

Auditory processing, speech perception and phonological ability in pre-school children at high-risk for dyslexia: A longitudinal study of the auditory temporal processing theory

Bart Boets^{a,b,*}, Jan Wouters^b, Astrid van Wieringen^b, Pol Ghesquière^a

^a Centre for Disability, Special Needs Education and Child Care, Katholieke Universiteit Leuven, Belgium

^b ExpORL, Department Neurosciences, Katholieke Universiteit Leuven, Belgium

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Abstract

This study investigates whether the core bottleneck of literacy-impairment should be situated at the phonological level or at a more basic sensory level, as postulated by supporters of the auditory temporal processing theory. Phonological ability, speech perception and low-level auditory processing were assessed in a group of 5-year-old pre-school children at high-family risk for dyslexia, compared to a group of well-matched low-risk control children. Based on family risk status and first grade literacy achievement children were categorized in groups and pre-school data were retrospectively reanalyzed. On average, children showing both increased family risk and literacy-impairment at the end of first grade, presented significant pre-school deficits in phonological awareness, rapid automatized naming, speech-in-noise perception and frequency modulation detection. The concurrent presence of these deficits before receiving any formal reading instruction, might suggest a causal relation with problematic literacy development. However, a closer inspection of the individual data indicates that the core of the literacy problem is situated at the level of higher-order phonological processing. Although auditory and speech perception problems are relatively over-represented in literacy-impaired subjects and might possibly aggravate the phonological and literacy problem, it is unlikely that they would be at the basis of these problems. At a neurobiological level, results are interpreted as evidence for dysfunctional processing along the auditory-to-articulation stream that is implied in phonological processing, in combination with a relatively intact or inconsistently impaired functioning of the auditory-to-meaning stream that subserves auditory processing and speech perception.

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1. Introduction

Developmental dyslexia is a specific learning disability that affects around 5–10 percent of children and adults. It is characterized by severe reading and spelling difficulties that are persistent and resistant to usual teaching methods and remedial efforts (Gersons-Wolfensberger & Ruijsenaars, 1997). Historically, there has been a longstanding discussion about the etiology of these specific literacy problems. The origin has been sought in the visual, the auditory as well as in the cognitive-

linguistic domain (Vellutino, Fletcher, Snowling, & Scanlon, 2004).

The phonological deficit theory, which is the predominant etiological view on dyslexia, postulates that literacy problems originate from a cognitive deficit that is specific to the representation and processing of speech sounds (Snowling, 2000). Phonological deficits have been demonstrated in three broad areas (Wagner & Torgesen, 1987): phonological awareness (e.g. Liberman & Shankweiler, 1985; Mann & Liberman, 1984), retrieval of phonological codes from long-term memory (rapid automatized naming) (e.g. Bowers & Swanson, 1991), and verbal short-term memory (e.g. Catts, 1989; Mann & Liberman, 1984). Moreover, several prospective longitudinal studies have suggested a causal link between sensitivity to the phonological structure of words and later progress in reading acquisition (e.g. Bradley & Bryant, 1983; Wagner, Torgesen, & Rashotte,

* Corresponding author at: Centre for Disability, Special Needs Education and Child Care, Faculty of Psychology and Educational Science, University of Leuven, Vesaliusstraat 2, B-3000 Leuven, Belgium. Tel.: +32 16 32 61 82; fax: +32 16 32 59 33.

E-mail address: bart.boets@ped.kuleuven.be (B. Boets).

1994), although this relation is probably reciprocal (e.g. Bentin & Leshem, 1993; Morais, Bertelson, Carey, & Alegria, 1979).

Research in the underlying neurological dysfunction of dyslexia suggests that the phonological problems may result from a more fundamental deficit in the basic perceptual mechanisms that are responsible for auditory temporal information processing. Dyslexics tend to have difficulties processing linguistic and non-linguistic stimuli that are short and enter the nervous system in rapid succession (for reviews see, e.g. Farmer & Klein, 1995; McArthur & Bishop, 2001). Recent studies in this context focus more specifically on an impaired perception of dynamic aspects in the auditory signal itself, like amplitude and frequency modulations (Menell, McAnally, & Stein, 1999; Talcott et al., 1999, 2000, 2002; Van Ingelghem et al., 2005; Witton et al., 1998). Besides, subjects with reading impairments also tend to have difficulties with speech perception, in particular with the perception of degraded speech or speech-in-noise (for reviews see Boets, Ghesquière, van Wieringen, & Wouters, in press; McBride-Chang, 1995). It has been hypothesized that the basic deficit in perceiving auditory temporal cues causes a problem for the accurate detection of the rapid acoustical changes in speech. Consequently, the speech perception problem causes a cascade of effects, starting with the disruption of normal development of the phonological system and resulting in problems learning to read and spell (Talcott & Witton, 2002; Tallal, 1980; Wright et al., 1997). In this way, the supporters of the auditory temporal processing deficit theory do not deny the existence of the phonological deficit, but rather see it as secondary to a more basic auditory impairment (Ramus, 2003).

At a neurobiological level, two parallel pathways have been described that are implicated in the processing of auditory stimuli, speech perception, and higher-level phonological and linguistic information (Hickok & Poeppel, 2000; Scott & Johnsrude, 2003).

The first is an antero-ventral *auditory-to-meaning* pathway, linking areas in bilateral dorsal superior temporal gyrus (STG), presumably involved in the analysis of physical features of speech and complex non-speech sounds, to areas in the left anterior superior temporal sulcus (STS) and middle temporal gyrus (MTG) that are engaged in lexical-semantic and higher-level linguistic processing (Liebenthal, Binder, Spitzer, Possing, & Medler, 2005; Scott, 2003; Scott & Wise, 2003). Processing along this pathway happens in a hierarchically organized way: whereas the primary auditory cortex preferentially responds to pure tones, the antero-lateral association cortex (bilateral dorsal STG and STS) is responsive to progressively more complex spectro-temporal signals like amplitude modulations (AM) and frequency modulations (FM) (e.g. Giraud et al., 2000; Hall et al., 2002; Hart, Palmer, & Hall, 2003; for reviews see Scott, 2003; Scott & Wise, 2003). In primate studies it was demonstrated that neurons in this area were highly selective to the rate and direction of FM sweeps (Tian & Rauschecker, 2004). These temporal changes in amplitude and spectral shape are very important aspects of the speech signal and have been argued to form the basis of speech perception, which is accomplished along the left middle and anterior STS (i.e. phonemic perception and perception of intelligible speech; Liebenthal et al., 2005; Scott & Wise,

2003). It is still a matter of debate whether the left hemisphere specialization for speech perception relies upon its specialization for complex acoustic stimuli with rapid spectro-temporal variations (e.g. Belin et al., 1998; Zatorre & Belin, 2001), or whether it rather depends upon its functional specialization for familiar sounds for which category representations have been developed (Démonet, Thierry, & Cardebat, 2005; Gandour et al., 2002; Liebenthal et al., 2005).

The second pathway involved in speech processing is an *auditory-to-motor* stream that connects posterior temporal cortex to inferior parietal (i.e. angular gyrus and supra-marginal gyrus) and inferior frontal regions when assessing sub-lexical speech segments such as phonemes and syllables (Hickok & Poeppel, 2000). This parietal-frontal network, predominantly in the left hemisphere, interfaces auditory and articulatory representations of speech (Démonet et al., 2005), and also appears to be recruited in lexical retrieval (e.g. Misra, Katzir, Wolf, & Poldrack, 2004), grapheme-to-phoneme mapping (e.g. Jobard, Crivello, & Tzourio-Mazoyer, 2003; Simos et al., 2002), phonological working memory and phonological storage (e.g. Becker, MacAndrew, & Fiez, 1999; Démonet et al., 2005). The frontal areas have long been associated with articulation and naming (i.e. inferior frontal gyrus or Broca's area) (Fiez & Peterson, 1998; Murphy et al., 1997), but more recently they have also been shown to be implicated in subvocal rehearsal (Smith & Jonides, 1999), overt segmentation of speech (Burton, Small, & Blumstein, 2000) and higher level processes involved in the extraction of phonological elements (Gandour et al., 2002).

Although never stated as explicitly by its proponents, one might assume that the phonological theory of dyslexia would situate the core neurobiological deficit in the auditory-to-motor stream. In contrast, the auditory temporal processing theory would primarily situate it in the auditory-to-meaning pattern recognition stream, somewhere at the level where phonemic representations have to be extracted from the acoustic features in the speech signal. Evidently, according to the auditory temporal processing theory, dysfunctional processing should also be observed along the auditory-to-motor pathway as a secondary consequence of these aberrant phonemic representations.

Neuroimaging studies of *phonological processing* in dyslexic adults and children have consistently reported a reduction of activity in left temporoparietal language regions, often in combination with an increased – and probably compensating – activity in inferior frontal gyrus (Broca's area) (for reviews see Eden & Zeffiro, 1998; Habib, 2000; Temple, 2002). With regard to *speech perception and auditory temporal processing*, several ERP and MEG studies demonstrated deficient neurophysiologic processing in dyslexic adults and children (for review see Lyytinen et al., 2005; for a review of mismatch negativity studies see Kujala & Näätänen, 2001), and even in infants at family risk for dyslexia (e.g. Leppänen & Lyytinen, 1997; Lyytinen et al., 2005; Molfese, 2000) or language learning impairment (e.g. Benasich et al., 2006). The most consistent finding across these latter studies is the differential hemispheric activation between groups, where infants at-risk for dyslexia do not present the typical left hemisphere dominance in response to speech or rapid auditory signals (Lyytinen et al., 2005). Although most

ERP and MEG studies were not primarily concerned about source localisation, Neville and colleagues (Neville, Coffey, Holcomb, & Tallal, 1993) reported abnormal auditory ERP responses linked to processing in STG in a subgroup of reading and language impaired children that displayed abnormal performance on an auditory temporal discrimination task. However, somehow unexpectedly, the few imaging studies presenting auditory temporal stimuli to dyslexic subjects did not reveal dysfunctional processing in the auditory-to-meaning stream, but reported reduced activity in left frontal areas (Ruff, Cardebat, Marie, & Démonet, 2002; Temple, 2002). This is in line with neuroimaging studies in normal subjects that also reported the involvement of inferior frontal gyrus in the processing of rapidly changing speech and non-speech stimuli (Fiez et al., 1995; Joanisse & Gati, 2003; Johnsrude, Zatorre, Milner, & Evans, 1997; Müller, Kleinhans, & Courchesne, 2001). Yet, it remains unclear to what extent this frontal activation is specific for auditory temporal processing or rather is a general result of increasing task demands, placing a greater demand on secondary capacities such as working memory (Joanisse & Gati, 2003).

An additional anatomical argument in favour of the auditory temporal processing theory is the observation that the dyslexic brain does not only display a reduced left > right asymmetry of the planum temporale (Galaburda, Sherman, Rosen, Aboitiz, & Geschwind, 1985), but that the left posterior temporal cortex of dyslexics also presents a disorganization and smaller volume of white matter, as demonstrated by diffusion tensor imaging (Klingberg et al., 2000; Niogi & McCandliss, 2006). Moreover, this regional white matter diffusion anisotropy was significantly related to reading scores in children as well as in adults (see also Beaulieu et al., 2005; Deutsch et al., 2005). In normal subjects, it has been shown that the white matter volume of the auditory cortex is typically greater on the left as on the right, which could be a consequence of a greater number or denser myelination of fibres (Penhune, Zatorre, MacDonald, & Evans, 1996). The latter interpretation is supported by post-mortem tissue analysis of the posterior superior temporal lobe, which confirmed a greater volume of white matter on the left than on the right, and showed that this was due to greater myelin sheath thickness on the left (Anderson, Southern, & Powers, 1999). As axons with thicker myelin sheaths conduct faster and require a greater volume, these results suggest that asymmetry of myelination explains both the left hemisphere dominance for rapid sensory signal processing and the typically larger left planum temporale (Anderson et al., 1999; Zatorre & Belin, 2001). Given that dyslexic subjects present (a) a reduced left > right asymmetry of the auditory cortex, (b) an inverse hemispheric specialization in response to auditory and speech stimuli (Lyytinen et al., 2005), and (c) white matter anomalies in superior temporal cortex, these studies suggest that dyslexics' problems in auditory temporal processing and speech perception might be a consequence of reduced myelination in brain areas along the auditory-to-meaning speech-stream that is responsible for detailed spectro-temporal pattern analysis. Yet, no studies have been undertaken that directly relate white matter qualities to rapid auditory processing or speech perception.

Although theoretically attractive, the auditory temporal processing hypothesis has been hotly debated and has been facing growing criticism in recent years (e.g. Blomert & Mitterer, 2004; Denenberg, 1999; Mody, Studdert-Kennedy, & Brady, 1997; Nittrouer, 1999; Ramus, 2003; Rosen & Manganari, 2001). A first line of criticism emphasizes that most supporting studies only focused upon a particular skill (either low-level auditory processing, or speech perception or phonological processing), but did not assess the entire postulated pathway at all levels. Second, most of these studies only reported group data, hence suggesting that the observed deficits would be largely uniformly distributed among reading impaired subjects. However, more recent studies that also reported individual data, demonstrated that the lower-level sensory problems are usually only observed in a relatively small proportion of dyslexics. Of course, this makes it questionable whether the auditory problem could be regarded as *the* principle cause of reading problems (Ramus, 2003; Ramus et al., 2003; Rosen, 2003; White et al., 2006). Third, the observed auditory deficit does not always seem to imply rapid or temporal processing. Rather, it appears to encompass a broad range of complex spectro-temporal processing abilities that cannot easily be characterized in a coherent way (e.g. Amitay, Ahissar, & Nelken, 2002; Ramus et al., 2003; Rosen & Manganari, 2001).

Finally, a more general critique concerns the lack of longitudinal evidence on the intrinsic developmental and causal aspect of the auditory temporal processing theory. For example, few studies have assessed pre-school auditory processing and/or speech perception in children that will ultimately present reading difficulties (Heath & Hogben, 2004; Share, Jorm, Maclean, & Matthews, 2002), yielding mixed results on the relationship between pre-school thresholds and reading development. Although prospective neurophysiologic studies are underway (e.g. Jyväskylä longitudinal study of dyslexia, for review see Lyytinen et al., 2005), no systematic data have been reported contrasting normal versus reading impaired subjects. Thus, the auditory temporal processing theory has almost exclusively been based upon cross-sectional adult and school-aged data. Such data cannot discern whether the observed sensory deficits are the result rather than the cause of differences in reading ability. In this respect, Talcott and Witton (2002) suggested that it would not be too far-fetched to expect auditory skills of good readers to be more finely tuned than those of dyslexics by virtue of their more highly trained phonological systems.

With this longitudinal study we aim to investigate the different levels of the auditory pathway hypothesized to cause reading and spelling problems, while taking into account the aforementioned issues. We did not apply neurophysiologic measures, but we administered psychophysical and cognitive-linguistic tasks in order to determine whether the core bottleneck of literacy-impairment would be situated at the sensory level or at a higher-order phonological level. Indirectly, these data will also inform us about the level of the underlying neurological deficit.

Specifically, we assessed basic auditory skills, speech perception and phonological ability in a group of 5-year-old pre-school children at family risk for dyslexia, compared to

a group of well-matched control children. Auditory processing was assessed by means of three psychophysical tests: gap-in-noise detection (GAP), 2 Hz FM-detection (FM) and tone-in-noise detection (TN). With the GAP-detection task we tested ‘rapid and brief’ temporal processing. With the FM-detection task we assessed the processing of ‘dynamic stimuli’. With the TN task, a non-temporal control task, we verified the temporal specificity of any observed auditory deficit. To evaluate speech perception, a speech-in-noise perception task was administered. Phonological processing was assessed by a broad test battery comprising tasks for rapid automatized naming, verbal short-term memory and phonological awareness.

In the first place, all pre-school data were analyzed comparing the familial high-risk (HR) versus low-risk (LR) group. The results of this group comparison have been described in two previous papers (Boets, Wouters, van Wieringen, & Ghesquière, 2006a; Boets et al., *in press*) and can be summarized as follows. On every task the HR group demonstrated lower performance than the LR group, with the group difference being statistically significant for phonological awareness and speech-in-noise perception. For the other tasks the difference was in the expected direction, but did not reach statistical significance. Of course, the lack of a significant group difference for the auditory measures might be attributed to the fact that we did not study a well-defined clinical group but only a risk group that still might show substantial overlap with the non-affected control group. To clarify this aspect, we followed up these children through first grade of primary school and assessed their reading and spelling skills to determine whether or not they present literacy difficulties. Accordingly, in this paper we will describe the crucial retrospective analysis comparing pre-school (future) reading impaired children versus pre-school (future) normal readers. By measuring these diverse abilities in the same subjects, and by analyzing data both at a group level and at an individual level, we can verify whether literacy-impaired subjects present a consistent pattern of deficiencies across auditory, speech perception and phonological skills. Moreover, by assessing these abilities at a pre-school age and by applying a prospective design, we can verify whether the postulated problems precede the literacy problem.

2. Methods

2.1. Participants

Sixty-two children were included in the study (36 boys/26 girls) and followed from 1 year before the onset of formal reading instruction till 1 year into reading instruction. Half of the participants were children of ‘dyslexic families’, the so-called high-risk group (HR); the other half were control children of ‘normal reading families’, the so-called low-risk group (LR). All children were native Dutch speakers without any history of brain damage, long-term hearing loss or visual problems. Additionally, at the moment of data collection they did not present any gross deficiencies in visual acuity (Landolt-C single optotypes Snellen acuity > .85) and/or audiology (audiometric pure-tone average < 25 dB HL). The HR children were selected on a basis of having at least one first-degree relative with a diagnosis of dyslexia. The LR children showed no history of speech or language problems and none of their family members reported any learning or language problem. For every individual HR child we selected the best matching LR control child based on five criteria: (1) educational environment,

i.e. same school, (2) gender, (3) age, (4) non-verbal intelligence, and (5) parental educational level. Non-verbal intelligence was assessed by an adapted version of the Raven coloured progressive matrices (Raven, Court, & Raven, 1984), a collective non-verbal intelligence test measuring spatial reasoning. Parental educational level was assessed using the ISCED-scale (International Standard Classification of Education by UNESCO, 1997), by converting classifications on the original seven-point scale to a three-point scale. At the time of collecting the first kindergarten data, the mean age for both the HR and LR group was 5 years and 4 months, not being statistically different [paired $t(30) = 0.22, p = .83$]. The non-verbal IQ scores were slightly above population average (107 for HR group and 111 for LR group) and did not differ significantly [paired $t(30) = 1.88, p = .07$]. Both risk groups represented children from relatively higher educated parents, with Fisher’s exact test confirming that both groups did not differ in frequency distribution of the different parental educational categories ($p = .71$ for maternal and $p = .43$ for paternal educational level). Further details about the participants and the selection procedure are described in Boets et al. (2006a).

Full written consent was obtained from the parents of all children involved in the study. The study was approved by the local Ethics Committee.

2.2. Measures at pre-school age

2.2.1. Phonological tests

A broad test battery reflecting the three traditional phonological domains was administered. *Phonological awareness* was measured by three sound identity tasks and a rhyme task. *Verbal short-term memory* was measured by a digit span test and a non-word repetition test. *Rapid automatized naming* was assessed by a color and an object rapid naming task. A detailed description of the phonological tasks can be found in a foregoing paper (Boets et al., 2006a).

2.2.2. Tests for low-level auditory processing

Auditory processing was assessed by means of three psychophysical threshold tests. In the *GAP-detection test*, subjects had to detect a silent interval (gap) in a white noise stimulus. Threshold was defined as the minimum gap length required for detecting the silent interval. In the *FM-detection test* participants had to detect a 2 Hz sinusoidal frequency modulation of a 1 kHz carrier tone with varying modulation depth. Threshold was defined as the minimum depth of frequency deviation required to detect the modulation. In the *TN-detection task* participants had to detect two pure tone pulses (1 kHz, length = 440 ms) within a one-octave noise signal, centered around 1 kHz (from 707 to 1414 Hz, length = 1620 ms). Threshold was defined as the lowest signal-to-noise ratio (SNR) required for detecting the tone pulses. For all three auditory tests, thresholds were estimated using a three-interval forced-choice oddity paradigm embedded within an interactive computer game with animation movies (Laneau, Boets, Moonen, van Wieringen, & Wouters, 2005). The length of the gap, the depth of modulation and the amplitude of the sinusoidal pulses were adjusted adaptively using a two-down, one-up rule, which targeted the threshold corresponding to 70.7 percent correct responses. A threshold run was terminated after eight reversals and the threshold for an individual run was calculated by the geometric mean of the values of the last four reversals. After a short period of practice, comprising supra-threshold trials to familiarize the participants with the stimuli and the task, three threshold estimates were determined for every experiment. For the data we present here, the average of the best and second best threshold was used as an indicator of auditory sensitivity. A more detailed description of the stimuli, procedure and equipment can be found in Boets et al. (2006a).

2.2.3. Speech-in-noise perception task

In the speech-in-noise perception task seven lists of 10 high frequent monosyllables were presented monaurally with an inter-stimulus interval of 7 s. Simultaneously a continuous stationary speech noise, with an identical spectrum as the average spectrum of the word lists, was presented to the same ear, at a fixed level of 70 dB SPL. Words were presented at -1 , -4 and -7 dB signal-to-noise ratio (SNR). Before administration of the six test lists (3×2), one list was presented at an SNR of $+4$ dB as a practice list. The child’s task was to repeat the words as accurately as possible, resulting in a percentage correct word score for every test list.

2.3. Literacy measures at the end of first grade

To assess literacy skills, a standardized spelling test (Dudal, 1997) and six standardized reading tests were administered at the end of Grade 1 (after receiving 1 year of formal reading instruction). The one-minute real-word reading test and the pseudo-word reading test (van den Bos, Spelberg, Scheepstra, & De Vries, 1994) measure respectively real-word reading and non-word reading. To further discern between reading accuracy and reading speed, we constructed and administered four additional reading tests based on the description provided by de Jong and Wolters (2002): the real-word reading accuracy test, the real-word reading speed test, the non-word reading accuracy test and the non-word reading speed test. To make these tests applicable for the diagnostic process, age norms were collected in a large-scale pilot study (Peeters, Ghesquière & Boets, 2005).

2.4. Data collection

Data collection was carried out by qualified psychologists and audiologists. All tests were administered individually during several sessions adding up to approximately 10 h of testing for every subject. After every subtest children were rewarded with little gadgets. Testing took place in a quiet room at the children's school. Since the LR child was selected from the HR child's classmates, both children could always be tested in exactly the same circumstances. Phonological and auditory data were collected during the first trimester of kindergarten; speech perception data were collected during the second trimester. Literacy measures were collected in the last month of Grade 1.

2.5. Statistical analysis

Prior to analysis, all data were individually checked for outliers, resulting in the removal of the non-word repetition and rapid automatized picture naming score of one subject (subject 61). As this subject presented severe articulation problems, it was impossible to judge whether the poor non-word repetition score reflected weak verbal short-term memory or rather was a consequence of difficulties in pronunciation. During rapid automatized picture naming this same subject was interrupted twice by a teacher entering the test room, resulting in a much poorer score for picture naming as for colour naming. For another subject (25) the speech-in-noise perception data were discarded as a consequence of technical problems.

To obtain a normal distribution, thresholds for GAP and FM were log₁₀-transformed. Generally, all data were analyzed using mixed model analysis (MMA) (Littell, Stroup, & Freund, 2002), taking into account the clustered nature of the data (i.e. matched pairs attending the same school). Although the original HR and LR group as well as the reading defined groups (see below) did not differ for age, non-verbal intelligence or parental educational level, these variables were additionally controlled for in our analyses. Concretely, a series of (repeated) MMA's was calculated with school (=pair number) as a random variable and participant group as the fixed between-subject variable. Age, non-verbal IQ and educational level of both mother and father were added as fixed (co)variables. All post hoc analyses were corrected for multiple comparisons using the Tukey procedure ($\alpha = .05$).

Table 1

Mean performance (and standard deviations) on literacy achievement tests at the end of Grade 1 for the high-risk and low-risk group

	HR (<i>n</i> = 31)		LR (<i>n</i> = 31)	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Spelling	46	13	53	5
One-minute real-word reading	16	8	22	9
Pseudo-word reading	17	9	22	9
Real-word reading accuracy	22	13	28	10
Real-word reading speed	60	33	78	32
Non-word reading accuracy	14	11	21	11
Non-word reading speed	40	23	54	26

3. Results

3.1. Defining literacy groups at the end of Grade 1

Descriptive statistics summarizing the performance of the HR and LR group on the reading and spelling measures are shown in Table 1. A paired-wise MMA incorporating age, non-verbal intelligence and parental educational level as covariates revealed a significant group difference for every single literacy measure ($p < .01$).

The contemporary definition of dyslexia for the Dutch language area (Committee on Dyslexia of the Health Council of The Netherlands, see Gersons-Wolfensberger & Ruijsenaars, 1997) emphasizes that the diagnosis does not only depend upon the observation of severe reading and spelling problems (below the 10th percentile), but also requires these problems to be persistent and resistant to the usual teaching methods and remedial efforts. However, after 1 year of formal reading instruction, this additional criterion of persistence cannot yet be verified. As a consequence, we will refer to the children currently demonstrating impaired reading and spelling ability as 'literacy impaired' instead of 'dyslexic'. To determine the reading groups, a LITERACY composite score was calculated averaging the standardized results (relative to population data) of all reading and spelling tests. In line with the <Pc10 criterion postulated by the Health Council of The Netherlands, a cut-off point of -1.3 *SD*'s on the LITERACY composite was taken as a criterion to delineate the literacy-impaired group. Applying this criterion resulted in three literacy-impaired subjects in the LR group (3/31 = 9 percent), and 9 in the HR group (9/31 = 29 percent), indicating a

Table 2
Characteristics of the participants

	HR-LI (<i>n</i> = 9)		HR-LN (<i>n</i> = 22)		LR-LN (<i>n</i> = 28)	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Age in months	63	3	64	3	64	3
Non-verbal IQ	108	7	106	16	111	14
Maternal educational level	2.4	0.7	2.6	0.7	2.6	0.6
Paternal educational level	2.4	0.5	2.1	0.8	2.4	0.6

Note. ANOVA revealed that there were no significant group differences for any of the subject characteristics (Tukey contrasts, $p < .05$). Parental educational level was calculated from ordinal data; the correspondence in frequency distribution of the different educational categories was also confirmed using Fisher exact test.

relatively increased risk of 3.0 for the genetically at risk subjects [$\chi^2(1) = 3.7, p < .05$]. Hence, on the basis of familial risk status and current literacy achievement, we defined four groups: a HR normal literacy (HR-LN) and a HR literacy-impaired (HR-LI) group, and a LR normal literacy (LR-LN) and a LR literacy-impaired (LR-LI) group. In the further analyses we will compare the results of the HR-LI, HR-LN and LR-LN groups. Data of the LR-LI participants ($n = 3$) were excluded because we wanted to restrict our clinical group to children showing both (1) the family risk for dyslexia and (2) already presenting a significant impairment in reading and spelling development.

Table 2 provides descriptive statistics for the three groups, indicating that they did not differ significantly regarding age, non-verbal intelligence and parental educational level. Since the groups were defined based on literacy achievement, it is evident that for all reading and spelling measures the HR-LI group differed significantly ($p < .0001$) from the two other groups (HR-LN and LR-LN), who themselves did not differ from each other.

3.2. Group comparisons for pre-school measures

Table 3 shows the performance of the three groups of children on the phonological tests. For every test, except the first-sound identity task and the digit span task, the HR-LI group scored significantly below the LR-LN group. Furthermore, it is interesting to note that the HR-LN group, in spite of a hitherto normal reading and spelling development, scored in between both other groups. For the end-sound identity task and the non-word repetition test, the difference between the HR-LN and the

LR-LN group was even significant, suggesting that familial risk is continuous rather than discrete.

A principal component factor analysis with varimax rotation confirmed that the test battery excellently reflected the traditional three-fold phonological structure: (a) the *phonological awareness factor* had high loadings of the three sound identity tasks and the simple rhyme task, (b) the *rapid automatized naming factor* had high loadings of both the color and object rapid naming tasks, and (c) the *verbal short-term memory factor* was completely determined by high loadings of the non-word repetition and the digit span task (for details, see Boets et al., 2006a). Consequently, for every phonological factor summarizing composite scores were calculated averaging the standardized scores of their constituent tests (AWARENESS, RAN and VSTM). In addition, a general phonological composite averaging the standardized scores of all phonological tests was also calculated (PHONOLOGY). Statistics for these four phonological composites are displayed in Table 3. To assist in the interpretation of the results, composite values were transformed to effect sizes relatively to the mean and standard deviation of the LR-LN group. As can be seen, the HR-LI group scored significantly lower than the LR-LN group on AWARENESS, RAN and PHONOLOGY, with the HR-LN group again scoring in between. For VSTM group differences did not reach significance.

Results on the low-level auditory measures are displayed in Table 3. For FM detection the HR-LI group scored significantly worse than the two other groups, who themselves did not differ from each other. Also for GAP and TN detection the thresholds of the HR-LI group tended to be increased, but this difference did not reach significance.

Table 3

Mean performance (and standard deviations) on pre-school phonological ability, auditory processing and speech perception for the high-risk literacy-impaired, high-risk literacy-normal and low-risk literacy-normal groups

	HR-LI		HR-LN		LR-LN	
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>
Phonological awareness						
Simple rhyme	5.1 a	2.8	6.6 ab	2.2	7.3 b	1.8
Rhyme identity	8.0 a	3.2	9.0 ab	2.4	10.1 b	1.5
First-sound identity	3.9	1.8	4.9	2.2	5.9	2.3
End-sound identity	4.0 a	1.4	4.7 a	2.4	6.3 b	2.3
Rapid automatic naming						
Colour naming	0.58 a	0.06	0.65 ab	0.13	0.71 b	0.16
Picture naming	0.58 a	0.09	0.65 ab	0.13	0.71 b	0.16
Verbal short-term memory						
Digit span	6.4	1.5	7.3	1.6	6.9	1.6
Non-word repetition test	16.1 a	3.9	17.1 a	5.8	21.3 b	6.5
Composite AWARENESS	−1.49 a	1.5	−0.75 ab	1.3	0.00 b	1.0
Composite RAN	−0.87 a	0.4	−0.49 ab	0.9	0.00 b	1.0
Composite VSTM	−0.62	0.6	−0.31	0.9	0.00	1.0
Composite PHONOLOGY	−1.65 a	1.3	−0.85 a	1.2	0.00 b	1.0
AV GAP (ms)	7.4	4.3	5.1	4.3	5.1	3.8
AV FM (Hz)	11.1 a	5.8	7.0 b	3.4	6.9 b	3.8
AV TN (dB SNR)	−6.2	2.1	−8.0	2.2	−7.8	1.5
Speech perception: SRT (dB)	−2.8 a	1.7	−3.6 b	1.6	−3.9 b	1.2

Note. Pairs with different letters differ significantly (MMA controlled for non-verbal IQ, age, parental educational level and school environment; Tukey contrasts, $p < .05$). For SRT weighted group means and *SD*'s are presented.

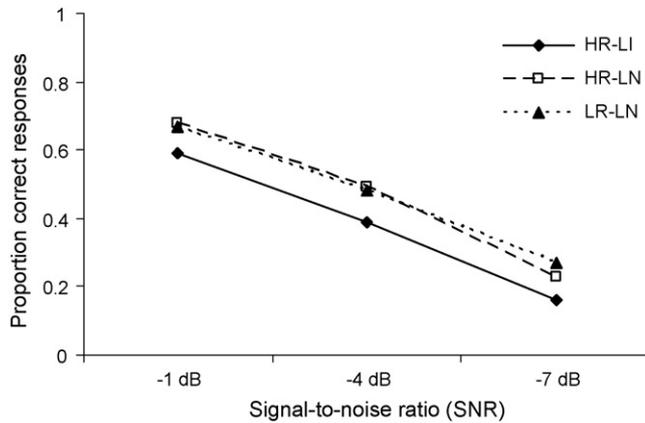


Fig. 1. Speech-in-noise perception: mean scores relating the proportion correctly perceived words to the relative level of the presented words (SNR).

Average results of the speech-in-noise perception test are depicted in Fig. 1. A repeated measures MMA with proportion correctly perceived words as dependent variable, group as between-subject variable, SNR as within-subject variable and with the same covariates as mentioned above, revealed a significant main effect for group ($p = .004$) and SNR ($p < .0001$) with the group \times SNR interaction being insignificant ($p = .50$). Post hoc analysis revealed that the HR-LI group differed significantly from both other groups, who themselves did not differ from each other. Additionally, to estimate the speech reception threshold (SRT: the signal level required for 50 percent correct responses), for every subject a logistic function was fitted to the data. In order to take into account the variable quality of the fits, the inverse standard error of the estimated parameters was added to the model as a weight variable (for details see Boets et al., in press). Table 3 shows weighted group means for SRT. As was the case with the repeated measures analysis, MMA with the weight variable and the same covariates as mentioned above demonstrated that the HR-LI group required a significantly easier signal-to-noise ratio than the two other groups to perceive 50 percent of the presented words correctly.

3.3. Individual deviance analysis

Since one of the goals of this study was to explore early indicators of reading and spelling impairment, and in view of the fact that group comparisons might mask significant individual differences, we also carried out analyses at the subject level. To decide which individual did and did not show abnormal performance, we adopted the two-step criterion as suggested by Ramus and colleagues (Ramus et al., 2003). Applying this procedure, the criterion for deviance was placed at 1.65 standard deviations of the 'purified' mean of the LR-LN group (one-sided in the direction indicative of deficiency), after first having excluded all deviant LR-LN subjects (by applying a similar 1.65 SD criterion, typically resulting in the removal of one or two deviant LR-LN subjects for each measure). A distribution analysis on the data of the 'restricted' LR-LN group confirmed their normality, indicating that the 1.65 SD criterion corresponds to the fifth percentile. Individual scores for the auditory and speech

Table 4

Proportion of deviant subjects for each reading group

	HR-LI ($n = 9$) (percent)	HR-LN ($n = 22$) (percent)	LR-LN ($n = 28$) (percent)
PHONOLOGY	78	45	14
GAP-detection	33	23	14
FM-detection	44	14	7
TN-detection	11	9	4
Speech (SRT)	33	5	4

perception measures and for the PHONOLOGY composite are plotted in Fig. 2, with Table 4 presenting the number of deviant subjects in each group (see Appendix A for an overview of all individual deviancy data). Inspection of the individual data reveals three main findings. First, for all measures and in particular for PHONOLOGY, it is evident that the HR-LI group showed an increased proportion of deviant subjects. Second, it is clear that not all HR-LI subjects scored in the deviant range. For PHONOLOGY this applied for about 80 percent of the group. For the auditory and speech perception measures this typically applied for about one third of the group. Third, inspection of the scores of the children of the HR-LI group reveals no straightforward pattern relating deficiencies across the different levels of processing skills. Although there might be a tendency for children with auditory deficiencies to present also more severe phonological deficits, this pattern does not extend to speech perception. On the contrary, the two HR-LI subjects without significant phonological deficiencies were among the ones presenting the most severe speech perception problems. Thus, in sum, we have to conclude that we are not able to demonstrate a consistent pattern of deficits across auditory, speech perception and phonological processing abilities for the subjects of the HR-LI group.

4. Discussion

In this study we investigated low-level auditory processing, speech perception and phonological ability in 5-year-old pre-school subjects who did not yet receive any formal reading instruction. Data were retrospectively analyzed, comparing three groups of subjects defined by first grade literacy achievement and family risk for dyslexia. Since it is questionable to firmly assess dyslexia in subjects that have received only 1 year of reading instruction, we preferred to use the term literacy-impaired instead of dyslexic. However, evidence for the validity of defining literacy groups at the end of first grade has been provided by research demonstrating that differences among children in reading and spelling (dis)abilities are quite stable over time, and that the majority of those identified as having reading difficulties in first grade continue to read poorly throughout their school years and beyond (McCardle, Scarborough, & Catts, 2001). Moreover, our clinical group was defined by showing both the actual literacy impairment and the increased family risk. In this respect, recent longitudinal research demonstrated that the proportion of HR subjects with reading problems increased from first to second grade (van Otterloo, Regtvoort & van der Leij, 2006). This

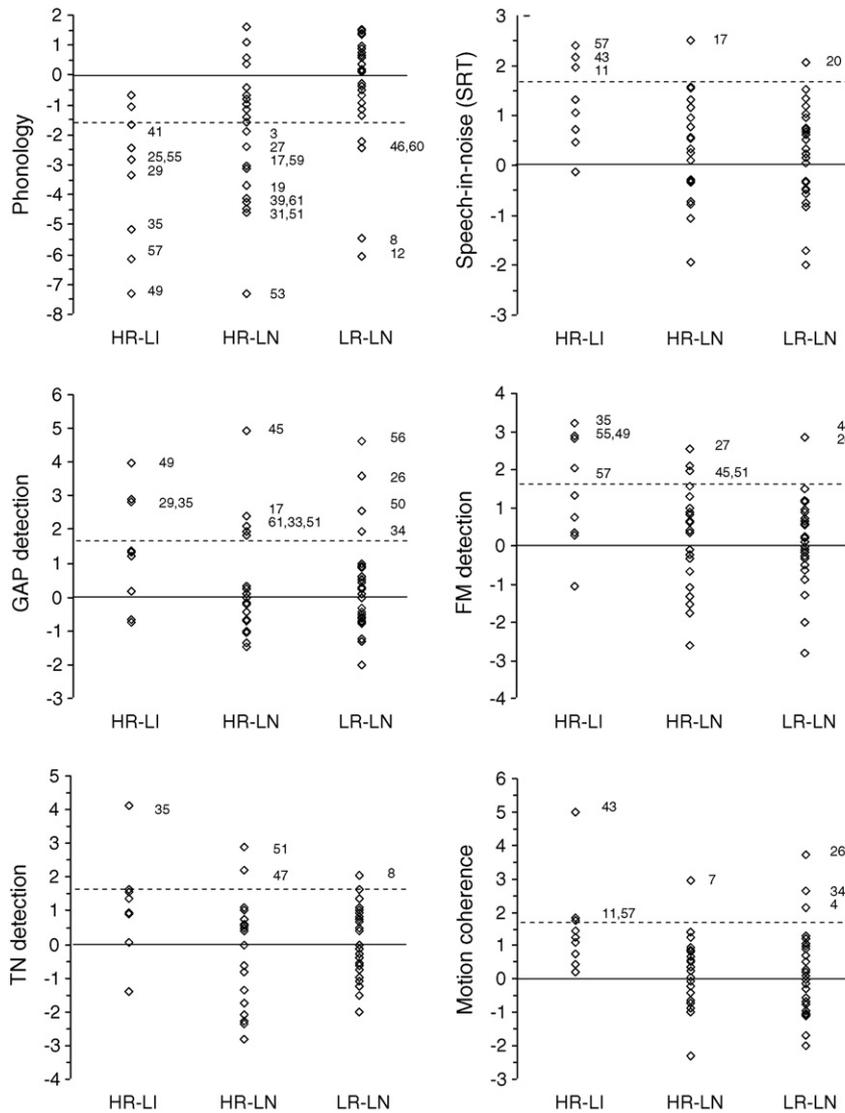


Fig. 2. Individual Z-scores for the PHONOLOGY composite and for all sensory measures. The solid line indicates the mean for all LR-LN subjects above Pc5; the dashed line indicates the chosen deviance criterion (1.65 SD deviating of the 'restricted' LR-LN mean). Deviant individuals are identified. *Note.* For PHONOLOGY a higher Z-score indicates better processing; in contrast, for all other measures a higher Z-score indicates reduced sensory sensitivity. For details regarding motion coherence data, see Boets et al. (2006b). For an interpretation of these data, see Section 4.

suggests that children of our HR-LI group will be diagnosed as dyslexic, if we retest them in second or third grade.

Comparing the HR-LI group with the LR-LN group, we observed a significant difference to the advantage of the LR-LN group for phonological ability, FM-detection and speech-in-noise perception. With respect to the phonological data, these results are consistent with the phonological deficit hypothesis and are in line with other prospective longitudinal studies revealing similar deficits in genetically at risk children (Scarborough, 1998).

Regarding the auditory data, the significant group difference for FM-detection is consistent with a whole series of psychophysical studies demonstrating that this measure differentiates reliably between adult and school-aged dyslexic and normal reading subjects (e.g. Stein & McAnally, 1995; Talcott et al., 2000; Talcott & Witton, 2002; Van Ingelghem et al., 2005; Witton et al., 1998). For the two other auditory tasks,

GAP- and TN-detection, the thresholds of the HR-LI subjects were increased but did not differ significantly from the HR-LN group. For TN-detection this lack of a difference was expected according to the auditory temporal deficit hypothesis since TN-detection was merely taken up as a non-temporal control task. For GAP-detection in contrast, both the theory and previous empirical evidence (McCroskey & Kidder, 1980; Van Ingelghem et al., 2005) predicted that the dyslexia-prone HR-LI group should show a significant deficit. Nevertheless, our failure replicating a significant deficit in GAP-detection does not stand alone: several other studies also failed to demonstrate a GAP-detection deficit in dyslexia, suggesting the task might not reliably tap the underlying processes that differentiate between dyslexic and normal reading subjects (Adlard & Hazan, 1998; McAnally & Stein, 1996; Schulte-Korne, Deimel, Bartling, & Remschmidt, 1998). In line with these findings, Phillips and colleagues proposed that a between-channel variant of the GAP-

detection task (where the pre- and post-gap markers occupy different critical bands) might be a more appropriate measure to probe the perceptual mechanisms involved in stop consonant speech discrimination (Phillips, Taylor, Hall, Carr, & Mossop, 1997).

With respect to speech perception, the considerable deficit in speech-in-noise perception is remarkable, but generally corroborates studies on adult and school-aged dyslexic subjects (e.g. Bradlow, Kraus, & Hayes, 2003; Brady, Shankweiler, & Mann, 1983). Regardless of the actual signal-to-noise ratio, children of the HR-LI group perceived on average about 10 percent fewer words than their peers. Applied to a regular classroom situation, typically characterized by a lot of background noise, this implicates that a substantial amount of communication and instruction is missed by these children. Undoubtedly, this might have far-reaching implications for scholar development in general and language and phonological development in particular.

Taken together, we observe that the HR-LI children *as a group* present significant concurrent difficulties in dynamic auditory processing, speech perception and phonological ability. Since these measures have been administered before children received any formal reading instruction, it is clear that the observed deficits precede the literacy problem and are not merely the result of lacking reading experience. This makes it very tempting to consider them as potential causal factors of the literacy problem. Moreover, in a foregoing paper we demonstrated that dynamic auditory processing is related to speech perception, which on its turn is related to phonological awareness and subsequent literacy development (see Boets et al., *in press*). Integration of all these measures within one causal path analysis also yielded a fairly satisfying fit of the proposed model (see Boets, Wouters, van Wieringen, De Smedt, & Ghesquière, *submitted for publication*).

So, from a global perspective our data appear to be quite in favour of the auditory temporal processing theory. However, if we *inspect the data at the level of the individual subjects*, we notice several findings that are not in line with the theory. First, it is clear that not all literacy-impaired subjects demonstrate an auditory and/or speech perception deficit. Of course, it could be argued that we did not assess *all* crucial aspects of sensory processing or that our psychophysical tasks might have lacked differentiating sensitivity. Yet, we would like to emphasize that the test battery that we administered is among the broadest applied in this research tradition, including several tasks that have proven to differentiate reliably between normal and reading-impaired subjects. The observation that only a relatively small proportion of HR-LI subjects demonstrate an auditory and/or speech perception deficit indicates that deficient sensory processing is not a necessary condition to develop reading or spelling problems—a conclusion that clearly conflicts with the assumption that the sensory problem would be at the basis of the reading problem (see Bishop, Carlyon, Deeks, & Bishop, 1999 for a similar conclusion regarding specific language impairment).

Second, some normal reading subjects do also show auditory and/or speech perception problems. This indicates that deficient sensory processing is neither a sufficient condition to develop reading or spelling problems (again see Bishop et al.,

1999; Bishop and Carlyon et al., 1999 for a similar conclusion regarding specific language impairment). Apparently, children growing up with quite severe auditory and speech perception problems can still develop normal literacy skills. This could imply that either lower-level sensory processing only plays a limited role in the development of literacy ability or that some of these sensory-impaired children can rely upon compensatory mechanisms to overcome their basic impairments.

Third, it is not possible to demonstrate a consistent pattern of deficiencies across auditory processing, speech perception and phonological abilities, neither for individual subjects of the HR-LI group, nor for individual subjects of the normal reading groups. Actually, even at a group level it is not possible to demonstrate such a consistent pattern. For instance, if we focus upon the group results of the children at family risk for dyslexia that do not (yet) present an actual literacy problem (HR-LN), it becomes clear that these children do present a mild phonological deficit but they do not present the lower-level auditory or speech perception deficit. Obviously, the observation of this partial dissociation between phonological and sensory deficits poses a problem for the auditory temporal processing theory, since it demonstrates that phonological impairments are not necessarily secondary to lower-level auditory or speech perception problems. In particular, these data suggest that the occurrence of a phonological problem is largely genetically determined (or at least by familial risk status) and is partly irrespective of actual literacy achievement. In contrast, the presence of a sensory problem is less genetically determined, but seems to be related to other factors that co-occur with literacy problems. This conclusion is well in line with data of Bishop and colleagues (Bishop et al., 1999; Bishop and Carlyon et al., 1999) and Olson and Datta (2002) who demonstrated in twin studies that in contrast to the highly heritable phonological skills, sensory skills depend less on genetic and more on environmental influences. Within the framework of an integrative neurobiological theory of dyslexia, Ramus (2004b) recently suggested that these co-occurring sensori(motor) problems might be the consequence of elevated levels of foetal testosterone that are mostly influenced by non-genetic factors.

It is evident that the aforementioned objections question the validity of the auditory temporal processing theory. However, it is important to note that the same objections also partially apply to the phonological theory. Indeed, also with regard to phonology some HR-LI subjects presented relatively preserved skills, whereas some normal reading subjects demonstrated extremely severe phonological deficits (even within the low-risk group). This implies that even the more established phonological theory is not able to explain the whole story of literacy impairment. In this respect, it is striking that the two HR-LI subjects without significant phonological problems, were among the ones presenting the most severe problems in visual coherent motion detection (see Fig. 2 and Appendix A; for details see Boets, 2006; Boets, Wouters, van Wieringen & Ghesquière, 2006b). This task has been suggested to be a sensitive measure of visual magnocellular processing, which on its turn has been postulated to play an important role in the development of orthographic skills and subsequent reading and spelling skills (e.g. Stein,

2001, 2003; Stein & Walsh, 1997). Hence, our data seem to confirm that some subjects (with relatively intact phonological abilities) develop literacy problems as a consequence of a specific visual dysfunction. On the other hand, our data also reveal that a substantial number of subjects presents quite severe pre-school deficits in auditory processing, speech perception or phonological processing and nevertheless is still able to develop normal literacy skills. Thus, although deficient sensory processing and in particular deficient phonological processing tends to be a risk factor to develop literacy problems, neither of both is a sufficient nor a necessary condition to cause these problems. This suggests that whether one develops reading problems or not, still depends upon additional factors executing an aggravating or protective influence. Actually, recent work has suggested that general language ability might be an important moderator variable in the process of literacy achievement. For instance, Snowling and colleagues demonstrated that children at risk for dyslexia because of speech difficulties (Nathan, Stackhouse, Goulandris, & Snowling, 2004) or because of a family history of dyslexia (Snowling, Gallagher, & Frith, 2003) can overcome their emerging phonological problems and yet develop normal literacy skills under condition that they can rely upon strong compensatory language abilities.

Overall, these findings substantiate that reading and writing is a complex multifaceted activity that involves a dynamic interplay of multiple sensory and cognitive-linguistic processes, moderated by various unspecified environmental or higher-order cognitive influences. Deficits at any level might interfere with normal literacy development. Comprehensive theories like the phonological theory, the auditory temporal processing theory or the visual magnocellular theory are nevertheless important and necessary to guide and stimulate scientific research, but it is an illusion to expect them to explain the full complexity of literacy development. Together with the growing awareness that there does not exist one uniform manifestation of dyslexia, researchers also start to realise that no single all-embracing cause or theory will be found. In line with this evolution, one can also notice a broader conceptual change from a deterministic single-cause model of developmental and learning disorders towards a probabilistic and multifactor model (Pennington, 2006).

Given the evidence for a multifactor account of literacy problems, it is difficult to relate our data to a uniform and localized brain deficit. In this respect, it should be noted that many areas of the brain have been found to be ‘different’ in literacy-impaired subjects on average (e.g. Habib, 2000), but the functional significance has not been established (Ramus, 2004a). Notwithstanding, with respect to the two cortical pathways described in the introduction of this paper, our data suggest that the majority of literacy-impaired subjects would present neurophysiologic dysfunctions along the auditory-to-motor stream implied in various aspects of phonological processing and grapheme-to-phoneme mapping. This is in line with the bulk of imaging studies confirming dysfunctional parieto-temporal processing in reading-impaired subjects during phonological tasks (for review, see Temple, 2002). Given that only a minority of literacy-impaired subjects presented auditory and speech perception problems (and mostly following a rather inconsistent

pattern), we would not expect to observe major neurophysiologic dysfunctions along the auditory-to-meaning stream (or only in specific subgroups of subjects). This is in contrast with the main hypothesis of the auditory temporal processing theory, but is supported by findings of the few imaging studies that investigated auditory temporal processing in reading-impaired subjects (Temple et al., 2002; Ruff et al., 2002). Finally, in view of the evidence that a small subgroup of subjects presented literacy problems that are probably related to a deficit in visual magnocellular processing, one might expect to observe small anomalies along the dorsal visual stream which is particularly involved in magnocellular processing. This was indeed confirmed by fMRI studies demonstrating reduced activity in literacy-impaired subjects in area V5/MT in response to coherent motion stimuli (Demb, Boynton, & Heeger, 1997; Eden et al., 1996).

To summarize, the present study calls for a multifactor account of literacy development. There is most evidence to situate the core of the reading and spelling problem at the level of higher-order phonological processing. Low-level auditory and speech perception problems are relatively over-represented in the group of literacy-impaired subjects and they might possibly aggravate the phonological and literacy problem. However, it is unlikely that they would be at the basis of these problems. Finally, there is also evidence that a particular visual magnocellular problem might independently contribute to the development of literacy problems in a subgroup of subjects.

It should be noted that this conclusion, which is based upon a retrospective analysis of data collected in pre-school children, is particularly well in line with findings of Ramus et al. (2003) and White et al. (2006) who adopted a similar design to study a group of adults and school-aged children with dyslexia.

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Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at doi:10.1016/j.neuropsychologia.2007.01.009.

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