

RESEARCH ARTICLE

Exploring the phonological profiles of children with reading difficulties: A multiple case study

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Studies of group differences have established that the phonological profiles of people with reading difficulties contain both strengths and weaknesses. The current study extends this work by exploring individual differences in phonological ability using a multiple case study approach. A heterogeneous sample of 56 children (M age = 9 years) with reading difficulties completed a battery of tasks measuring literacy, phonological processing, expressive vocabulary and general ability. The phonological tasks included measures of phonological awareness (PA), phonological memory (PM), and rapid naming (RAN). A majority—although not all—of the children had phonological processing impairments. However, there was also substantial variability in the nature of children's phonological difficulties. While multiple impairments encompassing two or more phonological domains were most common, impairments that were specific to PA, PM or RAN also occurred frequently. Even within the domain of PA, where children completed three well-matched tasks, individual children were rarely impaired across all three measures and a number of different profiles were observed. Additional, group-level analyses indicated that PA was a significant predictor of decoding while RAN was a significant predictor of automatic word recognition and comprehension. Findings are discussed with reference

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to conceptual models of phonological processing and implications for assessment.

KEYWORDS

dyslexia, multiple case study, phonological awareness, phonological memory, RAN

Practitioner points

- Our findings demonstrate substantial heterogeneity in the scope and nature of children's phonological deficits
- Phonological awareness (PA), phonological memory (PM) and rapid naming (RAN) are specific abilities and deficits may occur together or independently
- PA deficits are predictive of decoding and/or diffuse reading difficulties while RAN may indicate lexical impairments and slow reading speed
- Even within the domain of PA children with reading impairments may show differing profiles of strengths and weaknesses
- This study also replicates previous findings that reading difficulties may sometimes occur in the absence of any phonological impairment

A number of cognitive deficits have been associated with reading difficulties and this has given rise to competing models of Developmental Dyslexia (e.g., White et al., 2006, plus associated commentaries). The multiple case study (e.g., Carroll, Solity, & Shapiro, 2016; Gathercole et al., 2016; Ramus et al., 2003; White et al., 2006), in which participants' individual patterns of performance across the tasks in an assessment battery are examined and compared, has proven fruitful in exploring these competing theories and evaluating the relative contributions of phonological, auditory, visual, motor and other skills to reading impairment. Taken together, these and many other studies have helped inform the development of a probabilistic, multiple-risk account of Dyslexia (Moll, Loff, & Snowling, 2013; Pennington, 2006; Snowling, 2008). This approach contrasts with earlier models which often sought to explain all cases of reading impairment by appealing to a single "core" deficit localised within a specific cognitive domain (see Ramus et al., 2003, for review).

Phonology retains a central role in contemporary multiple-risk models of Dyslexia due to an overwhelming body of research indicating its causal role in reading development (e.g., Hulme, Bowyer-Crane, Carroll, Duff, & Snowling, 2012). However, only in recent years have researchers begun to explore the phonological skills of children and adults with Dyslexia in finer detail. Studies have suggested that phonological processing may be composed of several related but distinct abilities (e.g., Anthony, Williams, McDonald, & Francis, 2007; de Jong & van der Leij, 1999; Nelson, Lindstrom, Lindstrom, & Denis, 2012; Powell, Stainthorp, Stuart, Garwood, & Quinlan, 2007) and, while some of these abilities may be impaired in people with Dyslexia, many others appear to remain intact (Ramus & Ahissar, 2012). In addition, researchers have also presented data suggesting that the scope of the phonological deficit may change during the course of development (Boets, 2014) and that subtle differences exist in the phonological impairments associated with Dyslexia and other, often comorbid developmental disorders (Carroll & Breadmore, 2018; De Groot, Van den Bos, Van der Meulen, & Minnaert, 2015; Marshall, Ramus, & van der Lely, 2011; Nithart et al., 2009; Ramus, Marshall, Rosen, & van der Lely, 2013).

Wagner and colleagues (Wagner et al., 1987) proposed several models of phonological processing, including the General Ability Model, in which phonological processing is conceptualised as a single, undifferentiated construct and the Specific Ability Model, in which phonological awareness (PA), phonological memory (PM), and rapid naming (RAN) are held to be distinct but related abilities. The Specific Ability Model has received support from studies applying confirmatory factor analysis to data from large samples of typically developing children (see Nelson et al., 2012, for review). Importantly though, these models of phonological processing do not only make predictions regarding which language tasks should load together in large, multivariate analyses, they also lead to different predictions regarding the kinds of phonological deficits that should and should not occur in individual children. For example, the General Ability Model would only allow for global deficits that encompass all three areas of phonological processing. In contrast, the Specific Ability Model would allow for the occurrence of PA-, PM-, or RAN-only deficits in some children. Although controversial (see Vukovic & Siegel, 2006, for review) the Double Deficit Hypothesis of Dyslexia (Wolf & Bowers, 1999) also proposes a framework in which RAN and PA are separable components of phonological ability. There is evidence to suggest that these represent distinct sources of impairment in Dyslexia (Wolf et al., 2002), relate to different aspects of reading ability (Georgiou, Parrila, & Kirby, 2009; Warmington & Hulme, 2012), and make their strongest contributions to reading ability at different developmental stages (Kirby, Parrila, & Pfeiffer, 2003). A multiple case study systematically examining the phonological processing deficits of children with reading impairments offers an opportunity to evaluate models of phonological processing from a new perspective.

Thus far studies seeking to explore the nature of the phonological deficit in Dyslexia have focused on group-level data. This is potentially limiting because, as Carroll et al. (2016) point out, averaging across groups of impaired readers can mask individual differences in a particular domain of impairment. For example, a deficit observed at the group level may be the result of a small number of affected individuals with relatively extreme scores on the measure in question. Conversely, group-level data may also obscure more subtle impairments that apply only to certain individuals within the sample. Individual differences in the occurrence of auditory and visual impairments in Dyslexia are well documented (e.g., Carroll et al., 2016; Ramus et al., 2003; White et al., 2006) but, in contrast, potential heterogeneity in the nature of children's phonological impairments has not been fully explored. The current investigation aims to address this issue by mapping individual differences in the phonological profiles of children with reading difficulties using the multiple case study approach. In particular, we address research questions regarding the extent to which phonological deficits shown by these children are universal (i.e., observed in all cases), all-encompassing (i.e., observed on all phonological processing tasks) and uniform (i.e., consistent in nature across different individuals).

1 | ARE PHONOLOGICAL PROCESSING DEFICITS UNIVERSAL?

In the multiple case study of dyslexic adults published by Ramus et al. (2003), a phonological deficit was observed in all of the reading-impaired participants. Likewise, subsequent multiple case studies of children with Dyslexia reported a phonological deficit in more than 50% of individuals and found phonology to be by far the most common area of difficulty for those with reading impairments (Carroll et al., 2016; White et al., 2006). However, substantial numbers of children in these samples (41 and 47% respectively) were not significantly impaired in the domain of phonological processing. The apparent absence of phonological difficulties in large numbers of struggling readers, at least in childhood, merits further investigation at the level of individual cases.

Furthermore, existing multiple case studies of children and adults have generally aimed to pit phonological processing against other factors as putative causes of Dyslexia and thus they have tended to treat phonological processing as a unitary ability (Ramus et al., 2003; White et al., 2006). Carroll et al. (2016, p. 755) provided a more fine-grained analysis of children's phonological abilities by contrasting PA, verbal short-term memory (VSTM) and RAN, with findings indicating that VSTM was the most commonly occurring phonological deficit in poor readers.

However, this study subsequently combined PA and VSTM when exploring different phonological profiles and the analyses do not indicate how frequently deficits in the three core phonological processing domains were observed in isolation or how frequently different types of phonological processing deficit co-occurred. The current study aims to address this issue by exploring the occurrence and co-occurrence of PA, PM and RAN deficits in a sample of struggling readers. The study is also the first to map individual differences in patterns of impairment observed within the specific domain of phonological awareness by contrasting performance in phoneme isolation, blending, and elision.

2 | ARE PHONOLOGICAL PROCESSING DEFICITS ALL-ENCOMPASSING?

Research comparing groups of individuals with reading impairments to unimpaired control participants has recently begun to emphasise the multifaceted nature of phonological processing by highlighting areas of both intact and impaired performance across different phonological tasks (see Ramus & Ahissar, 2012, for review). For example, despite experiencing persistent difficulties with phonological awareness tasks, adults with Dyslexia have been observed to show phonological similarity effects in short-term memory that are equal in magnitude to those of control participants, as well as performing normally in repetition priming paradigms, and in tasks that assess the acquisition of phonological grammar (Ramus and Szenkovits, 2008). This pattern of performance has been broadly replicated in the domain of suprasegmental phonology, with participants with Dyslexia demonstrating impairments on prosodic analogues of phonological awareness tasks but not on perceptual or priming tasks tapping the underlying representation of linguistic stress (see Mundy & Carroll, 2016, for review). Ramus and Szenkovits (2008) argued that the phonological difficulties of people with Dyslexia are only observed on tasks that impose specific processing demands, such as a higher short-term memory load, the need for speeded, sequential retrieval, or conscious manipulation of phonological representations. They proposed the Phonological Access Hypothesis to account for these findings, suggesting that the phonological deficit in Dyslexia may arise due to difficulties accessing phonological representations, rather than the quality of the representations themselves. This theory has continued to gain support from studies of children (Mengisidou & Marshall, 2019) and adults (Boets et al., 2013). Bearing in mind some of the issues with averaging across groups of participants when studying heterogeneous developmental disorders (Carroll et al., 2016), the current study aims to complement these group-level findings by exploring the different patterns of strengths and weaknesses that may occur within the phonological profiles of individual children.

3 | ARE PHONOLOGICAL DEFICITS UNIFORM IN NATURE?

While some studies have suggested that different aspects of phonological processing may be more or less impaired in those with reading difficulties, others have argued that the nature of the phonological impairment may vary across the course of development. Specifically, it has been suggested that phonological representations may be impaired early in childhood, producing a more generalised deficit in phonological processing, but recover by the time children enter primary school, leaving only the more specific pattern of impairment observed in adults and older children (Boets, 2014; Boets et al., 2011). In addition, other researchers have explored subtle contrasts in the type of phonological deficits associated with different developmental disorders. One such contrast is that between Dyslexia and Developmental Language Disorder (DLD: formerly known as Specific Language Impairment, or SLI). Ramus et al. (2013) compared groups of children with Dyslexia-only, SLI-only, and comorbid diagnoses to typically developing controls on a large battery of language tasks. Using principal components analysis, the researchers were able to identify two latent constructs underlying the phonological tasks in the assessment battery. The first of these incorporated measures of PA (e.g., Spoonerisms) and RAN, as well as digit span and was taken to reflect children's phonological skills, that is, their ability to access, retrieve, process and manipulate phonological information. The second

construct was comprised of tasks that required perceptual discrimination or simple encoding and production of phonological information. These tasks were taken to reflect the integrity of children's underlying phonological representations. Children with SLI showed impairments of approximately equal magnitude in their phonological skills and their underlying phonological representations while, consistent with the Phonological Access Hypothesis (Ramus & Szenkovits, 2008), the children with Dyslexia performed significantly better on the phonological representations component relative to the phonological skills component. Other studies have also suggested a more specific form of phonological impairment in children with Dyslexia relative to those with other diagnoses. For example, children with Dyslexia, unlike their peers with DLD, appear to be unimpaired in correcting for the effects of place assimilation in speech (Marshall et al., 2011) and in phoneme discrimination (Nithart et al., 2009). Findings from other studies have suggested that the ability to manipulate phonemes (Carroll & Breadmore, 2018) and RAN (De Groot et al., 2015) may be key points of difference between the phonological deficits associated with Dyslexia and those associated with recurrent ear infections and DLD respectively, with children with Dyslexia more likely to be impaired in these areas than other groups. The current study again seeks to complement and extend these findings by exploring the relative frequency with which different aspects of phonological processing are impaired in individual children with reading difficulties.

4 | THE CURRENT STUDY

In summary, studies have demonstrated that phonological processing is multifaceted, that individuals with Dyslexia may have difficulty with some phonological tasks and not others, and that there may be multiple phonological deficits each associated with different types of literacy and language impairment and/or different stages of development. All of this sits comfortably within the probabilistic, multiple-risk approach to understanding reading difficulties and their overlap with other developmental disorders. Indeed, phonological awareness has come to be considered an endophenotype of Dyslexia, rather than a cognitive deficit that leads to reading impairment in a simple, deterministic fashion (Moll et al., 2013; Snowling, 2008). With these recent developments in mind, we adopt the multiple case study approach used to good effect by other researchers in order to explore variation in the nature of phonological impairments associated with reading difficulties in childhood.

While existing multiple case studies have proved valuable in exploring different patterns of impairment across cognitive domains, they are yet to fully explore different patterns of performance within the domain of phonological processing or investigate task-specific variation in phonological awareness specifically. The multiple case study reported here addresses these issues by exploring individual differences in the phonological profiles of 56 children with reading impairments, focusing on the aspects of phonological processing that are most consistently associated with reading difficulties (PA, PM and RAN: Wagner et al., 1987). As stated by Ramus and Ahissar (2012), the question of whether deficits in these three areas emerge from a common underlying source or reflect distinct deficits that may occur independently of one another remains open for debate. Furthermore, it is common practice to use composite scores as an indicator of a child's ability in a particular area (e.g., phonological awareness), but if phonological abilities and the tasks used to measure them are more heterogeneous than previously appreciated, reliance on composite scores has the potential to mask the specific difficulties being experienced by some children. The frequency with which different phonological tasks reveal deficits in children with reading problems is therefore an important question for research, assessment, and intervention in the field of reading difficulties and has the potential to help inform the ongoing theoretical debate concerning the nature of phonological processing and its relationship to reading as well as providing useful insights for educators working with children on a one-to-one basis. As outlined in the subsections above, our three research questions ask to what extent the phonological deficits of the children in the sample are universal (i.e., observed in all cases), all-encompassing (i.e., observed on all phonological processing tasks) and uniform (i.e., consistent in nature across different individuals).

5 | METHOD

5.1 | Design and participants

The phonological profiles of 56 children (22 females, M age = 9.9 years, SD = 1.7, range: 6.2–14.6) were explored using a multiple case study design. Participants had all been referred for Special Educational Needs (SEN) assessments with the second author on the basis of concerns raised by the child's school and/or their parents or guardians. In some instances, concerns related to general difficulties accessing the curriculum and a failure to make academic progress as expected, while in other instances children were referred due to difficulties in specific areas, such as reading or mathematics. The 56 children were selected for inclusion in the study because they achieved a standard score ≤ 85 (i.e., one SD below the population mean) on one or more of the literacy measures administered during the SEN assessment. A further 21 children were referred for assessment but were excluded from the study after obtaining scores in the average range on all of the literacy measures. All of the literacy measures administered during the SEN assessment are described below.

All of the children were native English speakers and none had English as an additional language. A parental questionnaire indicated that 16 of the children (28.6% of the sample) had experienced one or more middle ear infection (otitis media) and three children (5.4%) were deaf in one ear. In addition, 46 of the children (82.4%) had one or more immediate relative with a history of reading difficulties or Dyslexia. At the time of the assessments all of the children were already receiving some degree of additional reading instruction in school. The children were all attending mainstream state or independent schools in the East and West Midlands regions of the UK. Measures of socio-economic status were not obtained for individual children. However, reports produced by the Office for Standards in Education (OFSTED) showed that Pupil Premiums (Department for Education, 2020) received by the participating schools were below the national average. This indicates that the number of children from disadvantaged socioeconomic backgrounds attending the schools was also below average.

5.2 | Measures

Background and literacy measures. The Wide Range Intelligence Test (WRIT: Glutting, Adams, & Sheslow, 2000) was administered to assess both verbal and non-verbal IQ. Verbal ability was assessed using the verbal analogies subtest and non-verbal ability was assessed using the matrices subtest. The WRIT has been standardised on a sample of participants aged 4;0 to 85;0 years and shows good levels of reliability (Cronbach's α = .84 and .90 for analogies and matrices respectively). Mean scores for both measures were within the average range but the children were heterogeneous in terms of their general ability. As a group, they scored significantly below the population Mean for non-verbal ability (M = 93.02, SD = 16.22, p < .001) but not for verbal ability (M = 97.75, SD = 13.65, p = .262).

Children also completed several literacy measures. The York Assessment of Reading Comprehension (YARC: Snowling et al., 2009) provided separate standardised scores for reading accuracy, reading rate, and reading comprehension. During the task children read ability appropriate passages of text aloud and answer associated comprehension questions. The YARC has been standardised on a sample of participants aged between 4.0 and 12.4 years and generally has good levels of reliability (Cronbach's α = .75, .90 and .64 for reading accuracy, rate, and comprehension respectively). Two of the children were beyond the upper age limit for the YARC and completed the Gray Oral Reading Tests, 5th Edition (GORT-5: Bryant & Wiederholt, 2011) as an alternative. These children were excluded from group-level analyses involving the YARC to avoid complications associated with aggregating data from different assessments. In addition, the Test of Word Reading Efficiency, 2nd Edition (TOWRE-2: Wagner, Torgesen, & Rashotte, 2011) provided standardised scores for sight-word reading and phonological decoding (i.e., non-word reading). During this task, children read aloud words and non-words arranged in printed columns as quickly and accurately as possible. Scores are based on the total number of items read correctly in 45 seconds. TOWRE-2 has been standardised on a sample of participants aged 6.0 to 23.11 and has good levels of reliability (Cronbach's α = .91

TABLE 1 Descriptive statistics and one-sample Z-tests for literacy measures

Measure	One-sample Z-test ($\mu = 100, \sigma = 15$)		
	Mean (SD)	<i>p</i>	Cohen's <i>d</i>
YARC accuracy	86.26 (9.25)	<.001	−0.92
YARC rate	90.12 (11.49)	<.001	−0.66
YARC comprehension	97.69 (11.11)	=.271	−0.15
TOWRE-2 sight-words	83.74 (13.66)	<.001	−1.08
TOWRE-2 decoding	80.98 (11.18)	<.001	−1.27
WRAT-4 word reading	90.64 (11.89)	<.001	−0.62
HAST-2 spelling	77.96 (12.22)	<.001	−1.47

Note: Cohen's *d* effect sizes have been calculated using the sample mean, population mean and population SD.

and .92 for sight-word reading and phonological decoding respectively). The Wide Range Achievement Test, 4th Edition (WRAT-4; Wilkinson & Robertson, 2006) was also administered and this gave a standardised score for single-word reading. WRAT-4 has been standardised on a sample of participants aged 5.0 to 94.0 years and has good levels of reliability (Cronbach's $\alpha = .92$). Finally, the Helen Arkell Spelling Test, 2nd Edition (HAST-2; Caplan, Bark, & McLean, 2000) was also administered to provide a standardised spelling score. This is a spelling dictation task in which children listen to words of increasing difficulty spoken aloud by the examiner. Each target word is presented in the context of a short sentence. HAST-2 has been standardised on a sample of participants aged 5.0 to 18.11 years and has good reliability (Cronbach's $\alpha = .87$).

Sample characteristics for the literacy measures are provided in Table 1. As a group, the children's literacy impairments were most pronounced for timed tasks (TOWRE-2) and single-word spelling (HAST-2) indicating particular difficulties with decoding and knowledge of spelling-sound mappings. The only measure for which there was not a significant, group-level impairment relative to the population mean was YARC reading comprehension.

Phonological processing. Children completed the Comprehensive Test of Phonological Processing, 2nd Edition (CTOPP-2; Wagner, Torgesen, Rashotte, & Pearson, 2013) which incorporates a number of subtests designed to assess different aspects of phonological ability. PA was assessed with three of the CTOPP-2 subtests; Elision, Blending, and Isolation. In the elision task, children listen to individual words spoken aloud by the examiner and are required to repeat the words while omitting a target sound (e.g., say snail without the /n/ -> "sail"). In the blending task, children listen to a sequence of pre-recorded sounds and are required to blend them together to form a word (e.g., /sh/ + /e/ -> she). Finally, in the isolation task, children listen to individual words spoken aloud by the examiner and identify a target sound from within the word (e.g., what is the first sound in fan? -> /f/). PM was assessed with two further CTOPP-2 subtests; Digit Span and Non-word Repetition (NWR). During these tasks, children listen respectively to pre-recorded digit sequences and non-words of increasing length and are required to repeat them back to the examiner. Finally, phonological retrieval was assessed with the RAN Letters and RAN Digits subtests. During these tasks, children are presented with 36 printed letters and 36 printed digits respectively and required to name them aloud as quickly as possible. The CTOPP-2 has been standardised on a sample of participants aged between 4.0 years and 24.11 years and has good levels of reliability (Cronbach's $\alpha = .92, .85$, and $.92$ for phonological awareness, phonological memory and naming respectively).

Expressive vocabulary. The vocabulary subtest of the WRIT (Glutting et al., 2000) was administered to assess children's broader language ability. In this task, children are required to provide definitions for words spoken aloud by the examiner (Cronbach's $\alpha = .91$).

Procedure

Ethical approval for the study was obtained from the Research Ethics Committee at Coventry University. Before completion of their child's requested SEN assessment, parents or guardians had the option to give informed consent

permitting the anonymised test scores to be pooled with assessment data from other children and used for the purposes of academic research. Children also gave verbal assent before taking part in the different tasks. Testing took place in a quiet room at the child's school. All tasks were administered by the second author, a qualified assessor holding an Assessment Practising Certificate (APC). The SEN assessment was conducted in a single session lasting approximately 150 min. Statistical analyses were conducted in JASP, version 0.9.1 (JASP Team, 2018) and Jamovi, version 1.2 (The Jamovi Project, 2019).

Analysis plan.

The primary aim of the current study was to address three research questions regarding the extent to which a phonological deficit is universal in children with reading difficulties (i.e., observed in all cases), all-encompassing (i.e., observed on all phonological processing tasks), and uniform (i.e., consistent in nature across different individuals) using a multiple case study approach. Participants completed a battery of standardised cognitive tasks as part of their SEN assessment (in each case $\mu = 100$, $\sigma = 15$). A subset of these tasks (described above) measuring various aspects of phonological processing, vocabulary, reading, spelling and general cognitive ability was selected on theoretical grounds and, in accordance with the research questions set-out above, children's pattern of performance across these measures was explored. Analyses recorded the frequency of deficits in the three broad areas of phonological processing assessed by the CTOPP-2 (Wagner et al., 2013)—PA, PM, and RAN—as well as the frequency of different deficits (elision, blending and isolation) within the domain of phonological awareness (the area of ability most strongly and consistently associated with reading problems). For the purposes of these analyses, a child was considered to have a deficit if they achieved a standard score ≤ 85 on the relevant composite or subtest.

6 | RESULTS

6.1 | Group-level data

As a group the children were significantly impaired on nearly all of the phonological processing and language measures (Figure 1). One-sample z-tests revealed that the most marked deficits, relative to the population mean, were for blending ($p < .001$, $d = -1.31$), elision ($p < .001$, $d = -.96$), and RAN letters ($p < .001$, $d = -1.10$). There were also small-to-moderate impairments for non-word repetition ($p < .001$, $d = -.53$), digit span ($p < .001$, $d = -.48$), RAN digits ($p < .001$, $d = -.51$), and vocabulary ($p = .013$, $d = -.33$). The only measure on which the children were not impaired at the group level was phoneme isolation ($p = .688$, $d = -.05$).

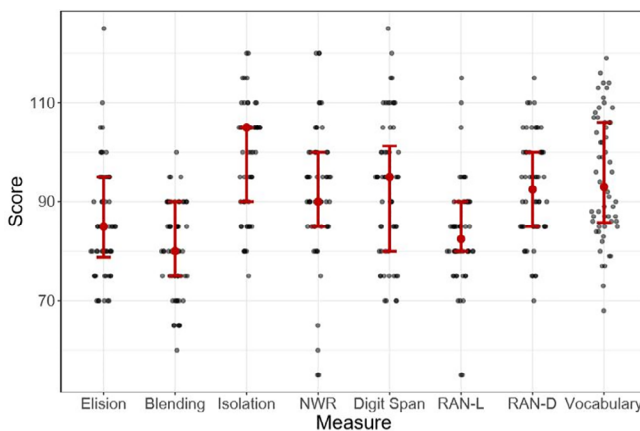


FIGURE 1 Standard scores ($\mu = 100$, $\sigma = 15$) obtained by children on the phonological processing and vocabulary measures [Colour figure can be viewed at [wileyonlinelibrary.com](https://onlinelibrary.wiley.com)]

FIGURE 2 Frequency of deficits in phonological awareness (PA), phonological memory (PM) and rapid naming (RAN) composite scores

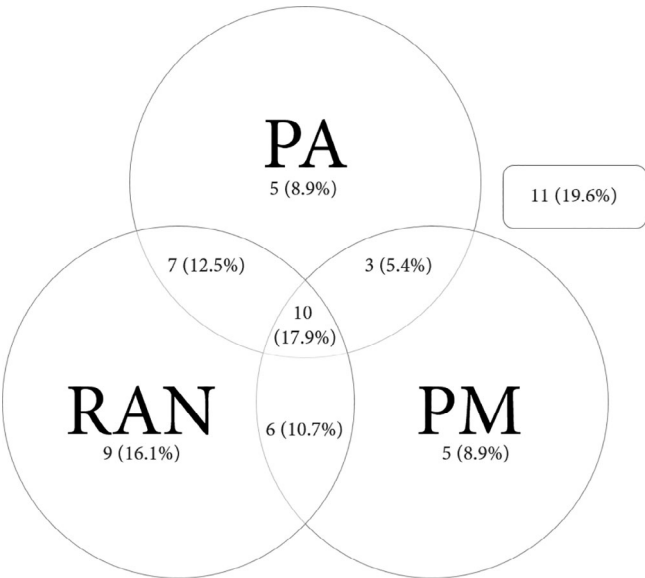
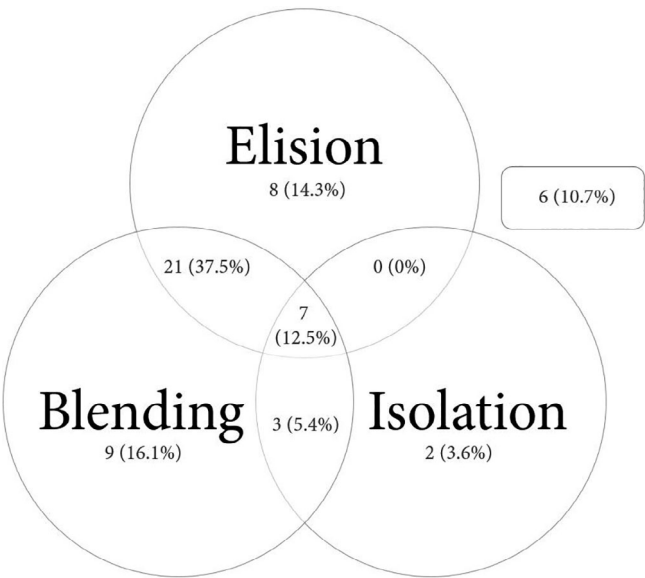


FIGURE 3 Frequency of deficits in elision, blending and isolation subtests



As anticipated, the group level analyses masked substantial heterogeneity in performance. The children in the sample achieved a wide range of scores on all of the tasks and the performance of individual children also varied markedly from task to task. These individual differences in performance were subsequently examined using the multiple case study approach.

6.2 | Phonological profiles

In relation to the first research question, phonological impairments were not universal in the sample. Analysis of the CTOPP-2 composite scores for PA, PM and RAN (Figure 2) showed a total of 11 children (19.6% of the sample) who

were apparently unimpaired in all three domains of phonological processing. However, more fine-grained analysis of the phonological awareness subtests detected a larger number of difficulties (Figure 3). Interestingly, seven of the 11 children who were unimpaired in terms of their composite scores were found have significant problems with the blending and/or elision subtests. In these cases, comparatively good performance on the isolation subtest had resulted in a PA composite score greater than 85. There were six children in the sample (10.7%) who were unimpaired across all three of the phonological awareness subtests. Four of these children were unimpaired in all areas of phonological processing, one child had a RAN-only deficit, and another had a PM-only deficit.

Regarding the second and third research questions, when phonological impairments did occur they were rarely all-encompassing and, rather than a uniform deficit, there was substantial variation in the nature of the phonological impairments observed across individuals (Figure 2). According to the CTOPP-2 composite scores, a total of 25 children (44.6% of the sample) were impaired in the domain of phonological awareness and five of these children (8.9% of the sample) showed phonological deficits that were unique to this area. Likewise, a total of 32 children (57.1%) showed RAN deficits, with nine cases (16.1%) of a RAN-only impairment. Finally, 24 children (42.9%) had a PM deficit in their profile and five children (8.9%) had impairments that were unique to this area alone.

Even within the domain of phonological awareness, impairments that encompassed all of the relevant measures (elision, blending and isolation) were quite rare and different children showed contrasting patterns of impairment across tasks (Figure 3). A total of 36 children (64.3% of the sample) showed deficits on the elision task, for example, and eight children (14.3% of the sample) were impaired on this PA task alone. Similarly, 40 children (71.4%) had a blending deficit as part of their profile and nine children (16.1%) were not impaired on any of the other phonological awareness measures. Finally, 12 of the children (21.4%) showed deficits on the phoneme isolation task and there were two cases (3.6%) of PA impairments that were unique to this measure.

Given that children within the sample varied widely in age and some studies have reported qualitative differences in the phonological deficits observed at different stages of development (e.g., Boets, 2014; Boets et al., 2011) the ages of individual children exhibiting the different phonological profiles were also explored.

There was a wide range of ages observed in all of the profile groups and children with no phonological deficits also varied widely in age (Figure 4). Robust ANOVA analyses (Wilcox, 2017) using Median ages confirmed that there was no evidence of any systematic variation in children's phonological processing ($p = .785$) or phonological awareness ($p = .647$) profiles as a function of their age.

6.3 | Phonology-literacy relationships

Finally, to gain an indication of how aspects of a child's phonological processing profile might be expected to impact their reading, additional ad-hoc analyses were conducted to explore how the different measures of phonological processing related to decoding, word reading accuracy, reading speed and reading comprehension. The two children who completed the GORT-5 as an alternative to the YARC were excluded from these analyses along with six further children who had missing data on one or more of the literacy measures. Composite literacy measures were then created through exploratory factor analysis. Bartlett's test of sphericity ($\chi^2 [15] = 117.63, p < .001$) and the Kaiser-Meyer-Olkin (KMO) measure of sampling adequacy (0.760) confirmed that the data were suitable for factor analysis. Factor extraction was conducted using the ordinary least squares method with oblimin rotation and factors were selected using parallel analysis. The analysis suggested two factors underlying the correlation matrix. The first factor was comprised of TOWRE sight-word reading ($\lambda = 0.883$), YARC reading rate ($\lambda = 0.780$), and YARC reading comprehension ($\lambda = 0.684$) and thus seemed to reflect fast, automatic word recognition of the kind that supports fluent reading and makes cognitive resources available for comprehension processes. The second factor was comprised of YARC reading accuracy ($\lambda = 0.776$), TOWRE decoding ($\lambda = 0.720$), and HAST single-word spelling ($\lambda = 0.515$) and thus seemed to reflect knowledge of spelling-sound mappings. The WRAT single-word reading measure was removed from the analysis due to it cross-loading on both factors and significantly reducing the fit of the model

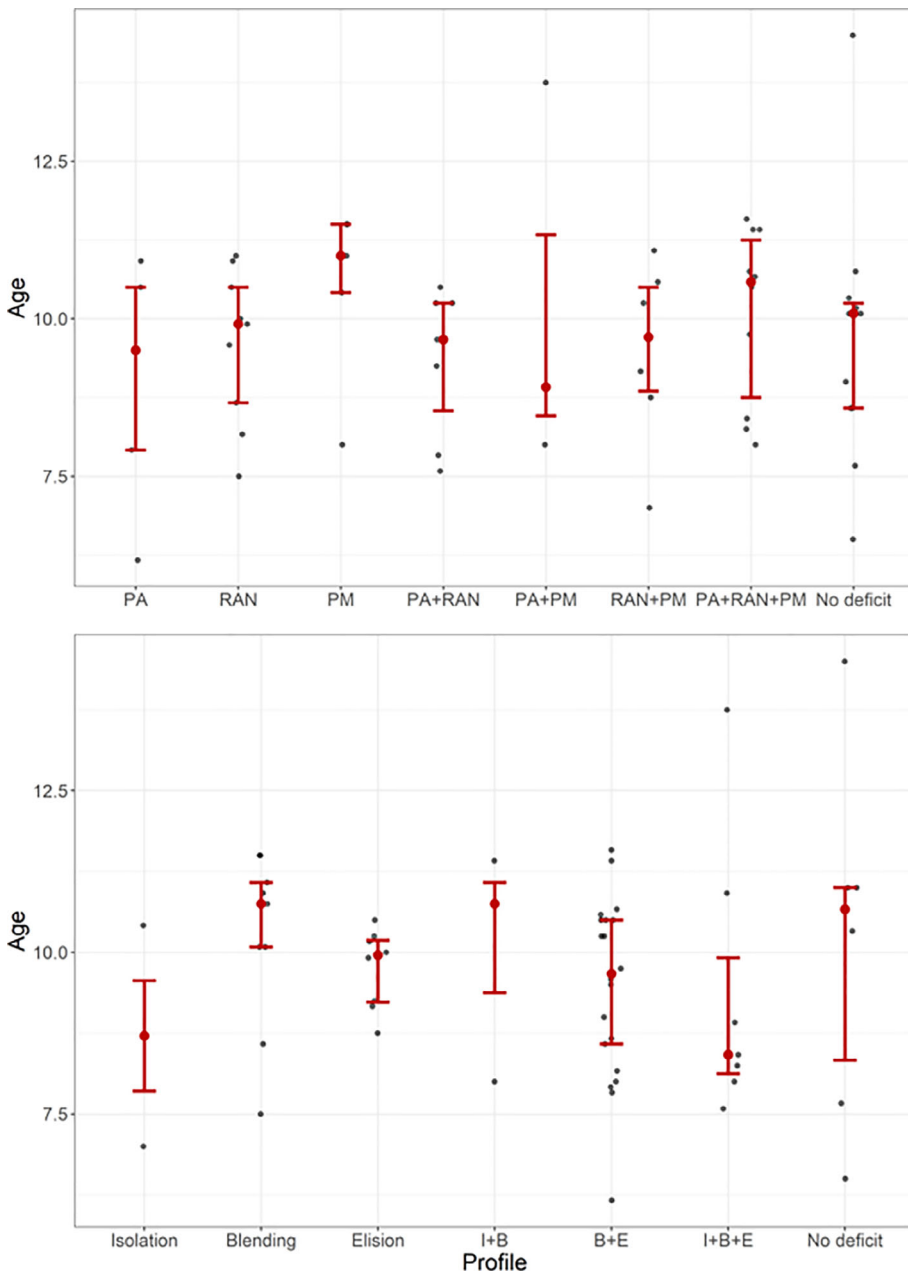


FIGURE 4 Chronological ages of children with different phonological processing (upper panel) and phonological awareness (lower panel) profiles [Colour figure can be viewed at wileyonlinelibrary.com]

($\lambda = 0.594$ and 0.451 on factor one and factor two respectively). The final two-factor model was a satisfactory fit for the data ($\chi^2 [4] = 5.64$, $p = .227$, RMSEA = 0.098 , TLI = 0.948) and there were no other cross-loadings (λ for non-dominant factors < 0.2 in each case).

Regression analyses were subsequently conducted with the literacy factors serving as outcome variables and the CTOPP-2 composite scores for PM and RAN as predictors. Children's average scores across the blending and elision subtests were also entered as a predictor and used to capture levels of phonological awareness. This approach

was preferred to the CTOPP-2 PA composite due to unexpectedly strong performance on the isolation task masking some children's PA difficulties (see above). Together, the three predictors accounted for approximately 22% of the variance in the word recognition and comprehension factor ($F [3, 43] = 3.94, p = .014, R^2 = 0.216$) and approximately 36% of the variance in the spelling-sound knowledge factor ($F [3, 43] = 7.90, p < .001, R^2 = 0.364$). RAN ($\beta = .456, p = .003$) was found to be the only significant predictor of word recognition and comprehension (PA: $\beta = -.060, p = .672$, PM: $\beta = .064, p = .653$) while phonological awareness ($\beta = .544, p < .001$) was the only significant predictor of children's spelling-sound knowledge (PM: $\beta = -.034, p = .794$, RAN: $\beta = .143, p = .290$).

7 | DISCUSSION

The current multiple case study aimed to explore individual differences in the phonological profiles of children with literacy impairments. In particular, the study sought to investigate the extent to which a phonological deficit is universal (i.e., observed in all cases of reading impairment), all-encompassing (i.e., observed on all phonological processing tasks) and uniform (i.e., consistent in nature across different individuals). The analyses suggest that a small proportion of children in the sample showed no evidence of any phonological deficits. Furthermore, when phonological processing deficits were observed, they varied in nature quite substantially across different individuals. Adopting the multiple case study approach has underlined the probabilistic nature of the relationship between phonology and reading and emphasised that the precise nature of the phonological deficit may vary from person to person.

The non-universality of PA impairments and phonological processing impairments more broadly is consistent with previous multiple case studies of children with reading difficulties (Carroll et al., 2016; White et al., 2006) and also sits comfortably with the idea that phonological deficits are an endophenotype of dyslexia, that is, a risk factor strongly related to reading difficulties but in a probabilistic rather than deterministic fashion (Moll et al., 2013; Snowling, 2008). The fact that heterogeneous phonological profiles were observed, as opposed to an all-encompassing, uniform phonological deficit, is also consistent with this view and the presence of PA-only, PM-only and RAN-only deficits in the sample also offers support for the Specific Ability Model of phonological processing (Wagner et al., 1987). The fact that children may be impaired in one particular area, or indeed on one particular task, while performing normally in others suggests that PA, PM and RAN represent related but distinct sets of abilities. De Groot et al. (2015) have previously argued that RAN impairments may be more characteristic of the phonological difficulties associated with Dyslexia than those associated with SLI/DLD. Consistent with this, of the children found to have phonological processing impairments in the current study, almost 90% (40/45) had a RAN deficit in their profile. In addition, RAN-only deficits were more common than PM-only and PA-only deficits within the sample. Overall, though, mixed profiles with deficits in multiple areas were more common than those that were restricted to a single domain or task. Finally, the finding that RAN was predictive of faster reading speed and automatic word recognition while phoneme blending and elision were more closely related to decoding and spelling is also consistent with past research (e.g., Georgiou et al., 2009; Warmington & Hulme, 2012; Wolf et al., 2002) and again emphasises the distinct nature of these abilities and their relationships to reading. Given the average age of the children in the sample, these findings are also consistent with longitudinal research exploring the developmental trajectories of language-literacy relationships which has indicated that RAN typically emerges as a stronger predictor of literacy ability later in childhood (Kirby et al., 2003).

It is perhaps surprising that the analyses reported in the current study did not suggest any systematic influence of age on the occurrence of different phonological profiles. Given findings reported elsewhere in the literature, we may have expected to observe certain patterns in the frequencies of different phonological profiles as a function of children's ages. For example, the work of Boets and colleagues (Boets, 2014; Boets et al., 2011) has suggested that phonological impairments may become narrower and more focused on the specific domain of phonological awareness with age. Intuitively, we may also expect older children to experience less difficulty with some of the simpler phonological measures (e.g., phoneme isolation) and demonstrate narrower profiles as a result. Other researchers

have observed that reading difficulties may manifest in different ways as children grow older. For example, Kirby et al. (2003) reported that RAN accounted for increasingly more, and PA gradually less, variance in several measures of children's reading ability with age. Findings such as this may lead to the prediction that RAN deficits would be observed more frequently among older children in cases of reading impairment. While the sample for the multiple case study had a wide age range, the *SD* of children's ages was actually rather small and approximately 82% of the sample were aged between eight and 11 years. It is possible that a sample selected to represent a wider range of ages in approximately equal numbers would reveal more about the role of age in shaping children's phonological difficulties.

The current findings also touch upon the debate regarding the measurement and assessment of phonological processing. Researchers have argued that the cognitive demands associated with measures of phonological processing, and auditory processing more generally, are not well understood (Protopapas, 2014) and that even superficially simple tasks can tap a number of underlying processes. Tasks that ostensibly belong to the same domain can also differ in terms of their memory load, response format, time constraints, and metalinguistic demands. These issues create challenges for researchers seeking to interpret relationships between phonological tasks and literacy performance. Furthermore, some of these processes and task demands are likely to be more crucial than others in understanding the relationship between performance in phonological tasks and literacy ability. Cunningham and colleagues (Cunningham, Witton, Talcott, Burgess, & Shapiro, 2015) used a matched task design to demonstrate that task demands such as requiring the production of a verbal response and engagement with complex as opposed to simple linguistic stimuli (phonemes vs. pseudowords) accounted for unique variance in children's decoding ability. The results obtained in the current study further emphasise these challenges. While previous studies have demonstrated variability in performance across different phonological tasks in groups of people with Dyslexia (e.g., Ramus et al., 2013; Ramus & Szenkovits, 2008), the current study suggests there may also be substantial differences in the pattern of impairment seen in different individuals. For example, children completed three tasks considered to be relatively pure measures of phonological awareness (isolation, elision and blending) that were matched in terms of their response format (all required a verbal response) and the complexity of the stimuli (all involved single words analysed at the level of individual phonemes). Despite this, children were very rarely impaired across all three of the PA tasks, there were many cases in which deficits were specific to one particular task and, as a group, the children performed far better on one of the tasks—phoneme isolation—than the others. The latter finding is consistent with the results of Cunningham et al. (2015) who found children's isolation performance to be superior to their performance in phoneme deletion and repetition. It is not immediately obvious why performance differences like this should emerge between tasks that are superficially well matched. One possibility is that they reflect differences in more general task demands, such as working memory. It may be possible to isolate an initial phoneme without going on to encode the entirety of the stimulus, but such a strategy would not allow successful completion of a blending or deletion task where the correct verbal response must contain multiple phonemes. Further detailed investigations of the processing demands associated with different tasks—even tasks from within a particular domain of phonological processing—will be necessary in order to shed light on such findings and better understand how specific tasks relate to reading ability. This in turn will allow researchers and practitioners to make more informed decisions about the phonological processing measures that they use in their work (Cunningham et al., 2015; Protopapas, 2014; Ramus & Ahissar, 2012). The potential that we have noted for performance differences between tasks to bias composite scores and thus mask more specific difficulties that children are experiencing has particular implications for practitioners undertaking language assessments.

The adoption of a multiple case study approach has provided a novel perspective on debates regarding the structure and measurement of phonological processing as well as its relationship to reading ability. Overall, our findings are still highly consistent with an intervention approach that emphasises phonics and phonological processing. However, they do underline the heterogeneity that exists among children with reading difficulties. Therefore, arguably the main implications of the case study findings for teachers and other practitioners is the need to tailor any interventions—including phonological interventions—according to the child's specific profile of

strengths and weaknesses. In addition, in terms of assessment, the findings warn against over-reliance on phonological awareness composites as these may obscure task-specific difficulties that are having a negative impact on a child's reading.

Some limitations to our approach should also be acknowledged. Firstly, due to the diversity of the phonological profiles observed in the study, the numbers of children demonstrating certain profiles were sometimes quite small. While we have been able to make some tentative observations, this has generally made it difficult to draw strong conclusions from crosstabulation of children's phonological and literacy deficits and this issue has instead been explored at the group level. Children with multiple phonological deficits did tend to have more diffuse reading difficulties (i.e., difficulties that registered across a range of different measures), those with the "classic" blending and elision deficits generally showed decoding difficulties, and those with RAN and/or, interestingly, isolation deficits, appeared more likely to struggle with the lexical reading measures and/or their reading rate. Future research may pursue more detailed exploration and cross-referencing of children's literacy and phonological profiles in order to determine whether more fine-grained differences in children's phonological skills have implications for their literacy performance. Similarly, future studies may also seek to explore the roles of FRD and hearing status in relation to children's individual phonological profiles more systematically. The most frequently observed phonological profiles remained unchanged regardless of children's FRD and hearing status and children with these additional risk factors did not appear more or less likely to demonstrate any particular profile of phonological impairment. However, the present study can again only offer tentative observations on this issue as the vast majority of the children had FRD and less than one third had a history of otitis media.

The current study has also adopted a broad definition of reading impairment. Children were included in the sample if they scored more than one *SD* below the population mean on one or more of the reading measures administered during their SEN assessment. As such, the children's reading difficulties are heterogeneous in nature and severity. Furthermore, there were no exclusion criteria relating to general ability and as such, while the children all have reading impairments of some kind, they may not all meet stricter diagnostic criteria for Dyslexia. Finally, vocabulary was the only available measure of wider language abilities, so it is unclear how many of the children in the sample may also have met criteria for a broader language impairment. Bearing these issues in mind, it would be wise to interpret the findings of the current study with reference to reading impairment as a broad term, rather than Dyslexia in a more specific, narrow sense. Future research may also seek to explore the heterogeneity of both phonological and non-phonological difficulties that occur in cases of reading impairment. However, the value of the approach adopted in the current study is that it maximises the available sample size for the multiple case study as well as reflecting the heterogeneity and complexity of the reading impairment referrals seen by practitioners.

In summary, the current study has demonstrated the substantial individual differences in phonological processing ability that may exist within samples of children with reading difficulties. These individual differences are relevant for theorists aiming to determine the structure of phonological processing as well as researchers and practitioners seeking to interpret the results of different phonological assessments and understand how they may relate to a child's literacy ability. Further research is required, particularly in relation to the latter point, but a more fine-grained understanding of the differences in children's phonological profiles may provide future opportunities for more individual tailoring of phonological interventions for struggling readers.

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CONFLICT OF INTEREST

We have no known conflicts of interest to disclose.

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