Abstract

Executive functions (EFs) refer to a set of cognitive processes, specifically shifting, inhibition, updating of working memory, and are involved in the cognitive control of behavior. Conflicting results have been reported regarding impairments of EFs in Primary Progressive Aphasia (PPA). We performed a multi-level meta-analysis to confirm whether deficits of EFs exist in this population, focusing on a common EFs composite, and the components shifting, inhibition and updating separately. We included 141 studies that report on 294 EFs tasks. The overall mean weighted effect size was large (d = -1,28), indicating poorer EFs in PPA as compared to age-matched cognitively healthy controls. Differences between effect sizes of the EFs components were not significant, indicating all components are affected similarly. Overall, moderator analysis revealed that PPA variant and disease duration were significant moderators of performance, while task modality and years of education were not. The non-fluent/agrammatic PPA and the logopenic PPA variants were similarly affected, but the semantic variant was affected to a lesser extent. We discuss implications for clinical and research settings, and future research.

Keywords

Primary Progressive Aphasia, Executive Functions, Meta-Analysis, Neuropsychology, Dementia

Manuscript

1. Introduction

Primary Progressive Aphasia (PPA) is a neurodegenerative syndrome, differentiated from other dementia syndromes by early and predominant speech and language symptoms. There are currently three clinical variants of PPA according to the most recent consensus (e.g., Gorno-Tempini et al., 2011): a nonfluent/agrammatic variant PPA (NfvPPA), a logopenic variant PPA (LvPPA) and a semantic variant PPA (SvPPA) (see Table 1). A diagnosis of PPA and its classification into three clinical variants is based on patterns of atrophy, underlying pathology and, for the most part, on the relative presence and/or absence of speech and language features. According to the current diagnostic guidelines, the speech and language symptoms are the main factors causing impairment of daily activities at onset and during the early stages of the disease. In these early stages, other cognitive functions, such as memory, visuospatial skills and executive functions (EFs) are supposed to remain relatively intact (Mesulam, 2001, 2003; Mesulam et al., 2012). Thus, EFs have long been assumed to be unimpaired in patients with PPA (at least until late stages of the disease) and as such have rarely been the main subject of investigation in this population. However, studies providing data on EFs in PPA have in fact reported deficits relating to EFs in all variants of PPA (Foxe et al., 2021; Hutchinson & Mathias, 2007; Wicklund et al., 2007), even in earlier stages of the disease.

Table 1. *Features of PPA variants, according to (Gorno-Tempini et al., 2011)*

	NfvPPA	LvPPA	SvPPA
Clinical features	At least one of the following primary features must be present: • Agrammatism in language production • Effortful, halting speech (e.g., apraxia of speech) Additionally, at least two of the following features must be present: • Impaired comprehension of syntactically complex sentences • Spared single-word comprehension • Spared object knowledge	Both of the following primary features must be present: • Impaired single-word retrieval in spontaneous speech and naming • Impaired repetition of sentences and phrases Additionally, at least 3 of the following features must be present: • Phonologic errors in spontaneous speech and naming • Spared single-word comprehension and object knowledge • Spared motor speech • Absence of frank agrammatism	Both of the following primary features must be present: • Impaired confrontation naming • Impaired single-word comprehension Additionally, at least three of the following features must be present: • Impaired object knowledge, particularly for low-frequency items • Surface dyslexia or dysgraphia • Spared repetition • Spared speech production

Most prominent location of atrophy*	• left posterior fronto-insular region	• left posterior perisylvian or parietal region	• anterior temporal lobe
Most commonly associated pathology	• FTLD-tau (52%) (Mesulam et al., 2008)	• AD (50-60%) (Mesulam et al., 2008)	• FTLD-TDP-43 (69-83%) (Gorno-Tempini et al., 2011)
PPA= Primary Progressive Aphasia; LvPPA= logopenic/phonological variant PPA; SvPPA= semantic variant PPA; NfvPPA= nonfluent/agrammatic variant PPA; AD= Alzheimer's Disease; FTLD-TDP-43= Frontotemporal Lobar Dementia with ubiquitin and Transactive response DNA binding Protein kDa (TDP-43) pathology; FTLD-tau= Frontotemporal Lobar Dementia with tau-positive pathology; *Disease epicenters. Damage can progress and become more widespread, including white matter (Acosta-Cabronero et al., 2011; Agosta et al., 2012; Galantucci et al., 2011; Mahoney et al., 2013) and functional connectivity (Agosta et al., 2014; Bonakdarpour et al., 2019; Guo et al., 2013; Yuan Tao et al., 2020; Whitwell et al., 2015) alterations.			

1.1 Executive Functions and their assessment: theoretical framework

"Executive functions" are considered a multidimensional construct, referring to higher-level cognitive functions (i.e., planning, reasoning, problem solving, working memory, inhibitory control, cognitive flexibility) involved in the control and regulation of lower-level cognitive processes and independent, goal-directed behavior (Alvarez & Emory, 2006). Within the field of cognition and neuropsychology, conceptualizations vary regarding which cognitive processes or components actually define EFs as a construct. The present meta-analysis focuses on three established components of EFs, based on the seminal factor model of 'unity and diversity' by Miyake et al. (2000), pp.54-58: (1) 'inhibition of prepotent responses' (inhibition), (2) 'shifting between mental states' (shifting), and (3) 'updating and monitoring of working memory representations' (updating). Inhibition refers to one's ability to deliberately inhibit dominant, automatic, or prepotent responses when necessary (Miyake et al., 2000) and is often evaluated with for instance the Stroop task. Shifting involves shifting back and forth between multiple tasks, operations or mental sets (Kiesel et al., 2010; Monsell, 1996), often assessed with shifting tasks such as Trail Making Test B. Updating involves monitoring and coding incoming information for relevance to the task at hand and appropriately revising the items held in working memory by replacing old, no longer relevant information with new, more relevant information (Morris & Jones, 1990). Importantly, updating is considered to require active manipulation of relevant information in working memory, next to storing and monitoring information (e.g., operation span tasks). The confirmatory factor analysis by Miyake et al. (2000) showed that the three EFs components are separable ('diversity'), but share at least some commonality and are thus not considered to be completely independent ('unity'). In their later work, 'unity' is described by a 'Common EF' latent variable on which tasks for all EFs components load (Friedman et al., 2008). Importantly, this theoretical framework of the unity and diversity model allows for a classification of eleven widely used tasks for EFs into the three EFs components, which can be used for a well-grounded comparison of EFs across studies and will also be the basis for examining EFs in PPA in this meta-analysis. (See

Supplementary Material 3 Appendix A for further explanation and see methods section 3.1.2 for our task classification).

Neuroanatomically, EFs are for a large part sustained by the frontal and prefrontal cortices and their connections to the sub-cortex, with lesion studies linking the dorsolateral prefrontal cortex (DLPFC) to response inhibition, set shifting, and updating (Bettcher et al., 2016; Stuss, 2011). Interestingly, these regions can be affected in PPA(Agosta et al., 2014; Mandelli et al., 2018; Y. Tao et al., 2020).

1.2 Executive Functions and language

The idea of a reciprocal relationship between EFs and language abilities is long-standing (Baddeley & Hitch, 1974; Ullman, 2001), and is validated by several lines of research.

In cognitively healthy populations, associations between lexical-semantic processing and inhibition (Bilenko et al., 2009; Khanna & Boland, 2010), and updating of working memory have been suggested (Gathercole & Baddeley, 1990; Khanna & Boland, 2010). Syntactic processing and sentence production has been linked to inhibition (Kaushanskaya et al., 2017) and updating of the working memory (Daneman & Carpenter, 1980; Slevc, 2011).

Further, the relationship between EFs and language has often been examined in populations with language impairments. Children with language impairments have been found to perform worse on tasks for inhibition (Im-Bolter et al., 2006; Pauls & Archibald, 2016), shifting (Marton, 2008; Pauls & Archibald, 2016) and updating of the working memory (Henry et al., 2012). In adults, this has mainly been investigated in patients with post-stroke aphasia, with people with aphasia performing worse on tasks for shifting (Frankel et al., 2007), inhibition (McCarthy & Kartsounis, 2000; Murray et al., 1997) , and updating of the working memory (Frankel et al., 2007). This research provides evidence that overlap of neural domains of language and domain-general cognitive processes (i.e., EFs) has been found to lead to deficits in both domains in post-stroke aphasia (Purdy, 2002).

The notion of a connection between language and EFs is further exemplified in the literature on bilingualism and EFs, where bilingual language control is the mechanism allowing bilingual speakers to correctly speak in one language without interference of the other language (e.g., (De Baene et al., 2015; Declerck et al., 2021). This language control may be exerted by engaging a network of cortical and subcortical brain areas closely related to EFs (Abutalebi & Green, 2007; Green & Abutalebi, 2013).

1.3 Executive Functions impaired in PPA?

While EFs processes have often been considered to be intact in the PPA population, impairments have been found across all of the PPA variants as the disease progresses, even in the earlier stages of the disease (Bozeat et al., 2000; Macoir et al., 2017; Rohrer et al., 2010). Two recent meta-analyses investigated measures of EFs in PPA, next to a broad spectrum of other neuropsychological functions (Kamath et al., 2019; Kamath et al., 2020). The authors defined the subdomains of EFs as response inhibition, visual set-shifting and concept

formation. In the first meta-analysis, a comparison was made between patients with NfvPPA, SvPPA and behavioral variant frontotemporal dementia, which is a type of dementia marked by executive dysfunctions. The authors found comparable effect sizes for deficits on EFs between NfvPPA and behavioral variant frontotemporal dementia participants, while the SvPPA group had a significant lower effect size (Kamath et al., 2019). The second meta-analysis focused on LvPPA, where deficits in the EFs subdomain of visual set-shifting were found to be as prominent as their language difficulties. Not enough studies were included to evaluate other subdomains of EFs or to compare with the other PPA variants (Kamath et al., 2020).

Next to EFs, impairments of other cognitive functions other than language have been reported in PPA. A recent meta-analysis focusing on memory found episodic and working memory deficits in all PPA variants compared to healthy controls. The authors suggest that one contributing factor to these memory problems could be executive dysfunction affecting encoding and retrieval of memory (Eikelboom et al., 2018).

While these meta-analyses provide valuable insights into the connections between EFs and PPA, they do not systematically investigate the three components of EFs that are typically considered to be the subcomponents of EFs, and/or other aspects of EFs that are less well-understood. Regarding the components shifting, inhibition and updating, some studies show the existence of impairments (Y. Chen et al., 2018; Clark et al., 2015; Foxe et al., 2013; Magnin et al., 2013), while others do not (Benhamou et al., 2020; Borghesani et al., 2020; Ramanan et al., 2021). Because of these conflicting results, we cannot conclusively say that EFs are impacted in PPA. Shedding light on this matter is the main objective of the current meta-analysis.

Moreover, the meta-analyses discussed above reinforce preliminary indications that aspects of EFs dysfunction may be distinct according to PPA variant (Benhamou et al., 2020; Y. Chen et al., 2018; Kumfor et al., 2011; Macoir et al., 2017). The current meta-analysis can help elucidate on differences in EFs between the PPA variants.

1.4 The complexity of assessing Executive Functions in PPA

As is generally the case for EFs assessment (see Supplementary material 3, Appendix A), research in PPA has used an extensive variety of tests purporting to measure EFs. Few studies have focused on EFs in PPA specifically, so most reporting on EFs in the PPA literature comes from basic neuropsychological assessments, usually with the goal of establishing baseline cognitive performance, (e.g., (Tsapkini & Hillis, 2013; Wilson et al., 2016). This is often done by use of standardized test batteries (e.g., Addenbrooke's cognitive examination-Revised (So et al., 2018), Frontal Assessment Battery (Dubois et al., 2000), Frontal Systems Behavior Scale (Grace, 2001), Wechsler Adult Intelligence Scale (Wechsler, 1981)).

These test batteries also often include measures for EFs, but typically do not include tests related to all three components of EFs. Importantly, results are often presented as collapsed summary statistics for a neuropsychological score on these batteries, not specifically for EFs or the EFs components (e.g., (Alladi et al., 2017; Johnen et al., 2018; Petersen et al., 2019; Raczka et al., 2010; Zamboni et al., 2008). Neuropsychological assessments frequently

include tests in line with tests for EFs according to Miyake et al. (2000), such as the Stroop test or Trail Making Test Part B, measuring inhibition and shifting, respectively. Other frequently reported EFs tests in PPA studies however show a weaker and less pure association with EFs, such as digit span and verbal fluency tests (e.g., (Adlam et al., 2006; Libon et al., 2009; Savage et al., 2013; Wood et al., 2020). Digit span tests are often classified as tests for 'phonological short term memory' or '(updating of) working memory' (e.g., (Eikelboom et al., 2018; Meyer et al., 2015) and verbal fluency tests as tests for 'inhibition', 'mental flexibility' or 'processing speed' (e.g., (Libon et al., 2007). Regarding memory span tasks however, some consider only complex memory span tasks to be related to actual updating abilities. As such, forward and backward digit span tests are considered to be limited to (passive) storage capacity of short-term memory, while complex span tasks add a more explicit processing requirement (Miyake, 2001; Unsworth & Engle, 2007). Verbal fluency task performance on its part, is dependent on more cognitive abilities than just EFs.
Studies in healthy subjects have shown performance on verbal fluency tests to be impacted by language-related factors such as vocabulary size (Shao et al., 2014; Whiteside et al., 2016) and lexical access speed (Shao et al., 2014). In PPA, performance on fluency tasks has previously shown to be negatively affected by semantic memory impairment in SvPPA (Laisney et al., 2009; Riello et al., 2021). Further, the outcome measure of verbal fluency tasks provided in PPA studies is usually the number of words generated, while other measures may more fully capture the underlying processes. The quantitative clustering (generated words belonging to the same category) score is considered to be an executive-linguistic subprocess, while the qualitative switching score (number of effective switches from one subcategory to another), is thought to carry a larger executive aspect than clustering does (Pagliarin et al., 2022). Although very often used as a measure for EFs in PPA, verbal fluency tests may have limited usefulness in this regard due to their relation to several other cognitive abilities. See section 3.1.2 for our task inclusion criteria, and Supplementary Material 1 for all studies and tasks included, categorized for EFs component. In conclusion, there is a mismatch regarding EFs and their assessment in PPA literature at the moment, with more concern required for this cognitive process in this population.

Investigations into the interaction between language decline and executive function performance in patients with PPA is further complicated by the observation that many tests for EFs are mediated by language processing, namely by requiring a verbal response (e.g., verbal fluency tests), and/or through the use of verbal instructions or verbal stimuli. This makes assessment of EFs in patients with PPA particularly complex, given the linguistic deficits in these patients. These deficits may negatively affect performance on verbally mediated tests for EFs. The identification of impairments of EFs or other cognitive aspects measured by using verbal tasks in PPA, has been attributed to simply reflecting decline of these language abilities, adding to the idea that EFs remain relatively intact (Kertesz et al., 2003; Machulda et al., 2013; Wicklund et al., 2007). While the contribution of language impairment to substandard performance on verbal tests of EFs has been generally accepted, studies often still assess EFs with verbal tests in PPA. On the other hand, seemingly affected EFs can lead to an overestimation of executive dysfunction, and to a misdiagnosis of having another type of dementia (e.g., behavioral variant frontotemporal dementia). Thus, there is a need to clarify to

what extent difficulties on EFs assessments can be attributed to language deficits or can be considered as an independent deficit of EFs. To this end, the use of nonverbal tests of EFs can provide valuable information, as these tests try to detach EFs and language (e.g., WCST, Tower of London, antisaccade task). However, even tasks with a nonverbal output still require verbal input.

There have indeed been discrepancies between results on verbal or nonverbal tests of EFs in PPA. In an early meta-analysis, patients with PPA were impaired on verbally mediated tasks of cognitive flexibility (i.e., phonemic fluency), but were not distinguishable from age-matched HC on the nonverbal WCST (Zakzanis, 1999). Wicklund and colleagues (2004) provided additional evidence regarding intact nonverbal EFs, where PPA patients with PPA (mainly NfvPPA) performed comparably to age-matched HC on a nonverbal shifting task, but better than patients with AD or behavioral variant frontotemporal dementia (Wicklund et al., 2004).

However, not all impaired performance of patients with PPA on EF test can simply be explained by verbal characteristics of the tests administered: several studies have indicated executive deficits as measured by nonverbal tests. WCST results have indicated impairments in patients with NfvPPA (Knibb et al., 2009; Nestor et al., 2003), and severe deficits have also been reported as measured by Trail Making Test Part B in NfvPPA (Ash et al., 2010; Bejanin et al., 2020; Green et al., 1990) and SvPPA (Bejanin et al., 2020).

2. Research questions and aims

Given the unclear evidence that EFs are affected by PPA, the question arises whether PPA patients suffer from executive deficits, and if so, whether these affect EFs in general, or specific subcomponents.

In the present meta-analysis, we aim to answer the following research questions based on the 'unity and diversity' model of EFs (Miyake et al., 2000):

1) Do patients with PPA score significantly worse than age-matched cognitively healthy controls (HC) on tests for EFs?
2) If patients with PPA score significantly worse than HC on tests for EFs, is there a difference in the EF components: inhibition, shifting, updating or is it rather a 'common EFs factor' deficit?

Subquestions of these main research questions are: How do PPA variant, task modality (verbal/non-verbal neuropsychological tests for EFs) and disease duration affect both main research questions?

To our knowledge, this is the first meta-analysis to date focusing solely on EFs in a PPA population, giving opportunity to an extensive analysis and discussion of EFs specifically. Further, previous meta-analyses that focused on PPA and bv-FTD, have not looked at the three components that are typically considered to be the subcomponents of EFs (Kamath et al., 2019; Kamath et al., 2020). Lastly, the tasks we include as tasks for EFs differ from others, as we apply stringent task inclusion criteria in accordance to the latent variable analysis by Miyake et al., (2000) (see section 3.1.2), to ensure a solid theoretical framework to operate in.

We believe this comprehensive assessment of EFs in PPA is highly desirable to:

(1) Identify cognitive changes besides language impairments, in early or advanced stages, to support clinical care decisions, treatment planning and anticipate future needs.
(2) Shed light on the informative value of verbal or nonverbal tests of EFs in patients with PPA.
(3) To gain further insight into the debate concerning a unity or diversity governing EF components.
(4) Further establish whether assessments of EFs can help differentiate between PPA variants or other syndromes of dementia.

3. Methods

3.1 Identification of articles

3.1.1 Search strategies

We first conducted a systematic review, according to the Preferred Reporting for Systematic Reviews and Meta-analyses guidelines (PRISMA; (Moher et al., 2015). We adhered to the PRISMA checklist in preparation of the protocol and in writing the systematic review and meta-analysis (Supplementary Material 2). Three electronic databases (Pubmed, ScienceDirect, Web of Science) were searched for records using combinations of the following search terms and Boolean key operators: (["FTD" OR "FTLD" OR "semantic dementia" OR "semantic variant PPA" OR "progressive nonfluent aphasia" OR "PNFA" OR "primary progressive aphasia" OR "progressive aphasia" OR "logopenic"] AND ["executive function*" OR "executive control" OR "inhibit*" OR "update" OR "shifting" OR "switching" OR "neuropsych*" OR "cognitive control" OR "working memory"]). There were no restrictions made concerning time span[1]. The electronic search scanned each title and abstract, retrieving articles potentially reporting on executive functions in PPA. The reference list of each publication was reviewed to identify additional sources. The inclusion criteria listed below were applied to the retrieved database of papers.

3.1.2 Inclusion criteria and study selection

Classification of tasks according to components of executive function

Tasks were classified according to the three components of EFs proposed by the 'unity and diversity' model: shifting, inhibition and updating. Task inclusion was based on whether task characteristics align with the tasks proposed by the model of Miyake et al. (2000), rather than on the definition given by the authors.

Tasks were classified into the **shifting** component if it was a valid measure tapping the shifting component according to Miyake et al. (2000): the plus-minus task (Jersild, 1927), the number-letter task (Rogers & Monsell, 1995), the local-global task (Navon, 1977), and the

[1] Studies between 2000 and 2004 may not recognize the logopenic phenotype as a variant of PPA. To account for this, we have also run the meta-analysis without studies prior to 2004, of which we included four. There were no differences with the current meta-analysis found regarding significance of effects.

WCST (Berg, 1948). Further, we included tasks of shifting if their characteristics were in line with the definition of shifting tasks given by Miyake et al. (2000), such as the Trail Making Test part B (Reitan, 1955).

Tasks were classified into the **updating** component if it was a valid measure tapping the updating component according to Miyake et al. (2000): the keep track task (Yntema, 1963), the letter memory task (Morris & Jones, 1990), the *n*-back task (Jonides & Smith, 1997), the tone monitoring task (Larson et al., 1988), and complex working memory span tasks such as the operation span task (Turner & Engle, 1989). We did not include simple working memory span tasks such as the digit span task, as they are not shown to be related to the three executive function components from the 'unity and diversity' model (Miyake et al., 2000), and can be considered a storage-related task rather than a storage *and* processing-related task of working memory, as discussed in section 1.2.1.

Tasks were classified into the **inhibition** component if it was a valid measure tapping the inhibition component according to Miyake et al. (2000): the antisaccade task (Hallett, 1978), the stop-signal task (Logan, 1994), the Stroop task (Stroop, 1935), and the Tower of Hanoi (Arnett et al., 1997), or if it was one of the tasks that taps into the 'Resistance to Distractor Interference' inhibition-related component researched in a subsequent study (Friedman & Miyake, 2004), such as the flanker task (Eriksen & Eriksen, 1974) the Attention Network Test ((Fan et al., 2002), and go-no go tasks (Gordon & Caramazza, 1982).

Further, we did not include tasks for verbal fluency, as there is unclarity on how these are related to the three EF components according to the 'unity and diversity' model (Miyake et al., 2000), and performance on this measure is, most probably, affected by language (impairments), as discussed in section 1.2.1. See Supplementary Material 1 for all studies and tasks included in this meta-analysis, categorized into EFs components.

Study inclusion criteria

Studies selected for inclusion in this meta-analysis adhere to the following criteria:

1) The study population included Primary Progressive Aphasia patients and a cognitively healthy age-matched control group (HC).
2) The materials used in the study included at least one task of executive functioning, meeting the criteria described in the previous paragraph.
3) Studies using normative data were excluded, as well as case-studies. Case-series were included if HC data was provided.
4) The study reported sufficient data to generate an effect size (e.g., mean and standard deviation of tests for patients with PPA and HC).
5) Language of the peer reviewed paper was understood by the first author of this meta-analysis, being English, Dutch or French.

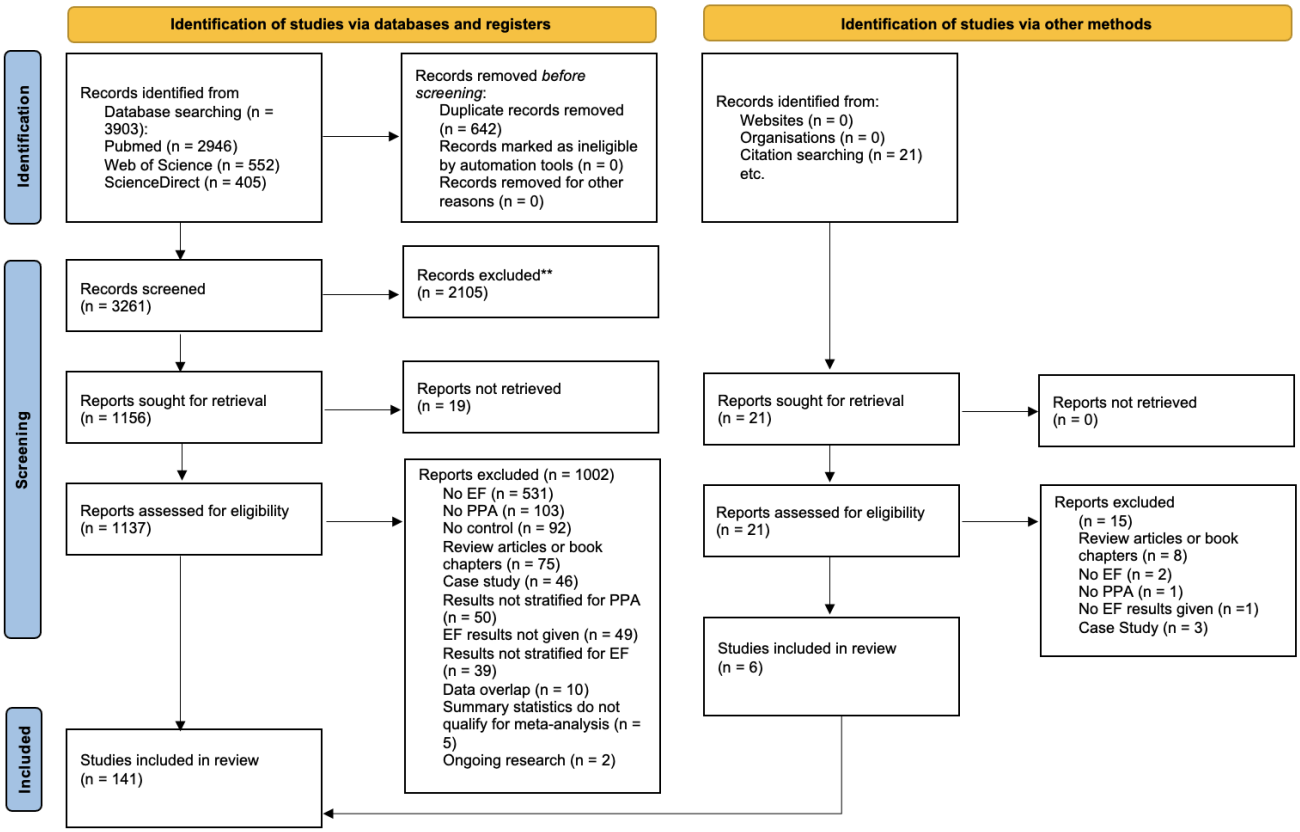
Final set of studies

See Fig. 1 for a summary of the search process. The initial database search was completed on December 16th, 2020, and was updated on November 17th, 2021. The electronic database search yielded 4095 studies. Duplicates were removed (n= 642) and titles and abstracts were screened (n= 3453) to exclude studies (n= 2105) that clearly did not meet our inclusion criteria. The full texts and reference lists of the remaining articles (n=1348) were screened for more detailed

information related to the inclusion criteria and to identify other eligible studies (n=21). Records were excluded after reading of full-texts because: (1) no tasks for EF, or none that met our EF task inclusion criteria (see section 3.1.3), were conducted (n= 635), (2) no participants with PPA were included (n=133), (3) no HC participants were included (n=114), (4) articles were review articles or book chapters (n=113), (5) articles were case-studies (n=59), tasks for EF were conducted, but (6) results on these tasks were not stratified for patients with PPA (e.g., total FTLD group results) (n=54), (7) EF task results were not published (n=48), (8) results were not stratified for EF task (e.g., 'total EF score') (n=36), (9) there was data overlap with previous articles (e.g., overlapping sample) (n=9), (10) summary statistics did not qualify for conducting a meta-analysis (e.g. no median and/or range) (n=5), (11) the research was still ongoing (n=2). The remaining 142 articles were included in the meta-analysis.

In case of overlapping study samples, the publication with the largest sample size was used. In longitudinal studies, baseline data were used. Authors were contacted to request data in case of here above-mentioned exclusion criteria numbers (6), (7), (8) and (10). When multiple outcome measure scores from the same test were reported to measure the same underlying cognitive construct, only one representative outcome was chosen for the effect size calculation. See Supplementary Material 3 for the full reference of each included study.

Figure 1. *PRISMA 2020 flow diagram for new systematic reviews which included searches of databases, registers and other sources*



3.2 Data analysis

3.2.1 Data coding

Data on study characteristics (authors, publication year, number of participants, study design), demographic and clinical characteristics (mean control age, mean patient age, mean control years of education, mean patient years of education, mean disease duration or time since disease onset), type(s) of task, type(s) of outcome measure) and data needed to calculate effect sizes (mean scores, standard deviation, sample sizes, or estimation of means and standard deviation from medians, ranges, confidence intervals, standard errors) were extracted from each individual study. When studies provided their results in subgroups that were not relevant for this study, subgroups were combined (e.g., patients with LvPPA with heterogenous underlying pathologies were combined in one single group).

3.2.2 Statistical Analysis

For each EFs task included in the studies, we calculated the standardized mean differences (SMD) effect size using Hedges' g (= one 'case'). Effect size directions were inverted for tasks where larger scores indicate poorer performance (e.g., Trail Making Test B, Completion Time measure). Combined measures were calculated for group PPA (all variants) and for PPA variants separately. To account for differences in study sample size, studies were weighted according to their inverse variance of effect sizes. A multilevel random-effects model was fitted for the calculation of the combined effect sizes and the moderator analyses, to account for dependency of study variables. Dependency in the set of studies included in this

meta-analysis mainly stems from: (1) multiple measurements for EFs in one study (e.g., a Stroop interference and a TMT-B test is performed in the same study), and/or (2) measurement of EFs in multiple PPA variants in one study (e.g., Stroop interference test measures for all PPA variants in the same study). Comparing a multilevel approach to traditional meta-analytical procedures, Van Den Noortgate and Onghena (2003) found that the maximum likelihood multilevel approach can be considered to be superior to fixed-effect approaches used in traditional meta-analyses. We used a three-level meta-analytic model to analyze the data, to model three sources of variance: sampling variance of the effect sizes (level 1), variance between effect sizes within a study (level 2), and variance between studies (level 3).
For the statistical analyses we used the function "rma.mv" of the metafor package (Viechtbauer, 2010) in the R environment (R Core Team, 2021). The t-distribution was used for testing individual regression coefficients and for calculating corresponding confidence intervals. The F-distribution was used to assess the omnibus test of the null hypothesis that all group mean effect sizes are equal when models were extended with categorical moderators of three or more categories. All model parameters were estimated using the restricted maximum likelihood estimation method, and each continuous variable was centered around its mean before moderator analyses were performed, and all categorical variables were treated as factors. The log-likelihood-ratio-tests were performed one-tailed and all other tests were performed two-tailed. We considered p-values <.05 as statistically significant. Two separate one-tailed log-likelihood-ratio-tests were performed to determine whether the within-study variance (level 2) and between-study variance (level 3) were significant, in which the deviance of the full model was compared to the deviance of a model excluding one of the variance parameters.

First, we estimated the overall effect and 95% confidence intervals by fitting a multilevel random-effects models to investigate whether there were differences in performance on EFs tasks between PPA participants and healthy controls. Next, we looked at heterogeneity of within – and between study variance, and the distribution of the total variance over the three levels in our multilevel analysis. Further, a subgroup analysis was performed to check for significant differences between the three components of EFs: shifting, inhibition and updating. Moderator analysis was performed to explore whether heterogeneity in performance may be affected by variables of interest: (1) PPA variant, as evidence indicates EFs dysfunction may differ between the variants (see section 1.3), (2) task modality (verbal or nonverbal), to examine whether performance on tasks for EFs may be mediated by language skills in these language-impaired patients, and to help clarify whether low performance on tests for EFs can be attributed to their language impairments, (3) disease duration, as we hypothesize impairments to increase as disease progresses, and (4) patient age at time of assessment and (5) patient years of education, both demographic variables that can help interpret the results. Both the latter variables are assumed to affect EFs, as older and less educated people tend to score worse on EFs tasks (Bento-Torres et al., 2017). This was firstly done for the common EFs component, and a model with significant moderators is fitted, after which residual heterogeneity is checked. Moderator analysis was then repeated for each of the EF components separately. Missing data can reduce the statistical power of a study and can produce biased estimates, leading to invalid conclusions. Therefore, to account for missing data (see section 4.1), we imputed incomplete moderators through the two-level predicting mean matching method (2l.*pmm*) from the mice

v3.14.0 package (van Buuren & Groothuis-Oudshoorn, 2011). Multiple imputation is an approach to handle the problem of missing data, creating several different plausible imputed datasets, and combining the results obtained from each of these created filled-in datasets (Sterne et al., 2009).
The parameters of substantive interest were estimated in each imputed dataset separately and combined using Rubin's rules. We used multiple imputation to create and analyze 40 multiply imputed datasets.

To measure inconsistency of studies' results, we calculated Cochran's Q and I2. To identify potential outliers, we employed the 'leave one out' method, where the results of our meta-analysis are recalculated K-1 times, each time leaving out one study (Viechtbauer & Cheung, 2010). To examine the effect of publication bias, adjusted rank-correlation tests and a funnel plot were generated (See Supplementary Material 3, Appendix C).

4. Results

4.1 Descriptives, variability and assessment of missing data

A total of 141 peer-reviewed published studies were included in the meta-analysis reporting on N=4864 participants of whom n=2337 had SvPPA, n=1272 had NfvPPA, n=871 had LVPPA and n=384 were not classified into which variant they belonged to. Within these 141 studies, a total of 294 tests for EFs were performed in a PPA and HC population. Descriptive information on the demographic and clinical details of the sample used for the analysis is summarized in Table 2, for the overall 'common EFs' composite as well as for shifting, inhibition and updating separately. The overall median age of the participants was 66.1 years (IQR 63.6 to 68.0). All extracted data of the cases is summarized in Supplementary Material 1, and Supplementary Material 3, Appendix D contains basic study characteristics of all studies included in this meta-analysis.

A Kruskal-Wallis Test was conducted to examine whether the variants differed significantly for patient age and disease duration, which was significant for both variables, with patient age: chi-square = 55.944, p <.001, df = 2, and disease duration: chi-square = 31.40, p <.001, df = 2. Next, we conducted multiple comparison post hoc tests using the Dunn's Test to determine which pairs were different. For patient age, the difference was significant between all three variants: LvPPA-NfvPPA, p = .007; LvPPA-SvPPA, p = .005; NfvPPA-SvPPA, p < .001, with mean patient ages: LvPPA = 67.0±4.2 years, NfvPPA = 67.9±4.1 years, SvPPA = 64.7±2.73 years. For disease duration, the difference was significant between SvPPA and the other variants, but not between LvPPA and NfvPPA: LvPPA-NfvPPA, p = .659; LvPPA-SvPPA, p <.001, NfvPPA-SvPPA, p <.001, with mean disease duration: LvPPA = 3.87±1.07 years, NfvPPA = 4.06±1.21 years, SvPPA = 5.05±1.1 years.

Table 2. *Descriptive Characteristics*

	EFs composite (whole group)	Shifting	Inhibition	Updating

N participants	4864	3342	1469	53
N effect sizes	294	203	85	6
Age, year total group median (IQR)	66.1 (63.6-68.0)	65.7 (63.3-67.8)	66.8 (64.6-68.5)	64.7 (61.1-65.6)
Age, year SvPPA median (IQR)	64.6 (63.0-66.7)	64.0 (62.6-66.0)	66.1 (64.0-67.1)	64.7 (61.0-65.5)
Age, year NfvPPA median (IQR)	67.9 (66.4 -70.5)	67.9 (66.4 -71.0)	68.0 (61.1-68.0)	NA
Age, year LvPPA median (IQR)	66.1 (63.6-68.8)	65.8 (63.6 -67.6)	70.2 (63.5-74.4)	NA
Education, year median (IQR)	14.9 (12.9-16.5)	14.9 (12.9-16.4)	14.6 (12.9-16.4)	11.2 (10.1-13.5)
Disease duration, year total group median (IQR)	4.5 (3.5-5.5)	4.5 (3.5-5.3)	4.8 (3.7-5.6)	3.4 (3.4-3.5)
Disease duration, year SvPPA median (IQR)	5.2 (4.4-5.9)	5.1 (4.6-5.8)	5.4 (4.9-6.0)	3.4 (3.4-3.5)
Disease duration, year NfvPPA median (IQR)	3.8 (3.1-4.8)	3.8 (3.1-4.9)	1.8 (1.4-2.9)	NA
Disease duration, year LvPPA median (IQR)	3.9 (3.4-4.4)	3.9 (3.4-4.4)	4.2 (3.3-5.0)	NA
N (%) SvPPA	2337 (48.9%)	1567 (46.9%)	717 (48.8%)	53 (100%)
N (%) NfvPPA	1272 (26.2%)	933 (27.9%)	339 (23.1%)	0 (0%)
N (%) LvPPA	871 (17.9%)	673 (20.1%)	198 (13.5%)	0 (0%)
N (%) PPA	384 (7.9%)	169 (5.0%)	215 (14.6%)	0 (0%)
N Verbal tests	65 (22.1)	0 (0%)	62 (72.9%)	3 (50%)
N Nonverbal tests	229 (77.9%)	203 (100%)	23 (27.1%)	3 (50%)

Note. # studies= number of studies; # ES =number of effect sizes; mean d=mean effect size (d); CI= confidence interval; Level 2 variance = variance between effect sizes from the same study; Level 3 variance= variance between studies.

We evaluated the amount of missing information. The variables disease duration, patient years of education and patient age were not reported on in respectively 37%, 17% and 1% of the cases. For other variables, no missing data was found. We performed multiple imputations to

impute data for the variables with missing data. By evaluating the fraction of missing information (fmi), we determined the process of fill-in was to be performed 40 times (*m*=40) (See Supplementary Material 4).

Table 3. *Results for the overall mean effect size and subgroup analysis by executive function components*

	# Studies	# ES	Mean d (SE)	95 % CI	p-value	% Var at level 1	Level 2 variance	% Var at level 2	Level 3 variance	% Var at level 3
Overall	141	294	-1.28 (0.08)	-1.44; -1.12	<.001	9.54	0.97	68.03	0.32	22.44
Executive function components										
Shifting	128	203	-1.29 (0.07)	-1.43; -1.16	<.001		0.40		0.19	
Inhibition	50	85	-1.17 (0.25)	-1.67; -0.68	<.001		1.38		1.92	
Updating	5	6	-0.80 (0.34)	-1.67; 0.08	0.066		0		0.38	

An overview of the overall mean effects size and subgroup analysis by EFs components is presented in Table 3. The overall composite effect size was large, d = -1.278 and significantly lower than zero (p <.001), evidencing participants with PPA score significantly lower on tests for EFs than HC. The overall effects in the shifting and inhibition component were significant, and large (d = 1.294; d = 1.172, respectively). The overall effect in updating was not significant (p = .066). However, the one-way ANOVA analysis of variance between the components is not significant (F (2,291) = 1.066, p = .35), indicating there are no differences in performance between the EFs components in PPA.

The results of the likelihood-ratio test showed there was significant within-study variance (at level 2, $X^2_{(1)} = 772.98, p<.001$) as well as significant between-study variance (at level 3, $X^{22}_{(1)} = 14.64, p<.001$). This implied that there was more variability in effect sizes (within and between studies) than may be expected based on sampling variance alone. 9.54 percent of the total variance could be attributed to variance at level 1 (i.e., the typical within-study sampling variance); 68.03 percent of the total variance to differences between effect sizes within studies at level 2; and 22.44 percent of the total variance could be attributed to differences between studies at level 3. Therefore, we conducted moderator analyses to examine variables that could explain within- and/or between-study variance.

4.2 Moderator analyses

We performed moderator analysis for the common EFs composite, as well as for shifting and inhibition. Moderator analysis was not possible for the updating component, as

moderator analysis with $k < 10$, may not be reliable (k updating = 5) (Schwarzer et al., 2015). The results of all moderator analyses are presented in Table 4.

Table 4. *Results for continuous and categorical moderators (univariate models)*

Moderator variables	Mean *d* (SE)	95 % CI	*F* (df1, df2)[a]	*p*-value	Level 2 variance	Level 3 variance
Overall EF						
Primary Progressive Aphasia (PPA)			F(2, 281) = 28.88	<.001***	.79	.26
Logopenic (reference)	-1.97 (.17)	-2.31; -1.63		<.001***		
Semantic variant	1.10 (.19)	.73; 1.47		<.001***		
Nonfluent variant	-0.177 (.20)	-.21; .57		.376		
Task modality			F(1, 292) = 0.22	.636	.98	.32
Non-verbal (reference)	-1.26 (.09)	-1.44; -1.09		<.001***		
Verbal	-.08 (.17)	-.41; .25		.636		
Disease duration (in years)	.16 (.06)	.04; .27	F(1, 292) = 7.47	.007**	.92	.34
Age of patient	-.09 (.02)	-.13; -.05	F(1, 292) = 18.59	<.001***	.88	.34
Patient years of education	.03 (.03)	-.04; .10	F(1, 292) = .86	.355	.97	.32
Shifting						
Primary Progressive Aphasia (PPA)			F(2, 194) = 52.76	<.001***	.15	.17
Logopenic (reference)	-2.07 (.12)	-2.31; -1.84		<.001***		
Semantic variant	1.17 (.13)	.92; 1.42		<.001***		
Nonfluent variant	0.46 (.13)	.20; .72		<.001***		
Disease duration (in years)	.12 (.05)	.02; .21	F(1, 201) = 5.63	.019*	.38	.19
Age of patient	-.09 (.02)	-.12; -.06	F(1, 201) = 31.21	<.001***	.30	.21
Patient years of education	.02 (.03)	-.03; .07	F(1, 201) = 0.55	.46	.40	.19
Inhibition						
Primary Progressive Aphasia (PPA)			F(2, 78) = 9.40	<.001***	1.12	1.80
Logopenic (reference)	-1.39 (0.52)	-2.42; -.36		.009**		
Semantic variant	.65 (0.51)	-.37; 1.67		.206		
Nonfluent variant	-.82 (0.51)	-1.84; .20		.113		
Task modality			F(1, 83) = 2.21	.141	1.38	1.83
Non-verbal (reference)	-.67 (.42)	-1.50; .15		.109		
Verbal	-.64 (.43)	-1.49; .22		.141		
Disease duration (in years)	.31 (.14)	.02; .59	F(1, 83) = 4.66	.034*	1.17	2.10
Age of patient	-.09 (.05)	-.20; .01	F(1, 83) = 3.28	.073	1.22	2.10
Patient years of education	.00 (0.10)	-.21; .21	F(1, 83) = 0.00	.989	1.40	1.94

Note. mean *d* = mean effect size (*d*); CI = confidence interval; Level 2 variance = variance between effect sizes from the same study; Level 3 variance = variance between studies.

[a]Omnibus test of all regression coefficients in the model.

∗ $p < .05$.

∗∗ $p < .01$.

∗∗∗ $p < .001$.

4.2.1 Common EF composite

PPA variant (p <.001), disease duration (p =.007) and age of patient (p <.001) were significant moderators of overall EFs performance (see Table 4). Task modality (p =.636) and years of education (p = .355) were not significant. For PPA variant, the effect for all three variants was significant, with LvPPA: d = -1.97, p < .001; NfvPPA: d = -1.79, p < .001; SvPPA: d =-0.87, p <.001; indicating that all three variants perform worse than healthy controls on tests for EFs (effect sizes: Table 4, p-values: Supplementary Material 4). The mean difference in effect sizes between LvPPA and SvPPA was significant (p <.001), whereas the mean difference between effect sizes of LvPPA and NfvPPA was not significant (p = .376) (Table 4). The effect sizes of LvPPA (d = -1.9689) and NfvPPA (d = -1.7933) were significantly larger (p < .001) than the effect size of SvPPA (d = -.868) (Supplementary Material 4).

4.2.2 Shifting

PPA variant (p <.001), disease duration (p = .019,) and age of patient (p <.001) were significant moderators of shifting. Years of education was non-significant (p = .460) and Task Modality could not be analyzed as a moderator for shifting, as only non-verbal tasks were used in the included studies. For PPA variant, the effect for all three variants was significant, with LvPPA: d = - 2.074, p <.001; NfvPPA: d = -1.614, p <.001; SvPPA: d = - 0.903, p <.001; indicating that all three variants perform worse than healthy controls on tests for shifting (effect sizes: Table 4, p-values: Supplementary Material 4). The mean difference in effect sizes between LvPPA and NfvPPA (p <.001) and between LvPPA and SvPPA (p <.001) was significant (Table 4). The mean difference between all three variants was significant (p <.001) (Supplementary Material 4).

4.2.3 Inhibition

PPA variant (p < .001), and disease duration (p = .034) but not age of patient (p = .074) were significant moderators of inhibition. Task modality, (p = 0.141), and years of education (p = .989) were not significant moderators. For PPA variant, the effect for all three variants was significant, with LvPPA: d = -1.391, p <.001; NfvPPA: d = - 2.211, p <.001; SvPPA: d = -.740 p <.001 (effect sizes: Table 4, p-values: Supplementary Material 4). The mean difference in effect sizes between LvPPA and the other variants (SvPPA, p = .206; NfvPPA, p = .113) was not significant (Table 4). The mean difference in effect sizes between NfvPPA and SvPPA was significant (p <.001) (Supplementary Material 4).

4.3 Multivariable model

Lastly, to check for residual heterogeneity, we fitted a model with significant moderator variables for the overall 'common EFs' composite (Table 5).

From the multiple moderator models, we derived that, in the overall EF model, at least one of the regression coefficients of the moderators were significantly different from zero, as

the omnibus test showed a significant result (F (4, 279) = 14.63, p < .001); the mean difference in the SvPPA variant was significant (p < .001) while effects age of patient, disease duration and the mean difference in the NfvPPA variant were not significant. Tests of the significance of the residual within-study and between-study variance after adjusting for significant moderators showed that there was still variability in effect sizes within and between studies ($X^2_{(1)}$ = 663.5, p <.001 and $X^2_{(1)}$ = 16.83, p <.001 respectively).

Table 5. *Results for continuous and categorical moderators (multivariable model)*

Moderator variables	Mean d (SE)	95 % CI	F (df1, df2)	p-value	Level 2 variance	Level 3 variance
Overall EF						
Sign. moderator variables			F(4, 279) = 14,63	<.001	0.79	0.27
Intercept	-1.94 (.18)	-2.29; -1.59		<.001		
Semantic variant	1.03 (.20)	0.63; 1.43		<.001		
Nonfluent variant	.21 (.20)	-0.19; 0.60		.308		
Disease duration	.02 (.06)	-0.10; 0.14		.737		
Age of patient	-.02 (.02)	-0.07; 0.02		.307		

5 Discussion

5.1 Are EFs affected by PPA?

The principal aim of this meta-analysis was to determine whether patients with PPA show deficits in EFs, by aggregating the sometimes-conflicting results on EFs tasks in PPA. Overall, the present meta-analysis showed that patients with PPA performed significantly poorer on tasks assessing EFs when compared to cognitively healthy, age-matched controls (HC). These results confirm the hypothesis that EFs is a deficit in patients with PPA, settling ongoing controversy of presence of executive dysfunction (Macoir et al., 2017) in this population. In a staggering majority of the studies included, the assessment of EFs was not specifically the scope of the study. The present meta-analysis shows that further exploration of this domain is warranted, and that the identification of EFs deficits in clinical settings does not rule out a diagnosis of PPA.

5.2 Is there a difference in the EFs components of shifting, inhibition and updating?

A second aim was to determine whether patients with PPA have a profile of relative weaknesses (and possibly strengths) concerning specific EFs abilities, or whether the different EFs may be equally affected by PPA. This is the first study characterizing deficits in the components of EFs in PPA according to a cognitive theoretical framework. The summary effect sizes indicate poorer performance not only on the overall EFs composite, but also separately for inhibition and shifting. The differences between EFs components are not significant, so deficits in PPA are not limited to one of the multiple EF components. This suggests that there is no specific pattern of deficits in patients with PPA, and that all components are affected to a similar extent, suggesting a higher importance for unity than diversity of EF components in a PPA context

(Miyake et al., 2000). Interestingly, when looking at PPA variant as a moderator, this variable does moderate the EFs components slightly differently, as is discussed in 5.3.1.1.

This corresponds to some extent with the current literature of EFs components in the domain of healthy cognitive aging. Often based on the seminal study of (Miyake et al., 2000), attempts have now been made to characterize EFs components in older populations, by use of factor analysis and structural equation modeling. A very recent study reports a confirmatory factor analysis of a three-factor (shifting, inhibition, updating) model of EF, comparing young and older adults (Glisky et al., 2021). In the older groups, a reduction from the three EFs to two EFs was demonstrated: a shifting factor and an updating/inhibition factor. Further, increasing age was accompanied by a pattern of increased correlations between the two EFs factors, and a greater reliance on general control processes. This confirms previous analyses finding support for a two-factor model in older adults (Hull et al., 2008; Karr et al., 2018). These studies suggest the presence of age-related differences in the relative contributions of the EFs components, which in an older population may involve recruitment of less 'diverse', less separable EFs processes. This also aligns with evidence of lateralization reduction in older adults. These studies support a higher importance for 'unity' in older adults compared to younger populations, indicating that more unification of functions could be a sign of aging (Learmonth et al., 2017).

Our current meta-analysis gives an indication of similar deficits in all three EFs components in PPA, while more exploration on this topic is needed. For instance, for the updating component only six effect sizes were included, as few studies met our task inclusion criteria for this component. Research using tests that tap into the different EFs components can help to more elaborately assess the generality or specificity of an EFs deficit. This can be relevant in a clinical setting for these patients, to help understand the nature of their cognitive deficits. From a fundamental perspective, this can provide additional information on how EFs may change in neurodegenerative disease, and how this may or may not correspond to what is found in healthy aging.

5.3 What influences the deficits of EFs in PPA?

Next, we looked at potential moderators of performance on EFs tasks, in the overall 'common EFs' composite as well as for the shifting and inhibition components. Unfortunately, there were not enough studies of updating to be able to perform moderator analysis.

5.3.1 PPA variant

We looked at PPA variant as a potential moderator of performance, as previous studies have indicated differences may exist between PPA variants (see section 1.3) (K. Chen et al., 2018; Kumfor et al., 2011; Leyton et al., 2016). Our analysis shows that PPA variant is a significant moderator of the overall EFs composite, as well as for shifting and inhibition. For the overall EFs composite, effect sizes were significant for all variants, indicating that they perform worse than healthy controls on tests for EFs. However, performance of LvPPA and NfvPPA was more aligned, with no significant difference between the two, while their performance was significantly worse than that of SvPPA. These different results according to PPA variant are in line with two recent meta-analyses investigating a broad spectrum of

neuropsychological functions in PPA, including measures of EFs (Kamath et al., 2019; Kamath et al., 2020). In the first meta-analysis, (not including LvPPA), the authors found comparable effects sizes for deficits on EFs between NfvPPA and behavioral variant frontotemporal dementia participants (a type of dementia marked by executive dysfunctions), while the SvPPA group had a statistically significant lower effect size (Kamath et al., 2019). Their second meta-analysis, focusing on LvPPA, showed that patient's deficits in the EFs subdomain that they evaluated, visual set-shifting, were as prominent as their language difficulties (Kamath et al., 2020). The current meta-analysis confirms, and adds to this, by looking at the components of EFs separately (see below). These difference between the PPA variants, wherein LvPPA and NfvPPA seem to be more aligned than SvPPA concerning EFs deficit, could be due to the neuroanatomical regions implicated in each variant (Table 1).

Patients with PPA suffer from (pre)frontal cortex damage, with abnormalities in the left fronto-insular region, and frontal portions of dorsal language tracts, often including the inferior frontal gyrus, insula, premotor and supplementary motor areas (Gorno-Tempini et al., 2004; Gorno-Tempini et al., 2011). These regions partially overlap neuroanatomical correlations of EFs, wherein the (pre)frontal lobes have long been known to be involved (Robinson et al., 2014; Smolker et al., 2018). As such, we expected to find EFs to be affected in this variant, possibly more than in the other two variants. Patients with LvPPA however appear to be affected even to the same extent as NfvPPA patients. These patients mainly suffer from damage to left inferior parietal lobule, and lateral temporal and perisylvian cortical regions surrounding the left temporal gyrus (Gorno-Tempini et al., 2008; Gorno-Tempini et al., 2011). While these regions are generally considered to be less implicated in EFs than frontal regions, temporoparietal disruption is associated with damage to the phonological loop, causing one of the core problems in LvPPA: deficits of auditory short-term memory. Further, the phonological loop is important for working memory processes (Baddeley & Hitch, 1974), which are found to be impaired in LvPPA (Eikelboom et al., 2018). As such, we might expect deficits in the EFs component updating of the working memory, but unfortunately for this component not enough studies could be included in this meta-analysis. The executive deficits however extend to the common EF model, and shifting and inhibition components. Considering hypotheses on the neuroanatomical correlations of this extensive damage of EFs in LvPPA, this could support the notion that while executive dysfunctions are considered to be 'frontal symptoms', they may also be elicited from damage to non-frontal regions often implicated in LvPPA (Bettcher et al., 2016). In healthy adults, the role of fronto-parietal and parietal lobes in EFs has been shown (Brass et al., 2005; Cole et al., 2013), as well as in patient populations, i.e., EFs deficits caused by parietal tumors (Teixidor et al., 2007) and posterior lesions (Vilkki et al., 2002). On the other hand, atrophy in LvPPA can progress to affect medial parietal and temporal cortices, as well as fronto-insular regions (Galantucci et al., 2011; Rogalski et al., 2011; Rohrer et al., 2013), i.e., frontal regions known to be implicated in EFs. Indeed, a recent neuroimaging study investigating correlations between atrophy patters and deficits in EFs in LvPPA found that patients with poorer scores on EFs tasks generally demonstrated more right-hemisphere temporoparietal and prefrontal atrophy (Ramanan et al., 2020). Further, previous studies of functional connectivity in PPA have found loss of fronto-parietal hubs in all three variants. Deficits in performance may not only be due to atrophy, but also to functional connectivity

impairments, which have even been suggested to be a precursor of atrophy (Tao & Rapp, 2020) Future (longitudinal) studies can help shed light on both the possible role of temporoparietal damage in EFs dysfunction, as well as on the effect of atrophy extending to prefrontal areas in causing deficits in LvPPA.

In patients with SvPPA, neuroanatomical damage is usually most prominent in left and right anterior temporal lobe regions (Gorno-Tempini et al., 2011), regions less implicated in EFs. Therefore, controversy regarding existence of executive dysfunction has been greatest for this variant (Macoir et al., 2017). Importantly, atrophy in these patients can become more widespread, and white matter has been shown to be disrupted in a number of major tracts, including ventral as well as dorsal tracts, connecting multiple cortical (i.e., temporal, parietal and frontal) areas (Schwindt et al., 2013). Even though SvPPA seems to suffer from less EFs impairments than the other variants, effect sizes for EFs impairments variant were also significant.

A cause for differences between variants may be the underlying neuropathologies causing specific patterns of brain atrophy (see Table 1): NfvPPA and SvPPA are more frequently associated with frontotemporal lobar dementia (FTLD), respectively with FTLD-tau (Mesulam et al., 2008) and FTLD TDP-43 (Gorno-Tempini et al., 2011) pathology, while LvPPA is mostly associated with Alzheimer's disease (Mesulam et al., 2008). AD pathology in PPA has been linked to much more widespread atrophy patterns than FTLD variants (Preiß et al., 2019). This can help explain the EFs deficits found LvPPA, next to numerous symptoms other than language problems that have been described in this variant: E.g., loss of empathy (Hazelton et al., 2017), problems with emotion detection abilities (Multani et al., 2017), verbal working memory and episodic memory (Eikelboom et al., 2018) and executive dysfunction (Magnin et al., 2013). Importantly, this is not a one-to-one relationship: all variants have been associated with the different pathologies, and brain patterns can vary greatly between individuals.

Regarding the EFs components, we find similar patterns: while all variants score significantly more poorly on tests for EFs than HC, this seems to be more so the case for LvPPA and NFvPPA compared to SvPPA. For inhibition, effects sizes are largest in NfvPPA and LvPPA, who do not differ significantly from each other, but are both significantly worse than SvPPA. For shifting, effects sizes are largest in LvPPA, followed by NfvPPA and then SvPPA, with significant differences between all three variants. The remarkably poor performance of LvPPA on shifting measures is in line with the findings of Kamath et al., (2020). Our present meta-analysis additionally indicates that patients with this variant have significant problems with inhibition tasks. As mentioned above, as language impairments in LvPPA are considered to be caused by a phonological short-term memory deficit, we may also expect them to perform poorly on tasks for updating, although no results on updating tasks (meeting our task inclusion criteria) in LvPPA have been reported in the literature yet.

Findings of differences in EFs deficits may be very relevant in a clinical setting, by aiding the differential diagnosis of PPA variants. Assessments of EFs can contribute to help differentiate between SvPPA and the other variants. Further, our findings give preliminary indications of differences between PPA variants on the different EFs components, which future research may provide more in-depth information on. As such, tests for shifting may aid in differential diagnosis of LvPPA.

In general, we can conclude that all variants suffer from EFs weakness, but this is more strongly the case for LvPPA and NfvPPA variants.

5.3.2 Task modality

Another key potential moderator of interest was task modality (i.e., verbal vs. nonverbal tasks). EFs tasks are often verbally mediated, and in patients with PPA, weak performance on EFs tasks has been suggested to be a consequence of their language impairments (Gunawardena et al., 2010; Kertesz et al., 2003; Machulda et al., 2013; Wicklund et al., 2007). Thus, it is necessary to clarify how task modality moderates EFs performance. Importantly, shifting tasks included in the present meta-analysis all required nonverbal output, and thus we could not examine whether this component is differently affected by task modality. For the updating component, not enough studies were included to be able to perform moderator analysis. Thus, we performed moderator analysis of task modality for the overall EFs composite, as well as for the inhibition component, finding that for both, task modality was not a significant moderator of performance. This is a very important finding, suggesting that task modality does not have the previously suggested confounding role in EFs task performance in PPA and that low performance can possibly be attributed to an independent deficit of EFs, rather than reflecting existing language deficits. Further, this is suggestive of domain-general control involvement in PPA, with deficits found in performance, irrespective of the verbal (or nonverbal) nature of the response. Aside from confirming existence of EFs deficits in PPA, this also has practical implications for clinical and research settings, suggesting that both verbal and nonverbal tests of EFs can shed light to the deficits of patients with PPA.

5.3.3 Disease duration and age

Disease duration was a moderator for the overall EFs composite, as well as for shifting and inhibition. This suggests that performance on tests for EFs gets worse over time, in a linear fashion. This is not surprising, considering the progressive nature of the disease, and can be relevant information when considering the disease course of PPA. The presence and potential worsening of EFs deficits is something that might need to be taken into account when anticipating future needs for patients with PPA. The range of time post disease onset included in this meta-analysis is 1.7- 8.9 years. While in this range, disease duration can be considered to be a moderator of EFs performance, we cannot stipulate to what extent these deficits may already be present at time of diagnosis. The median and mean disease duration for patients with SvPPA included in this meta-analysis was significantly longer than for the other variants, while no significantly different disease duration was found between NfvPPA and LvPPA. Nonetheless, patients with SvPPA performed significantly higher than the other variants on tests for EFs, meaning patients with SvPPA appear to suffer less from executive dysfunction even after a longer period of disease. This strengthens the idea that EFs are more/longer spared than in SvPPA than in the other variants.

When looking at patient age, this was a significant moderator for the overall EFs composite, and for shifting, although not for inhibition. Age as a significant moderator suggests that older patients with PPA performed more poorly on tests for EFs, which is in line with some of the aging literature on EFs with healthy adults (e.g.,(Hirsch et al., 2016; Maldonado et al., 2020).

5.4 Limitations and future directions

The current meta-analysis has limitations that should be considered. To study the profile of EFs in PPA, the number of studies included and effect sizes to analyze has to be large enough. The number of available effect sizes (k = 6) for the updating component was too low for moderator analysis (Schwarzer et al., 2015). This indicates a pressing need for more research on updating in PPA, however, also stipulates another limitation: while the work of Miyake et al. (2000) provides a solid theoretical framework to work with, stringently adhering to their task criteria excludes some tasks often for EFs. For instance, while the backwards digit span might not be considered to be an adequate 'complex' span task by some (Unsworth & Engle, 2007), one can argue that this test does require an element of manipulation, and is not purely storage-related. Further, when we looked at PPA variant as a moderator of effects, studies that reported on mixed-PPA or group composites of PPA, could not be included in the analysis. Our moderator analyses were also limited to which characteristics are reported in the studies, and how often. For example, time since disease onset/disease duration of the sample was not reported in about 40% of the studies. If this information is known, providing this data in research articles can help to examine the trajectory of unravelling EFs, among other cognitive processes, in PPA. Additionally, there was a large degree of heterogeneity among the effect sizes. While moderators such as PPA variant and disease duration accounted for some of the heterogeneity, a large portion of the heterogeneity is still unexplained. Variability among tasks measuring the same EFs can be a potential source of heterogeneity, as for each component an array of different tasks and scoring criteria exist. Other factors that can introduce variability can include small sample sizes, inclusion and exclusion criteria of the studies, recruitment practices, etc. Future research can help account for residual heterogeneity found in the present meta-analysis.

Importantly, as we found that patients suffering from PPA suffer from weakness in EFs, across all components, as well as in verbal and nonverbal task modalities, it is pivotal that more research on EFs is conducted in this population. The overwhelming majority of studies included in this meta-analysis conducted tasks for EFs as part of a general patient characteristics assessment, but elaborate research of EFs in PPA specifically is still heavily lacking in the literature. Many aspects are yet to be explored. Regarding PPA variant, it would be very relevant to further look into differences between the variants, and possible relations with neuroanatomical damage in PPA. Further, questions remain regarding the correlation between the progressive nature of both the aphasia as well as EFs deficits: do they decline at a similar pace, or deteriorate independently of the other? Concerning possible correlations between aphasia and the EFs deficits, research should also explore how the EFs (deficits) might influence performance on language tasks. Further, regarding task modality, we have looked at how task modality may influence language performance, finding that in both modalities patients show EFs deficits. However, it would be relevant to investigate how auditory comprehension performance of these patients may influence EFs task performance, as most nonverbal tasks included in this analysis still required verbal instructions (and thus auditory comprehension skills). Lastly, none of the included articles were conducted on bilingual patients with PPA. Considering the relationship between EFs and language (see section 1.1), and the role EFs play in bilingual language control, assessment of EFs should not be overlooked in clinical as well as research settings in this population.

6 Conclusion

The current meta-analysis showed that patients with PPA suffer from a decline in EFs, compared to healthy, age-matched controls. This is the case for a common EFs composite, as well as for the three subcomponents (shifting, inhibition, updating) separately. It is also demonstrated that the magnitude of this difference is moderated by PPA variant and disease duration, with patients with LvPPA and NfvPPA showing more deficits than patients with SvPPA. Further, task modality is not a moderator of effects, evidencing that low EFs task performance represents true deficits of EFs in these patients, rather than their language impairments. Our findings call for more research on this subject, to better understand the profile of EFs impairments in PPA variants, and the implications this may have on clinical practice with regard to for instance differential diagnosis or treatment of EFs as an intervention goal.

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Competing interests

The authors have no competing interests to declare.

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Figures