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Multivariate predictive model for dyslexia diagnosis

Guylaine Le Jan · Régine Le Bouquin-Jeannès · Nathalie Costet · Nolwenn Trolès · Pascal Scalart · Dominique Pichancourt · Gérard Faucon · Jean-Emile Gombert

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Abstract Dyslexia is a specific disorder of language development that mainly affects reading. Etiological researches have led to multiple hypotheses which induced various diagnosis methods and rehabilitation treatments so that many different tests are used by practitioners to identify dyslexia symptoms. Our purpose is to determine a subset of the most efficient ones by integrating them into a multivariate predictive model. A set of screening tasks that are the most commonly used and representative of the different cognitive aspects of dyslexia was proposed to 78 children from elementary school (mean age=9 years±7 months) exempt from identified reading difficulties and to 35 dyslexic children attending a specialized consultation for dyslexia. We proposed a multi-step procedure: within each category, we first selected the most representative tasks using principal component analysis and then we implemented logistic regression models on the preselected variables. Spelling and reading tasks were considered separately. The model with the best predictive performance includes eight variables from four categories of tasks and classifies correctly 94% of the children. The sensitivity (91%) and the specificity (95%) are both high. Forty minutes are necessary to complete the test.

Keywords Diagnosis · Dyslexia · Logistic regression · Multivariate analysis · Predictive model

G. Le Jan · R. Le Bouquin-Jeannès (✉) · N. Costet · G. Faucon
INSERM, U 642, Rennes 35000, France
e-mail: Regine.Le-Bouquin-Jeannes@univ-rennes1.fr
URL: www.ltsi.univ-rennes1.fr

G. Le Jan · R. Le Bouquin-Jeannès · N. Costet · G. Faucon
Université de Rennes 1, LTSI, Rennes 35000, France

N. Trolès · J.-E. Gombert
Université de Rennes 2, CRPCC, Campus de Villejean, 35000 Rennes, France

P. Scalart
IRISA/ENSSAT, 6 Rue de Kérampont, B.P. 447, 22305 Lannion Cedex, France

D. Pichancourt
Service MPR Pédiatrique du Centre Hospitalier Lannion-Trestel, rue Trestel, 22660 Trévou-Tréguignec, France

Introduction

Developmental dyslexia affects about 5% of school age children in France. It is traditionally defined as an enduring and heavy impairment of reading ability in spite of normal intelligence and adequate educational opportunities. Dyslexics have a specific disorder of written language and can have some associated deficits such as attention deficit, visuo-attentional deficit, auditory and memory deficits. Research on the origin of dyslexia has led to multiple theories. The most common hypothesis is the phonological theory which suggests that difficulties experienced by dyslexics in reading new words are generally explained in terms of their poor phonological skills (Casalis, 1995; Sprenger-Charolles, Cole, Larcet, & Serniclaes 2000; Casalis, 2003; Grainger, Bouttevin, Truc, Bastien, & Ziegler 2003), especially phonemic awareness, phonological short-term memory (Snowling, 2000) and more recently, the speed of lexical access (Wolf and Bowers, 1999). Other hypotheses showed that morphological awareness had an important part in reading acquisition (Colé, Marec-Breton, Royer, & Gombert 2003; Marec-Breton, Gombert, & Colé 2005) and dyslexic children seem to over-use this capacity to compensate their difficulties in reading. Visuo-attentional hypothesis (Ans, Carbonnel, & Valdois 1998; Valdois, Bosse, Ans, Carbonnel, Zorman, & David 2003) considers that a visuo-attentional deficit can also be present in dyslexia and reduce the perceptual span. The visuo-attentional processing may serve to correctly extract graphemes from words and match them to abstract categories stored in lexical memory. A deficit in visuo-attentional processing would be implicated in poor performance of speed and visual information processing. The cerebellar theory (Fawcett, Nicolson, & Dean, 1996; Nicolson, Fawcett, & Dean 2001) suggests that dyslexics have motor problems (equilibrium, motor coordination, automatism deficits, etc.). It would explain their phonological deficit as a motor deficit affecting articulatory coding and these deficits would be associated to the dysfunction of cerebellum. Finally, an auditory deficit, which would be the basis of a categorical perception deficit, is supported by Tallal's findings (Tallal, 1980). As a matter of fact, dyslexics' performance is lower than that of control children in non-speech auditory tasks such as judgments of temporal order between acoustic stimuli. Even if performance on these tasks does not have a straightforward implication in speech perception (Serniclaes, Sprenger-Charolles, Carré, & Démonet 2001) several studies suggest that children with dyslexia present less categorical capacities than the average reader in the way they perceive phonetic contrasts. These multiple theories led to various diagnosis methods and treatments which can be inadequate (equilibrium exercises (Reynolds, Nicolson, & Hambly 2003), eye occlusion (Stein & Fawler, 1985; Stein, Richardson, & Fowler 2000), training of the awareness phonological (McPhillips, Hepper, & Mulhern 2000)).

The review of literature on dyslexia (INSERM, Expertise collective, 2007) shows that (1) on the one hand, reading and spelling tasks are good predictors of dyslexia, (2) on the other hand, some garden-variety children may have poor reading/spelling as dyslexic children but cannot be "labelled" as dyslexics. These somewhat opposite observations guided our approach to propose a rough model of dyslexia that can be called an "interim diagnostic tool", to help clinicians in their diagnosis in a manageable amount of time before investigating more deeply the specific causes for a future rehabilitation. We want to evaluate whether, once reading and spelling difficulties were identified in a subgroup of children, there is possibility to identify, with a time-limited test, primary underlying causes of the trouble through a condensed diagnostic assessment that would provide clinicians information on areas requiring more detailed and thorough investigation for intervention planning. In this way, the proposal of a model could help in intervention development. Such an assessment tool is intended to be recommended as a priority to children already presenting great difficulties in reading/spelling and for whom dyslexia is suspected. Our objective is to develop an “interim diagnostic tool” which:

– Integrates the heterogeneity in dyslexia etiology
– Evaluates, for children who have been identified as struggling readers (through the reading and spelling tasks), the specific nature of the underlying cause for reading difficulties, in order to prescribe appropriate interventions

In this sense, the tasks of reading and spelling are viewed as upstream stages in the diagnosis, since reading/spelling abilities alone do not differentiate poor readers from dyslexics (the case of poor readers will be discussed in the following).

Practically, an extensive set of screening tasks assessing cognitive impairments involved in dyslexia have been integrated in a software platform developed in a previous work (Le Jan, Troles, Le Bouquin Jeannès, Faucon, Gombert, Scalart et al. 2008). These tasks were organized in ten categories according to the cognitive deficit addressed: reading, spelling, memory, attention, metaphonology, phonological automatism, morphology, visual attention, motor and auditory tasks and were tested on 91 garden-variety children and 35 dyslexic children. The purpose of our study consists in deriving a screening model of dyslexia based on a limited number of tasks, selected among the original complete set of tasks.

This contribution is organized as follows: first of all, we describe the database and the screening tasks. After examining the reading/spelling tasks to verify their prominent role in the analysis, we retain the most contributing variables to derive the most predictive model.

Methods

Participants

Ninety-one children from five classes of French elementary school (four classes of grade 3 and one class of grade 4) took part in this study during class hours. Their reading level was estimated by the “Alouette test” (Lefavrais, 1965) which indicates a lexical age (i.e. reading level) from a reading test of 3 minutes. The level is evaluated by the speed and the accuracy of reading. In order to clearly differentiate the performance of dyslexics from those of normal readers, it was important to avoid the presence of dyslexics among the sample of normal readers. Among the ninety-one children tested in schools, 13 exhibited a reading age which was more than 18 months below their chronological age. We were unaware of the origin of their reading difficulties and, as it was very likely that some of them were dyslexics, we decided to exclude these thirteen children from the study. The mean age of the 78 remaining normal readers (38 boys and 40 girls) was 9 years (SD=7 months), their mean lexical age being 9 years and 3 months (SD=1 year and 4 months).

Thirty-five dyslexic children (22 boys and 13 girls) were diagnosed during a specialized hospital consultation. Their mean age was 9 years and 7 months (SD=7 months), their mean lexical age being 7 years and 6 months (SD=12 months). They were provided with standard education despite persistent difficulties in school work and daily activities. The dyslexia diagnosis was made by a multidisciplinary team of medical practitioners and speech therapists, using the routine protocol of the hospital (according to the guidelines produced by the National Institute for Health and Medical Research; INSERM, Expertise collective, 2007). The criteria used for diagnosis referred to the definition of dyslexia from the ICD-10 (International Classification of Diseases) and the DSM IV (Diagnostic and

Statistical Manual of Mental Disorders): persistent troubles in learning reading and spelling in comparison to a normal population despite normal intelligence, sensory acuity, and instruction. The selection procedure to classify individuals as dyslexics was as follows:

1 All of the children were given the Odedys 2 battery of tests (Laboratoire des Sciences et de l'Education—UMPF, 2005) which includes the "Alouette test" to confirm that they exhibited more than a two-year lag in reading ability. A complete medical check-up was then performed including neurological, visual and audiometric examinations in order to exclude children with physiological deficiencies.
2 An additional multidisciplinary evaluation was conducted:

(a) The intellectual development was assessed using WISC III (Wechsler, 1991). All the children included in the database had an Intellectual Quotient (IQ) above 70 points.
(b) The oral language and spelling difficulties were evaluated with the following normed assessments: ELO (evaluation of oral language; Khomsi, 2001), L2MA (assessment of oral and written language, memory and attention; Chevrier-Muller, Simon, & Fournier 1997), DRA (rapid denomination; Plaza, Chauvin, Lanthier, Rigoard, Roustit, Thibault et al. 2002), NEEL (new tasks for language evaluation; Chevrier-Muller & Plaza, 2001), BELEC (Reading Tasks; Mousty, Leybaert, Alegria, Content, & Morais 1994).
(c) Some additional specific examinations were done when some major causal associated disorders were suspected from the previous steps: neuropsychological evaluation was realized in order to measure memory function (CMS, Cohen, 2001) and/or attentional, sensorimotor and executive functions (NEPSY, Korkman, Kirk, & Kemp 2003). Children with major deficits of attention, oral language, and/or motor functions were excluded from the protocol.

A synthesis was realized by a multidisciplinary team including a speech therapist, a medical practitioner, an occupational therapist, and a neuropsychologist. Children were classified as dyslexic when their scores in reading tasks were at least two standard deviations below the expected level according to their IQ and their chronological age under the condition that these children did not present other predominant cognitive deficit.

This protocol to classify children as dyslexic was independent from the battery of tasks used in this research.

The ethnicity of the children was not collected in the protocol, but the study took place in the North of Brittany (in the north-west of France) where the geographical origin of the population is very homogeneous and predominantly Caucasian. All children were native French speakers.

Procedures

Screening tasks were organized in ten categories based on the cognitive domains considered. These categories represent all the cognitive domains potentially affected in dyslexia and describe the cognitive development of the children. Within each category, some tasks were selected according to experts' advices (practitioners, neuropsychologists, researchers). The majority of the tasks are routinely used by speech therapists. We designed completely auditory tasks (signal generation and processing). The tasks were administered to dyslexic children by neuropsychologists. Normal readers were tested individually at school by our research team which was trained by neuropsychologists to administer the measures.

Among the ten categories of tasks, the spelling and reading ones play a major role in dyslexia detection because they are the first general symptoms that suggest a problem of dyslexia. The other categories of tasks explore more precisely specific limitations of the dyslexic children, according to the different hypotheses about the origin of dyslexia. These tasks are described below; the time frame of each category is indicated in parentheses.

Reading tasks (10 min) Children read four sheets of 20 isolated words which were presented together according to their frequency and regularity[1] (frequent regular words, frequent irregular words, infrequent regular words and infrequent irregular words) and two sheets of 20 isolated pseudo-words[2] and near phonological pseudo-words. For each sheet, reading speeds were calculated (number of correct answers divided by the reading time) and, for each sheet with irregular words, the number of regularizations[3] was noted, which results in a total of eight scores.

These tasks are commonly used to estimate the level of reading and to identify developmental dyslexia subtype (surface dyslexia, phonological dyslexia, or mixed dyslexia) according to the type of mistake. Surface dyslexia concerns subjects who can read regular words and pseudo-words but have deficit in reading irregular words (Coltheart, Mastersin, Bying, Prior, & Riddoch 1983). Phonological dyslexia concerns individuals who can read aloud both known regular and irregular words but have difficulties with non-words and with connecting sounds to symbols or with sounding words (Beauvois and Derouesné, 1979). Mixed developmental dyslexia integrates both preceding subtypes of dyslexia.

Spelling tasks (30 min) A dictation was extracted from the BELEC (Mousty et al., 1994). The children had to complete sentences with specific words, the goal being to determine which spelling mechanism they set up. The specific words include phonemes that belong to five categories of spelling: the spelling of the phoneme is systematic and independent of the context (acontextual consistency; one score out of 29 points), the spelling of the phoneme is systematic and depends on the context (contextual consistency; two scores out of 6 points for, respectively, frequent and infrequent words), the spelling of the phoneme is not systematic and depends on the context (contextual inconsistency; four scores out of 9 points for, respectively, frequent and infrequent words with dominant rules and frequent and infrequent words with minority rules), the spelling of the phoneme is derived from the morphology[4] (two scores out of 6 points for, respectively, frequent and infrequent words), and, finally, the spelling of the phoneme is not derived from the morphology; two scores out of 6 points for, respectively, frequent and infrequent words). So, 11 scores are derived from the spelling tasks, corresponding to the category of spelling, the frequency of word and the type of rules (minority or dominant) for the spelling of the phoneme depending on the context.

Memory tasks (6 min) Some studies about the development of verbal short-term memory in children (Sprenger-Charolles et al., 2000) argue that dyslexics have a poorer short-term memory than normal readers. It seems to be an additional indicator of dyslexia. Verbal short-term memory and working memory were evaluated by two tasks: forward and backward verbal span. Several lists of numbers were read to the child who had to memorize them and repeat them. The size of the list was progressively increased, the objective being

[1] Irregular words are words that do not obey to graphophonological rules of pronunciation.

[2] Pseudo-words are words that look like in sound like real words but do not exist.

[3] Regularization occurs when the child does not modify the usual pronunciation of a phoneme to read correctly an irregular word.

[4] Example in French: in the word "délicat" the last phoneme /t/ is not pronounced but is written to derive the feminine form of the word "délicate".

to see how many numbers the child was able to memorize. This number was noted. For the forward memory span, numbers had to be repeated in the same order as they were heard, for the backward span they had to be repeated in the inverse order. Working memory was calculated by the difference between forward and backward span scores. Visual short-term memory was tested by a forward visual span in using Corsi's blocks: a series of cubes with written numbers was pointed in a specific order by the examiner and the child had to point them in the same order. An increasing number of cubes were pointed until the child could not memorize the series anymore. Finally, for the memory tasks, three scores were derived: one score for verbal forward span (indicating how many numbers were repeated by the child), the working memory score and a forward visual span score (indicating how many cubes were memorized by the child).

Attention tasks (2 minutes) Some studies report a deficit of attention in dyslexic children and particularly an impairment of selective attention (Iles, Walsh, & Richardson 2000) which is essential to select relevant information. Attention tasks were included and extracted from the BREV ("Batterie Rapide d'EValuation des fonctions cognitives") (Billard, Gillet, Galloux, Piller, Livet, Motte et al. 2000): children had to cross out as quickly as possible all the "3" present among a table of integers between 1 and 9. The number of "3" crossed out during 20 and 60 s was noted.

Metaphonological tasks (25 min) The phonological hypothesis (Snowling, 2000) postulates that dyslexics have a specific impairment in phonological awareness. They have difficulties to deal with phonemic units of speech whereas this capacity is essential in learning to read and, similarly, learning to read increases the phonological awareness. In order to evaluate the capacity of children to identify and deal with phonemic units, four different tasks were assessed: phonemic segmentation task (split the word into phonemes), spoonerism task (switch syllables), initial phoneme elision task (delete the first phoneme of each word presented) and a task of judgment of rhymes (find the word which does not rime with three other words). These tasks are respectively graded out of 16, 10, 12, and 8 points (four scores).

Phonological automatism tasks (10 min) This category tests the phonological automatism set up by children. It includes naming speed (denominate as quickly as possible a set of letters and a set of colors) (Denckla & Rudel, 1976) and lexical discrimination (recognize whether the pronunciation of two words is the same or not). Three variables have been retained: naming speed of letters and colors and a score of lexical discrimination out of 20 points (one point for each correct answer).

Morphological task (5 min) The decomposition of words in morphological units is commonly used when learning to read, but some studies showed that dyslexic children seem to over-use this capacity to compensate their difficulties in reading, when compared to normal reader children of the same lexical age. A morphological task was therefore proposed where children had to identify a pseudo-affixed word among a list of affixed words[5]. Different lists mixing affixed and pseudo-affixed words were proposed. For this task, a global score out of 6 points (one point for each correct answer) was derived.

[5] An affixed word is composed of a core word and a prefix or a suffix (example: to rewrite). A pseudo-affixed word is a word that begins with a syllable looking like a prefix but which is not a prefix (example: to regulate). In French, an example is: "recoller" (affixed) and regretter (pseudo-affixed).

Motor task (10 min) Motor deficit hypothesis in dyslexic children is based on the cerebellum theory derived from the observation of a poorer performance of dyslexics in a large number of motor tasks. The cerebellum plays a role in speech articulation and a dysfunctional articulation may lead to deficient phonological representations.

Moreover, cerebellum is implicated in the automation of over-learned tasks such as reading. A weak capacity to automate may affect the learning of correspondences between graphemes and phonemes. Two motor tasks were extracted from NEPSY ("bilan NEuroPSYchologique"; Korkman et al., 2003): motor sequences where children had to execute manual motor sequences noted out of 60 points (one point for each correct manual motor sequence) and secondly, a "tapping" exercise which evaluates the digital sleight and motor speed. The speed of digital sleight was noted.

Visuo-attentional task (10 min) Dyslexics may have difficulties in the treatment of visual information when this information is presented rapidly (Valdois et al., 2003). These difficulties of visual temporal treatment may reduce the attentional window and may disturb reading. Visuo-attentional capacities were tested with a task of partial report of letters: the child had to fix a central point on a computer screen, then a series of five letters appeared during 250 ms and a dash came under one of the letters and disappeared. The child had to indicate which letter has been pointed. Several series were proposed where the position of the dash varied. Each position of the dash (for each position of letters) is indicated ten times. For each position (five positions), the number of correct answers was graded out of 10 points.

Auditory tasks (40 min) Some studies demonstrate that dyslexics may have a poorer categorical perception of some phonemic contrasts as /ba/ versus /pa/ (Serniclaes & Sprenger-Charolles, 2003). To evaluate categorical perception skills in dyslexic children, two exercises were proposed: an identification task where children had to identify a syllable, /ba/ or /pa/, and a discrimination task where they had to discriminate a pair of /ba/–/pa/ syllables along a voice onset time (VOT) continuum. VOT is the time between the release of the consonant and the start of vocal fold vibration (voicing); it is measured in milliseconds and quantifies the degree of phonetic voicing. The extremities of VOT continuum were constituted of two syllables /ba/ and /pa/ which differ by their VOT. Intermediate syllables allow to link the extremities using a progressive variation of 10 ms. By convention, when voicing starts before the release of the consonant, VOT is negative; when voicing and consonant release happen simultaneously, VOT equals 0 ms; when voicing starts after the release of the consonant, VOT is positive. When the syllables belong to distinct phonemic categories, a minimal difference of 20 ms between VOT values of two syllables is necessary to distinguish them (Liberman, Harris, Hoffman, & Griffith 1957). In this study, the continuum ranged from −40 ms to 40 ms and was generated using three reference French natural syllables: /ba/, /pa/, and /pha/ with, respectively, a VOT of −117 ms, +13 ms, and +70 ms (see Fig. 1).

To make a positive VOT, a part of /pha/ VOT was selected and inserted in /pa/ syllable. For the negative VOT, a part of /ba/ VOT was introduced in the beginning of the /pa/ syllable. The 0 point of continuum was realized in deleting the burst of /pa/ syllable. Nine stimuli with respectively a VOT of −40, −30, −20, −10, 0, +10, +20, +30, and +40 ms were created. In the identification task, the nine stimuli of the continuum were randomly presented ten times to the listener. For each stimulus, the listener indicated if he (she) heard /ba/ or /pa/ syllable. In the discrimination task, seven pairs of syllables which differ by a VOT of 20 ms and nine pairs of syllables which have the same VOT value were randomly

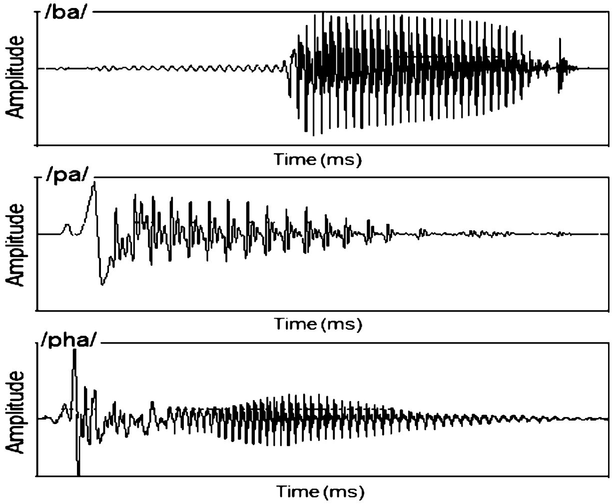


Fig. 1 Sound analysis of the French natural syllables, respectively /ba/, /pa/, and /pha/

presented eight times. For each pair, the listener indicated whether the syllables were identical or not. The identification task was evaluated through two variables: at first, the identification function given by the percentage of /pa/ responses was adjusted by a sigmoid function, then we computed its slope at the inflexion point VOT* and the identification threshold given by the difference between the values of the sigmoid function computed at the two abscissas: VOT* and VOT*+10 ms (Fig. 2).

The categorical perception is evaluated by the difference between the percentages of correct discrimination directly observed from the discrimination task and those expected according to the results of the identification task. In this way, we obtain seven variables evaluating the difference between observed and expected percentages of correct discrimination for each pair of syllables (seven pairs of syllables).

Three sessions of 50 min each were necessary to complete the whole set of tasks.

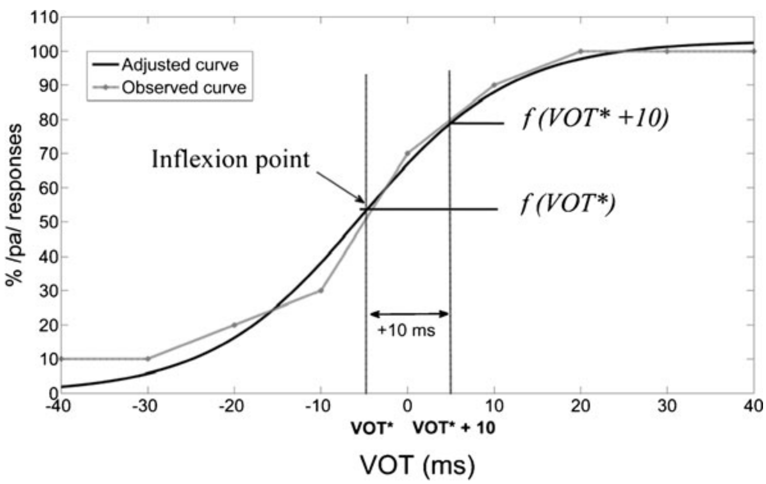


Fig. 2 Observed identification function and adjusted curve by a sigmoid function (slope and identification threshold)

Data analysis

Since reading and spelling tasks play a prominent role in screening dyslexia, we first studied these two tasks to compare the reading and spelling levels of the two groups (dyslexics and normal readers) and identify specific deficits involved in poor performance levels in the dyslexic group. The poor capacities of dyslexic children in these literacy tasks may be comparable to that of "poor readers" without cognitive impairments. A PCA was applied on all the variables and put in evidence the dominance of reading and spelling on the first factor: the reading and spelling variables explained more than 75% of the variance of the first factor. In our study, we want to underline the significance of cognitive variables as added-value in the determination of dyslexic and normal reader profiles. Consequently, it was of great importance that the effect of these variables should not be hidden by the predominance of reading and spelling variables. If the latter are crucial for the identification of children with potential dyslexia, the other variables could enable us to refine the diagnosis (dyslexics vs. normal readers). In order to underline the significance of cognitive variables, the variables representing the reading and spelling tasks were excluded for the next step.

The determination of the best predictive model for dyslexia screening proceeded then in two steps: (i) principal component analysis (PCA), (ii) selection of the best model based on logistic regression.

As the initial number of covariates was high (29), the number of possible models among which the best one has to be identified is huge. A direct automatic selection method was not reasonable in our case, since the number of cases of dyslexia compared to the number of variables is low (35 dyslexics/29 variables) (Steyerberg, Eijkemans, & Habbema 1999; Steyerberg, Eijkemans, Harrel, & Habbema 2000). A pre-selection among the initial variables was done using PCA in order to reduce the number of variables to be analyzed in the second step.

PCA provides factors ("components") defined as linear combinations of the original variables. The factors are successively obtained by performing a diagonalization of the initial correlation matrix between variables. The first factor is the most representative of the total variance of the original data set (Jollife, 2002; Duffi, Valencia, Mcanulty, & Waber 2001). Each extracted factor can be interpreted through the loadings of the initial variables on this factor. The highest loadings indicate that the factor is highly influenced by these variables.

The 29 screening variables belonging to the eight remaining categories correspond to eight cognitive domains involved in dyslexia. Separate PCAs were applied within each category in order to identify the most representative tasks of each category and to keep all categories represented before the modeling step.

In each PCA (within each category), the first factor was analyzed and the most contributing variables were selected. The selection criterion was the relative contribution (RCT) of the variables, defined as follows:

$$\mathrm{RCT}_j\left(v_{ij}\right) = \frac{\left[\mathrm{coordinate}\left(v_{ij}\right)\right]^2}{\mathrm{variance}\left(\mathrm{first\ principal\ component}\right)} \tag{1}$$

where j=1,...,8 and i=1,…, N_j where N_j is the number of variables in the category j.

The variable v_{ij} was considered to be representative of the first component if the value $\mathrm{RCT}_j(v_{ij})$ was greater than the average RCT_j, defined as follows:

$$\overline{\mathrm{RCT}}_j = \frac{1}{N_j}$$

where N_j is the number of variables in the category. $\overline{\text{RCT}_j}$ corresponds to the situation where all the variables equally contribute to the variance of the factor.

If $\text{RCT}_j(v_{ij}) < \frac{1}{N_j}$, the corresponding variable is excluded for the next step of the analysis. This method allowed us to select, within each category, the most representative tasks which will be used in the model-based final selection.

Logistic regression models and selection of the best model

The logistic model (Hosmer & Lemeshow, 2000) was chosen for modeling the probability of being dyslexic: the logit of the conditional probability of being dyslexic was modeled by a linear combination of the variables (scores obtained to the different tasks):

$$\log\left[\frac{P(\text{dyslexic}/X)}{1-P(\text{dyslexic}/X)}\right] = \alpha + \beta^t X + \varepsilon \tag{2}$$

where $X = [x_1, \ldots, x_p]^t$ is a vector composed of the p variables retained in the previous step, α is the intercept, β is the vector of p real coefficients to be estimated and ε the error.

This model is robust to non-normal distributions of explanatory variables (X) and can deal simultaneously with different types of variables (continuous, normally distributed or not, categorical variables). By default, we first introduced all the variables as continuous in the models. But as some of them had a highly skewed distribution, a lack of precision of their estimated parameters appeared. In such a case, an alternative categorical coding based on the quartiles was proposed: the raw value of the variable was replaced by the number of the corresponding quartile. The quartiles were defined within the normal readers sub-sample. The estimated relationship between the logit of the risk of being dyslexic and the variable was thus ordinal and no more linear. For each subject, the probability of being dyslexic was estimated from the model as follows:

$$P(\text{dyslexic}/X) = \frac{\exp[\alpha + \beta^t X]}{1 + \exp[\alpha + \beta^t X]} \tag{3}$$

A classification rule was derived: if the probability to be dyslexic is greater than 0.5, the subject is classified as dyslexic, otherwise he (or she) is classified as non-dyslexic. The performance of the logistic model was estimated by comparing the predicted status of each subject (dyslexic or not) to the real one. Since this resubstitution estimation of the classification performance is biased, we used a cross-validation method ("leave-one-out" method) to estimate the overall percentage of children correctly classified (hit ratio), the sensitivity (correct detection rate within dyslexic children), the specificity (correct detection rate within non-dyslexic children), the false-positive rate (percentage of children classified as dyslexic who were actually not dyslexic) and the false-negative rate (percentage of children classified as non-dyslexic who were actually dyslexic). The cross-validation method is only used to evaluate the classification performance of the model: the logistic model is run as many times as there are observations. Each time, one observation is left out from the sample, the model is estimated without it and the excluded observation is then classified using this model. The observation is thus identified as a well-classified observation or a classification error. At the end of the successive models and classifications, a non-biased classification matrix can be calculated. The receiver-operating characteristic (ROC) curve of the model was plotted and the area under the ROC curve (area under curve, AUC; Swets, 1988) was calculated. The ROC curve plots the sensitivity as a function of the false-positive rate (100−specificity (in percent (%)) for different cut-off points. A test with perfect discrimination has a ROC curve that passes through the upper left corner (100% sensitivity, 100% specificity). Therefore, the closer the ROC plot is to the upper left corner, the higher the overall accuracy of the test. On the contrary, a non discriminative test has a straight line between the lower left corner and the upper right corner as a ROC curve. This corresponds to the situation where the observations are randomly classified, independently from their scores on the screening tests. The area under the observed ROC curve quantifies the global discrimination performance of the function.

As the objective of this work was to select the most predictive but also parsimonious model, different models including more or less covariates were compared according to two criteria to maximize: first, the sensitivity and secondly the specificity of the derived classification rule. These criteria are not used in the common stepwise procedures where the usual stopping rules for inclusion (or exclusion) of variables in multivariate models are the significance level of their coefficients and/or the global AIC. These criteria cannot be formalized as a function to maximize or minimize but can only be estimated ad hoc as an output of the models. That is the reason why we used an approach where all the possible models, including all the possible subsets of variables, were compared (without embedment constraints).

Statistical analysis used the R software (ade4, ipred, klaR, DAAG, class, lspls, kpi, ROCR packages).

Results

Reading and spelling tasks

Performance in reading speeds and spelling scores obtained from normal readers and dyslexics are summarized in Table 1. Means and standard deviations for the two groups are presented. The effect of the regularity of the words on the reading speed is significant in both groups and the reading of pseudo-words is less accurate and less rapid than that of words. However, all reading speeds in dyslexic group are largely lower than those of the normal readers. Moreover, the scores in the dyslexic group are fairly similar for the reading speed of four types of words: infrequent regular words, frequent irregular words, near phonologically pseudo-words, and pseudo-words reading speed. The poor performance in reading pseudo-words and the presence of a regularity effect can therefore be attributed to the poor phonological representations in dyslexic readers. Reading pseudo-words requires good phonological skills and is especially problematic for dyslexics. These results suggest the existence of phonological and lexical access deficit in dyslexic children. For the spelling task, all the scores of the dyslexic group are inferior to those of the normal readers, except in the case of contextual inconsistency with dominant rules. These results show that dyslexic children are able to apply the correspondences between grapheme and phoneme provided that these correspondences depend on the context and the existence of a dominant rule for the phoneme to write.

A PCA was applied to the reading and spelling scores presented in Table 1. This analysis extracted 19 independent factors, with a maximum percentage of inertia of 56.58% on the first factor. The plot of the coordinates of the subjects on the first factorial plan is presented on Fig. 3. The dyslexic children are all projected on the positive part of the first factor, whereas the majority of normal readers are projected on the negative part. Factor 2 does not discriminate between dyslexics and normal readers.

In Fig. 4, the graph representing the coordinates of the variables indicates which variables are the most representative of the first factor. For all the tasks, the lowest level

Table 1 Comparison of normal readers and dyslexics on reading and spelling tasks

Variables	Normal readers (n=78), mean (SD)		Dyslexics (n=35), mean (SD)	
	Frequent words	Infrequent words	Frequent words	Infrequent words
Reading				
Reading speed of regular words[b]	1.69 (0.33)	0.95 (0.33)	0.97 (0.42)	0.36 (0.20)
Reading speed of irregular words[b]	1.20 (0.44)	0.55 (0.34)	0.40 (0.27)	0.12 (0.09)
Numbers of regularization mistakes[b]	0.78 (1.05)	6.47 (3.89)	2.37 (1.83)	10.47 (3.32)
Reading speed of near phonologically pseudo words[a,b]	0.73 (0.27)		0.34 (0.15)	
Reading speed of pseudo words[a,b]	0.71 (0.25)		0.31 (0.18)	
Spelling				
Acontextual consistency spelling[a,b]	27.88 (1.76)		25.71 (2.72)	
Contextual consistency spelling[b]	4.27 (1.66)	5.05 (1.36)	1.29 (1.36)	2.00 (1.51)
Contextual inconsistency				
Contextual inconsistency with dominant rules	6.97 (1.46)	8.68 (0.58)	7.35 (1.23)	8.32 (0.90)
Contextual inconsistency with minority rules[b]	4.05 (1.96)	7.61 (1.48)	1.38 (1.24)	4.91 (2.52)
Spelling derived by morphology				
Derived[b]	1.99 (1.48)	4.30 (1.42)	0.09 (0.28)	1.71 (1.36)
Not derived[b]	1.73 (1.30)	4.34 (1.13)	0.47 (0.55)	2.41 (1.37)

[a] Frequent and infrequent words are not distinguished for these categories

[b] Mann–Whitney test, p value<0.05

seems to be systematically projected on the positive part of the first factor, the best scores being projected on the opposite part. The graph indicates high correlations between reading and spelling tasks, except for the number of regularizations (R3 and R6) projected on the negative part, which is logical considering the way these scores are defined. The variable W4 corresponding to the spelling of frequent words with contextual inconsistency and dominant rules is the only variable which is not correlated with the other spelling and reading tasks.

Analyzing jointly children and variable's position on the first factor indicates that dyslexic children seem to cumulate a poor performance in all reading and spelling scores.

All these scores are highly correlated and highly discriminative to evaluate reading performance in dyslexic children but poor performance in reading is not specific to dyslexic children. Some more specific tasks are necessary to evaluate other limitations involved in dyslexia and the next step focused on these specific tasks, represented by 29 variables.

PCA within cognitive domains

The goal of the following analyses was to select the most representative tasks within each category. Tables 2 and 3 display the results for all categories of tasks for normal readers and dyslexics, except for reading and spelling tasks. The percentage of inertia of the first factor is high for all categories, except for the auditory one. We considered that the first factor summarized efficiently the cognitive domain underlying each category of variables and we used it to select the most relevant variables within each cognitive category using their relative contribution to this factor.

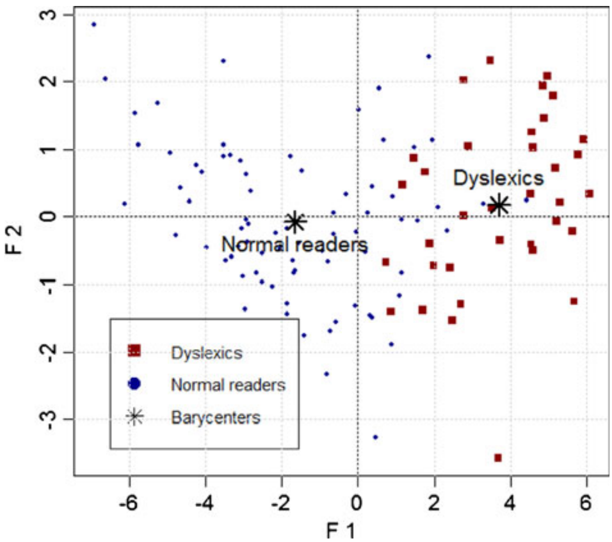


Fig. 3 PCA on reading and spelling tasks: plot of the coordinates of the children on the first factorial plan

As an example, within the metaphonological category, three variables are representative of the first factor (spoonerism, segmentation, and phoneme elision) while one variable (judgment of rhymes) shows a low correlation with the other scores of this category. The plot of the subjects on the first factor indicates that a majority of dyslexic children are projected on the negative part of the first factor, while the majority of normal readers are projected on the positive part. These results suggest that the three metaphonological variables of the first factor are "good candidates" to predict dyslexia.

The selected variables using PCA method are indicated by "*" in Tables 2 and 3. Finally, 20 variables representing every category were selected for the modeling.

Detection model by logistic regression

The selection procedure based on logistic regression allowed us to retain eight variables which are presented in Table 4 with the estimated parameters of the model (estimate, odds-ratio (OR), standard error (Std. Error) and *p* value). A significant *p* value (0.1 level) is found for four variables: spoonerism, morphology, partial report of letters and difference of VOT (10 ms; 30 ms) variables. These results suggest that these variables have a high contribution in the detection model and have a high capacity to explain dyslexia. The parameters of the model indicate lower scores of dyslexics in phoneme elision, spoonerism and partial report of letters (negative estimates) and a better performance of dyslexic children in the morphological task (positive estimate). The parameter of the identification slope is positive which suggests that after adjustment for the other tasks, the dyslexic children may have a good capacity in phoneme identification. Nevertheless, this estimate is very close to 0 and non-significant (p value=0.98). The parameters of the remaining auditory variables (VOT) are positive, except for the difference of VOT (10 ms; 30 ms). These results suggest that the difference between expected and observed scores is higher in dyslexic children at the extremities of the continuum (VOT(−40 ms; −20 ms) and VOT (20 ms; 40 ms)) than at intermediate positions.

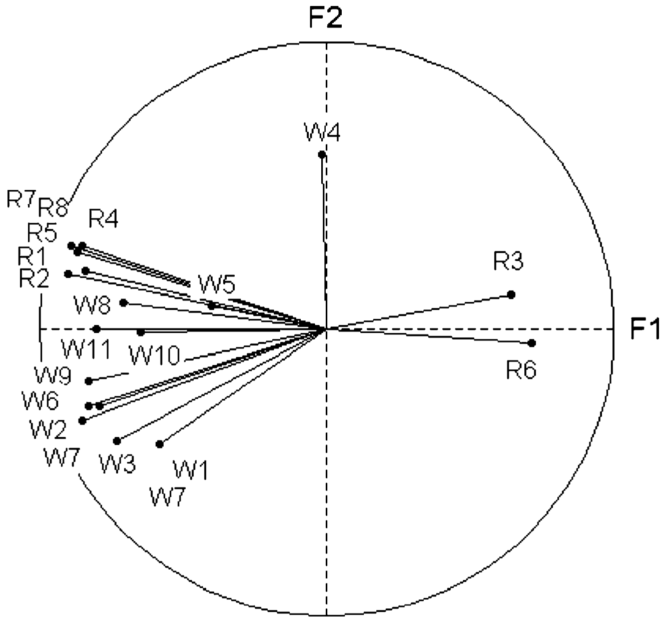


Fig. 4 PCA on reading and spelling tasks: plot of the coordinates of the variables on the first factorial plan. W1: Acontextual consistency spelling; W2: Contextual consistency spelling (frequent); W3: Contextual consistency spelling (infrequent); W4: Contextual inconsistency with dominant rules (frequent); W5: Contextual inconsistency with dominant rules (infrequent); W6: Contextual inconsistency with minority rules (frequent); W7: Contextual inconsistency with minority rules (infrequent); W8: Spelling derived by morphology (frequent); W9: Spelling derived by morphology (infrequent); W10: Spelling not derived by morphology (frequent); W11: Spelling not derived by morphology (infrequent); R1: Regular words (frequent); R2: Irregular words (frequent); R3: Number of regularization (frequent); R4: Regular words (infrequent); R5: Regular words (infrequent); R6: Number of regularization (infrequent); R7: Near phonologically pseudo words; R8: Pseudo words

The global predictive accuracy of the selected model (estimated by cross validation) is very high (94% of individuals correctly classified). The sensitivity and the specificity are also high (91% of dyslexics and 95% of normal readers correctly classified). The false-positive rate is 9% and the false-negative rate is 5%.

The ROC curve (Fig. 5) differs highly from the random ROC curve and the AUC is 0.95, which indicates a very high accuracy (Swets' interpretative guidelines (Swets, 1988)).

The selected variables belong to four categories of tasks (metaphonological, morphology knowledge, visuo-attentional and auditory tasks), and the administered measures require around 40 min to be completed by children.

Discussion

The PCA results report a selection of discriminatory variables by category which allows to keep a global representation of the cognitive domains frequently involved in dyslexia. The high percentage of inertia of the first factor within the majority of the categories shows good internal consistencies of the tasks to describe dyslexia.

In the predictive model, the two metaphonological variables (phoneme elision and spoonerism) highlight the prevalence of phonological deficits in developmental dyslexia. In the literature, a number of studies (Ramus, Rosen, Dakin, Day, Castellote, White et al. 2003;

Table 2 Selection of variables by PCA within categories of tasks (memory, attention, metaphonological, phonological automatisms, morphology knowledge)

Category	Normal readers (n=78), mean (SD)	Dyslexics (n=35), mean (SD)
Memory		
Verbal forward span[a]	5.28 (1.02)	4.66 (0.94)
Working memory span[a]	1.42 (0.99)	1.46 (1.01)
Visual span	5.00 (0.77)	5.14 (0.77)
Attention		
"3" crossing out (20 s)[a]	10.90 (3.00)	9.66 (2.76)
"3" crossing out (60 s)[a]	24.23 (3.65)	24.30 (3.93)
Metaphonology		
Segmentation[a]	11.19 (3.03)	9.03 (3.95)
Phoneme elision[a]	10.81 (1.44)	8.34 (2.72)
Judgment of rhymes	7.11 (1.06)	6.66 (1.28)
Spoonerism[a]	7.76 (1.89)	3.37 (1.97)
Phonological automatisms		
Lexical discrimination	18.62 (1.58)	18.97 (1.25)
Naming speed of letters[a]	1.72 (0.41)	1.41 (0.33)
Naming speed of colors[a]	1.08 (0.19)	0.93 (0.20)
Morphology knowledge		
Morphology[a]	4.83 (1.17)	4.83 (1.15)

[a] The variable contributes significantly to the first factor.

White, Milne, Rosen, Hansen, Swettenham, Frith et al. 2006) confirm this result and report that 70% of the dyslexics have a phonological deficit. This deficit is characterized by specific disorders in treating the correspondence between graphemes and phonemes and an inability to use the representation and the storage of sounds. Moreover, dyslexic children would not be able to use explicit learning because phonological capacities are inalienable (Gombert, 2003). All studies agree on the fact that phonological deficit is central in dyslexia, but some researchers suggest that it would be only the visible part of a more global underlying dysfunction. In order to detect most of the dyslexics, the predictive model proposes to link the two metaphonological variables to morphological, visuo-attentional and auditory variables. As for the morphological variable, results are in line with the idea defended by Casalis, Colé, & Sopo (2004) indicating that some dyslexics use the morphological regularities of the language to decode more efficiently the words while reading. The visuo-attentional variable in the predictive model supports the visuo-attentional hypothesis reported by Valdois, Bosse, & Tainturier (2004). The author based her theory on the difficulties the dyslexics meet to deal with fast, complex visual information. These difficulties result in a reduction of the width of visuo-attentional window also called visuo-attentional span, which corresponds to the number of items that can be simultaneously treated. This reduction would have an impact on the lexical spelling acquisition and would disrupt the learning of reading. In our visuo-attentional task (partial report of letters), the variable we retained corresponds to the identification score of the letter in the fourth position. In the dyslexic group, the number of correct identifications of this letter is lower than in the normal reader group. Some studies (Valdois et al., 2003) reported that phonological and visual processing impairment can dissociate in developmental dyslexia. A pattern of surface dyslexia may be associated to a visual processing disorder (poor

Table 3 Selection of variables by PCA within categories of tasks (motor task, visuo-attentional, auditory tasks)

Category	Normal readers (*n*=78), mean (SD)	Dyslexics (*n*=35), mean (SD)
Motor tasks		
Manual motor sequences[a]	51.85 (5.62)	48.42 (3.82)
Tapping[a]	93.34 (29.85)	86.72 (9.99)
Visuo-attentional		
Partial report of letters (position 1)[a]	8.32 (1.99)	7.66 (2.21)
Partial report of letters (position 2)[a]	7.21 (2.19)	5.71 (2.69)
Partial report of letters (position 3)	8.32 (1.48)	7 (2.25)
Partial report of letters (position 4)[a]	7.32 (2.02)	5.66 (2.53)
Partial report of letters (position 5)	7.77 (2.44)	7.06 (2.47)
Auditory		
Slope identification of speech sound[a]	0.11 (0.18)	0.11 (0.18)
Identification threshold[a]	3.48 (1.17)	2.83 (1.41)
VOT (-40 ms; -20 ms)[a]	0.05 (0.05)	0.07 (0.06)
VOT (−30 ms; −10 ms)	0.07 (0.07)	0.09 (0.09)
VOT (−20 ms; +0 ms)	0.11 (0.10)	0.08 (0.09)
VOT (−10 ms; +10 ms)	0.15 (0.14)	0.13 (0.10)
VOT (+0 ms; +20 ms)	0.11 (0.18)	0.10 (0.09)
VOT (+10 ms; +30 ms)[a]	0.062 (0.063)	0.05 (0.07)
VOT (+20 ms; +40 ms)[a]	0.063 (0.058)	0.07 (0.05)

[a] The variable contributes significantly to the first factor

scores for the positions 4 and 5 of letters in the partial report task; Valdois et al., 2004). The predictive model suggests to consider metaphonological and visuo-attentional variables, so that phonological, surface or mixed dyslexia can be addressed. Finally, four auditory variables appear in the predictive model. The association of auditory and phonological measures could suggest that the impairments in phonological processes may be related to the difficulties in manipulating verbal stimuli (difficulties in identifying and discriminating brief sounds of syllables). These suggestions are supported by other studies (Serniclaes et al., 2001; Serniclaes & Sprenger-Charolles, 2003; Tallal, Miller, & Fitch 1993).

It is of note that a very high performance of detection was obtained without the spelling and reading tasks. These tasks were excluded from the model development because of their potentially huge discriminative power. Moreover, spelling and reading tasks are the usual first indicators of a problem of dyslexia, but are not specific to that symptom: if all the dyslexic children have spelling and reading difficulties, children with spelling and reading difficulties are not all dyslexic. We decided to focus on the detection of the specific underlying causes of reading and spelling difficulties of the dyslexic children. The variables of the predictive model describe more precisely the most common difficulties of children with dyslexia so that the model can also help practitioners to clarify the type of dyslexia and identify the appropriate remediation to consider. It would also be interesting to define some profiles, using the variables of the prediction model, the variables of reading and spelling tasks, and some complementary variables as attention, memory and naming speed. In a limited time (40 minutes), the health professional can have the basic information in order to suggest other investigations and/or define the remediation.

Table 4 Multivariate predictive model for dyslexia: selected variables and estimated parameters from the logistic regression model

Variable	Estimate	Odds ratio (OR)	Std. ERROR	*p* value
Metaphonological				
Phoneme elision	−0.19	0.829	0.18	0.3
Spoonerism	−1.45	0.233	0.36	<0.0001
Morphology knowledge				
Morphology	0.65	1.922	0.4	0.1
Visuo-attentional				
Partial report of letters (position 4)	−0.3	0.743	0.16	0.065
Auditory				
Slope identification of speech sound[a]	0.0073	1.007	0.37	0.98
VOT (−40 ms; −20 ms)[a]	0.0640	1.066	0.39	0.87
VOT (10 ms; 30 ms)[a]	−1.1	0.332	0.45	0.01
VOT (20 ms; 40 ms)[a]	0.18	1.197	0.34	0.6
Intercept	9.6	1.5E04	3.49	0.0059

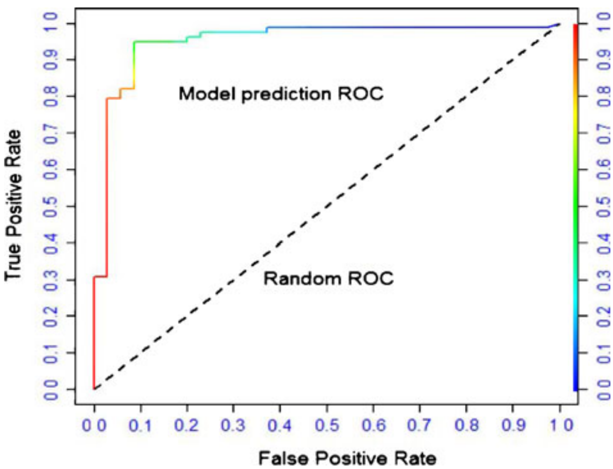
[a] Recoded variable to obtain an ordinal trend

The poor readers were excluded from our analysis, because their real status regarding dyslexia was unknown. However, we applied the predictive model to classify them afterwards. Eight of them were classified as dyslexics and the other five as normal readers. We do not know whether they are well classified and they could not be used to validate the predictive model. Some of them seem to share common characteristics with dyslexic children, whereas some others seem to be only poor readers.

To complete this research and to address some limitations of the study, we plan to validate our findings by two approaches:

– Firstly, we will check the validity of the predictive model in applying it to a larger population of children from the same education level (third and fourth grades students), using the same protocol of experiment and analysis. The objective is to test the stability of our predictive

Fig. 5 ROC curve of the logistic model for dyslexia detection



model in terms of cognitive categories and tasks that are finally selected. In this validation study, more investigation will be done to know the real status of the poor readers.

- Secondly, we will extend this study to children from three other class levels (first, second, and fifth grades). In order to adapt the screening test to the spelling and reading levels that can be expected for each class level, some of the screening tasks will have to be modified and the predictive model will be fitted accordingly.
- Finally, we will develop an integrated software to help the practitioner along the process of dyslexia diagnosis. It will include reading and spelling tasks, the predictive model and some additional tasks of the initial battery in order to specify the subtype of dyslexia. Diagnosis would progress in three steps. After completion of reading and spelling tasks, the predictive model will help the practitioner to identify the more defective cognitive functions of the child. On the basis of these findings, he/she will administer other relevant tasks included in the software. Consequently, the complete profile of the child will be established and remediation planned.

Conclusion

Different theories try to explain dyslexia origins and consequently generate multiple screening tasks and rehabilitation strategies. The first step of our research was to develop a protocol including the most representative tasks in each category of the deficits implied in developmental dyslexia. Then, we developed a method for selecting the most relevant screening tasks and identified an efficient predictive model which evaluates the percentage of similarity with dyslexia symptoms. The performance of the model was estimated on 113 children using cross-validation. It proved to be very high (more than 90% in sensitivity and specificity), independently of the reading and spelling variables, which were considered as the first but non-specific symptoms of dyslexia. The predictive model includes 8 variables from 4 cognitive categories (metaphonological, morphology knowledge, visuo-attentional and audition) and appears relevant to the clinicians involved in the study. More generally, this integrated approach should meet practitioners' expectations insofar as they need to evaluate the importance of deficits and to propose other evaluations and/or rehabilitation tasks in a quite limited time.

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