with 35 patients in total), with some positive results in both trained and untrained words, as well as different types of dysgraphia (Beeson et al., 2003; Beeson & Egnor, 2006; Clausen & Besson, 2003; Rapp, 2005; Rapp & Kane, 2002; Raymer et al., 2010; Thiel & Conroy, 2014; Thiel et al., 2016). Only one study showed benefits with a phonological treatment (Kiran, 2005), and two others showed significant outcomes with interactive treatments (Beeson et al., 2010; Kim et al., 2015). The latter were also used to support compensatory problem-solving recovery, but the extant literature still shows little evidence for conclusive findings (Thiel et al., 2015).

Importantly, some studies directly evaluated the relation between types of dysgraphia and written rehabilitation protocols. In one study, lexical treatment with spell–study–spell protocol resulted in improvements in all patients with spelling difficulties, but only those with a deficit in the graphemic buffer—and notably, none with a deficit in the orthographic output lexicon—demonstrated a generalization to untrained words (Rapp, 2005; Rapp & Kane, 2002). This finding may suggest a possible supporting involvement of semantic or sublexical preserved abilities. Phonological exercises are theoretically considered suitable for phonological dysgraphia (Beeson et al., 2010), but from the limited experimental evidence, it appears that they are also appropriate for individuals with deep dysgraphia (Kiran, 2005). Finally, in interactive treatments, patients with surface dysgraphia (e.g., damage to the semantic-lexical pathway) also seemed to benefit from combining phonological treatment and semantic tasks (i.e., reading and writing tasks that strengthen orthographic representation; Kim et al., 2015). Considering the presence of semantic-lexical damage, the phonological treatment was identified as mainly supporting sublexical processing (see Figure 1). However, no strong conclusions could be drawn because of the lack of evidence for phonological and interactive treatments.

Furthermore, some authors hypothesized that a dysgraphia treatment might also be suitable and beneficial for aphasic syndromes. Indeed, all patients had a lesion in the left hemisphere, which is mainly related to language deficits. Although the neurological etiology involved only vascular diseases and little details were reported about the types of aphasia in these patients, our review seems to confirm a trend toward the application of dysgraphia treatments in presence of aphasia (Beeson & Rapcsak, 2002). However, only one study with lexical treatment (i.e., CART protocol) and oral repetition task directly examined and reported significant positive effects on spoken lexical impairment in case of anomic and conduction aphasias, hypothesizing a potential involvement of residual phonological skills (Beeson & Egnor, 2006). Furthermore, descriptive evaluations indicate that lexical treatments with the CART protocol were considered appropriate for patients with severe nonfluent aphasia (Beeson et al., 2003; Clausen & Besson, 2003; Raymer et al., 2010). No other studies involving other treatments analyzed their effects among different aphasia syndromes. Therefore, it is not easy to predict the success of therapy in all aphasic patients. However, the potential increase of known written words (i.e., through exercise and retention of stored knowledge during treatment) could be an alternative way to communicate simple and personal relevant concepts effectively, especially when spoken language is critically impaired (Beeson & Egnor, 2006; Beeson et al., 2010).

With regard to clinical considerations in rehabilitation, it is relevant to address aspects of the treatments that

Table 1. Collected data on the included studies.

Study reference	Participants	Linguistic characteristics	Control group	Type of treatment
Lexical treatments				
Rapp & Kane, 2002	*N* = 2 (one woman, one man); age range: 58–67 years; education not specified; etiology: LH CVA; time of onset: > 2.5 years	Type of aphasia: not specified Type of dysgraphia: orthographic output lexicon deficit (*n* = 1), graphemic buffer deficit (*n* = 1)	No	One type: spell–study–spell treatment protocol
Beeson et al., 2003	*N* = 8 (three women, five men); age range: 65–79 years; education: 12–21 years; etiology: LH CVA; time of onset: > 2 years	Type of aphasia: nonfluent severe Broca (*n* = 7) and fluent Wernicke (*n* = 1) Type of dysgraphia: severe impairment of writing in written naming and in writing to dictation tasks (*N* = 8)	No	One type: CART protocol
Clausen & Besson, 2003	*N* = 4 (one woman, three men); age range: 61–72 years; education: 12–20 years; etiology: LH CVA; time of onset: > 6 years	Type of aphasia: nonfluent severe Broca (*N* = 4) Type of dysgraphia: not specified	No	One type: It is made of (a) individual treatment session (CART protocol), (b) group sessions of oral language, and (c) new person interaction
Rapp, 2005	*N* = 3 (two women, one man); age range: 58–67 years; education not specified; etiology: LH CVA; time of onset: > 2 years	Type of aphasia: not specified Type of dysgraphia: orthographic output lexicon deficit (*n* = 1), graphemic buffer deficit (*n* = 2)	No	One type: spell–study–spell treatment protocol
Beeson & Egnor, 2006	*N* = 2 (one woman, one man); age range: 60–72 years; education: 7–14 years; etiology: LH CVA; time of onset: > 61 months	Type of aphasia: fluent conduction (*n* = 1), nonfluent anomic (*n* = 1) Type of dysgraphia: severe impairment of writing in written naming and in writing to dictation tasks (*N* = 2)	No	Two types: (a) CART + repetition of target words and (b) only repetition of target words. In each session, the participant received both treatments; the order alternated from session to session.
Raymer et al., 2010	*N* = 4 (two women, two men); age range: 39–65 years; education: 12 years; etiology: CVA; time of onset: > 2 months	Type of aphasia: nonfluent anomic aphasia (*n* = 2), moderately severe nonfluent aphasia (*n* = 2) Type of dysgraphia: phonological dysgraphia (*n* = 2); severe impairments in sublexical, orthographic, and buffer processes (*n* = 1); deep dysgraphia (*n* = 1)	Two groups of two participants with cross-over design	Two types: (a) errorless treatment (CART adaptation) and (b) errorful treatment

Table 1. *(Continued).*

Study reference	Intervention (duration, frequency, and follow-up)	Outcomes	Main results
Lexical treatments			
Rapp & Kane, 2002	8–13 weeks of bi-weekly sessions. Treatment ended when the participant reached the criterion. Follow-up after 8, 12, and 20 weeks	Spelling accuracy of trained, repeated, and untrained words	Significant reduction of errors for both participants on treated words (P1, $\chi^2 = 45.0$, $p < .0001$; P2, $\chi^2 = 25.9$, $p < .0001$) and on repeated words (P1, $\chi^2 = 13.9$, $p < .0001$; P2, $\chi^2 = 7.5$, $p < .006$). Signs of generalized learning just for one patient (P1, $\chi^2 = 1.8$, $p > .18$ *ns*; P2, $\chi^2 = 28.8$, $p < .0001$). At follow-up, significant decrements for one patient for both conditions (P1 treated, $\chi^2 = 17.5$, $p < .0001$; P1 repeated, $\chi^2 = 4.6$, $p < .03$); for the other, decrements were significant just for treated items (P1 treated, $\chi^2 = 7.5$, $p < .006$; P1 repeated, $\chi^2 = 2.4$, $p > .12$ *ns*).
Beeson et al., 2003	8–14 weeks of weekly 1-hr sessions, daily homework, and 1 month of practice at home after the treatment. Treatment ended when the participant reached the criterion. One month of maintenance of just homework. No follow-up	Spelling accuracy of trained words Increasing use of written language to support communication	Large effect sizes (P1, $f = 3.20$; P2, $f = 1.03$; P3, $f = 0.97$; P4, $f = 0.84$; P5, $f = 1.19$; P6, $f = 0.99$) in written naming of trained items for six participants; however, one of them failed to reach the criterion, and another one had a modification to the treatment protocol. Small effect size for one participant (P7, $f = 0.23$) and no effect for one participant.
Clausen & Besson, 2003	13 weeks of weekly 1-hr individual sessions, with a month-long break; 15 weeks of weekly 1-hr–long group sessions; and daily homework. One month of maintenance of just homework. No follow-up	Spelling accuracy of trained words Increasing use of written language to support communication	Large effect sizes for all participants (P1, $f = 1.23$; P2, $f = 0.62$; P3, $f = 0.95$; P4, $f = 0.59$) on spelling of trained words following individual and group treatment; data were collected during the group setting.
Rapp, 2005	7–11 weeks of bi-weekly sessions. Treatment ended when the participant reached the criterion. Periodic follow-up after 8 to 112 weeks	Spelling accuracy of trained, repeated, and untrained words	Significant reduction of errors for all the participants on treated words (P1, $\chi^2 = 61.5$, $p < .001$; P2, $\chi^2 = 52.3$, $p < .001$; P3, $\chi^2 = 46.9$, $p < .001$) and on repeated words (P1, $\chi^2 = 18.2$, $p < .001$; P2, $\chi^2 = 22.2$, $p < .001$; P3, $\chi^2 = 9.9$, $p < .01$). Signs of generalized learning for the two individuals with graphemic buffer deficits (P1, $\chi^2 = 2.1$ *ns*; P2, $\chi^2 = 41.2$, $p < .001$; P3, $\chi^2 = 5.3$, $p < .05$). At follow-up, significant long-term benefits (P1 at 40 weeks treated, $\chi^2 = 12.4$, $p < .001$; P1 at 40 weeks repeated, $\chi^2 = 3.1$, $p < .05$; P2 at 2 years treated, $\chi^2 = 23.3$, $p < .001$; P2 at 2 years repeated, $\chi^2 = 15.2$, $p < .01$; P3 at 90 weeks treated, $\chi^2 = 6.63$, $p < .01$; P3 at 90 weeks repeated, $\chi^2 = 0.05$, $p < .05$).
Beeson & Egnor, 2006	10 weeks of bi-weekly sessions and daily homework. One month of maintenance of just homework. No follow-up	Oral naming and spelling accuracy of trained and untrained words	Large effect sizes for written naming (P1, $d = 9.49$; P2, $d = 16.62$) for both and one large and one small effect size (P1, $d = 17.41$; P2, $d = 3.46$) for spoken naming of trained words following CART with repetition; gains only in spoken naming (P1, $d = 6.51$; P2, $d = 2.62$) following repetition therapy; little change for untrained items (statistics are not reported for this measure)
Raymer et al., 2010	2 weeks of two to three 90-min sessions per week and homework 3 times a day, 5 days per week. Treatment ended when the participant reached the criterion. Follow-up after 1 month	Spelling accuracy of trained words	Positive effect sizes for trained words following each therapy: errorful training phase, three patients had large effect sizes (P1, $d = 21.4$; P2, $d = 15.54$; P4, $d = 13.14$), one positive (P3, $d = 4.0$); errorless training phase, three patients had large effect sizes (P1, $d = 13.0$; P3, $d = 5.92$; P4, $d = 7.41$), one positive (P2, $d = 5.17$). Advantages of errorful therapy in three participants. No notable changes for control words (errorful: P1, $d = 0.87$; P2, $d = 1.02$; P3, $d = 1.99$; P4, $d = 0.74$; errorless: P1, $d = -0.99$; P2, $d = 0$; P3, $d = 1.85$; P4, $d = 0.57$). At 1-month follow-up, there is maintenance for three participants for spelling of errorful trained words (P1, $d = 16.12$; P2, $d = 11.30$; P4, $d = 8.61$) whereas not for one (P3, $d = 2.96$) and for two for spelling errorless training words (P1, $d = 6.5$; P4, $d = 7.22$), but not for the other two (P2, $d = 1.94$; P3, $d = 5.36$); again, advantages of errorful training in three participants.

Table 1. *(Continued).*

Study reference	Participants	Linguistic characteristics	Control group	Type of treatment
Thiel & Conroy, 2014	*N* = 4 (one woman, three men); age range: 55–74 years; education: 10–12 years; etiology: CVA; time of onset: > 20 months	Type of aphasia: severe nonfluent aphasia (*n* = 1), severe expressive aphasia (*n* = 1), fluent aphasia (*n* = 2) Type of dysgraphia: deep dysgraphia (*n* = 2), phonological dysgraphia (*n* = 2)	No	Two types: (a) errorless treatment and (b) errorful treatment. In each session, participants received both conditions; the order alternated from session to session.
Thiel et al., 2016	*N* = 8 (three women, five men); age range: 47–80 years; education: 9–16 years; etiology: LH CVA; time of onset: > 6 months	Type of aphasia: nonfluent (*n* = 3), fluent (*n* = 5) Type of dysgraphia: deep dysgraphia (*n* = 2), surface dysgraphia (*n* = 2), phonological dysgraphia (*n* = 3), graphemic buffer disorder (*n* = 1)	Two groups of four participants each with cross-over design	Two types: (a) unimodal therapy and (b) multimodal therapy
Phonological treatments				
Kiran, 2005	*N* = 3 (three men); age range: 59–67 years; education: 16–22 years; etiology: LH CVA; time of onset: > 2 years	Type of aphasia: nonfluent motor transcortical (*n* = 1), nonfluent Broca (*n* = 1), nonfluent anomic (*n* = 1) Type of dysgraphia: impairments in orthographic output lexicon/orthographic level, impairment in phoneme to grapheme and grapheme to phoneme conversion (*n* = 2); deep dyslexia and dysgraphia with impairments in phoneme to grapheme and grapheme to phoneme conversion (*n* = 1)	No	One type: phonological treatment
		Interactive treatments		
Beeson et al., 2010	*N* = 2 (two women); age range: 46–76 years; education: 12–14 years; etiology: LH CVA; time of onset: > 5 years	Type of aphasia: fluent conduction (*n* = 1), nonfluent anomic (*n* = 1) Type of dysgraphia: phonological dysgraphia (*N* = 2)	No	One type: It is made of (a) phonological treatment and (b) interactive treatment.

Table 1. *(Continued).*

Study reference	Intervention (duration, frequency, and follow-up)	Outcomes	Main results
Thiel & Conroy, 2014	4 weeks of twice-weekly 90-min-long sessions. Follow-up after 5 weeks	Spelling accuracy posttherapy of trained and untrained words, and speed of spelling Increasing use of written language to support communication	Significantly improved spelling accuracy of treated and untreated words following both approaches for all participants ($p < .003$ for all). Only one participant showed an advantage of errorless over errorful learning ($\chi^2 = 4.17$, $df = 1$, $p = .04$); otherwise, there had been no differences between therapies ($\chi^2 = 0.20$, $df = 1$, $p = .66$). At follow-up, therapy effects overall had been maintained (errorless: $\chi^2 = 0.25$, $df = 1$, $p = .62$; errorful: $\chi^2 = 0.08$, $df = 1$, $p = .78$): accuracy scores did not change significantly for any condition. There is no effect of therapy type on spelling speed, $F(1, 169) = 0.80$, $p = .37$.
Thiel et al., 2016	3 weeks of five 1-hr-long sessions for each therapy (6 weeks in total), with a 2-week break between the two types of therapy. Follow-up at 6 weeks	Spelling accuracy of trained and untrained words immediately posttherapy	Accuracy of trained items posttherapy significantly improved on unimodal and multimodal therapy ($p < .01$ for all participants); there is no significant difference between the two treatments ($p = .31$). Accuracy of untreated items significantly improved immediately after both therapies ($p = .01$ for all participants); only one participant showed an advantage of multimodal over unimodal therapy (P1, $p = .03$); otherwise, there had been no differences between therapies (P3, $p = .78$; P4, $p = .46$; P7, $p = .80$; P2, $p = .20$; P5, $p = 1.00$; P8, $p = 1.00$; P6, $p = 1.00$). Scores of both treatments decreased significantly at follow-up (unimodal: $p = .02$; multimodal: $p = .01$), but individual results were mixed: three of them maintained both their multimodal and their unimodal scores (multimodal: P2, $p = 1.00$; P5 and P8, $p = .06$; unimodal: P2, $p = .13$; P5, $p = .13$; P8, $p = .06$).
Phonological treatments			
Kiran, 2005	From 10 to 20 bi-weekly 2-hr sessions (P1: 20 sessions; P2: 18 sessions; and P3: 10 sessions). Treatment ended when the participant reached the criterion. No follow-up	Accuracy in writing to dictation, written naming, oral spelling, oral naming of trained and untrained regular words, and 10 irregular words	Significantly improved writing to dictation of trained (P1, C = 0.916, s = 4.597, $p = .001$; P3, C = 0.681, s = 2.577, $p = .005$) and untrained words (P1, C = 0.705, s = 3.537, $p = .001$; P3, C = 0.811, s = 3.068, $p = .001$), written naming of trained (P1, C = 0.889, s = 4.460, $p = .001$; P3, C = 0.737, s = 2.788, $p = .002$) and untrained words (P1, C = 0.763, s = 3.829, $p = .001$; P3, C = 0.540, s = 2.044, $p < .05$), and oral spelling of trained words (P1, C = 0.909, s = 4.562, $p = .001$; P3, C = 0.631, s = 2.387, $p = .008$) for two participants. No improvements in oral naming. No changes in performances on irregular words. No significant improvements for one participant in all of the outcomes (he does not reach the criterion).
Interactive treatments			
Beeson et al., 2010	12–14 weeks of 1-hr sessions 2 or 3 times per week, and daily homework. Treatment ended when the participant reached the criterion. No follow-up	Sound–letter and letter–sound correspondences, and untrained nonword spelling accuracy	Significantly improved phonological processing, with positive effect sizes (letter–sound P1 consonants, $d = 2.62$, letter–sound P1 vowels, $d = 3.99$; letter–sound P2 vowels, $d = 0.95$; sound–letter P1 consonants, $d = 1.29$, sound–letter P1 vowels, $d = 2.52$; sound–letter P2 vowels, $d = 6.48$); improved spelling and reading via the sublexical route with positive effect size (P1, $d = 2.31$; P2, $d = 3.01$ on nonwords spelling); significantly improved spelling of untrained regular and irregular words for both participants when using the electronic speller (P1, $p = .0001$; P2, $p = .06$), for one participant also without (P1, $p = .021$; P2, $p = .113$ *ns*); significantly improved spelling accuracy (P1, $p = .0107$) and reading accuracy (P3, $p = .0107$) of treated nonword just for one participant.

Table 1. *(Continued).*

Study reference	Participants	Linguistic characteristics	Control group	Type of treatment
Kim et al., 2015	*N* = 3 (one woman, two men); age range: 72–80 years; education: 12–14 years; etiology: LH CVA (PCA); time of onset: > 2 weeks	Type of aphasia: nonfluent anomic aphasia Type of dysgraphia: surface dysgraphia and alexia (*n* = 2), surface dysgraphia and letter-by-letter reading (*n* = 1).	No	One type: It is made of (a) reading treatment (MOR approach) and (b) spelling treatment with a three-step problem-solving strategy.

can be generalized to predict long-term maintenance of improvements. Although a previous review found that lexical treatments had only item-specific improvements (Thiel et al., 2015), it can be expected that the effects could be generalized to untreated items, particularly for the few patients with phonological and interactive treatments. However, the type of dysgraphia could influence generalization. Moreover, long-term skill maintenance was assessed only for lexical treatments with positive but heterogeneous results. These aspects require further research. Finally, since these studies are based on a single-subject design, which is a valid methodological approach for smaller samples, recommendations for treatment selection are suggested at a single-patient level. In their overview of the interventions, the reported studies emphasized the promising gains and negligible or nonexistent harm that could follow the administration of dysgraphia treatments. The lexical treatments seemed to be the most common and have the most positive results; they also appeared to be the most promising in terms of generalized and long-term effects. Lexical treatment may be suitable for various types of dysgraphia and even applied in combination with phonological exercises, for example, in interactive treatments for compensatory problem-solving recovery (Thiel et al., 2015; see Figure 4). Additionally, the lexical treatments may be considered appropriate for patients with severe nonfluent aphasia, providing possible improvements in oral language (Beeson et al., 2003) and communication (Beeson et al., 2010; see Figure 4).

Additional Clinical Considerations for Application

Some papers focused on comparing different treatment modalities, which allows treatments to be tailored for individualized clinical purposes. Thus, some authors investigated the effects of multimodal versus unimodal methods on lexical treatments, achieving some improvements (Thiel et al., 2016). In addition, patients during multimodal treatments (considering studies with crossover design) appeared more flexible in both trained tasks and the proactive use of strategies, as observed during behavioral treatments (Thiel et al., 2016). Furthermore, in the only study with effects on aphasia, the authors combined a written lexical treatment and repetition tasks in a multimodal method (Beeson & Egnor, 2006). These considerations may support the effectiveness of this flexible approach, with potential adaptation to more complex language deficits.

Regarding the frequency of treatments, high intensity (i.e., from biweekly to daily sessions, including sessions with the clinician and at home) seems to be recommended for successful therapy (Beeson et al., 2003). However, the optimal frequency and intensity of treatment may depend on participants' characteristics, environmental factors, type of intervention, specific stimuli, and required responses (Cherney, 2012). Finally, the duration of the intervention varied between 4 and 14 weeks, with most treatments ending when patients reached the established criteria.

Our considerations appear to be in line with the indications described in previous literature (Thiel et al., 2015). In fact, they may demonstrate that treatments can be flexible and adaptable in terms of intensity and duration and, thus, tailored to individual patient's goals. In conclusion, the results suggest an encouraging relevance of the multimodal approach for its flexible administration and possible support in the recovery of patients with aphasia.

Limitations

Studies on speech and language therapy of acquired central dysgraphia are commonly characterized by small

Table 1. *(Continued).*

Study reference	Intervention (duration, frequency, and follow-up)	Outcomes	Main results
Kim et al., 2015	6 weeks of bi-weekly 1-hr sessions, and daily homework. One participant underwent 32 sessions of treatment over a 6-month period because of a fall and subsequent hospitalization. No follow-up	Reading rate on trained passages, reading rate and accuracy on a novel passage, reading reaction time, and spelling accuracy of trained and untrained irregular words	Significantly improved reading rate on trained passages, P1, t(4) = 3.40, p = .014; P2, t(16) = 9.88, p < .001; P3, t(8) = 2.39, p = .022); it seems there is generalization in reading rate and accuracy on a new novel, rate for the GORT-3: P1, t(10) = 7.02, p < .001; P2, t(7) = 3, p = .010; P3, t(9) = 3.30, p = .005). Significantly improved single-word speed reading, P1, t(144) = 4.12, p < .001; P2, t(143) = 4.73, p < .001; P3, t(123) = 9.57, p < .001, but word-length effect is not significant (P1, r = .774, p = .113; P2, r = .974, p = .013; P3, r = .796, p = .102). Significantly improved spelling accuracy, P1, $\chi^2(1)$ = 6.13, p = .013; P2, $\chi^2(1)$ = 8.1, p = .004; P3, $\chi^2(1)$ = 6.13, p = .013), also on untrained irregular words, P1, $\chi^2(1)$ = 5.14, p = .023; P2, $\chi^2(1)$ = 5.14, p = .023; P3 not reported.).

Note. Significant results are those with a p value of < .05. LH = left hemisphere; CVA = cerebrovascular accident; CART = Copy and Recall Treatment; PCA = posterior cerebral artery; MOR = multiple oral re-reading; GORT-3 = Gray Oral Reading Tests–Third Edition.

sample sizes, wide variability in dysgraphia and/or aphasia syndromes, and heterogeneity in types of assessments, treatments, and outcomes, and these limitations also affected the selected studies. Moreover, with regard to the assessment of methodological quality, only a few studies reported inclusion and exclusion criteria (in the reporting subscale) or recruitment procedures (for external validity bias). These aspects of the studies highlight their limitations in terms of replicability and representation of the population. This was partially overcome by a comprehensive description of the patients' deficits.

Furthermore, only trials written in English and single-subject design studies could be considered for the purpose of this review. It could be relevant to await further methodological implementation of writing treatment studies, including those in different languages.

In addition to the positive therapeutic outcomes, this review highlights the need for further research on the specific factors involved in the recovery of patients with writing deficits or their responsiveness to treatment and generalization (e.g., pharmacological therapies, severity/extent of the lesion, severity of aphasia, time of onset and age, perceptual and spatial processes), following the trend in neurological rehabilitation research.

Conclusions

This study provides a detailed summary of speech and language therapies for neurological patients with acquired central dysgraphia (see Table 2), including all lexical, phonological, and interactive treatments. Importantly, the majority of the reviewed studies showed positive gains following treatment targeting written language abilities, despite the variability in patients' linguistic and spelling abilities and the varying type, frequency, and duration of treatments. Nevertheless, these results are promising and indicate the clinical applicability of writing treatments for patients with left hemisphere damage.

However, it seems to be difficult to clearly establish a predominant therapy, as mentioned in a previous narrow review (Thiel et al., 2015), mainly due to a lack of studies and direct comparisons between treatment types and restricted evaluations of everyday life abilities. Linking the classification of dysgraphia with the treatments allowed to obtain a summary of interventions tailored to diagnosis (see Figure 4), while reading the original studies for ad hoc clinical applications, is recommended.

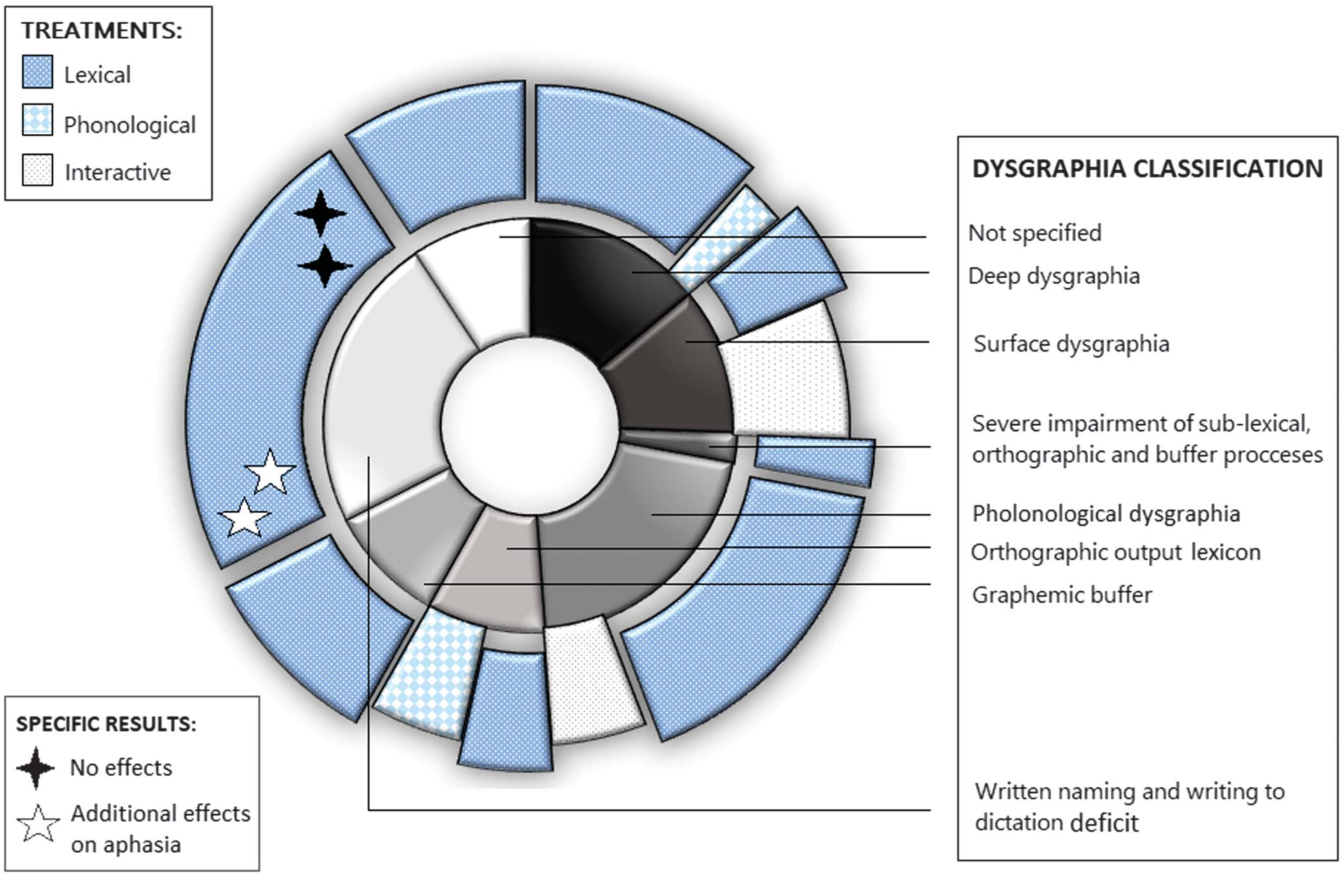
The lexical treatments appear to be the most frequently used and those that could bring greater improvements with different types of dysgraphia and in severe cases of nonfluent aphasia. To optimize therapeutic outcomes, treatments should allow adaptation and flexibility according to patients' needs. Adaptation of treatments can be achieved by monitoring and adjusting the intensity and duration of the sessions. In addition, flexibility can be obtained by applying a multimodal approach to encourage a variety of abilities and the use of strategies during training to maximize the potential for transferability of these skills to patients' daily life. Additionally, most treatments report a generalization of effects to untreated items. However, long-term maintenance of learned skills was confirmed only in studies with lexical treatments, although the mixed outcomes in relation to patients' type of dysgraphia suggest the need for further investigations.

Ultimately, the results of our systematic review suggest that rehabilitation in the field of dysgraphia is still at

Table 2. Description of treatments protocols.

Study reference	Lexical treatments
Copy and Recall Treatment (CART) (Beeson et al., 2003; Clausen & Besson, 2003)	Homework protocol that includes repeated copying and recall trials for target words. Each target word has to be copied at least 20 times per day, 6 days per week. The copying task is followed by a self-test of the recall for practiced words. The criterion for mastering of spelling is defined as a participant's ability to write at least four of five words in a set over a minimum of two consecutive sessions. After a participant met the criterion on one set of target words, training began on the next set. Continued practice with the mastered words was still included in homework practice. In the work of Beeson et al. (2003), modification to facilitate patients' participation was applied, as additional semantic training or treatment sessions.
Spell–study–spell treatment protocol (Rapp, 2005; Rapp & Kane, 2002)	Three sets of words (n = 30 for each test) are created: treated, repeated, and control sets. For the treated items, the participant hears a word, repeats it, and attempts to spell it. If the word is not spelled correctly, the experimenter shows a card with the correct spelling and says each letter aloud. The participant has no time restriction to study it. After that, a delayed copy of the target word is required. All the three sets are presented during the pretreatment assessment, the posttreatment evaluation, and the follow-up phase. Only the treated and the repeated sets were administered in a spelling-to-dictation task, during every session of the treatment. Treatment sessions were discontinued when subjects reached the treatment goal of stable performance with fewer than 5% letter errors on the treated words.
Repetition (Beeson & Egnor, 2006)	It involves repetition of the target words, in association with CART. The criterion for mastering of spelling was defined as a participant's ability to write at least four of five words in a set across two consecutive sessions.
Errorless treatment (Raymer et al., 2010; Thiel & Conroy, 2014)	This approach can be incorporated in different types of treatments (e.g., CART). The goal is to minimize errors as far as possible. In the work of Raymer et al. (2010), an adaptation of the CART protocol, incorporating systematic vanishing cues, was used. The treatment phase ended when the criterion of 90% correct across two probe sessions was met or when 2 weeks of training had elapsed. In the work of Thiel and Conroy (2014), the treatment was administered in eight sessions in 4 weeks.
Errorful treatment (Raymer et al., 2010; Thiel & Conroy, 2014)	This approach can be incorporated in different types of treatments and consists of using progressive orthographic cues in the task, revealing one letter at a time that the participant can copy and then attempt to continue spelling by memory. In the work of Raymer et al. (2010), the treatment phase ended when the criterion of 90% correct across two probe sessions was met or when 2 weeks of training had elapsed. On the contrary, in the work of Thiel and Conroy (2014), the treatment was administered in eight sessions in 4 weeks. In the work of Raymer et al. (2010), modification to facilitate a patient's participation was applied with repetition of training (i.e., a second errorful training phase for inconsistencies in previous homework).
Unimodal therapy (Thiel et al., 2016)	This approach consists of only one task at a time, for example, a word copy task.
Multimodal therapy (Thiel et al., 2016)	This approach of therapy involves multiple tasks, for example, for each target word, there can be a semantic distractors task, a phonological task, and an orthographic distractors task. Participants received 5-hr sessions of both unimodal and multimodal therapy (10 hr in total), which took place over 3 weeks with a 2-week break between the two types of therapy.
	Phonological treatments
Phonological treatment (Kiran, 2005)	Targeted to strength sound–letter correspondences. Participants have to write to dictation 10 regular words through a series of steps that emphasized phoneme to grapheme conversion rules: (a) writing to dictation of the word, (b) copying the word, (c) oral reading of the word, (d) selecting and writing the sounds of the target word from distractors in the accurate sequence, (e) writing phonemes of the target word presented orally but randomly, and (f) writing to dictation of the word. Treatment was discontinued when writing to dictation of trained items was 80% accurate over two consecutive sessions or when 10 sessions elapsed with no improvements. Modification to facilitate patients' participation was applied, by providing a tool (as an alphabet sheet) or training (starting session with training with the alphabet sheet) and homework practice.
	Interactive treatments
Interactive treatment (Beeson et al., 2010)	Treatment was implemented using a cueing hierarchy to retrain the relations between graphemes and phonemes for 20 consonants and 12 vowels before to train the spelling on sets of 20 regularly spelled words and 20 nonwords presented aurally. Training was implemented for each set of phonemes until the participant was able to correctly write and say each set with 80% accuracy over two sessions. Moreover, it uses an electronic device to check and correct spelling errors at the end of the word target writing.
Multiple oral rereading (MOR) approach (Kim et al., 2015)	Text-based treatment approach used to remediate letter-by-letter reading, improving word recognition and promoting interactive processing of all available information to support reading. The same text has to be read aloud until a criterion of 100 words per minute for passage was reached. After that, a new passage was assigned after four treatment sessions even if the criterion is not met to maintain interest and compliance. The goal of MOR treatment is to improve word recognition and to promote interactive processing of all available information to support reading (problem-solving strategy). Moreover, it uses an electronic device to check and correct spelling errors at the end of the word target writing. Modification to facilitate patients' participation was applied with additional training sessions (due to disruption of hospitalization).

Figure 4. Summary chart of frequency and combination of treatments for dysgraphia in the studies. All treatments reported positive significant effects, with the exception of two patients (out of 43). In addition, positive effects on aphasia were reported in two patients.



an early stage. Nevertheless, according to the hierarchical levels of evidence proposed by the American Speech-Language-Hearing Association (Taylor-Goh, 2005), all included papers were rated as IIb (well-designed quasi-experimental studies); therefore, the efficacy of the dysgraphia treatments has a low level of evidence. To reduce the risk of bias and improve external validity, future trials are needed, which employ larger sample sizes, more robust designs with a control group and randomized allocation, homogeneous groups of patients (e.g., in terms of time postonset and type of dysgraphia profile), and similar methods of assessment, including that of common outcomes and daily life skills.

In conclusion, while the rehabilitation of central dysgraphia still requires further investigation, the reported improvements across previous studies are a sign of its applicability and efficacy. This systematic review highlights the importance of considering the deficits of the patients and their expected outcomes in choosing adequate interventions. In particular, we encourage knowledge of the available treatments to provide insight in guiding clinical practices (see Table 2). Considering that the lexical treatments along with a multimodal approach were found to be the most efficient in addressing several writing difficulties (see Figure 4), tailoring the intervention to the patient may also be significant in maximizing improvements and autonomy. Writing performance is relevant to daily activities and positively impacts the communicative potential of aphasic patients (Beeson & Rapcsak, 2002; Clausen & Besson, 2003). In this regard, it is crucial for further research to also consider the impact of treatments on patients' quality of life, taking into account the written language skills required in everyday activities.

Statement of Ethics

The review protocol was registered in PROSPERO (International Prospective Register of Systematic Reviews) with registration number CRD42018084221. We followed the PRISMA 2020 guidelines for reporting. Published research complies with the guidelines for human studies and includes evidence that the research was conducted ethically in accordance with the World Medical Association Declaration of Helsinki.

Author Contributions

Federica Biddau: Conceptualization (Lead), Data curation (Lead), Writing – original draft (Lead), Writing – review & editing (Equal). **Camilla Brisotto:** Data curation (Lead), Writing – original draft (Equal), Writing – review & editing (Supporting). **Tiziano Innocenti:** Methodology (Lead). **Sara Ranaldi:** Data curation (Supporting), Writing – original draft (Supporting). **Francesca Meneghello:** Conceptualization (Equal), Supervision (Supporting). **Daniela D'Imperio:** Methodology (Supporting), Supervision (Lead), Writing – review & editing (Lead). **Sara Nordio:** Conceptualization (Lead), Data curation (Lead), Writing – original draft (Lead), Writing – review & editing (Lead).

Data Availability Statement

All data generated or analyzed during this study are included in this published article.

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Weekes, B. S. (2005). Acquired disorders of reading and writing: Cross-script comparisons. *Behavioural Neurology, 16*(2–3), 51–57. https://doi.org/10.1155/2005/492935

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Appendix A

Search Terms and Strategies Specific for Each Database

PubMed:

(((((treatment) OR “Treatment Outcome”[Mesh]) OR “Speech Therapy”) OR “Speech Therapy”[Mesh])) AND (((((((“writing impairment”) OR “writing disorder”) OR “acquired dysgraphia”) OR “acquired agraphia”) OR “dysgraphia”) OR “Agraphia”) OR “Agraphia”[Mesh])

Filters: Adult: 19+ years

Embase:

‘dysgraphia’/exp OR dysgraphia OR ‘agraphia’/exp OR agraphia AND (‘treatment’/exp OR treatment OR ‘speech and language therapy’/exp OR ‘speech and language therapy’) AND ([adult]/lim OR [aged]/lim OR [middle aged]/lim OR [very elderly]/lim OR [young adult]/lim)

Web of Science:

Dysgraphia AND treatment

Cochrane Central Register of Controlled Trials (CENTRAL):

Dysgraphia AND treatment

Google Scholar:

Dysgraphia treatment

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Modified Downs and Black Checklist for the Assessment of the Methodological Quality of Single-Subject Designs

Item	Criteria	Possible answers
Reporting bias		
1	*Is the hypothesis/aim/objective of the study clearly described?*	Yes = 1 No = 0
2	*Are the main outcomes to be measured clearly described in the Introduction or Method section?* If the main outcomes are first mentioned in the Results section, the question should be answered *no*.	Yes = 1 No = 0
3	*Are the characteristics of the patients included in the study clearly described?* In cohort studies and trials, inclusion and/or exclusion criteria should be given. In case–control studies, a case definition and the source for controls should be given.	Yes = 1 No = 0
4	*Are the interventions of interest clearly described?* Treatments and placebo (where relevant) that are to be compared should be clearly described.	Yes = 1 No = 0
6	*Are the main findings of the study clearly described?* Simple outcome data (including denominators and numerators) should be reported for all major findings so that the reader can check the major analyses and conclusions. (This question does not cover statistical tests, which are considered below).	Yes = 1 No = 0
7	*Does the study provide estimates of the random variability in the data for the main outcomes?* In nonnormally distributed data, the interquartile range of results should be reported. In normally distributed data, the standard error, standard deviation, or confidence intervals should be reported. If the distribution of the data is not described, it must be assumed that the estimates used were appropriate and the question should be answered *yes*.	Yes = 1 No = 0
9	*Have the characteristics of patients lost to follow-up been described?* This should be answered *yes* where there were no losses to follow-up or where losses to follow-up were so small that findings would be unaffected by their inclusion. This should be answered *no*, where a study does not report the number of patients lost to follow-up.	Yes = 1 No = 0
10	*Have actual probability values been reported (e.g., 0.035 rather than < 0.05) for the main outcomes except where the probability value is less than 0.001?*	Yes = 1 No = 0
External validity – bias		
11	*Were the subjects asked to participate in the study representative of the entire population from which they were recruited?* The study must identify the source population for patients and describe how the patients were selected. Patients would be representative if they comprised the entire source population, an unselected sample of consecutive patients, or a random sample. Random sampling is only feasible where a list of all members of the relevant population exists. Where a study does not report the proportion of the source population from which the patients are derived, the question should be answered *unable to determine*.	Yes = 1 No = 0 Unable to determine = 0
13	*Were the staff, places, and facilities where the patients were treated representative of the treatment the majority of patients receive?* For the question to be answered *yes*, the study should demonstrate that the intervention was representative of that in use in the source population. The question should be answered *no* if, for example, the intervention was undertaken in a specialist center unrepresentative of the hospitals most of the source population would attend.	Yes = 1 No = 0 Unable to determine = 0
Internal validity – bias		
16	*If any of the results of the study were based on "data dredging," was this made clear?* Any analyses that had not been planned at the outset of the study should be clearly indicated. If no retrospective unplanned subgroup analyses were reported, then answer *yes*.	Yes = 1 No = 0 Unable to determine = 0
18	*Were the statistical tests used to assess the main outcomes appropriate?* The statistical techniques used must be appropriate to the data. For example, nonparametric methods should be used for small sample sizes. Where little statistical analysis has been undertaken but where there is no evidence of bias, the question should be answered *yes*. If the distribution of the data (normal or not) is not described, it must be assumed that the estimates used were appropriate and the question should be answered *yes*.	Yes = 1 No = 0 Unable to determine = 0
20	*Were the main outcome measures used accurate (valid and reliable)?* For studies where the outcome measures are clearly described, the question should be answered *yes*. For studies that refer to other work or demonstrate that the outcome measures are accurate, the question should be answered *yes*.	Yes = 1 No = 0 Unable to determine = 0

(*table continues*)

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Modified Downs and Black Checklist for the Assessment of the Methodological Quality of Single-Subject Designs

Item	Criteria	Possible answers
Internal validity – confounding (selection bias)		
21	*Were the patients in different intervention groups (trials and cohort studies) or were the cases and controls (case–control studies) recruited from the same population?* For example, patients for all comparison groups should be selected from the same hospital. The question should be answered *unable to determine* for cohort and case–control studies where there is no information concerning the source of patients included in the study.	Yes = 1 No = 0 Unable to determine = 0
22	*Were study subjects in different intervention groups (trials and cohort studies) or were the cases and controls (case–control studies) recruited over the same period of time?* For a study, which does not specify the time period over which patients were recruited, the question should be answered *unable to determine*.	Yes = 1 No = 0 Unable to determine = 0
26	*Were losses of patients to follow-up taken into account?* If the numbers of patients lost to follow-up are not reported, the question should be answered *unable to determine*. If the proportion lost to follow-up was too small to affect the main findings, the question should be answered *yes*.	Yes = 1 No = 0 Unable to determine = 0

Note. Items are reported with original numbers, and missing numbers are those excluded.

Appendix C

Excluded Studies With Specified Rejection Justifications

Authors	Title	Rejection criteria
Seron et al., 1980	A computer-based therapy for the treatment of aphasic subjects with writing disorders	It doesn't fit selection criteria in terms of etiology (one patient is cancer)
Basso et al., 1982	Pattern of recovery of oral and written expression and comprehension in aphasic patients	Different aim (the aim is to find the correlation between the four areas of language; there is no specific treatment)
Beeson et al., 2000	Problem-solving approach to agraphia treatment: interactive use of lexical and sublexical spelling routes	It does not fit selection criteria in terms of design of the study (it is a case series)
Luzzatti et al., 2000	Rehabilitation of spelling along the subword-level routine	Different aim (the aim is to analyze the relationship between recovery in oral and written expression and comprehension)
Papathanasiou et al., 2003	Plasticity of motor cortex excitability induced by rehabilitation therapy for writing	Different aim (the aim is to investigate how mechanisms of rehabilitation can induce plasticity in motor system)
Krajenbrink et al., 2017	No effect of orthographic neighborhood in treatment with two cases of acquired dysgraphia	Different aim (the aim is to further examine the role of orthographic neighborhood size in both treatment effects and generalization)
Thiel et al., 2016	The role of learning in improving functional writing in stroke aphasia	Different aim (the aim is to consider functional writing and perception of disability)
Tao & Rapp, 2019	The effects of lesion and treatment-related recovery on functional network modularity in poststroke dysgraphia	It does not fit selection criteria in terms of design of the study (it is a cohort study, without a control group)