

**Dyslexia**  
**Dysorthography**  
**Dyscalculia**

**Review of the scientific data**

Synthesis and recommendations

Inserm

Collective expert review

This document presents the synthesis and recommendations of the expert group convened by Inserm in the context of the collective expert review procedure to reply to the request of the Social Regimen for Free-Lance Workers (RSI), formerly known as the Canam, relating to dyslexia, dysorthography and dyscalculia. The document is based on the scientific data available as at the second half of 2006. Over 2,000 articles constituted the document base for this expert review.

The Inserm center for collective expert reviews ensured the scientific and editorial coordination of this collective expert review. In the context of the mission of accompanying collective expert reviews set up by Christian Bréchet, Director General of Inserm, the expert review benefited from the advice of Jean-Marie Danion, Professor of Psychiatry, representing the Directorate General of Inserm, Jean-Claude Ameisen, Chairman of the Inserm Ethics Committee, and members of the Department of Scientific Information and Communication directed by Pascale Sillard and Patrick Chanson.

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## Preface

I would like to thank the social regimen for free-lance workers (RSI, formerly the Canam) for having entrusted Inserm with implementation of the collective expert review on scholastic learning disabilities such as dyslexia, whose importance in public health terms gave rise to a national action plan in 2001.

I would particularly like to congratulate the expert group for having addressed a particularly difficult subject by means of an objective analysis of the literature available in the disciplinary fields in which progress has been most marked in recent years. I hope that the scientific light shed will be of value to professionals in the educational and health sectors in order to help children overcome their handicaps.

The expert review does not claim to have addressed all the aspects of the multiple-component disorders or to have answered all the questions. Research must continue in order to fill in the numerous gaps in our knowledge. The interdisciplinary approach is a key factor for success in that research, as the experts stress.

The expert review was implemented in the context of the ongoing process with respect to Inserm collective expert reviews designed to enhance the incorporation of the various points of view and modes of thought into the debate.

The representatives of patient associations, parent associations, educational professionals, and medical and paramedical professionals (speech therapists, neuropsychologists, psychologists, etc.) were able to discuss the work implemented by the experts prior to publication of this expert review and contribute their point of view, experience and know-how. Their contributions, reported herein, contribute to the quality of the work.

I would like to warmly thank my colleagues who contributed their advice during this work in the context of their mission of accompanying collective expert reviews, a mission that I recently set up. I would also like to thank all the scientists and professionals involved in learning and learning disabilities who, through critical rereading of all or part of the document, depending on their field of interest, have enabled further improvement.

I would like to stress Inserm's interest in the debate, which must take place after the experts' substantial but necessarily limited work. I invite the reader to take note of the three critical reading memoranda entrusted to authorities selected on the basis of their different and complementary approaches.

*Professor Christian Bréchet*  
*Director General of Inserm*

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# Foreword

Since the year 2000, the national organization for free-lance workers (Canam), which became the social regimen for free-lance workers (RSI) in 2005, has requested that Inserm implement several collective expert reviews relating to child health. The reviews have addressed, among other subjects, obesity, mental disorders and specific scholastic learning disabilities identified as public health priorities at national level.

Dyslexia, which interferes with learning to read, is the best known and best studied specific disorder. Dyscalculia and dysorthography have been the subject of much less research. Difficulties at school related to those disorders, if they are not managed, may lead to marginalization or even stigmatization of the children and the cumulative failures may give rise to difficulties in social life in adulthood.

The Canam (RSI) asked Inserm to conduct a review of scientific knowledge, using the collective expert review procedure, in order to further elucidate the specific disorders related to scholastic learning and assess the scientific publications with respect to the instruments and methodology for detecting and managing those disorders.

In order to respond to that request, Inserm convened a pluri-disciplinary group of 11 experts with skills in the field of cognitive psychology, developmental psychology, child neuropsychology, pediatrics, neurosciences, psycholinguistics, linguistics and epidemiology.

The impact that progress in the scientific knowledge of the management of dyslexia and other specific learning disabilities may have is one of the essential issues for the expert review. Numerous instruments for dyslexia remediation are now available, although their efficacies have not been evaluated and their theoretical references have not been explicitly stated. The situation provided the grounds for a review of our knowledge of dyslexia, dysorthography and dyscalculia and the methods of managing those disorders.

Dysphasia (specific disorder of spoken language) and dyspraxia (coordination acquisition disorder) have not been tackled in the context of this expert review targeting specific scholastic learning disabilities. Those disorders nonetheless have an important impact on scholastic learning. The disorders are mentioned in the section on associated disorders and should constitute the subject of an in-depth review in the course of a coming Inserm collective expert review.

The group structured its thinking in order to respond to the following questions:

- What is currently known about 'normal' language acquisition and learning to read, write and calculate, which may shed light on the specific learning disabilities?
- What definitions and classifications of specific learning disabilities were used in the various studies in order to determine frequency and demographics?
- What is dyslexia? What are the main signs and symptoms? How frequent is the disorder?
- What are the other specific learning disabilities (dyscalculia, dysorthography) and what are their relationships with dyslexia? What other disorders are frequently associated?

- What are the principal theories put forward to explain dyslexia and what has been the contribution of new scientific technologies (neuroimaging, genetics, cognitive neurosciences) enabling recent progress?
- What are the methods of remediation and how do they fit in with current theories? What is the current state of scientific research with regard to the indications for and evaluation of the proposed methods?
- What instruments and strategies are available for detecting, screening for, diagnosing and managing the disorders? What are the prospects from which teachers, families and the children themselves may procure benefit?

This expert review consists of four sections. In order to understand dyslexia, dysorthography and dyscalculia it was considered indispensable first to review succinctly the acquisition of spoken language, learning to read and write, spell and count in children not presenting with any particular difficulty. So doing constitutes the first section of this review.

The second section is devoted to presenting the specific scholastic learning disabilities (definition, prevalence, etc.). The diversity of the forms of dyslexia is illustrated through a case study analysis. The study of dyslexic child populations shows a high prevalence of specific cognitive deficiency (phonological deficiency). The published data on developmental dysorthography and dyscalculia remain few in number. The frequent association between 'dys' disorders and other psychological disorders points the way to considering shared pathophysiological mechanisms.

The various theories advanced to explain dyslexia (phonological deficiency, temporal auditory processing deficiency, visual theories, cerebellar function deficiency, magnocellular theory, etc.) constitute the subject of the third section. The link with other developmental disorders contributes to giving rise to new hypotheses. It should be observed that considerable research is under way in consequence. Neurobiological, neurological imaging and molecular genetic studies, while far from complete, open interesting approaches to the potential mechanisms underlying the deficiencies.

The last section of the review addresses identification, screening, diagnosis, prevention and management. Do the findings of the review support the general principles on which the professional practices and organizations for child management are based? In line with the methodology of the Inserm collective expert review, this section does not describe all the practices in France but analyzes the various remedial methods that have been documented by publications and their scientific evaluation. Generally speaking, should not the diagnostic approach and multidisciplinary management of those disorders consider the disorders related to a handicap?

Reports prior to this expert review (report by Ringard, 2000; report by Véber and Ringard, 2001) constituted the basis for a national plan of action for children presenting with a specific language disorder (Ministry of Health, Ministry of National Education, 2001). The plan reflects the commitment to acting to improve the situation of those children and their families. In the context of that plan, epidemiological studies and research programs particularly targeting the evaluation of care practices, pedagogic strategies and didactic contents were scheduled<sup>1</sup>. Five years after implementation, the measures taken in the context of the national plan in the field of health are the subject of an evaluation being conducted by the National Federation of Health Monitoring Stations under the auspices of the Directorate General for Health. In the context of that plan, an expert commission worked on the

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<sup>1</sup> <http://www.banquoutils.education.gouv.fr/>

language disorder screening instruments. The report was made public in 2006<sup>2</sup> when this expert review was being finalized. The findings have therefore not been incorporated in the analysis.

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<sup>2</sup> [www.sante.gouv.fr](http://www.sante.gouv.fr)- Theme: language

# Synthesis

Dyslexia, dysorthography and dyscalculia are specific scholastic learning disabilities whose etiologies are recognized to be neurodevelopmental. However, the disorders do not derive from patent deficits at sensory, motor or mental level. The disorders also do not derive from injury or a pervasive developmental disorder. Even though the definition of the disorders in the classifications excludes a cultural, social, economic, pedagogic or psychological etiology, this is not to say that such factors do not play a role.

The objective of this expert review was to take stock of the scientific work conducted in recent years with a view to identifying the knowledge that could contribute an enhanced manner of envisaging the prevention, identification and management of specific scholastic learning disabilities.

The progress of knowledge in the field has been particularly marked in recent years and covers several disciplines: psychology, neurosciences, cognitive sciences, linguistics, psycholinguistics, neurobiology and neuropsychology. Some of those disciplines have enabled differentiation and specification of the disorders focusing on spoken and written language, hand-writing, writing, orthography and arithmetic. Those disciplines have also enabled formulation of functional interpretations.

In certain fields, increasing knowledge has even enabled a relationship with the cerebral organization of information processing systems to be determined. However, the specific character of the disabilities which differentiates them radically from generalized retarded learning does not mean that they are monofactorial or isolated. If children are failing at school because of unfavorable social conditions or an inadequate educational level, this does not mean that specific disabilities do not coexist in the same children or that such factors are involved in the expression of the disorder. However, the fact that children with dyslexia are found in all social classes, including the most privileged, irrespective of the teaching methods used, means that solely sociological or pedagogical explanations of that type of disorder are invalid.

Some learning difficulties may be seen in the context of a patent psychological disease or in disturbed early interactions. It should however be noted that the psychological distress observed in many children with learning difficulties is frequently a consequence of their failure at school.

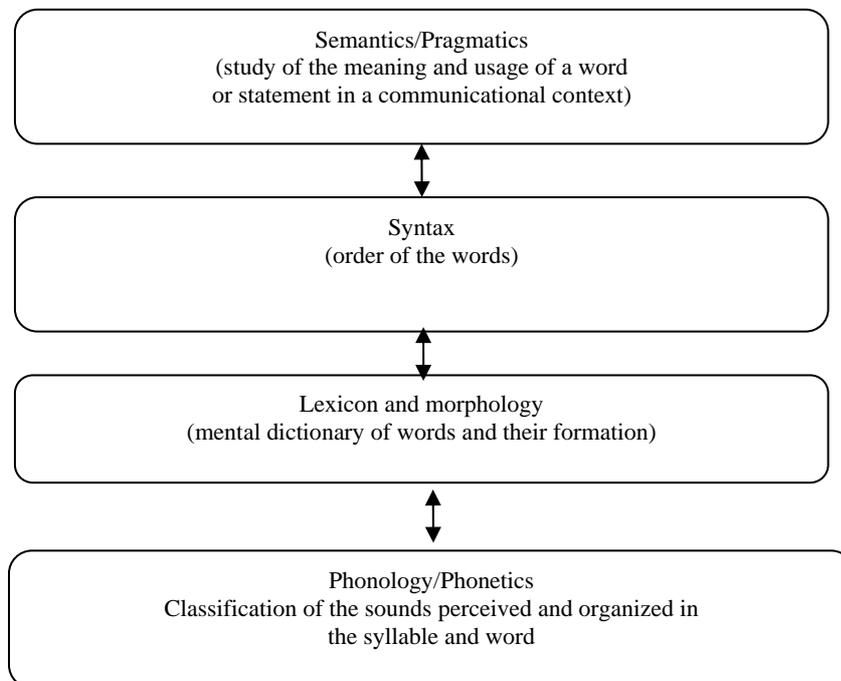
The involvement of socioeconomic, pedagogic, linguistic and psychological factors underlying the disorders does not seem in contradiction with a neurocognitive or genetic model if a model with more than one cause is accepted. In addition, the influence of the same factors on the ease with which the deficiencies are overcome may also be considered. Nonetheless, the literature on those aspects is still sparse and high-quality pluri-disciplinary studies are necessary.

Over the last 30 years, research mainly addressed the underlying cognitive mechanisms of dyslexia and resulted in various theories being advanced. The recent development of functional brain imaging and molecular genetic methods has shed complementary light on the relationships between dyslexia and its cerebral substrates. The scientific literature on those studies was reviewed in the context of the present expert review. Research has yet to address incorporation of the various theories in an overall concept explaining the manifestations of the deficiencies. Genetic factors, cognitive functions, psychological

structuring and the familial and social systems contribute together to the development of skills in the child. The complementarity of those approaches should enable overall management of the child in cognitive terms and in terms of the child's relationship with his/her environment.

## Spoken language acquisition: chronological benchmarks

The neonate's ability to learn his native language never fails to astonish. In a few years, the infant will master the complexity of the various constituents of the language. The linguistic and cognitive studies have tackled the question of speech and language acquisition by referring to a three-component system: form, content and usage. The formal aspects of the language are related to phonology (the limited set of sounds of a language which can be combined to form an infinite number of words) and the syntax (which organizes the order of the words). Content refers to the field of semantics (meaning of words and statements). Usage is a pragmatic field which studies the set of codes that regulate the speaker's communicational intentions.



### Language components

The studies of the emergence of acquisitions now enable a fairly clear concept of the very early specialization of perceived speech processing and the fast development trajectories with respect to lexicon and morphosyntax processing in the child. As of the first few months of life, the ability to perceive the sounds of speech enables the infant to discriminate, categorize the elemental sounds and recognize certain words of the language by prosody (the 'musical' envelope of speech with its aspect of rhythm, tempo, melody, accent and intonation). Towards the age of 7-8 months, the infant is able to recognize and memorize syllable forms of the 'word' type with clearly defined consonant-vowel sequences that are particular to the language. Toward age 9-10 months, the first words are acquired before the lexical explosion at about age 18 months. Assemblies of words occur at about age 24 months and grammatical expansion from about 30 months. While the between-individual variability

is very great, the period from 0 to 3 years is decisive in the rapid implementation of the process of speech and language acquisition in the child.

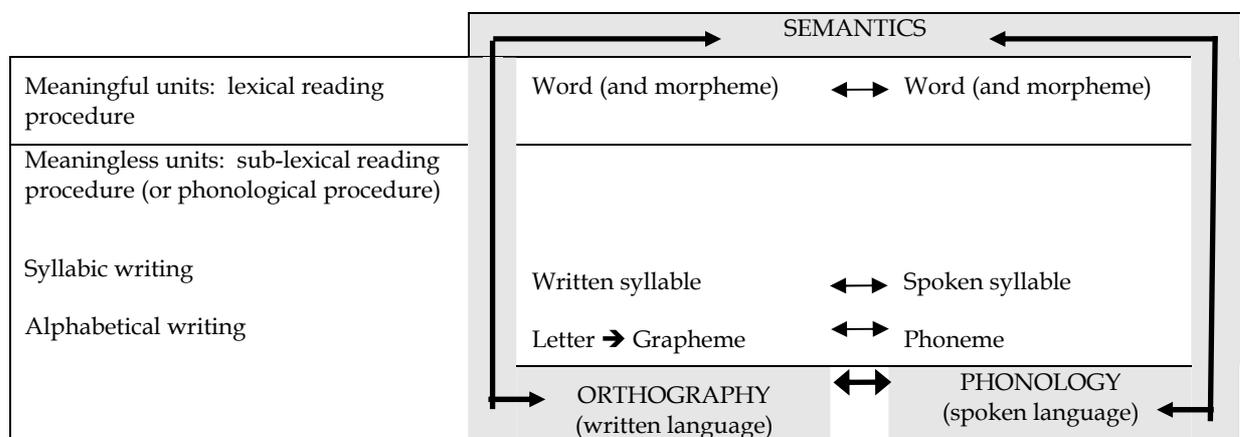
From about age 3 years, the child learns to construct a story. This consists first in a list of various states which includes adverbs such as 'here', 'there' and 'now'. At age 5 years, the child begins to make clearer and more varied links between the events taking place in a story. The child uses 'and then', 'when', 'after'. The accentuation of particular events only emerges gradually. At age 10-11 years, the child still does not have narrative skills similar to those of the adult.

The set of data on the chronology of speech and language acquisition and story construction should enable professionals (teachers and clinicians) to detect asynchronous development. Early evaluation of the ability to segment, the emergence of prattling, the emergence of the first words and the text based on the stories in their conceptual and linguistic dimensions is justified on the basis of the predictive value that those abilities have with respect to learning to read and write.

## Learning to read

The objective of reading is comprehension. To achieve that objective, the child must acquire a high degree of automatism in the identification of written words. By developing that skill, the child will achieve a level of reading comprehension equivalent to that of his/her spoken language comprehension.

In alphabetical writing, as in syllabic writing, two procedures enable identification of the written words: the sub-lexical procedure (phonological procedure or decoding procedure) and the lexical procedure (or orthographic procedure). The sub-lexical procedure is based on meaningless units, written syllables which, in syllabic writing, encode the spoken syllables or graphemes (consisting of one or several letters) and which, in alphabetical writing, encode the phonemes. The lexical procedure is based on units which have meaning: words. Decoding rapidly gradually becomes automatic. The procedure thus does not only refer to the slow and laborious reading of the beginner: the expert reader can very rapidly identify words that he/she does not know. Moreover, the lexical procedure is not based on a sight word strategy: the expert reader accesses the orthographic, phonological and semantic codes of written words in a few hundreds of milliseconds.



### Operations involved in written word identification in syllabic writing and alphabetical writing

The reader who learns to read alphabetical writing is faced with three types of problem. The first problem is that, before learning to read, the subject has no orthographic representations except for a few words learned by heart. Similarly, before learning to read, children reader does not necessarily have explicit access to certain phonological units, in particular the phoneme, which is the minimal distinctive feature enabling differentiation - in a given language - of two words, for instance 'cat' and 'mat' in English. Taking into account the phoneme, which is indispensable in order to understand the principle of alphabetical writing and hence read that type of writing, necessitates a focus on abstract spoken language components, phonemes. In additions, those components that are difficult to access for reasons of co-articulation (the word 'car' is pronounced /kar/ and not /k+a+r/, this renders identification of the three phonemes composing the word difficult). The second problem with which the beginning reader is faced is related to the degree of transparency of the relationships between the basic units of writing (graphemes) and spoken language (phonemes) which vary depending on the language in question. Thus, in Spanish, unlike English, the relationships are very regular. French is closer to Spanish than to English for the regularity between the graphemes and phonemes (used to read) while French is closer to English with regard to the relationships between phonemes and graphemes (used to write). The problems of transparency with regard to the relationships between the basic units of writing and speaking are explained by the history of languages (while the spoken language evolves, the written language is more conservative) but also by the fact that the letters of the alphabet do not enable transcription of the phonemes of the various languages. For example, there are only 5 or 6 letters for vowels. This does not enable transcription of the 16 vowels in French. A combination of letters therefore has to be used or a letter to which a specific mark has been added is used to transcribe certain vowels (for example, the 'é' in contrast 'è' and 'e' or 'ou' compared 'u' and the letters 'a, o, u, i' followed by 'n' which indicates a nasal vowel: 'an', 'on', 'un', 'in'). The basic unit of alphabetical writing is therefore not the letter but the grapheme which refers to the phoneme, the basic unit of the phonological system. The last problem is related to the size of the units. There are many more orthographic units to be learned in writing which uses large-scale units (e.g. the word) than in those which use small-scale units (e.g. the grapheme reflecting the phoneme). However, the large-scale units have a meaning and the small-scale units do not. The result is that small-scale units are less readily accessible than large-scale units<sup>3</sup>.

The research on learning to read has shown that the ease of learning depends on the degree of transparency of the grapheme-phoneme correspondences, which is very high in Spanish and very low in other languages such as English. French occupies an intermediate position. Accordingly, the worst reading scores are observed among English-speakers and the best among Spanish-speakers. For example, in France, in primary school (grade: CP, 6-7 years), the scores for reading regular words (e.g. 'table') and pseudo-words (regular words which do not exist, e.g. 'tople') do not differ and are higher than the scores observed for irregular words (e.g. 'sept'), which are very low. In a few months (at the end of CP), the picture changes markedly. The child improves for all items but the improvement is more marked for regular words, which are better read than pseudo-words, which are in turn better read than irregular words. The foregoing is probably due to the fact that regular words benefit from both their regularity and the frequency of exposure. Neither frequency of exposure alone nor regularity alone is sufficient as shown by the weaker progression for irregular words on the one hand and pseudo-words on the other. These data enable it to be understood why

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<sup>3</sup> In logographic writing, such as Chinese, the basic unit of writing is the word. It is noteworthy that in continental China children begin to read using an alphabetic system. This reflects the difficulties encountered by children who have to learn to read in a logographic system. Several thousands of different orthographic forms have to be memorized in order to read a text. This may take several years.

Spanish children who practically only encounter regular words make rapid progress while learning to read is slow and laborious for English children.

Linguistic factors induce not only quantitative but also qualitative differences in the procedures for written word identification. For instance, the lexical procedure is more used when the orthography is opaque. This is not the case when the orthography is more transparent.

Linguistic factors induce differences in the reading units used. For example, English promotes greater use of units such as word rhymes because, in that language, the pronunciation of vowels largely depends on the consonants that follow. This is not the case in languages which have clearly pronounced vowels such as Spanish, German and French.

In contrast, the (rare) studies in the field of morphology<sup>4</sup> show that, irrespective of the opaqueness of the orthography, children use morphological units when they read. However, that ability, which only seems to be acquired progressively, is dependent on phonological factors until a late period.

Lastly, irrespective of the deepness of the orthography, the grapheme-phoneme decoding ability determines the degree of success in learning to read. In fact, readers who are, from the outset, the best decoders are also those who progress the fastest, including with regard to reading irregular words and understanding text.

These findings enable other research results to be understood, particularly:

- why systematic and early teaching (as of the start of primary school, grade CP, 6-7 years) of grapheme-phoneme correspondences is the approach that most effectively helps pupils. When the method is introduced later, its impact is weaker. Moreover, that type of teaching is particularly beneficial for children who may experience difficulties in learning to read (future below average readers and dyslexics), irrespective of whether the risk factor is related to socioeconomic conditions, educational or linguistic level;
- why it is essentially training in phoneme analysis that has an effect on learning to read. The effect is, however, more marked in children at risk for reading acquisition (socioeconomic or linguistic factors) than in readers in difficulty, reflecting the fact that prevention is easier than remediation. The effect of that type of training is nonetheless more marked if, in addition, the children can handle the letters matching the phonemes;
- why the most reliable predictors of learning to read are the phoneme analysis abilities and the level of letter recognition, together with short-term phonological memory and rapid naming. Those predictors enable rapid detection (as of the end of the last section of nursery school, 5-6 years) of future below average readers and dyslexics, with a high degree of reliability. It may be considered that phoneme analysis abilities are of crucial importance at the start of learning to read in an alphabetical writing system because they enable the child to access the principle of that type of writing. The degree of intervention of the skills involved in completing tests which evaluate both the speed and accuracy of spoken lexicon access (rapid naming of images of objects, colors, etc.) is less clear. It may, however, be supposed that, initially, when the child is mainly using decoding, the ability enables rapid and precise access to the spoken word matching the series of letters decoded. This would facilitate the making of links between the orthographic code and phonological code for written words.

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<sup>4</sup>For example, the word 'orthodontist' is composed of 3 morphemes: 'ortho', 'donto' and 'list'

## Learning to write and orthography

Learning to write is a relatively long process and causes problems for all children. The latter are usually overcome towards the third year of primary school in the majority of children. During that learning setup phase for all children and beyond it for those who have difficulty making writing automatic, the difficulties experienced in writing impact on the quantity and, in a less sure manner, on the quality of written language production and on orthographic performance.

In all alphabetical writing systems, including French, the crucial stage in learning consists in understanding the fact that the sequences of letters have regular matches - sometimes complex - with sound sequences, in other words the alphabetical principle. In production, this means that the child must have achieved a certain degree of mastery of the native language so that the child can use it as an object to be observed in order to, for example:

- segment statements;
- retain the segmentation in memory while transcribing, even in a non-conventional manner;
- check afterwards, by reading, the (relative) accuracy of what the child has written.

In transparent orthographic systems, access to the alphabetical principle is practically enough to ensure transcription of all new words, whatever they may be. This does not apply in French since the relationships between phonemes and graphemes are irregular.

Since French cannot be transcribed by only referring to the associations between phonemes and graphemes, lexical information specific to the words (e.g. the spelling of the word '*thym*'), general orthographic information (e.g. no double consonants at the start or end of words) and information relating to morphology (e.g. the plural of nouns is made by adding an 's') need to be acquired and recalled, depending on the circumstances. This raises the question of acquisition.

From the very start of learning to read, children are able to memorize orthographic forms and at least some of their specificities without awaiting mastery of all the phoneme-grapheme matches. Little is known about between-individual differences, the number of exposures necessary in order to learn written words or the manner in which orthography becomes more conventional.

Also very early, as of the first year of primary school, children exposed to writing extract orthographic regularities which are not simply those reflecting phonological regularities or simple memorization of instances. Children are able to draw on those regularities in writing tasks, thus exhibiting very early acquisition of certain orthographic conventions which do not go so far as extracting rules, even when that would be possible. Too few of those regularities have been studied to enable determination of the conditions under which they are taken into account and the potential between-individual differences in learning them.

Traces of the use of morphology-related markers are also observed very early. However, studies in which the effects of graphotactic constraints were differentiated from those associated with morphology show that the use of morphology begins relatively late, at least if we consider that use corresponds to rules whose application does not depend on frequency. However, numerous examples show that the use of morphological markers is not initially independent of the frequency of item exposure. In addition, since morphology is correlated with other types of phonological, orthographic and semantic information, it may

be that the effects attributed to morphology are in fact due to other factors. A degree of caution is therefore required in the interpretation of the data generated by those studies alone.

Rare studies on the production of derivational morphology show the persistence of the effects of graphotactic constraints despite the possibility of using an orthographic rule. Thus, even after several years of writing, French pupils do not use a rule specifying how to write, for example /o/ when /o/ is a diminutive suffix (e.g. 'éléphanteau', 'lionceau'). This leads to questions on what happens when the rules are explicitly taught and on the possibility of doing so systematically, which is the case with flexional morphology.

Flexional morphology addresses a restricted number of fields: the gender and number agreement of nouns, adjectives and verbs ('accords'), the verbal system (conjugation). It is very productive: for example, nominal agreements ending in 's' apply to most nouns and adjectives. A limited number of markers for number and gender are used: 's', '-e', '-nt'. Then there are the verbal flexions which are more numerous and many of which do not have a phonological match ('-s', '-nt') or are associated with a given phonological form (e.g. /e/ associated with 'é', '-er', '-ait', etc.). In principle, the foregoing is systematically taught. Use seems to emerge late and very dependent on the teaching. It transits through stages that may erroneously suggest that the morphological rules have been mastered early.

Teaching consists in stating rules followed by exercises to apply the rules. The pupils learn to implement a procedure which, at the beginning, calls for time and attention. Overgeneralization errors sometimes occur: for instance, adding an 's' to plural verbs. Under the effects of frequent practice and teacher feed-back, the errors are eradicated and the procedure is applied more rapidly, decreasing the amount of attention required. When certain forms are frequent, they are memorized as such. The pupils thus retrieve them directly, leading, under certain conditions, to new types of error that may also occur in adults and consist, for instance, in writing 'il les timbres'. Awareness of the agreement rules thus occurs during checking production much more than during application of the procedure.

The data on learning to write show that several processes are involved. The first process relates to learning the associations between phonemes and graphemes: in consequence, it is highly linked to learning to read even though the relationships between writing and reading require further study. The second process also involves reading and, doubtless, writing practice. The frequent exposure to written words results in two acquisitions: first, certain words are memorized and thus directly retrievable; secondly, 'graphotactic' regularities, i.e. associations between letters or graphemes are extracted together with the contexts in which they occur by implicit learning (without being aware of having that knowledge). Thirdly, some of the regularities may be formalized and result in statement and application of rules associated, to a variable degree, with exceptions (e.g. all the diminutives ending in /o/ are written 'eau', except 'chiot'). However, such rules do not seem to be spontaneously accessible to pupils: awareness of them and generalization seems to require teaching. Fourthly, teaching the rules of flexional morphology leads to formation of procedures whose implementation requires marked attention rendering their application fragile, at least initially. Only regular and prolonged practice decreases the attention input. The practice, in reading as in writing, results in the most flexional forms being remembered and thus directly retrieved. Sometimes, this results in emergence of new errors. The availability of formal agreement rules enables production to be checked and errors to be corrected.

While the principal processes of orthographic learning have been identified, their interactions, particularly during schooling and as a function of what is or is not taught, have yet to be investigated.

## Learning arithmetic

The issues relating to acquisition of and then learning elementary arithmetic are situated at three levels. Firstly, infants, like animals, are able to mobilize two different systems to process quantities and the transformations affecting them (addition, subtraction). One system is precise but only applied to small discrete sets (1, 2 and 3); the other, which can be extended to very large quantities, operates on continuous dimensions or treats as such sets of discrete elements in order to formulate an approximate evaluation in which the errors increase with the size of the quantities estimated (Weber's law). The question of the specifically numerical character of the corresponding processes remains open as does the question of the characteristics of the representations on which the processes are implemented. The capabilities are only a point of departure but may constitute the basis of number semantics. The more complex mathematical knowledge that man has developed over his history go much beyond the foregoing and involve symbolic number systems.

Secondly, from the age of 12-18 months, infants appropriate the system specific to their culture and enabling precise determination of the number of things. The acquisition has two dimensions: first, that of the verbal naming of quantities specific to the infant's culture (when the culture has one); secondly, the practices of counting which may be based on universal principles. Acquisition is slow and difficult for reasons that have been partially elucidated.

The verbal systems are conventional systems based on two major principles:

- the lexicon which gives a cardinal number a single and unique name (five sixteen);
- combinatorial rules enabling elaboration of an infinity of complex formulations enabling naming of any cardinal number (six hundred seventy five million three hundred ten thousand two).

The rules enable combinations of the additive (one hundred three) or multiplicative (three hundred) types. The French spoken numerical system lexicalizes cardinal numbers up to sixteen, the tens from twenty to sixty, hundred, thousand, million and billion. Combination codes exclusively additive relations up to 79 (twenty five = twenty + five), then additive and multiplicative relationships (four hundred six = four x one hundred + six).

Quantities may be evaluated overall or precisely. In the first case, as previously indicated, the errors increase with the size of the collection to be evaluated: the errors are more rare for small collections (1, 2 or 3, or even 4) for which a specific processing mechanism may exist (subitizing). Subsequently, the errors increase. In the second case, counting is necessary, i.e. identifying each of the entities in the collection and processing it once and only once and assigning to it a verbal label and only one verbal label such that the last item's label is the cardinal number of the collection. Counting correctly requires attention, knowledge of the numerical lexicon and the ability to implement motor skills and coordinate them with memory retrieval of the verbal forms. Using counting, children manage to assign a single and precise cardinal number to each collection, irrespective of the perceptual form of the collection.

Thirdly, infants identify the changes in quantity associated with addition, subtraction or even fractionation very early. Mastery of counting enables children to precisely quantify those changes. They can therefore implement an action which is equivalent to an arithmetic operation although it has yet to become one. They do so by grouping or separating sets and by counting both the initial sets and the results of the changes. Only very gradually will they move from resolution by action to processing addressing the symbols only. It is possible that use of the fingers during that phase plays an important role insofar as the sets of fingers are

both analog and abstract (they are a substitute for all objects). Progress is reflected by the switch from external actions to interior actions, then processing conducted on verbal forms ( $4+3 \rightarrow 4, 5, 6, 7$ ). It is not easy to determine how infants realize that using the symbols alone yields results that are as reliable as those obtained by manipulation.

Fourthly, when the child goes to school, he/she is systematically taught a new code - the Indo-Arabic code - and the algorithms associated with it. This gives the resolution of the operations a power that the verbal code alone cannot ensure. Learning is both easy (the Indo-Arabic code only has ten items: 0,...9) and difficult, in particular due to the positional notation (the value of a digit changes with its position). Trans-coding, switching from the spoken to the Indo-Arabic code or the opposite, is initially based on verbal knowledge. This explains why, for instance in French, the transcription of quantities such as seventy five (*soixante-quinze*) gives rise to errors such as 6015. The algorithms concern the resolution of complex additions, subtractions and multiplications and, to a lesser degree in elementary school, divisions. The children must memorize certain associations between operands and results ( $3 + 2 = 5$ ) so that they no longer need to calculate them and can devote their attention to managing the algorithm. The algorithms also require a minimum of attention to and mastery of space. The algorithms need systematic and vigilant teaching in order to prevent the installation of procedural errors that are difficult to eradicate. The introduction of fractions and decimals raises new problems since, first, the representation of quantities changes (although fractionation of quantities occurs early) and, secondly, the operation processing algorithms differ from those applied to natural whole numbers (addition of fractions, addition of decimals, etc.).

Fifthly, the resolution of arithmetic problems remains a major problem as is shown by national and international evaluations. One of the difficulties is associated with reading comprehension more than the arithmetic processes themselves, at least at elementary school. Another difficulty is linked to the 'arithmetization' of situations. The children have to elaborate from a statement a representation of the situation described, then transition from that representation to an arithmetic formulation. Depending on the formulation, the familiarity with the field, the number of units involved and the frequency of exposure to the situations, the elaboration of that representation and its 'arithmetization' may be very difficult and lead to an erroneous interpretation and hence incorrect resolution. The essential question is thus that of finding situations enabling pupils to frequently encounter a variety of problem situations so that they familiarize themselves with the diversity of statement presentations, the diversity of the situations described and, lastly, the diversity of the modes of resolution.

In short, doubtless from birth, children have the quantitative processing capacities that are biologically determined. Subsequently, cultural practices are added. The latter exploit and amplify the initial capacities. School systems install new learning systems: knowledge (decimals), activities (problem solving), codes (Indo-Arabic code, operators) and processing procedures (problem solving algorithms). Each of the dimensions is associated with particular problems, some of which remain imperfectly understood.

## **Definitions and classifications of specific learning disabilities**

The disorders addressed herein are those related to learning in school: disorders of reading, writing and arithmetic. The disorders may emerge in a context of overall retardation or more specific retardation in the event of deficiencies restricted to certain cognitive processes. The following will mainly address the definitions of and criteria for 'Specific scholastic learning disabilities' present outside of overall developmental retardation.

The disorders are defined in the classifications as a set of learning difficulties that cannot be attributed to intellectual retardation, a sensory handicap or unfavorable environmental conditions. The difficulties are thus unexpected given the other aspects of development. The difficulties emerge very early in life, interfere with integration in school and social integration and frequently persist until adulthood.

The main criteria for specific learning disabilities (in reading, writing or arithmetic) which constitute the bases for the definitions proposed in the first years of life are as follows:

- the criterion of a 'discrepancy' between the difficulties experienced in tests related to the disorder in question and the good performance in other cognitive tests (frequently the IQ<sup>5</sup>);
- exclusion criterion: the primary cause of the disorder is not to be overall retardation, sensory handicap, educational deficiency (inappropriate teaching, inadequate socioeconomic level), a linguistic difficulty or mental disorder that has been clearly demonstrated;
- the disorder is due to factors intrinsic to the child (this point is directly derived from preceding two points and stresses the neurobiological origin of the disorders).

The criteria are used in the International classification of diseases, ICD-10<sup>6</sup> and in the fourth edition of the Diagnostic and Statistical Manual of mental disorders (DSM-IV)<sup>7</sup>. Both classifications are very widely used at international level.

#### **ICD-10 common diagnostic criteria for specific developmental disorders of scholastic skills**

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- A score in individually administered tests that is at least two standard errors of prediction below the level expected on the basis of the child's chronological age and IQ
  - The disturbance significantly interferes with academic achievement or activities of daily living
  - Not directly due to a sensory deficit
  - School experience is within the average expectable range
  - IQ greater than or equal to 70
- 

In line with a recent trend, the label 'specific learning disabilities' should not be allocated simply on the results of the various tests but should be restricted to children whose disorders are recalcitrant to appropriate treatment (pedagogic and/or speech therapy management). Moreover, progress in knowledge in recent years has led to the inclusion in the definitions of the neurobiological or even hereditary etiology of specific learning disabilities and their cognitive mechanisms even though those cognitive processes remain the subject of discussions and investigation. It should also be noted in the context of the research work that the criteria adopted may vary as a function of the number of subjects observed. Studies on large populations use, for feasibility reasons, criteria that are less rigorous and numerous than those used in studies on small groups of children.

Reading disorder or dyslexia is by far the most widely explored specific developmental disorder of scholastic skills. Generally speaking, it presents as a difficulty in learning to read despite appropriate teaching, adequate intelligence and a good socioeconomic environment.

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<sup>5</sup> Intelligence quotient (IQ) measured between 6 and 15 years using WISC-IV (D. Wechsler, Wechsler's intelligence scale for children and adolescents, 4th edition. ECPA, 2005)

<sup>6</sup> WHO. International classification of diseases. Chapter V (F): Mental and behavioral disorders. Diagnostic criteria. Masson, 1994

<sup>7</sup> AMERICAN PSYCHIATRIC ASSOCIATION. Learning disorders. DSM-IV-TR. Diagnostic and Statistical Manual of mental disorders. Masson, 2004

## ICD-10 diagnostic criteria for specific reading disorder

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Either 1 or 2:

1. A score in a standardized reading accuracy and/or comprehension test that is at least two standard errors of prediction below the level expected on the basis of the child's chronological age and general intelligence; with both reading skills and IQ assessed on an individually administered test standardized for the child's culture and educational system
  2. A history of serious reading difficulties or test scores that met criterion 1 at an earlier age plus a score on a spelling test that is at least two standard errors of prediction below the level expected on the basis of the child's chronological age and IQ
- 

In the event of concomitant disease, the diagnosis of specific reading disorder predominates over the other specific disorders (arithmetic and writing disorders) in ICD-10 while DSM-IV enables several diagnoses to be formulated.

Recent definitions of dyslexia address disturbed cognitive mechanisms (often a deficit in the phonological component of language) and the etiology of the disorders (neurobiological etiology).

The specific disorder of arithmetical skills or mathematics disorder or dyscalculia usually refers to a disorder of mathematical skills in children of normal intelligence. Despite indubitable progress in studies in recent years, the disturbances of cognitive mechanisms underlying dyscalculia continue to be studied even though numerous subtype classifications of dyscalculia have been proposed. The etiology is already at the core of the initial definitions of dyscalculia proposed, as are the genetic or congenital etiologies of the disorders.

## IDF-10 diagnostic criteria for specific disorder of arithmetical skills

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- A score on a standardized arithmetic test that is at least two standard errors of prediction below the level expected on the basis of the child's chronological age and general intelligence
  - Scores on reading accuracy and comprehension and on spelling that are within the normal range ( $\pm$  two standard deviations from the mean)
  - No history of significant reading or spelling difficulties
- 

In the definition of dyscalculia, as for the other specific learning disabilities, inappropriate teaching constitutes an exclusion criterion.

Since writing is a multidimensional process, several components may be responsible for the writing disorder: motor-related writing difficulty; written task completion difficulty; inadequate spelling; problems of writing composition such as planning, choice of words, sentence construction and text organization. The term dysgraphia, which rather suggests a handwriting disorder equally affecting the shaping of geometrical shapes, has sometimes been used to represent a general disorder in writing. However, in the literature, reference is more frequently made to dysorthography.

Dysorthography is a specific disorder of spelling which accompanies dyslexia; the cognitive dysfunction underlying the two disorders is probably common to both. In dysorthography, the spelling of words is highly deficient, a direct consequence of the phonological disorder in dyslexic children.

There is thus no single definition of writing disorder and this is clearly illustrated by the international classifications: the ICD-10 definition is that of dysorthography while the DSM-IV definition rather covers a more general disorder in written expression.

### **ICD-10 diagnostic criteria for specific spelling disorder**

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- A score on a standardized spelling test is at least two standard errors of prediction below the level expected on the basis of the child's chronological age and general intelligence
  - Scores on reading accuracy and comprehension and on arithmetic that are within the normal range ( $\pm$  two standard deviations from the mean)
  - No history of significant reading difficulties
- 

While insufficient and open to criticism, the international definitions and classifications (ICD-10 and DSM-IV) have the advantage of enabling comparison of studies conducted internationally and use of a common language. However, in the field, the international classifications are frequently ignored by both clinicians and researchers, who use their own classifications.

Thus, the choice of the cutoff defining the disorder (from 1 to 2 standard deviations below the mean) depends on the prevalence of the disorder. If 1 standard deviation is selected, in a normal distribution there will be 16% of subjects below the cutoff. If 2 standard deviations are selected (as proposed by the ICD-10 international classification), there will no longer be more than 2.5%. It may therefore be considered that the disorder is an entity that is in part arbitrary since dependent on the cutoff selected a priori. Nonetheless, in clinical practice, follow-up of each child and the use of numerous tests enable a real diagnosis to be formulated.

The questions raised by the concept of 'discrepancy' and the fairly frequent coexistence of several learning disabilities have prompted certain authors to question the concept of specificity. The DSM-IV classification, issued later than ICD-10, does not always use the term 'specific' without, however, changing the definitions of the disorders. It is considered important to compile a classification of learning disabilities based on common criteria shared by researchers, clinicians and educational professionals.

### **Prevalence data on specific scholastic learning disabilities**

Operational diagnosis of 'a case' requires several investigations and numerous tests repeated over several months or even several years. That approach cannot be used in the context of epidemiological studies of the overall population. Moreover, in epidemiological studies, the criteria may vary between authors, as may the cutoffs selected. It is therefore not surprising that the published prevalence data vary widely. Nonetheless, on the basis of the most methodologically rigorous international studies, mainly from English-speaking countries, it may be considered that dyslexia affects between 3 and 5% of children aged about 10 years.

Studies using the same methodology to compare populations with different languages have enabled evidencing characteristics specific to each language. Languages have different writing systems and, at present, variations in the form of dyslexia and its prevalence are considered dependent on factors such as transparency (regularity) or opacity with respect to the spelling (orthography) of each language. Thus, a study using three different definitions of dyslexia showed that the prevalence varied, as expected, as a function of the definition and strictness of the discrepancy criteria but also as a function of the language. Depending on the definition, the prevalence ranged from 3.6 to 8.5% in Italy and from 4.5 to 12% in the United States. The higher prevalences in the United States, compared to Italy, are related to the orthography: Italian spelling is transparent, unlike English spelling.

In France, in the absence of epidemiological data, the data available and complying with the criteria for characterization of dyslexia suggest that the proportion of children presenting with patent dyslexia is not different from that reported in the large-scale studies of English-speakers.

Although not all reading disorders, even when they are severe, may be considered dyslexia, the data generated by two recent epidemiological studies conducted in adults in France yielded results compatible with the estimates above: one study was conducted on young military recruits and the other on 10,000 adults aged 18 to 65 years (Insee). The studies found that 7% of adults aged 18 to 29 years experienced severe or marked difficulties in reading.

Questions may be asked with regard to the potential interactions between the school environment or educational level and the biological factors of dyslexia, and with respect to their effects on the manifestations of specific learning disabilities. By way of an example, studies conducted on the English populations of the Isle of Wight and London reflect geographic and social differences. Using the same definition of dyslexia, the prevalence was 3.6% on the Isle of Wight and 9.3% in London, where the subjects were generally of lower socioeconomic status.

The prevalence of dyscalculia is frequently considered to be equivalent to that of dyslexia. However, the most rigorous studies suggest that dyscalculia, as an isolated disorder, is rarer than dyslexia. To the authors' knowledge, no epidemiological study of dyscalculia has been conducted in France. There are few reasons, however, for thinking that the prevalence of dyscalculia differs markedly from that in other countries; the language seems to have little impact on the disorder as the convergence of the results generated in different countries shows.

Gender is one of the variation factors frequently suggested to be associated with specific learning disabilities. In general, the male/female sex ratio is estimated to be 2 for dyslexia while it is probable that dyscalculia affects girls as frequently as boys. It has also been observed that more boys than girls consult specialists for reading or writing disorders. However, the reality of a gender effect on specific scholastic learning disabilities remains a subject of debate and the reasons for any difference between boys and girls have yet to be investigated.

Longitudinal studies show that dyslexia persists with age and thus consists in a non-transient developmental deficiency even though long-term follow-up of dyslexic children raises the question of classification stability over time. Since the classifications are based on a cutoff, small changes in scores over a continuum may be sufficient for certain children to move from one side of the cutoff to the other.

Longitudinal studies also show dyscalculia to be a persistent disorder. However, the isolated forms of dyscalculia (not accompanied by dyslexia) appear to be less persistent and are considered by some authorities to be more related to developmental retardation than to a real individual difference.

In numerous studies, specific learning disability is considered an entity of genetic and cognitive etiology. It is regrettable that there are only a few studies on the interactions between genetic, cognitive and environmental factors and on the influence of the methods used to teach reading.

## **Dyslexia: studies of groups and multiple cases**

In order to understand a text, the child must acquire a high degree of automatism in the identification of written words. Development of that skill enables the child to achieve a reading comprehension level equivalent to that of his/her spoken language comprehension by freeing the child from slow and laborious decoding or recourse to risky contextual anticipation. One manifestation of dyslexia is in fact the inability to develop that type of skill.

The review of the data on dyslexia took into account studies conducted in various languages (including English, French and German). The studies were conducted either on undifferentiated groups of dyslexic children or on groups of dyslexic children with different profiles.

The results of the studies on undifferentiated groups of dyslexic children showed, first of all, that the phonological deficiencies were encountered in almost all the studies and in almost all the subjects both in reading and outside of reading.

For instance, the reading performance of dyslexic children is particularly deficient when the children cannot exploit their lexical knowledge, i.e., when they have to read rare words or non-existent words (pseudo-words). The deficiency in the sub-lexical (phonological) reading procedure has been detected in a convergent manner in almost all the studies, including comparisons with younger children with the same reading skill level. In addition, the deficiency in the sub-lexical reading procedure is more severe when the children are confronted with opaque writing (e.g. English compared to French). Lastly, when the orthography is transparent, the deficiency in the sub-lexical reading procedure is mainly observed in processing speed, and not in accuracy score.

In addition to the sub-lexical (phonological) reading procedure deficiency, deficiencies in the lexical reading procedure are also observed in children with dyslexia. This can be understood in the light of studies that have shown that the setup of the lexical procedure depends on the efficiency of the sub-lexical reading procedure. In consequence, children most frequently suffer from a double reading deficiency; the deficit of the sub-lexical reading procedure is, however, more severe than the deficit of the lexical reading procedure. The former is in fact the only deficiency observed compared to younger normal readers with the same reading level.

Dyslexic children also have particularly deficient skills with respect to tasks that involve phonological processing outside of reading: phoneme analysis; phonological short-term memory and naming tests which enable evaluation of the precision and rapidity of lexical accessing. As was the case with reading deficiencies, the deficiencies in those three fields have been reported in most of the studies and for most dyslexic children, including in the studies vs. younger children with the same reading skill level. Lastly, the skills in those three fields are the most reliable predictors of the future reading level of children. In comparison, the weight of non-verbal skills and sociocultural factors is less. The deficiencies observed in those three fields may interfere with the setup of the reading phonological procedure. In order to use that procedure, the written code must first be translated to the spoken code, which, in an alphabetical writing system necessitates associating graphemes with the corresponding phonemes. The units resulting from that decoding operation must then be assembled in order to access the words stored in the spoken language lexicon. The first operation requires phoneme analysis skills while the second involves the short-term phonological memory and the third precise and rapid access to the spoken language lexicon.

Other studies, also conducted on undifferentiated groups of dyslexic children, addressed the deficiencies in visual sequential processing. The results published to date are not conclusive for three main reasons. First, that type of deficiency is more marked for pseudo-words than

for words or for unpronounceable sequences of letters. In addition, the deficiencies in visual sequential processing may be the consequence of dyslexic reading difficulties. Indeed, in almost all the studies that have drawn attention to that type of deficiency (unlike those having pointed to phonological deficiencies), the dyslexic children were only compared with average readers of the same chronological age (not with average readers of the same reading level). Lastly, in most of the studies, the children's visual skills were evaluated by taking into account processing speed and/or in terms of tasks involving time constraints (very brief exposure to the stimuli). In contrast, only accuracy scores were examined for the assessment of phonological skills, and not processing speed. The results may therefore be explained by differences in measures. One of the rare studies in which visual sequential processing deficiencies and phonological processing deficiencies were intensively evaluated using a comparable methodology showed that dyslexic children who suffer selectively from visual deficiencies of that type are few in number.

A number of recent studies have evaluated a new hypothesis according to which the typical slowness problems observed in non-English-speaking dyslexic children are explained by their difficulty in memorizing the visual form of words, while the response precision deficiency observed in English-speaking children is related to a phonological deficiency. It is, however, difficult to imagine that the dyslexic phenotype may be fundamentally different depending on the transparency of the writing system and the measurement used. Moreover, the results of those studies, based on analysis of eye movements, do not enable validation of the new hypothesis given that the dyslexic children were compared to average readers of the same chronological age: the specificity of their eye movements may therefore be a consequence of their reading difficulties. Lastly, some of the results of those studies are compatible with the phonological hypothesis: for instance, the negative impact of the opacity of the orthographic system on the duration of ocular fixation; the fact that the most marked differences between the dyslexic and average reader children were observed with respect to pseudo-words; and the presence, in certain of those studies, of phonological deficiencies in future dyslexics, compared to future normal readers, before learning to read.

In addition to the studies of undifferentiated groups of dyslexic children, other studies have examined subjects considered to present differentiated dyslexic profiles such as phonological dyslexia (characterized by a selective deficiency in the phonological reading procedure, among other characteristics), surface dyslexia (characterized by a selective deficiency in the lexical reading procedure, among other deficiencies) and mixed profiles with a double deficiency. The published studies show that practically all dyslexic subjects have (or have had) a phonological deficiency, including the subjects presenting a surface profile. That is evidenced by longitudinal studies in particular. Moreover, in the comparison with younger average readers with the same reading level, only the proportion of phonological dyslexic children remained high, suggesting that most surface dyslexics simply suffer from a developmental lag, since they behave like younger normal readers. On the other hand, the phonological dyslexic children have, in the majority of cases, an atypical developmental trajectory.

The compensatory strategies developed by dyslexics may, however, mask certain realities. In particular, the expert reader has been shown to identify written words by a quasi reflex action which is little influenced by the contextual information. The least accomplished readers, particularly dyslexic subjects, use the context more in order to identify written words. It is likely that such strategies enable dyslexic subjects to overcome their phonological deficiency. The fact that dyslexic subjects read words they have already encountered better than new words is probably the result of compensatory strategies. Indirect evidence of the progressive adoption of compensatory strategies has been provided by the data generated by longitudinal studies. Most older dyslexic subjects in whom no phonological disorder could

be demonstrated in fact suffered from such disorders in the early periods of their development.

In conclusion, the studies reviewed indicate that a phonological deficiency is the major characteristic of dyslexia. The fact that that type of deficiency is the most frequently reported, including compared to younger normal readers with the same reading level, is the sign of atypical development. In addition, the opacity of the orthography is an exacerbating environmental factor. In the current state of the research, the evidence in favor of a visual deficiency underlying dyslexia (or certain forms of dyslexia) remains fragile.

### **Dyslexia subtypes: single-case studies**

Most of the neuropsychological and neuroscience or genetic studies on the specific disorders of reading acquisition concluded that the dyslexic population was highly heterogeneous. However, the heterogeneity has not been evidenced in the context of studies of groups designed to detect the characteristic general trends of the dyslexic population without taking into account the variability observed in that population. Numerous case studies have nonetheless been published and reflect the diversity of the forms of dyslexia. The studies not only enable the cognitive profile of a given individual to be defined but also the cognitive component(s) whose dysfunction accounts for the deficient performance observed. The studies are therefore particularly pertinent for healthcare professionals confronted with specific cases and needing to formulate hypotheses on the nature of the underlying cognitive deficiency on the basis of neuropsychological assessment data with a view to orienting therapeutic strategy. In theoretical terms, case studies are indispensable for validation of the theoretical models. They may also enable identification of new cognitive components that are potentially involved in dyslexic disorders and whose pertinence can subsequently be tested in larger populations.

Several clearly distinct forms of dyslexia have been observed in children. Although very rarely described, the peripheral development dyslexic forms - visual dyslexia (essentially characterized by preferential production of visual errors) and negligence dyslexia (errors on the first or last part of the word independently of its linguistic characteristics) - suggest that disorders of visual processing at attentional component level may interfere with learning to read.

Phonological dyslexia is characterized by a selective disorder in reading pseudo-words and the production of phoneme errors. Phonological dyslexia is accompanied by dysorthography of the same type characterized by weak performance in taking dictation of pseudo-words. All the cases reported in the studies presented with an associated disorder of meta-phonological capability and several demonstrated a deficiency in short-term verbal memory. Those forms of dyslexia therefore seem to be associated with a phonological dysfunction probably underlying the children's learning disabilities. Case studies of phonological dyslexia thus corroborate the conventional hypothesis that phonological deficiency gives rise to dyslexic disorders. However, certain prototypical cases call for modulation of the concept that phonological skills enable, via the analytical procedure, establishment of memorized orthographic representations. Cases showing excellent performance in reading regular and irregular words despite a massive phonological disorder and a highly deficient analytical procedure suggest that mechanisms of another nature also contribute to memorizing lexical knowledge.

In rare cases, a child may present with an extreme profile of surface dyslexia with no phonological deficiency. The dysorthography is massive: the words are written as they are

pronounced without taking into account their orthographic characteristics. The children seem not to have any memorized information on the orthographic sequences of words. In that type of case, other dysfunctions may be suspected, for instance visual and attentional dysfunctions.

Mixed dyslexia is characterized by difficulties with regard to all the types of item. Mixed dyslexia has been very little studied despite its frequency in the dyslexic population. Two cases described suggest a dysfunction that is either phonological or visual and attentional may underlie this type of disorder. This does not exclude the possibility of encountering two associated deficiencies in certain subjects.

## **Disorders of learning to write**

While the French orthography is particularly difficult to produce (but not to perceive) and it has social value and is socially defended, few in-depth studies on the difficulties associated with it and the disorders affecting it are available. Review of the few studies available and conducted with no attempt at standardization clearly shows the existence of 'fragility zones' 'inducing', in a manner of speaking, the errors. However, the mechanisms involved in error occurrence and the interventions liable to reduce error frequency have been very little elucidated or studied.

The literature available on the difficulties and disorders of orthographic production is rare with respect to written French. Currently, it does not enable rigorous differentiation of the errors encountered in the various dimensions involved in mastering orthographic production. In consequence, it is currently impossible to determine whether differentiated error patterns exist or whether certain error patterns characterize certain handicaps. The only sufficiently numerous data available are for dyslexic subjects. Once again, the data remain general (to the authors' knowledge, no study has addressed error types).

The data available were thus mainly generated by studies conducted on other orthographic systems. Acquisition of the alphabetical principle is problematic but possible as shown by the data generated by transparent orthographic systems (Spanish, Italian, etc.): dyslexic children manage to write words. The difficulties are greater with opaque systems due to the confusion induced by the phoneme-grapheme associations (one phoneme may have several graphemes). Opaque systems do not only encode the phonology but also the lexical and morphological dimensions. Some subjects have difficulty compiling an orthographic lexicon. Sometimes, the lexicon is very limited. Sometimes the difficulties are related not to lexicon size but to its approximate nature or the erroneous nature of the memorized orthographic forms. To the authors' knowledge, no data on the distribution of those scenarios are available. Similarly, no data on their prevalence or concomitant morbidities are available. The extraction of orthographic regularities (double consonants, frequent associations of letters, etc.) has not (yet) given rise to public research. It is therefore not known whether certain disorders are particularly involved. Difficulties relating to use of morphology affect all the population to various degrees: even the leading newspapers contain agreement errors made by eminent authors. The frequency of those errors in the overall population is not known; nor is their distribution which would enable disorders to be suspected in given subjects and related to other characteristics of the subjects involved. Since those errors are also very sensitive to the degree of attention, it is not known whether the errors occur because of misunderstanding, incorrect knowledge or implementation difficulties (for instance because of the attention load associated with writing in young children). Almost everything has still to be done.

Overall, in the absence of precise data on the errors committed by the overall child or adult population, it is difficult to determine the degree to which the number and nature of errors reflect normal performances requiring, for example, more frequent and regular practice of certain exercises or, on the contrary, constitute the grounds for suspecting a disorder necessitating specific management. This is no doubt the cause of two facts that emerged from the review of the literature on orthographic production difficulties and disorders. First, no study of the prevalence of writing learning disabilities is available: the study of those disorders is almost systematically associated with study of reading disorders (dyslexia). At least a priori, that situation leads to other hypotheses being neglected. Secondly, the studies addressing potential associations (concomitant diseases) with disorders other than reading disorders are extremely rare: one study with arithmetic disorders, a few studies with attention disorders. It should be added that, in contrast to what applies for other fields of learning, certain specificities of French orthography (the rareness of spoken language morphological markers and their systematic nature in the written language) prohibit waiting for the indispensable research to be conducted on other writing systems, enabling the solutions to the problems to be, in a manner of speaking, imported. In other words, if we wish to maintain the state of French orthography, it would appear urgent to make a radical change requiring political decisions and to address the nature and frequency of the difficulties, in a non-normative context, and the mechanisms involved in learning and implementing the various components of orthography. It is equally urgent to address the characteristics of the teaching given and evaluate its effects in the short and medium term. Lastly, prevalence and concomitant disorder studies should be conducted without considering, a priori, that the difficulties and disorders of orthographic production are, if not uniquely, at least preferentially observed in dyslexic children. Even if that is true, it is not possible to exclude the existence of disorders in other sub-populations.

In short, the difficulties involved in French orthographic production are such that one would have expected that research on those difficulties, their distributions, and underlying mechanisms and on the effects of the teaching given, including in sub-populations presenting with specific disorders, would be numerous and precise. The data show that that is not the case. The rare data are recent. Moreover, the data mainly cover descriptive aspects: study of cognitive mechanisms and their determinants, and, above all, studies of learning are still largely lacking.

## **Dyscalculia**

Studies of dyscalculia are only beginning and our knowledge of the field cannot be compared to that accumulated with regard to dyslexia. There is no definition or diagnostic criterion universally accepted for dyscalculia. Most frequently, children whose performances are markedly lower than the mean for children of their age in a standardized assessment evaluation and when the intellectual level does not deviate excessively from the normal are considered to present with dyscalculia. These criteria do not enable dyscalculia to be distinguished from what other authors call learning disabilities in mathematics. The analysis of the prevalence studies suggests that while children presenting with difficulties in arithmetic are approximately as numerous as those presenting with difficulties in reading, specific arithmetic disorders are more rarely encountered than isolated reading disorders. In more than one case out of two and even in children with normal intellectual quotients, the difficulties in arithmetic are accompanied by difficulties in reading. In contrast to what seems to hold for dyslexia, dyscalculia affects boys and girls equally.

The broadest consensus addresses the description of the disorder and sectors of arithmetic activities showing the greatest deficiency. Well before the start of systematic learning, children presenting with dyscalculia develop a poor understanding of the principles governing counting activities (counting by pointing with the finger at a set of objects), which constitute the basis on which all subsequent arithmetic skills will be built. Such children are distinguished from others by a more frequent and more prolonged use over development of immature counting procedures to accomplish simple arithmetic tasks. Above all, children presenting with dyscalculia are distinguished by atypical difficulties in memorizing arithmetic facts and learning addition and multiplication tables. Such children have recourse less frequently than others and in a less sure manner to direct retrieval of the result from the memory when doing arithmetic problems. The difficulty proves surprisingly persistent over those children's development. The disorders in the elementary activities have repercussions on the resolution of complex problems and operations. In the latter, use of carryovers remains difficult. The difficulties are all the more marked when the children also present with reading difficulties.

There is also agreement on the course of the disorder. Although few studies have addressed the issue, they all report that dyscalculia is a persistent disorder. However, the 'pure' forms in which arithmetic difficulties are the most isolated are also the most unstable, particularly in infants joining primary school. Thus, certain authors consider the isolated forms as developmental retardation rather than a true between-individual difference. During development, with the exception of the memory difficulties with respect to numerical facts, children presenting with dyscalculia manage to catch up with their peers with regard to the simplest arithmetical operations (additions).

In contrast, there is no consensus with regard to the cognitive profile accompanying dyscalculia. Apart from a few exceptions, most authors report low working memory capacities in children presenting with dyscalculia. Dyscalculia is frequently considered to also be accompanied by deficiencies in visual and spatial terms, although there is no unanimity on that question. However, those aspects are frequently deficient in children presenting with dyscalculia, but it has not been clearly established or universally recognized that they are specific to, or more pronounced in, that disorder. Children presenting with dyscalculia frequently have written language disorders affecting both reading and writing. Children presenting with dyscalculia also have attention disorders more frequently than others.

The uncertainties regarding the cognitive profile may be explained by the existence of several different subtypes of dyscalculia. Although several classifications have been put forward, they are sometimes markedly different from each other. Rarely founded on in-depth analysis of large populations, the pertinence of those classifications is dubious and their number argues against their validity. Even the most intuitively attractive classifications or distinctions have been compromised by rigorous analyses: for instance, it does not seem to be any qualitative differences between the difficulties encountered by children presenting with specific arithmetic disorders and those with associated reading disorders.

Similarly, the causes of dyscalculia have yet to be elucidated. It has not even been established whether dyscalculia is a primary disorder that can exist in an isolated manner or whether it is a manifestation of a more general disorder affecting a wider range of functions and activities. Among the general disorders which are reported to induce dyscalculia, low working memory capacity and a dysfunction of the right hemisphere inducing visual and spatial skill deficiency have been suggested. A more recent hypothesis suggests that dyscalculia may result from dysfunction of the cerebral structures specialized in numerical processing. Derived from evolution, those structures confer on human beings a 'sense' of

numbers and geometric relationships, which are missing in subjects presenting with dyscalculia. This attractive hypothesis nonetheless requires empirical evidence. Although we are not aware of the causes of dyscalculia, all the hypotheses put forward are in agreement on the fact that dyscalculia emerges markedly before the child begins systematic learning in primary school.

Studies of remedial interventions and programs are only just beginning. Addressing small populations, the studies are in general less well controlled than studies investigating the manifestations or causes of the disorder. Although some successes with interventions have been reported (children presenting with dyscalculia taking part show an improvement in arithmetic performance), it is still too early to pronounce on their respective merits and values.

In short, our knowledge of dyscalculia is fragmentary and uncertain. This is due to the limited number of studies compared to dyslexia, for example, but also to the amplitude and difficulty of the subject of study. Number and arithmetic, not to mention mathematics, cover very diverse activities involving a large number of different cognitive functions. However, the difficulties associated with studying dyscalculia may be offset by the fact that it is not only a learning disorder. As cognitive psychology has shown, in human beings, numerical activities and their understanding develop spontaneously. This means that before the child's first systematic learning experience, he/she has built a repertory of knowledge and know-how with regard to number and its uses. This characteristic makes possible not only the early detection of children who present with risks of subsequent learning disabilities, as is the case for reading, but also those who show atypical development in numerical skills, before any scholastic learning. It may be possible to remedy the state before dyscalculia emerges as a specific learning disability.

## Associated acquisition disorders

A shared characteristic of dyslexia, dysorthography and dyscalculia is that the entities are frequently associated in a given subject. For the clinician, rehabilitator or teacher, this provides evidence of the severity of the disorder and hence its repercussions in terms of rehabilitation and teaching. But the existence of disorders associated with dyslexia also constitutes a powerful theoretical tool for the researcher in that it provides potential research orientations.

In the context of a learning disability reference center, it is not surprising to observe a marked predominance of dyslexia or severe disorders of written language acquisition (since, theoretically, we can only term the disorder dyslexia after a certain duration of learning).

### **Inventory of the diagnoses formulated for 209 successive cases attending a reference center for learning disabilities (CHU de Marseille, after Habib, 2003\*)**

Diagnosis	Number of children affected (N = 209)
Dyslexia, dysorthography	177
Spoken language disorders	84
Dyscalculia	48
Dysgraphia	37
Attention deficit/hyperactivity disorder	32
Dysphasia	26
Intellectual precociousness	21

Dyspraxia	19
Behavioral disorders	11
Autism**	2
Dyschronia	45

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\* HABIB M. La dyslexie à livre ouvert. Résodys, Marseille, 2003 : 171 p

\*\* Children presenting with a pervasive development disorder (autism) will not be considered in this section since their management is not implemented in a learning disability reference center. The subject is addressed in the expert review 'Mental disorders: screening and prevention in children and adolescents', Inserm, 2002

Isolated dyscalculia rarely gives rise to consultation of a reference center. This means that the real frequency of dyscalculia in the overall population is higher than that indicated. Dyschronia (disorder of temporal orientation) is less known than the other syndromes, doubtless, once again, because the disorder does not, at least at first sight, constitute a real handicap with regard to scholastic progress. In any event, dyslexia is more frequently associated than isolated (only 10% in the population presented). However, there are no data enabling evaluation of the prevalence of concomitant disorders in the dyslexic population.

Dyslexia is, in over half of the cases, subsequent to spoken language disorders which have a variety of presentations. The term 'dysphasia' used in France for severe spoken language disorders is rarely used in the literature in English, which considers all spoken language disorders to be specific language impairment (SLI) with no preconception as to their severity.

Numerous children of normal intelligence experiencing difficulty in learning arithmetic also present with difficulties in learning written language. Certain authors have reported that in a population of children presenting with dyscalculia, 17 to 64% presented with dyslexia. However, dyscalculia also exists as an isolated disorder. It would appear that children presenting with specific difficulties in arithmetic are to be distinguished from those presenting with difficulties in the two fields. Most of the studies distinguishing the two populations observe differences in the severity of the disorders, the differences in the nature or extent of the difficulties being less certain. Most authors agree with the finding that children presenting with concomitant disorders together with reading disorders have a more marked handicap in arithmetic and in neurological test performance than children presenting with dyscalculia alone. However, the reasons for the frequent association of arithmetic and reading disorders remain obscure. In numerous studies, groups with associated disorders showed a lower intellectual level (at least descriptively and sometimes statistically significantly) than that of the groups with a simple deficiency.

Coordination disorders, which account for about 6% of the overall population with a marked predominance of boys, consist in various symptoms that are easy to recognize but are grouped together in a very variable manner. Broadly speaking, 'praxis' disorders, i.e. disorders in the ability to choose, plan, sequence and execute a movement, with consequences of variable degree on everyday life, are recognized. The incidence of those disorders on learning may be major. But dyspraxia does not simply consist in motor disorders or even sensory-motor coordination disorders: purely sensory disorders may also be incorporated in the concept of dyspraxia and may be more or less marked and affect to variable degrees the principal sensory systems involved in motoricity: the proprioceptive, visual and vestibular systems. In fact, all actions involving gestures and/or posture also require intact perceptual, visual and spatial capabilities, so that it is difficult to dissociate pure praxis phenomena from those related to perceptual capability.

There may be a 'comorbid basis' for developmental coordination disorders (DCD), attention deficit/hyperactivity disorder (ADHA), reading disorders and spoken language impairment (SLI). The question of the links between coordination disorders and reading difficulties is the

subject of particular interest. One of the major questions that have yet to be resolved is whether there is a particular form of dyslexia which specifically accompanies sensory-motor coordination disorders in subjects with dyspraxia. In most of the studies on the subject, over half the subjects with dyspraxia were also found to present with reading disorders. The coincidence between motor disorders and learning disabilities in general and reading in particular is one of the bases of the cerebellar theory, which constitutes one orientation in researchers' current quest to further elucidate the neurocognitive deficits underlying learning disabilities.

Clinical experience shows that it is rare for a child with dyspraxia to have intact writing skills. In contrast, many subjects with dysgraphia do not show clear signs of dyspraxia. The links between DCD and learning disabilities may reflect two scenarios: either dyspraxia with multiple and variable consequences on actions including writing or a more circumscribed disorder affecting writing itself and then closely linked to performances in the linguistic area.

Statistical studies of the association between precociousness and learning disabilities are rare. Practically no scientific publication has been devoted to the association between precociousness and learning disabilities, although, paradoxically, educational commentaries in non-scientific form are abundant. There are tens of articles or reports available on the Web. The lowest estimates indicate that 2 to 10% of the children enrolled in American studies on gifted children also suffer from specific learning disabilities. This is not significantly more frequent than in the overall population, but about 40% of those gifted school children do not undergo diagnosis before high school. Incontestably, the literature remains insufficiently precise in the field, preventing determination of the exact frequency of the association between precociousness and learning disabilities. Further studies are necessary. If the finding is confirmed, however, considering intellectual precociousness as a disorder concomitant with dyslexia and other learning disabilities would be of undeniable theoretical importance and constitute an important instrument for tackling the question of the underlying mechanisms. In that case, it would be necessary to image that a common process is capable of inducing both dysfunction of one module and enhanced function of the other. This places a considerable restraint on the potential explicatory models.

## **Associated behavioral or emotional disorders**

Several published studies provide significant evidence of a frequent association between learning disabilities and behavioral or emotional disorders. Emotional or behavioral disorders may then be secondary to failure at school and/or the social, familial or affective environmental conditions exacerbating the cognitive disorder, or, on the contrary, constitute a real concomitant disorder with a link, perhaps of a genetic nature, between the two diseases. Management of those two types of disorder is possible even though the question of the mechanism underlying their association has yet to be elucidated.

Large-scale child cohort studies using a methodology based on standardized questionnaires such as the Child Behavior Questionnaire (CBQ) show that children presenting with psychological disorders (externalized or internalized) have a reading and/or mathematical level lower than children free from those disorders. The studies do not describe the cognitive profile of the children or the severity indices of the learning disabilities. They do not enable recognition of a child population that, due to poorly managed dyslexia and dysorthography and failing at school, presents with secondary psychological disorders.

Anxiety, depressive mood, attention deficit / hyperactivity disorder (ADHD) and behavioral disorders are frequently observed in children who read poorly. The association was evidenced as of the first evaluation in longitudinal studies over 7 to 10 years. The possibility of early treatment may be envisaged. The association exists independently of the other familial and social environmental factors that may be causal in emotional and behavioral disorders.

Behavioral disorders are more frequent in dyslexic children or children presenting with dyscalculia than in children free from learning disabilities, but less frequent than in the child population referred for psychiatric treatment. Performance anxiety, difficulties in peer relations, family conflicts and low self-esteem are frequently encountered in that population. By way of an example, a French reference center for language disorders found, in the course of psychological and speech-therapy medical examinations, affective disorders in 28% of 173 children presenting with a learning disorder. This calls attention to the fact that the clinician must be attentive to the cognitive and affective or behavioral aspects in the diagnostic evaluation and management of learning disabilities.

Abundant literature on the association between learning disabilities (particularly dyslexia) and attention deficit with or without hyperactivity disorder is available. All the studies agree on the fact that a child with dyslexia or attention deficit / hyperactivity disorder (ADHD) has a high risk of having another disorder. However, the selection of measurement instruments, criteria defining ADHD and learning disabilities, child age and recruitment location may explain the disparity in the frequency of comorbidity. Several causality hypotheses have been advanced and the question of the mechanism underlying the association is far from being resolved. For certain authors, the behavioral disorders may be a consequence of failing at school, or, on the contrary, for others, the reading difficulties of hyperactive children may be explained by the attention deficit. For other authors, there is a link, doubtless of a genetic nature, between the two diseases as has been suggested by the study of twins conducted by the Colorado Learning Disabilities Research Center. The higher frequency of behavioral disorders in dyslexic subjects seems directly related to the comorbidity between dyslexia and hyperactivity and to be specific to boys, while the association between anxiety or mood disorders, on the one hand, and dyslexia, on the other, is independent of hyperactivity and more frequent in girls.

It would appear indispensable to evaluate the attentional ability and degree of hyperactivity together with the other associated psychopathological symptoms in children presenting with a learning disorder in order to enable complete management.

Several teams have reported their experience as pediatric psychiatrists, psychologists or psychoanalytically-oriented psychologists with regard to children presenting with learning disabilities. When requested by teachers or rehabilitators, the psychiatrists and psychoanalysts addressed the psychological functioning that provided the context for the child's learning difficulties in order to better elucidate the therapeutic orientations to be proposed. Studies using projective tests in dyslexic child populations do not show a univocal personality organization in those children, compared to dyslexia-free controls, excluding a unique causal relationship between personality and dyslexia. Nonetheless, when the child and his/her family are in distress and/or when rehabilitation does not yield the desired results, analysis of the child's psychological development and interactions with his/her environment using psychoanalytical approaches, may constitute a valuable complement to the cognitive approach. Providing the two approaches are not mutually exclusive, they enable management of the whole child in his/her diversity.

## Theories explaining dyslexia: phonological theory

Among the hypotheses advanced to explain dyslexia, the phonological theory is well supported by empirical studies.

The basis of the phonological theory consists in the fact that reading is a language activity. The theory is based on the finding that written language, which arose after spoken language in phylogenesis (the development of the human species) is also set up after spoken language in ontogenesis (individual development). Moreover, irrespective of the writing system, the written language is, intrinsically, a second system relative to the spoken language: even though perceiving writing depends on sight while perceiving the spoken language depends on hearing, the reader can always access the spoken form of the words he/she reads. Lastly, the phonological theory is based on the definition of dyslexia, which is a specific disorder of learning to read and does not result from patent sensory deficiencies.

Irrespective of the writing system, the word is the basic unit of writing. This explains why specific reading skills are based at the level of the procedures identifying written words. The development of automatism with respect to that identification enables the child to achieve a degree of reading comprehension equivalent to that of oral comprehension by freeing the child from slow and cumbersome decoding. In alphabetical writing, the identification may be obtained by decoding, based on the grapheme-phoneme correspondances, or by the lexical procedure which is based on the words. However, decoding does not merely consist in the beginners' laborious reading: the expert reader may identify words that he does not know in a few hundreds of milliseconds. Moreover, the lexical procedure is, which is not based on the silhouette of written words (their overall form), enables the expert to achieve access in a few hundreds of milliseconds, not only to the visual code but also to the phonological and semantic code.

The phonological hypothesis is based on the finding that the reading performances of dyslexic subjects are notoriously weak when those subjects cannot rely on their lexical knowledge, i.e. when they have to decode rare words or non-existent words (pseudo-words). All the studies of undifferentiated groups of dyslexic subjects have evidenced that deficiency, including relative to younger children with the same reading level. Moreover, the deficiency, which is more marked when dyslexic subjects are confronted with opaque writing, is mainly observed in the slowness of the response when the writing is transparent. Lastly, in most of the cases, the phonological reading skills of dyslexic subjects are inferior to those of younger children with the same reading level. The results suggest that deficiency in the phonological reading procedure is robust and prevalent.

The conventional phonological hypothesis explains the reading difficulties of dyslexic subjects by the weakness of their phonological skills outside of reading and including in phoneme analysis and phonological short-term memory. Those deficiencies may interfere with decoding given that, in order to use the procedure, the graphemes must be matched with the phonemes. This calls for phoneme analysis capability. The units resulting from decoding have then to be assembled in order to access the words. This requires solicitation of the short-term phonological memory. More recently, deficiencies in access to the spoken lexicon were reported for subjects with dyslexia. On the basis of that finding, some researchers assumed that the origin of reading deficiencies in dyslexic subjects is dual: one origin is related to analytical skills and phonological memory while the other is related to lexicon access, generally evaluated by the response time in tasks of fast naming of images of objects or colors. This hypothesis is supported by the fact that succeeding in that type of task explains a unique part of the variance in reading in addition to that explained by phonological capability. Moreover, phonological capability and the ability to rapidly name

are not correlated with the same reading skills. The former explain response precision while the latter explain processing time. Those results may nonetheless be due to the type of measurement used (accuracy scores for the phonological tasks, processing time for the others) and not to the type of task. It is currently admitted that fast naming tasks evaluate phonological skills which, when the child mainly uses decoding, enable him/her to access the spoken word matching the sequence of letters decoded rapidly and precisely.

The conventional phonological hypothesis is robust: in most of the studies conducted on groups, phonological deficiencies were found in the dyslexic subjects. In studies of individual data, a phonological deficiency was identified in most of the cases of dyslexia. Lastly, the phonological capabilities are the only capabilities that explain the reading performances of subjects with dyslexia with regard to both concomitant performance (as evidenced by regression analyses) and future performance (as evidenced by longitudinal studies). In addition, neuronal dysfunctions have mainly been detected in the brain areas involved in language processing (left perisylvian area). The dysfunctions mainly consist in hypo-activation of the areas dedicated to phonological processing, which is a strong argument in favor of the hypothesis. However, in those studies, the 'phonological' factor incorporates various capabilities. Deficiencies in those various fields could, in fact, be explained by an underlying factor: either a deficiency in rapid auditory processing or a deficiency in phoneme discrimination.

The auditory hypothesis stipulates that the phonological deficiency in dyslexia derives from deficient auditory perception affecting the processing of short sounds and fast temporal transitions, irrespective of whether or not the sounds of language are involved. In the current state of the research, the auditory disorders, when they are present, do not seem related to the rapidity of stimulus succession or to the order of appearance of the stimuli. In addition, compared to the conventional phonological deficiencies, the deficiencies are not frequent. Lastly, they do not enable explanation of the deficit in reading skills in dyslexic subjects when correlation and regression analyses are conducted.

Under the terms of another hypothesis, dyslexic subjects have difficulties discriminating between phonemes. This explanation is based on the fact that in order to map graphemes to phonemes, phonemes must not only be isolated but also discriminated. While numerous studies have addressed the links between learning to read and initial phonemic segmentation capability (as evaluated by counting or phoneme deletion tasks), few studies have addressed the incidence of the quality of phoneme representations on learning. However, the phoneme is the result of an arbitrary and specific slicing of language. In an acoustic continuum, we categorize, which means that we perceive a range of acoustically different sounds as /p/ and others as /t/ or /k/. Secondly, the phoneme repertory differs between languages. The phoneme is the minimal unit enabling differentiation of two words. Thus, /b/ and /v/ are two different phonemes in French which enable differentiation of 'bol' and 'vol', but not in Spanish. In contrast, the simple /r/ and rolled /r/ are two different phonemes in Spanish that enable distinction between '*pero*' ('but') and '*perro*' ('dog') while, in French, the two forms of /r/ are only dialect variants, allophones, of one phoneme. Neonates have been shown to perceive different phonetic oppositions liable to intervene in the world's languages. As a function of the neonate's linguistic environment, the repertory will very rapidly be restricted to the phoneme categories of value in processing the infant's language. This calls for a process of selection and restructuring of the initial phonetic categories. This process is considered not to have been completed in future dyslexic subjects or at least in some of them.

The studies available indicate that in dyslexic subjects the phoneme categories are not as clearly specified as in normal readers. Dyslexic subjects discriminate phonemes belonging to

different phoneme categories in their language less well (for instance, /b/ and /d/). In contrast, they have better perception of certain phonetic differences which are not of value in processing their language (allophonic perception). This double deficiency, which is not related to an auditory deficit, may only have minor consequences on spoken language acquisition. Access to the mental lexicon may take place using allophonic representations even though it may be less economical than if it were based on phoneme representations, at least in terms of the volume of information to be processed. In contrast, that type of deficiency may seriously interfere with the acquisition of written language: in order to match graphemes with the corresponding phonemes, clearly specified phoneme categories are necessary. These results may explain the difficulties of phoneme analysis, but also those of phonological short-term memory due to an increase in the memory load related to an expanded phonological repertory, that is allophonic rather than phoneme-based. The results may also explain the difficulties in accessing the lexicon, in particular those highlighted by serial rapid naming tasks. Hence a syndrome underlying the various deficiencies incorporated in the classic phonological explanatory framework for dyslexia may exist.

The results obtained with phoneme discrimination tasks provide further arguments in support of the explanatory hypothesis of phonological deficiency in dyslexia. However, complementary studies are necessary in order to evaluate, first, the frequency of the deficits, and, secondly, their explanatory power and the relationships they maintain with conventional phonological deficiencies (among others, phoneme segmentation and phonological short-term memory deficiencies).

## **The visual dimensions of dyslexia**

Another explanatory hypothesis for dyslexia covers the visual dimensions. Learning to read requires matching orthographic information derived from visual analysis of the sequence of the written word and phonological information derived from auditory processing of the corresponding spoken sequence. Numerous studies have addressed the nature of the visual processing involved in that learning and have formulated the hypothesis that certain dysfunctions in visual processing could induce dyslexic disorders.

Work conducted on that context suggests that certain subjects with dyslexia have, in fact, a deficiency in visual processing (independently of any perceptual impairment). It is important to note, however, that those visual disorders have never been evidenced by clinical tests and, in order to evidence them, computerized psychophysical tests are required.

The results of a number of empirical and neurophysiologic studies argue in favor of impairment of the magnocellular visual system in subjects (adults and children) presenting with developmental dyslexia. This conclusion was based on the fact that certain studies evidenced a disorder of sensitivity at low spatial frequencies and high temporal frequencies together with reduced sensitivity to moving points: the performance of dyslexic subjects is inferior to that of normal reading subjects with regard to detecting movement of a set of points. Dyslexics are less able to discriminate the difference in speeds of two moving targets. Some studies have also shown the existence of correlations between the performances obtained in psychophysical visual tests and various reading measurements. In addition, the magnocellular hypothesis, initially restricted to the visual system, has gradually evolved to tend toward the hypothesis of an amodal disorder of the magnocellular systems: dyslexic children are reported to have difficulty processing fast temporal information in both modalities (visual and auditory) subsequent to joint impairment of the visual and auditory magnocellular systems. The results of empirical and neurophysiologic studies would appear

to support the hypothesis and thus account for the concomitant occurrence of phonological and visual disorders in certain dyslexic subjects.

However, the magnocellular hypothesis is currently controversial. The methodological limits of certain studies that found a magnocellular visual deficiency have been stressed and the hypothesis has suffered from lack of result duplication. The amodal hypothesis is also to be reviewed in the light of results suggesting that only a small proportion of dyslexic subjects with a phonological disorder also have difficulty in processing the sounds of speech and difficulties in magnocellular visual processing. However, the research conducted in that context points to the extreme complexity of the type of disorder in question. It may now be considered that the disorder is manifested under certain particular experimental conditions that have largely still to be defined and is only observed in a subpopulation of dyslexic children that has also not been clearly defined.

The most recent studies suggest, in particular, that the magnocellular disorder could only become patent when the task involves specific attentional processing. This is in line with the results of a certain number of other studies showing the existence of visual attention disorders in contexts of dyslexia. Difficulties in attention focusing, attention disengagement and problems of automatic orientation of the attention giving rise to a left mini-negligence phenomenon have been described in dyslexic subjects. However, the hypothesis of a visual attention disorder like the hypothesis of magnocellular impairment is confronted with the fact that the deficiencies have most frequently been observed in association with phonological disorders. Thus, visual attention disorders are to be viewed in the context of an amodal disorder of attention processing extending to the auditory modalities and perhaps also haptic modalities in addition to the visual modality. It is, however, important to note that the deficiencies evidenced in dyslexic subjects are not situated in the context of a general attention disorder (e.g. ADHD). Few studies have provided any evidence elucidating the link between visual attention disorder and the reading profiles of dyslexic children.

Most frequently, the visual or visual attention dysfunctions have been described in the context of phonological disorders. Recent studies suggest, however, that a particular form of dysfunction, a disorder of the visual and attentional span, may be associated with certain forms of dyslexia and be observed independently of any phonological impairment. The concept of the visual and attentional span refers to the number of letters that can be simultaneously processed in the context of a word letter sequence at each glance. A reduction in the visual and attentional span has been evidenced in certain cases of surface dyslexia with no associated phonological disorder. Group studies suggest that the number of children presenting with that type of deficit is at least equal to the number of children presenting with an isolated phonological disorder in both the English- and French-speaking populations. Studies need to be continued in order to test the hypothesis that there is a causal relationship between visual and attentional span deficiency and difficulty in learning to read.

All the professionals agree that a diagnosis of dyslexia can only be formulated after having checked that no visual perception disorder is present: an ophthalmologic examination is therefore necessary in order to determine the child's visual acuity and rule out problems such as hypermetropia, myopia or astigmatism. It is also necessary to question the child on the feelings experienced when reading. Some dyslexic children have the impression that the letters move or overlap during reading. This reflects instability of binocular control. Any complaints of that type together with a number of warning signs (visual errors, difficulty following the lines, jumping from line to line) require complementary investigations (orthoptic examination and evaluation of the child's binocular focusing). The clinical examination must also rule out oculomotor disorders (e.g. nystagmus or exophoria).

Management methods such as colored lenses, prisms or temporary occlusion of one eye are not unanimously approved either by researchers or ophthalmologists. It must be noted that those 'treatments' have yet to be firmly validated and a convincing theoretical context has yet to be formulated. Instruments for the clinical diagnosis of magnocellular or visual and attentional disorders are lacking. Research needs to be pursued in order to develop the training necessary for management of those disorders.

## **Cerebellar hypothesis**

Again in an attempt to account for the complexity of the clinical symptoms observed and, in particular, the frequent association, within learning disabilities, of deficits in fields as varied as reading, language, arithmetic and even the sensory-motor systems, scientists have searched for explanation and proposed models tending to extract the shared characteristics of the various disturbed areas.

Thus, certain associations observed in dyslexic subjects have attracted the attention of researchers: a delay in the stages of motor development, sequential and temporal disorders (say the time, remember the month of the year) and, above all, the presence of motor coordination and balance disorders, all suggest dysfunction of the cerebellum. The latter organ, which was for many years considered to play a purely motor role, has been the subject of recent studies that have demonstrated its involvement in numerous cognitive processes and in learning in general.

Empirical evidence has supported a theory that was essentially based on clinical intuition: firstly, anatomic studies of the human brain *post mortem* and using various imaging methods have detected an anomaly of the cerebellum in dyslexic subjects.

Rather paradoxically, a study demonstrating cerebellar hypo-activation during purely motor tasks (learning a series of finger movements) in dyslexic adults was to draw attention to the cerebellar theory. In contrast, although the cerebellum is classically activated when a normal reader reads, almost no reports of cerebellar activation deficit when a dyslexic subject reads have been published.

Hence, the cerebellum may affect reading in different ways. The cerebellum is involved in control of eye movements, visual and spatial attention and peripheral vision, all essential components of reading. As a crucial structure in time management by the brain, the cerebellum may contribute to the sensory-motor coordination and intense integration problems observed in dyslexic subjects.

According to the advocates of the cerebellar theory, it supposes that the deficiency is present very early, as of birth, and interferes with the normal setup of auditory and articulatory skills necessary for constitution of the phonological system, including visual abilities such as eye movement and letter recognition, giving rise to both the phonological and orthographic difficulties characteristic of dyslexic children and adults.

As attractive as the theory may seem, it has been greatly criticized in recent years. The critics observe, first of all, that motor disorders are far from being the rule in all dyslexic patients and that many dyslexic subjects, even those with major difficulties in learning to read, show no motor difficulty or indeed are particularly gifted for motor activities both proximal and balance motoricity and distal motoricity. Certain authors have even suggested that the motor disorders sometimes observed in dyslexic subjects are only an artifact related to the coexistence of hyperactivity disorders. While several studies have thus minimized the incidence of such motor disorders, at least two recent studies, using sophisticated balance

and posture measurement systems, have contributed positive arguments in support of the cerebellar hypothesis. Dyslexic subjects were found to be significantly less able than controls to maintain balance on one foot, in particular with the eyes open. The performance of the dyslexic subjects in the postural test correlated with their reading and orthographic performance. Lastly, if, as has been reported, cerebellar-type disorders are less frequent in dyslexic adults than in dyslexic children, this perhaps means that the deficiencies initially observed in children stabilize over adolescence and have resolved by the time the dyslexic subject has become adult. In short, it is improbable that dyslexia can be explained by cerebellar dysfunction. However, the arguments are sufficiently numerous to encourage inclusion of the cerebellum among the brain systems, which are disturbed in at least some dyslexic subjects. In any event, the cerebellar hypothesis has had the merit of proposing a plausible alternative to the theses in which the reading disorder was the sole subject of interest to researchers. The cerebellar hypothesis has thus opened the way to taking into account comorbidity in the explanation of dyslexia.

## **The temporal processing hypothesis**

Not unrelated to the cerebellar theory, a hypothesis was proposed over 30 years ago by an American scientist, Paula Tallal, under the name: 'Temporal processing deficit theory.' The dyslexic child and, more generally, children suffering from specific learning disabilities, frequently have problems with time in general irrespective of whether they consist in problems with management of the temporal aspects of everyday life, consciousness and/or the perception of the duration of events or discrimination between brief events such as the events constituting human speech. Very many studies using a variety of approaches have addressed that strange characteristic with the aim of identifying a common point between the latter and learning difficulties.

According to the theory initially proposed by Tallal, the brain of a child suffering from spoken and written language disorders is constitutionally unable to specifically process the environmental stimuli characterized by briefness and rapid succession which are precisely the two characteristics of human speech. This finding becomes meaningful when we observe the auditory confusions to which many dyslexic children are subject. Dyslexic children tend to confuse acoustically close phonemes, in particular pairs such as /t/-/d/ and /ch/-/j/ which seem particularly difficult to distinguish for the auditory system of a dyslexic subject. Among the tests enabling evidencing of temporal auditory processing deficit, Tallal's 'repetition test' is doubtless the most widely used. The subject is presented with pairs of different sounds in random order. The child is to reproduce the sounds by successive pressures on two keys of a computer keyboard. By varying the interval between the two sounds from a few milliseconds up to several seconds, it is observed that the dyslexic subject experiences marked difficulties in reproducing the order but only for short intervals of less than 150 milliseconds. For longer intervals, the dyslexic child's performance becomes similar to that of the non-dyslexic controls. This disorder in temporal sequencing is found in children presenting with language disorders in general but also in at least some of the children presenting with dyslexia. In addition, the same effect can be demonstrated in dyslexic subjects by using pairs of syllables such as /ba/-/pa/ or /da/-/ga/. The simple test gave rise to a vast debate. The defenders of the temporal hypothesis consider it to demonstrate that dyslexic subjects suffer from a difficulty that is both perceptual and temporal, while the critics stress that the effect is not present in all dyslexic subjects and, above all, varies depending on the linguistic or non-linguistic nature of the stimuli. If children are required to fulfill a temporal order judgment (TOJ) task using pairs of phonemes

that are easier to discriminate, the deficit disappears. The same applies if the stimuli consist in artificial speech.

Another important consideration is related to the potential heterogeneity of the concepts based on the term 'temporal deficit'. While TOJ has been most widely studied and, in general, found to be deficient in dyslexic subjects, it has rarely been related to other aspects of temporal processing such as the judgment of the relative duration of a stimulus. In addition, it is important to consider the links between a potential temporal disorder and the intensity of the phonological disorder considered the crucial mechanism in learning to read. A recent study has shown that TOJ deficit is significantly correlated with the phonological disorder and a measurement of the judgment of auditory stimulus duration. However, the two deficits were not inter-correlated suggesting that they consist in two separable dimensions of the temporal deficit.

Another postulate of the temporal theory predicts that the deficit is to be found in different modalities. Several studies, including one by Tallal's team, have demonstrated that dyslexic subjects have difficulty discriminating between two stimuli in tactile mode (identifying which of two fingers of the same hand were touched simultaneously). More recently, various studies have evidenced judgment deficits with respect to visual and tactile stimuli in dyslexic subjects, including both in the same subject. This supports the idea of a supra-modal disorder, i.e. a disorder independent of the sensory modality. Lastly, several authors have recently stressed a particular difficulty in dyslexic subjects with regard to discriminating between stimuli not only in several perceptual systems in a given subject, but in tasks involving confrontation of several modalities in a given task, thus implementing an inter-modality transfer.

In both dyslexic children and adults, the protocols evidenced very marked differences for the majority of combinations studied. For the authors, these results show that, beyond the multi-modality impairment suspected by Tallal as of the initial formulations of her theory, there also exists a deficit in the temporal processing necessary for the pooling of information reaching the brain via different sensory channels in the dyslexic subjects. A recent study using a visual temporal order judgment paradigm associated with an auditory facilitator, suggested that dyslexic subjects have a problem with respect to the width of the 'temporal window'. The abnormal width of the temporal window in dyslexic subjects is considered to impair the process depending on rapid and precise pairing of two items of information derived from different modalities such as those involved in grapheme-phoneme conversion.

Thus, after a period in which the theories based on a supra-modal temporal disorder were sharply questioned, the more recent research, using refined study protocols, seems once again to argue in favor of a deficiency in certain very specific aspects of sensory information processing, in particular when two different items of information are to be related. The temporal characteristics of relating those items may be determinant. These findings are reminiscent of an already old theory of learning known as 'Hebb's synapse': when two neurons, A and B, are in proximity and neuron A discharges when B is activated, the reciprocal links between A and B are strengthened. Conversely, if A discharges when B is inactive, even if the discharge takes place within a few milliseconds, the links between the two neurons are inhibited. The general principle, which since it was first formulated in the 1940s has been confirmed empirically several times, could well prove, on the basis of the most recent data, a propitious framework for explaining the diverse and still poorly elucidated findings with regard to learning disabilities.

With regard to the auditory modality, a large number of studies have used the evoked potential method, of particular value in this context because of its temporal sensitivity: various studies have shown that the auditory cortex of the dyslexic subject imperfectly

processes auditory stimuli with, specifically, at least in certain cases, evidence of an anomaly in the processing of the temporal succession of the acoustic components of linguistic units. Thus, the acoustic difference between the phonemes /ba/ and /pa/ (the concept of voicing) is reflected in the electrical activity recorded opposite the auditory cortex by a subtle difference in the temporal succession of events strictly aligned temporally with the acoustic events. In dyslexic adults, the same stimulus is processed in a temporally anarchic manner such that the usual succession of the acoustic components is no longer recognized at electrical level. It may be presumed that a minimal disturbance in the simultaneity of activation of the various cell components of the system, by reducing the strength of their reciprocal connections, would be sufficient to prevent acquisition of specific perception of voicing simply because the latter is, among the characteristics of the human voice, the most dependent on the temporal organization of the stimuli.

## **The contribution of brain imaging**

The neurological approach to dyslexia in contemporary research began with the founding histological study by Galaburda et al. who showed the presence of micro-structural anomalies in the perisylvian cortex (ectopias and dysplasias) and a reduction in the size of the neurons in the geniculate nucleus. Those results suggest the existence of anomalies in neuronal migration and maturation in certain zones of the cortex essential for the automatism of reading mechanisms.

Given that basis, morphometric studies using magnetic resonance imaging (MRI) were conducted to detect the existence of macroscopic anomalies in cortical structure in dyslexic subjects. Targeting predefined regions of interest (parietal cortex, temporal cortex, inferior frontal cortex, cerebellum, corpus callosum), the studies detected structural anomalies but with no great degree of between-study reproducibility. Technological progress enabled analysis of the whole brain volume using voxel-based morphometry, a diffusion-tensor imaging method. The results obtained with the latter method showed particularities of the signal generated by a zone of white matter underlying the left temporoparietal junction suggesting abnormal connectivity between those zones of the cortex. In addition, correlations between anomaly intensity and reading score were observed.

Cerebral functional imaging (positron-emission tomography (PET) and functional magnetic-resonance imaging (fMRI)) and electromagnetic techniques (electroencephalographic-evoked potentials (EP), magnetoencephalography (MEG)) enabled detection of anomalies in the activity of the regions involved in reading and the associated cognitive processes in dyslexic subjects vs. normal readers. It should be noted however that the studies are subject to numerous experimental biases which may explain the discrepancies between the studies.

In the field of electrophysiology, anomalies have been reported in both the early and late potentials as a function of the experimental paradigms and visual or auditory modalities investigated.

Under PET or fMRI imaging, the normal functional anatomy of reading isolated words involves three major areas: the left temporo-occipital junction, the left temporoparietal junction and Broca's area. Activation of the 3 regions is abnormal in dyslexic subjects as is the functional connectivity between those regions. The two temporal and/or parietal regions are very generally hypo-functional. This has been confirmed by MEG studies. Hyperactivation of Broca's area in patients has frequently been considered to reflect premotor compensation for posterior cortex deficiency. Compensatory effects have also been suggested in order to explain the more marked activation of the right perisylvian cortex in dyslexic subjects.

The results in adults have broadly been confirmed by fMRI and MEG studies of children. Overall, the studies suggest an anomaly of connectivity with respect to the temporoparietal and frontal circuits which underlie language and particularly the circuits subjacent to the phonological loop in working memory and the interaction between the 'dorsal and ventral reading circuits'.

Numerous pathophysiological hypotheses with regard to the etiology of dyslexia have all been empirically supported by brain imaging studies confirming the existence of the predicted differential effects. The hypotheses may be divided into two types. The first type postulates the existence of a deficit affecting the phonological or lexical representations stored in long-term memory (phonological hypothesis). The second type covers numerous hypotheses suggesting a variety of anomalies in the sensory-motor processes for real-time processing of perceptual information.

The phonological hypothesis, which is predominant, has been greatly strengthened by the existence of activation deficits in the left perisylvian regions normally involved in phonological analysis and phonological working memory.

Certain studies have suggested a dysfunction of the magnocellular visual pathways but others have shown that those effects were only evidenced under certain empirical constraints. In the auditory field, several studies have shown a deficit in processing, of the magnocellular type, with respect to rapid acoustic or linguistic variations. However, recent data suggest that those effects are only present in subgroups of subjects and only involve certain sectors (left premotor) of the cortex while other sectors (left supramarginal) show constant hypo-activation irrespective of the variation rate of the speech signal.

Lastly, anomalies of right cerebellar cortex activation were observed during tasks consisting in learning a sequence of finger movements.

The neurological imaging methods were more recently applied to the study of the cerebral correlates of the improvement in performance induced by various interventional or intensive training methods for groups of dyslexic children. The greater influence on performance and neurofunctional signals of intensive phonological intervention, compared to conventional management, was demonstrated in some of those studies. In other studies, positive correlations between the increase in the fMRI activation signal generated by the left temporoparietal and right frontal regions and improvements in language and phonology scores, without generalization to the reading score, were observed. The various methods of functional imaging are undoubtedly sufficiently sensitive to evidence changes in cerebral activity following remediation but the specificity of the mechanisms underlying the effects obtained at cognitive and cerebral level are to be discussed.

So far, the functional or anatomical cerebral imaging studies have shown neurological correlates with the cognitive disorders. However, the direction of the causal relationship could be determined by longitudinal studies initiated in young children. Those studies would enable the effects of cerebral anomalies present from the outset to be distinguished from those resulting from adaptation to the handicap.

Lastly, functional cerebral imaging could play a role in the very early identification of the cerebral activity profile enabling prediction of the emergence of a handicap at learning to read level, particularly in families at genetic 'risk' of developing the disorder. An ethical assessment of that research orientation is to be conducted due to the risk of stigmatization when the resources necessary for handicap management are not ensured.

## Genetic factors

The hypothesis of the familial nature of dyslexia was first suggested long ago and circumstantial evidence collected over 20 years does indeed point to a genetic origin of dyslexia.

The best indicator of a familial component is an increase in risk in the relatives of an affected subject. The risk is a relative risk comparing the risk of relatives of healthy subjects to the risk of the disorder in the relatives of subjects suffering from it. In general, the first-degree relatives' (parents, brothers and sisters) history is used. For dyslexia, the risk of the disorder in a relative of an affected subject is 8- to 10-fold higher than the same risk for the relatives of an unaffected subject. This does not exclude the occurrence of other sporadic cases insofar as the same cerebral and cognitive dysfunctions may occur *de novo*. Family clustering suggests but does not prove a genetic origin. Families share not only some of their genes but also a certain environment. It may be considered that parents who do not read constitute an environment that is not conducive to learning to read for their children.

Studies of twins enable optimum assessment of the weight of the genetic factors compared to that of environmental factors. The studies are designed to calculate the heritability, in other words the percentage of the variance explained by genetic factors. In order to do so, the concordance between the impairments of monozygotic and dizygotic twins is measured. When a monozygotic twin is dyslexic, the probability of the other twin also being dyslexic is about 70%. In contrast, the probability is only 45% for dizygotic twins. Since monozygotic twins may be considered to share the same environmental factors as dizygotic twins, the difference in concordance is explained by the fact that monozygotic twins are 100% genetically similar while dizygotic twins are only 50% similar (for the genes which vary). The concordance data thus enable calculation of the heritability. This does not, however, indicate a direct causal relationship. A change in the environment of two true twins during their intrauterine development or during the perinatal period (lack of oxygen, maternal hormone, toxic product, etc.) may induce the same effect after birth through similar responses to an environmental change. The hypotheses deserved to be studied in the field of dyslexia.

The heritability of dyslexia is between 50 and 65% according to the larger scale studies on twins. Similar data were collected with regard to mathematical difficulties with an heritability of about 50%. With regard to dysorthography, the genetic studies did not truly distinguish it from dyslexia. Many studies used both orthographic and reading measurements to define the phenotype. For the time being, no study has specifically investigated for genetic factors which might distinguish dyslexia from dysorthography.

The data show that there is indeed a genetic contribution to specific learning disabilities but do not identify the genetic factors or explain their mode of action. That is the role of molecular and neurogenetic studies. At present, such studies have only begun generating results for dyslexia. With regard to the other specific learning disabilities, molecular genetic research is only at its inception.

In the case of dyslexia, numerous analyses of the genetic link have shown chromosomal regions that are significantly more frequently transmitted from the parents to children with disorders than to children without disorders. The link between dyslexia and regions on chromosomes 1, 2, 3, 6, 15 and 18 has been reported by independent teams on the basis of several studies. The multiplicity of the chromosomal sites identified suggests that the specific learning disabilities are, in most cases, deficiencies with a complex genetic component in which several genes are involved.

The linked chromosomal regions indicate regions of the genome in which the genes associated with dyslexia are very probably located. However, the regions may contain hundreds of genes that have been inventoried but that, in the majority of cases, have never been studied. There is thus an enormous gap between identifying a linked region and identifying the associated gene in that region. Progress in molecular genetics is reducing the magnitude of the gap day by day.

In the six regions, four candidate genes have been proposed. Two of them (*DYX1C1*, *ROBO1*) show mutations in rare cases or families but it is not known whether the alleles of those genes increase the risk of the more common forms of dyslexia. For the other two genes (*KIAA0319* and *DCDC2*), susceptibility haplotypes in large populations have been suggested. The haplotypes have nonetheless to be confirmed. Outside of the rare cases of a radical mutation impairing protein function (*DYX1C1* in a Finnish family), it would appear that susceptibility alleles exert their effects through alteration of the expression of the protein (certain haplotypes are, moreover, situated in regulatory regions). Nonetheless, the expression data remain fragmentary and require confirmation.

The most remarkable result with respect to the functional properties of the genes was generated by studies on developing animals. The studies show that four of the candidate genes for dyslexia are involved in the migration of neurons during cerebral development, three in the radial migration of cell bodies towards the cortex and one (*ROBO1*) in axon and dendrite migration. It seems highly unlikely that the convergence of functions is random. Moreover, the results enabled a link to be established between the genes associated with dyslexia and the cerebral abnormalities observed in the brains of certain dyslexic subjects.

It is important to note that while the genetic usage is such that the genes are referred to as 'genes for dyslexia', this is a misuse of language and in fact a shortcut for saying 'genes of which certain alleles increase the risk of dyslexia'. It is obvious that none of the genes is specific for dyslexia or is a gene for reading or even a gene for spoken language. Like almost all human genes, related forms are found in other mammals or even in *Drosophila* (*ROBO1*). In all animals the genes exert multiple functions and are expressed in numerous different organs during development and life. What links the genes more specifically to dyslexia is the fact that they are also involved in a particular stage of brain development and notably in the setup of certain brain areas which are later recruited for learning to read.

In certain rare cases, it would appear that mutation of a single gene is sufficient to induce dyslexia. The gene responsible may vary between cases. Most frequently, dyslexic subjects do not seem to carry a rare mutation but rather susceptibility alleles. The alleles are frequent in the normal population and do not in themselves constitute a sufficient cause of dyslexia. Each susceptibility allele slightly increases the risk of dyslexia. The disorder only emerges when particularly unfavorable combinations of susceptibility genes and/or interactions of the alleles with non-genetic factors combine in increasing the risk. A large number of environmental factors (biochemical, traumatic, linguistic, socio-educational, pedagogic) may modulate the expression of genetic factors in a positive or negative direction.

Thus, it is highly probable that in a certain number of cases the primary cause resides in non-genetic factors, for example factors inducing brain anomalies at birth.

Each dyslexic individual thus has his/her own cognitive profile and cerebral particularities, which are the result of the specific combination of genetic factors carried and the non-genetic factors to which the individual is exposed.

## Critical analysis of the various theories of dyslexia

There is great diversity in the theories advanced to explain dyslexia: phonological deficiency theory, temporal auditory processing theory, visual theories, cerebellar theory, magnocellular theory, etc. The diversity is due to several factors:

- there are undoubtedly several distinct causes of dyslexia and thus more than one theory could be correct, each theory accounting for a subset of the dyslexic population;
- the clinical presentation of dyslexia is complex and includes numerous symptoms other than reading, particularly phonological, auditory, visual, spatial, motor and other symptoms. Each of the symptoms has given rise to theoretical speculations;
- dyslexia is frequently a concomitant disorder with other developmental disorders (spoken language disorder or dysphasia, dyspraxia, attention disorders, etc.). In the studies of groups, the symptoms of other developmental disorders may thus seem linked to dyslexia giving rise to new theoretical hypotheses.

The wide variety of symptoms associated with dyslexia does not facilitate identification of the real causes and distinction of those causes from simple concomitant disorders. Nonetheless, on the basis of a very large number of studies, a few points have clearly emerged.

The vast majority of dyslexic children suffer from a specific cognitive deficit with regard to the representation and processing of the sounds of speech. This is known as a phonological deficiency. The deficiency interferes with learning, mastery and automation of the use of the grapheme-phoneme matches and, subsequently, with all of the process of learning to read, including the orthographic route. In general, it is therefore not appropriate to distinguish between phonological and surface dyslexia.

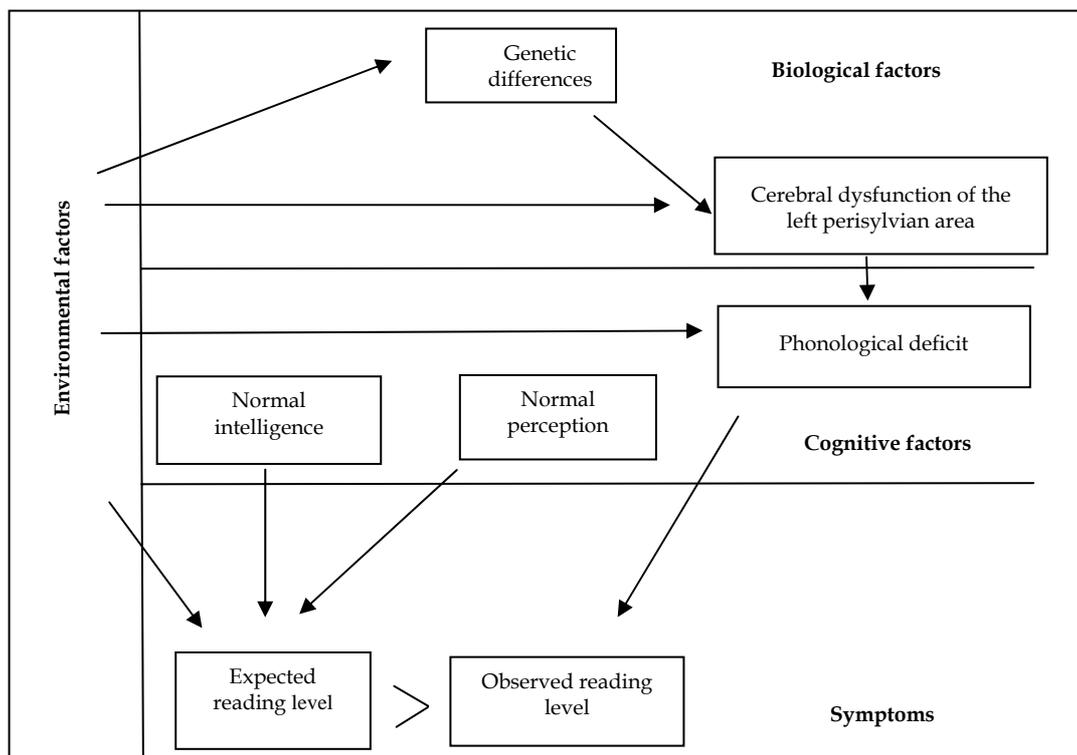
A minority of dyslexic children seem to present with visual disorders and no phonological deficiency. It would even appear that there are several subtypes of visual dyslexia. However, empirical studies in the field are for the time being insufficient and have not generated sufficiently comprehensive theories.

With regard to the neurobiological causes of cognitive deficiencies, two main types of explanation remain possible: those centered on the biological causes of the phonological deficiency that is directly responsible for reading disorders and those attempting to account for the association with sensory-motor disorders. The former postulate a specific impairment of early development of the left perisylvian area involved in language acquisition. The hypothesis was recently strengthened by genetic data. The ultimate source of dyslexia could thus reside in a certain number of susceptibility alleles on multiple genes. The alleles, acting alone or in combination, would increase the risk of interference with neuronal migration in the left perisylvian area.

In contrast, hypotheses such as the cerebellar theory or the temporal processing theory supply a more direct explanation of the associations of concomitant disorders. However, those hypotheses are currently insufficiently supported by empirical data and require new studies, in particular with regard to their ability to reflect the links between sensory deficits and reading disorder.

Moving beyond the discrepancies in the ultimate neurobiological causes, the two types of theory converge with respect to the presence of dysfunction in the development of the cerebral areas normally involved in the representation and processing of the sounds of speech ('phonology'). The dysfunction induces a cognitive deficit whose main manifestations are weak phonological consciousness, weak short-term verbal memory and slow retrieval of

phonological representations. The cognitive deficit in general has little influence on spoken language acquisition (except in the event of very marked severity or additional language disorders). In contrast, the cognitive deficit becomes patent with written language acquisition which recruits, in a particularly intense manner, the phonological skills. Lastly, the symptoms observed with respect to reading are the product of cerebral and cognitive factors and also numerous environmental factors such as the richness of the linguistic environment (which, in particular, influences vocabulary), the regularity of the writing system, the method of teaching reading and, undoubtedly, other environmental factors. The model proposed below reflects the interactions which are liable to occur between other environmental factors (injury, affective deficit, sociological factors, pedagogic factors, etc.), at various levels, and the biological, cognitive or even symptom expression factors. The model clearly shows that an exclusively biological or exclusively environmental (irrespective of its nature) determinism is not meaningful. However, further studies are required in that context. Studies would enable investigation of all the factors in a multi-scale and multi-field model (molecular bases, neuronal networks, cognitive psychology, developmental psychology, education) with a view to elucidating the learning and development mechanisms and their dysfunctions. Such studies would thus further the understanding of the links between the deficits observed at neuronal level by means of anatomical or functional imaging and the dynamic models underlying the interaction between individuals (parents-children, teacher-child). The objective of such studies would be to tend towards unifying the various partial models in a credible common synthesis ultimately enabling researchers and professionals to understand the same 'symptoms' using different and complementary approaches.



**Diagram of the various factors involved in dyslexia**

## Detection, screening and diagnosis

Detection, screening and diagnosis may be distinguished in that the systems, players and instruments differ. Unlike screening and diagnosis, detection may be implemented by persons other than healthcare professionals. The teacher, in liaison with the parents, is in the best position to detect a child who has learning difficulties. At nursery school, the teacher may detect spoken language disorders, risk factors for potential subsequent difficulties in written language acquisition, difficulties in writing or difficulty learning the numerical code. As of primary school, the difficulties in learning to read, spell, do arithmetic and write may be detected. The teacher is able to clearly define the acquisitions and gaps without being able to qualify what he/she has observed. The parents and family physician may also draw attention to the difficulties observed.

Screening is, in principle, a procedure that targets a given population. Since it targets all the children in a given age group, the school is the preferred site for screening professionals who use particular instruments to detect acquisition and learning problems. Article 85 of French act No. 2002-73 dated January 17, 2002, relating to social modernization provides for organization of language disorder screening during a mandatory assessment in the 6th year. Screening is, in principle, conducted by the National Education physicians. However, screening for scholastic learning disabilities can only begin when the children have begun to learn, in other words after the start of primary school class CP (6-7 years). Screening during the obligatory examination at age 6 years is thus most frequently a screening for risk factors for specific learning disabilities. Screening for risk factors (such as spoken language disorders) may also be conducted by the Maternal and Infantile Protection (PMI) physicians during the examination of infants in nursery school classes PS (3-4 years) or MS (4-5 years). Physicians with training in the field may be requested. During schooling, the network for specialized assistance to children in difficulty (Rased), a National Education in-house structure (consisting of a psychologist and specialist teachers) contributes to individual identification and screening.

Diagnosis frequently calls for the skills of various professionals working as a pluridisciplinary team given the complex nature of the disorders and the frequent existence of associated disorders. The open-care professional networks frequently offer that pluridisciplinary approach coordinated by a referring physician. Reference centers have been set up in the teaching hospitals (CHU). There are about 40 reference centers distributed through France. All the centers offer multidisciplinary consultations and, as a minimum, medical, speech therapy and psychological consultation and if necessary psychomotor and neuropsychological consultation for an overall assessment.

The instruments differ depending on whether they are intended to detect, screen or diagnose specific learning disabilities.

Before the start of scholastic learning (before 6 years), the instruments are designed to identify, screen or diagnose spoken language disorders and detect signs predictive of scholastic learning disabilities. After age 6 years, the instruments are designed to identify, screen or diagnose learning disabilities (most frequently written language disorders).

The first category of instruments enables identification of subjects at risk of subsequent reading disorders. Before age 5 years, the instruments (e.g. ERTL4: test to detect language and learning disabilities) are able to detect spoken language disorders. For children aged 5 to 6 years, the instruments (e.g. BSEDS: health assessment to evaluate development for school children aged 5-6 years) investigate spoken language disorders and identify the risk factors for dyslexia. The subjects at risk thus detected will not necessarily become dyslexic.

Management of the spoken language disorders (depending on severity) is in itself a means of preventing dyslexia since management acts on the risk factor.

Instruments for screening for written language acquisition disorders that can be collectively administered like Timé 2 from January in primary school class CP (6-7 years) to the end of CE1 (7-8 years), or individually administered like Odedys, calibrated on CE1 (7-8 years), or Batelem-R as of CP (6-7 years), or the BREV learning items instrument from CP (6-7 years) to CE2 (8-9 years).

A second type of instruments enables first-line neuropsychological clinical examination (e.g.: BREV, quick battery for the evaluation of cognitive functions). The instruments are of value in determining the reality of a disorder and its profile and seriousness, such as, for example, a specific spoken language disorder at age 5 years or a written language disorder at 7 1/2 years or writing disorder after 5 years. The instruments are thus used to define the complementary assessments necessary to confirm the diagnosis and the pedagogic and care operations that are necessary. They also enable the professional to assess the course of the disorder. The clinical examination is also designed to rule out a sensory disorder or other patent neurological or psychiatric impairment.

Lastly, the third type of instrument is designed to confirm a diagnosis based on the preceding instruments. The third type of instrument includes: N-EEL (new tests for the evaluation of language) and Elola 5 (battery for the evaluation of spoken language in aphasic children) for spoken language; Belec (battery for the evaluation of written language), Odedys or Evalec (battery for the diagnostic evaluation of dyslexia); and various neuropsychological batteries for attentional functions. The Weschler composite intelligence battery enables definition of the profile of the intellectual functions.

The third type of instrument, which is very specialized, is used by the professionals involved: psychological evaluation of behavioral, affective and cognitive functions by a psychologist; evaluation of spoken and written language by a speech therapist; evaluation of graphic and praxis functions by a psychomotrician or ergotherapist or neuropsychologist; evaluation of attentional and memory functions by a neuropsychologist, etc. For each of the specific learning disabilities, diagnostic instruments enable elucidation of the child's disorder by reference to recognized neuropsychological models. For example, for written language disorders, the instruments are not only to determine reading age but also the strategies used by the child as a function of the precision and rapidity of identification of regular and irregular words and pseudo-words and the underlying cognitive skills and comprehension.

Use of the various tools refers to key ages:

- as of age 3 years for the detection of a spoken language disorder that may lead to diagnosis of a secondary disorder (deafness, behavioral or communicational disorder, or even intellectual deficit) requiring specific management and not simply management the spoken language deficiency. This prerequisite is indispensable for the diagnosis of specific disorders. Up to the age of 4 1/2 years, only specific and severe disorders (one or several seriousness criteria: unintelligibility, agrammatism or comprehension deficit) necessitate detailed speech therapist evaluation and rehabilitation as soon as the child cooperates. Specific disorders without seriousness criteria are followed up with pedagogic adaptation and parental assistance;
- at age 5 years for screening for a writing disorder and/or dyspraxia requiring complementary psychological evaluation to confirm the specific nature of the disorder and, if the disorder is specific, psychomotor or ergotherapy assessment to determine the profile and seriousness and the care indicated;

- as of age 5-6 years for screening for numerical code acquisition difficulties (name and construction of numbers, numerical chant) and, in primary school years CP-CE1 (6-8 years) to screen for difficulties in accessing numerical facts (addition then multiplication tables) and number trans-coding. Screening may result in a psychological evaluation to confirm the specific nature of the disorder and a precise evaluation of the arithmetic difficulties;
- as of primary school class CP (6-7 years), to screen for disorders of written language acquisition. If those disorders are associated with a persistent disorder of spoken language, they require speech therapist evaluation associated or not associated with psychological evaluation;
- as of the second part of primary school class CP (6-7 years), to screen for severe disorders (non-acquisition of the process of decoding, no improvement after appropriate training, etc.) also requiring complementary evaluation of the cognitive capabilities, including those with respect to language.

The diversity of the scenarios depending on the key ages and symptoms determines the importance of the first-line screening examination.

Evaluations are indispensable in order to assess the time course of the disorder six months to one year after initiation of the educational and/or care project. The follow-up evaluations include, as a minimum, the tests whose scores were deficient in the initial evaluation in order to conduct objective quantitative and qualitative comparisons and reorient the educational and care program.

An expert commission was set up (decree dated February 8, 2002) in order to compile, at national level, recommendations on the instruments for the use of professionals in the context of the Action plan for children presenting with a specific language disability. The commission's report addresses the instruments cited above. The instruments have just been made available to professionals in the medical and childhood sectors and are available from the Ministry of Health and Solidarity website<sup>8</sup>. Since the findings were made public at the time of finalization of the work of this collective expert review, it was too late to incorporate the analysis in the report.

## **Prevention in the school environment**

Prevention in the school environment could target three populations of children: children identified as being at risk of presenting difficulties in learning to read in the GS class of nursery school (5-6 years); children experiencing great difficulty in reading in the CP class (6-7 years) of primary school; dyslexic children requiring special-needs teaching to promote learning in the preserved fields.

Numerous foreign studies have evaluated the effect of training with the objective of obtaining a pedagogic first-line response in school. Training targeted children at risk of difficulties in acquiring written language (from families at genetic risk) or, more frequently, children failing to learn to read. The prevention was not targeted on dyslexic children since it was implemented prior to any diagnosis.

The scientific bases for the type of training were the knowledge generated by basic research on learning to read. All the studies targeted English-speaking populations, i.e. populations learning to read in a language even less transparent than French.

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<sup>8</sup> [www.sante.gouv.fr](http://www.sante.gouv.fr)

The issues involved in those studies are essential: a pedagogic response in class can be implemented for all children without discrimination; the response constitutes a preventive action and does not induce health cost in contrast to a response consisting in care. It is thus indispensable to take the results of those studies into account: What is the most pertinent training? What children are involved? What is the quality and intensity of the effects? Nonetheless, the particularity of the French language and educational system will make French studies necessary.

The value of training is related to the need to prevent children who read poorly falling behind their peers. The child must therefore not be left in a vicious circle. It is necessary to act as quickly as possible to help poor decoders or children likely to become poor decoders. The questions are:

- which children need phonological training?
- should training address specific decoding deficiencies?
- are the effects generalized to understanding?

With regard to the training conditions (group size, type of training, duration, mode - individual or small group -) the tests and studies designed to determine the effectiveness of the various modalities have yielded very variable results.

The results of the main studies show that the effects of training in 'phonological consciousness' associating auditory and visual modalities has a marked statistical impact on development and a more moderate but statistically significant effect on reading and writing. Not only word identification but also understanding are enhanced. A more or less marked benefit with respect to reading was observed under all training conditions. The effects were positive for children learning to read normally, for children at risk of difficulties, and for those who had difficulties in nursery school or primary school class CP (6-7 years). Training also developed the writing skills of children in difficulty.

Thus, teaching phonological consciousness associated with the assembly route using intensive, specific and explicit training of small groups with similar needs promotes reading and writing in children experiencing decoding difficulties.

In France, preventive actions in the school environment were not inventoried in the context of this expert review and very few of those operations have given rise to studies published in the scientific literature.

A French training study using auditory and visual discrimination software was conducted on 'poor decoder' children in nursery school class GS (5-6 years). The training lasted 10 hours over 5 weeks at a frequency of 2 times 15 minutes daily, 4 days per week. It enabled the trained children to become better on average than the control children who decoded better at the outset. Another French controlled study addressed the effects of in-school training of 80 poor-decoder children in primary school class CE1 (7-8 years). Specific software was used for small groups. The study showed that trained children progressed more rapidly with respect to decoding than the untrained control group of poor decoders.

The software requires new rigorous evaluation in order to determine its specific effects and limits and the conditions for generalization of software use in the event that its beneficial effects are confirmed. The type of tool does not seem to replace a real teacher but to act as a complement.

However, the studies show that training in decoding does not improve or only improves a little the reading speed. The latter requires other types of training. According to the literature, the training recognized to be most effective with respect to fluidity (and hence speed of reading) is the technique of repeated reading: the letters, words and sentences read

are repeated until a certain speed is achieved. Several authors have described the effects of such training on text reading speed (training of duration 6 minutes daily for 6 to 9 months).

In terms of prevention, it must be stressed that teaching adaptation is absolutely indispensable. The adaptations are designed to enable children to overcome their handicaps. The adaptations consist in taking into account the difficulties encountered by the child due to his/her disorder and enabling the child to learn in his/her preserved fields without being handicapped by his/her disorder (e.g. read the problems or give extra time for mathematics in the event of dyslexia, decrease the writing load in the event of dysgraphia, etc.). Descriptions of the adaptations are available in several forms (awareness-promotion CD-Rom, guides issued by the academies of Grenoble and Rennes for the use of teachers, dyslexic child follow-up book). In France, reflection on the nature of the adaptations and on the criteria required in order for children to benefit with a view to harmonizing the measures is indispensable.

## **Assessment of rehabilitation methods**

There are a great variety of rehabilitation and training methods and some of them directly result from the various theories advanced to explain dyslexia. However, a large number of the recommended treatments for dyslexia have not undergone scientific study either from a theoretical viewpoint or from the viewpoint of evaluating the efficacy of treatment. The scientific criteria which enable treatment efficacy evaluation are not always totally fulfilled. The criteria include: studies of groups based on objective and quantified observations and supported by rigorous statistics (individual case reports do not constitute an objective evaluation); constitution of a control group which receives a placebo treatment of similar duration and intensity to that received by the test group; conventional procedure for a randomized, double-blind, controlled clinical trial; several independent studies conducted on a large number of subjects.

Most of the rehabilitation currently implemented is of the speech therapy type. Rehabilitation addresses: training the child with respect to phonological capability; reading rehabilitation with methods that are frequently different from those possible in class; setup of compensation strategies to enable the child to avoid the identified deficits.

While speech therapy rehabilitation as practiced in France is based on general principles derived from scientific knowledge acquired and validated in recent years, it has yet to undergo scientific assessment in the treatment of dyslexia. However, the fact that speech therapy is implemented individually with a specifically trained speech therapist enables development of an individualized rehabilitation program based on a precise assessment of the child's skills and weaknesses. The program can be implemented under optimum conditions of interactivity enabling real-time adaptation to the child's needs. That essential feature of speech therapist management can only, a priori, enhance efficacy compared to training programs administered to classes or groups as evaluated in the scientific literature. The international literature on training programs (computerized or not) applied to 'poor reader' children show that a degree of intensity is required in order to achieve a reasonable effectiveness (typically 4 to 5 sessions weekly). Given that intensity, good results can be obtained over relatively short rehabilitation durations (of the order of 6 weeks). Nonetheless, there is great variety in speech therapists' practices. Those practices should therefore be evaluated and compared taking into account the modalities of their implementation (of which, intensity, duration and use or non-use of computer tools) in order to further identify good practices and in order to enable orientation of speech therapy practice. A computer program in French involving audiovisual training in identifying spoken and written

syllables had a degree of efficacy. However, studies based on the criteria necessary for rigorous evaluation will be required in order to consider whether such training is of value as a complementary technique to speech therapy rehabilitation.

Among the various theories advanced to explain dyslexia, the theory in which the dyslexic (and dysphasic) subjects suffer from a deficit in temporal auditory processing has naturally given rise to training designed to rehabilitate auditory perception. This is the case of the US computer program Fast ForWord which includes training in discrimination of temporal auditory sequences and training in phonological skills. The special feature of the program is that it uses speech modified to render it more intelligible for children who have a temporal auditory processing deficit (short sounds and fast transitions amplified and prolonged in an adaptational manner). Nonetheless, the independent assessments of the program generated contradictory results and its efficacy on spoken and written language has yet to be clearly demonstrated.

The Tomatis method of auditory stimulation is based on a concept of audition that has no scientific basis. A meta-analysis of all the studies conducted did not show that the method had a positive effect.

The semiophonic method (or Lexiphone method) is an intensive rehabilitation method whose basic principle is auditory stimulation ('parametric sound') which is not based on any known scientific principle. The rehabilitation incorporates structured exposure to speech (syllables, words and pseudo-words), music and read texts, guided reading and writing. A clinical trial is underway in France.

Several rehabilitation methods for dyslexia address the visual capabilities and visual and attentional capabilities. Occlusion of one eye of children who were both dyslexic and had binocular instability was tested in the course of randomized, double-blind, controlled trials, which showed a significant improvement.

It has been suggested that wearing tinted glasses or lenses may improve the reading of dyslexic subjects with visual symptoms. A clinical trial showed modest beneficial effects on reading of the use of a transparency whose color was individually optimized for subjects suffering from visual stress.

A method known as 'specific hemispherical stimulation' consists in specifically stimulating the brain hemisphere which is presumed deficient in a given child by briefly (tachistoscopically) and repeatedly presenting words in the contralateral visual field. Experimental data suggest that the effects observed are non-specific (e.g. attentional).

The motor and proprioceptive systems have also been the subject of rehabilitation (motoricity, balance, archaic reflexes, proprioception). The data currently available are not sufficient to assess the efficacy of those methods in the treatment of reading disorders.

Several drugs and food supplements were not effective in clinical trials (methylphenidate, antihistamines, piracetam, polyunsaturated fatty acids).

No psychotherapy for dyslexia has been reported in the scientific literature. When a child presents with anxiety or depression or various behavioral disorders, in certain cases a difficulty in learning to read may result. But those disorders are of a very different nature to those encountered in the context of the definition of dyslexia. Nonetheless, many dyslexic children suffer from their failures at school and this may secondarily induce anxiety, depression or behavioral disorders. When that is the case, the disorders are to be diagnosed and appropriately managed in addition to the specific management of the learning disorder.

## Individual management in clinical practice

Care consists in individual management of the child determined by precise evaluation of the deficits in written language, arithmetic and writing and the associated disorders (e.g. spoken language, affective or attentional disorders). In France, management is conducted by specialized professionals depending on the diagnosis (speech therapists, neuropsychologists, psychologists, psychomotricians, ergotherapists, etc.) and in the context of pluri-disciplinary devices and networks for the management of specific learning disabilities (reference centers, Rased, etc.). The objective of this expert review is not to analyze the practices in France, which is the responsibility of other organizations in the context of their missions, but to present a few principles for action based on analysis of the literature on the care field.

Irrespective of the care prescribed, it must also be associated with pedagogic management in order to enable the child to continue learning in his/her areas of skill, despite his/her disorder and in order to offer teaching adapted to the child's requirements and possibilities in the deficient area.

Few scientific studies have reported objective data on the indications for speech therapy rehabilitation in the context of a written language disorder (At what age? For what disorder severity?), or on the orientations and instruments to be used or the frequency or duration.

The Anaes recommendations (1997) on speech therapy for specific written language disability essentially consist in recommendations based on professional agreements (with no scientific evidence). However, speech therapy rehabilitation is widely practiced and is of major value in enabling the child to alleviate his/her deficiency. The published data on the development of written language and the predictive factors, the increasing number of studies to evaluate specific training and the open studies on the benefits of intensive management of severely affected children will enable definition of at least some indications, orientations and practical conditions.

In the event of a specific disorder of written language acquisition, individual speech therapy rehabilitation is recommended as of primary school class CP (6-7 years) if a spoken language disorder persists or as of the end of CP if the initial adapted teaching response proves insufficient or in the event of signs of seriousness such as the absence of grapheme-phoneme or syllable matching (/b/a/ → /ba/), particularly if there is a familial history of language disorder or a personal history of delayed spoken language acquisition. Implementing reading and writing rehabilitation concomitantly seems preferable. The orientations are determined by the precise results of the individual evaluation of the deficient and preserved strategies, the cognitive functions underlying spoken language, phonological skills and visual processing, and quantitative and qualitative evaluation using standardized tests. Decoding and encoding by assembly and phonological skills constitute the first phase targeted in rehabilitation, if they are not sufficiently efficient and automatic. This is usual in most cases of dyslexia.

Visual, kinesthetic and semantic aids may be used to offset the auditory perceptual deficit. The orthographic stock is generally developed by differentiating the situations with no visual disorder and those with a visual processing deficit. In phonological dyslexia with no visual processing deficit, the decoding difficulties slow the constitution of the orthographic lexicon which is to be developed. In cases in which visual processing is disturbed, it is also rehabilitated to enable the child to enrich his/her orthographic lexicon. The use of that lexicon, once it has been developed, is to be promoted through writing then reading by working on the words in isolation or in context. The use of morphology is promising for dyslexic subjects with a view to helping the child develop orthographic knowledge independently of phonological skills. Lastly, fluid reading is developed by repeated reading

and, above all, comprehension skills are worked on in order to bring the written comprehension level at least up to the spoken-language comprehension level. The project generally defines a limited number of axes for a given time period. The most urgent as a function of the child's age and type of disorder take prevalence. For instance, the most urgent may be grapheme-phoneme matching and sound discrimination. The axes are reviewed by means of regular fractional quantitative and qualitative evaluations of the target deficient function.

Studies of the various types of training show that intensive training programs (half an hour daily, 4 days per week) over relatively short durations (5 to 10 weeks) procure specific benefits with respect to the trained function providing that they are precisely and specifically targeted on a deficient cognitive function. Those results may provide the grounds for reviewing current care practices. Intensive, specific, short-duration programs may be proposed for children recalcitrant to adapted pedagogic management and harmonized with the teaching which remains indispensable. Nonetheless, training on a precise function does not constitute the whole of speech therapy rehabilitation. The objective of the latter is to achieve functional reading and legible writing. The overall effects of rehabilitation need to be evaluated every 6 months to one year by using a new assessment employing similar tests to the initial assessment and enabling quantitative and qualitative assessment of the progress achieved and hence pursuit of the rehabilitation project. Depending on the time course of the child's skills, the decision may be to discontinue rehabilitation (in the event of score normalization or score stabilization with functional reading and legible writing), or continuation of rehabilitation at a rate to be determined as a function of the objectives to be achieved (in the event of improvement without normalization or perfectly functional reading or legible writing), or pluri-disciplinary evaluation, for example by a reference center with a view to detecting associated disorders in the event of an inadequate course.

After the end of rehabilitation, adaptation in the school environment remains indispensable and is to be based on the handicap in terms of reading speed and dysorthography. A computer may be of value: word processing (in the event of associated writing disorders), spell check, vocal dictation (in the event of marked sequelae) in order to offer the child text reading by the computer and enhance the legibility of the child's written production.

Writing disorders need to be precisely analyzed in order to ensure an appropriate response from the most pertinent professional (psychomotrician or ergotherapist). The aim is to determine, on the basis of the quantitative and qualitative results of specific tests, whether the disorder affects gesture coordination, perception and/or visual or visual and spatial production. Management may begin as of the end of nursery school, at the start of primary school if the disorders are severe and before age 8 years in the event of persistent disorders, before deviant strategies of letter sequencing become established. Like speech therapy rehabilitation, the management of handwriting is to be associated with appropriate teaching responses and is to be evaluated by progress assessment using standardized tests whose results will be compared with those of the initial assessment. Orthoptic management of a predominantly visual perception disorder may be indicated and the effects are to be evaluated. Learning word-processing or even vocal dictation depends on the comparative assessment determining the degree of persistent handicap given the school project.

The management of mathematical disorders has been very little studied. The instruments and trained professionals are only available in inadequate numbers. Nonetheless, disorders in the acquisition of the numerical code may be managed from the start of primary school, particularly since concomitant written language acquisition disorders are frequent, exacerbating the child's failure.

The disorders associated with written language acquisition disorders and affecting the spoken language require rehabilitation since the links between the spoken and written language are very close. With a view to preventing dyslexia, any specific spoken language disorder must be taken into account. Rehabilitation has two objectives: improving speech and language but also preparing for written language (work on assembly and phonological consciousness).

The associated behavioral and affective disorders require an appropriate care project. The assessment of learning in any child consulting for attention deficit/hyperactivity disorder and the evaluation of behavior and attention in any child consulting for a written language acquisition disorder will enable management of the two disorders if they are associated. Anxiety and mood disorders are to receive psychotherapeutic care. Psychotherapies addressing the child's psychological development are perfectly compatible with cognitive rehabilitation programs. The complementarity of the managements, providing that one does not exclude the other, addressing the whole child and his/her diversity at both cognitive and environmental relational level can in practice be implemented in the field with benefit for the child both with regard to the child's adaptation to his/her difficulties and his/her compliance with rehabilitation.

Care coordination and family counseling are indispensable and are the responsibility of the child's physician, trained referral professional and the various other professionals involved. Coordination is essential in order to define the objectives of the management programs, taking into account their feasibility and priority for the child, and also in order to assess the child's progress and readjust the objectives in consequence.

The harmonization of the therapeutic project and teaching project constitutes another fundamental aspect of management which can be based on the specialized teachers in the help and support networks and National Education physicians and psychologists. The choice of scholastic adaptation as a function of the child's course must enable the child to continue learning while minimizing the affective consequences of failure.

Overall, the variety of professional practices implemented in the context of care and the need for health-education coordination call for an analysis, under the responsibility of the High authority for health, with a view to compiling good practice recommendations.

## Principal findings

- The disorders addressed in this expert review are dyslexia, dysorthography and dyscalculia. The specific spoken language disorder known as 'dysphasia' and the motor coordination disorder known as 'dyspraxia' are not addressed in this expert review but may nonetheless interfere with learning in school.
- In accordance with the Inserm collective expert review procedure (appendix 1), the experts analyzed about 2,600 publications and referenced 1,500 articles in the course of the expert review. Of those articles, 40% had been published since the year 2000. The analyses implemented by the expert group fulfilled the specifications defined with the expert review sponsor but should not be considered to supply answers to all the questions in the field considered. The subjects that the experts were unable to address in the context of this expert review are not to be considered of less importance and certain of those subjects would indeed benefit from individualized expert review since the literature is abundant. For other subjects, publications are lacking and research needs to be intensified.
- The label 'specific' is applied to disorders whose etiology is known to be neurodevelopmental. The disorders are listed in the International classification of diseases (ICD-10) under headings F81.0 for specific reading disorder (dyslexia), F81.2 for specific mathematic disorder (dyscalculia) and F81.8 for the specific written expression disorder (dysorthography). Even though the criteria defining the specific disorders in that classification are unsatisfactory for researchers and clinicians, it is the only international standard available. When the criteria are applied, they have the value of constituting a basis for comparison of various studies. It is to be noted that a classification of disorders is not a classification of subjects. It is therefore appropriate to say 'a child presenting with dyslexia' rather than a 'dyslexic'.
- In order to elucidate the specific disorders of scholastic learning, the expert group considered it necessary to first review the chronology of spoken language acquisition and the mechanisms governing learning reading, writing and arithmetic in children. The review stresses the importance attributed to sharing the progress in knowledge of the functions exploited in learning to read, write and do arithmetic with teachers in the form of readily accessible instruments. It is also important that cooperation between teachers and researchers is pursued.
- The acquisition of speech and language between age 0 and 3 years has a strong influence on the development of scholastic learning. A specific spoken language disorder must therefore be taken into account before age 5 years and, if possible, before age 3 years.
- Comprehension motivates learning to read. In order to be able to read in an alphabetical writing system, a child must be able to master the matches between graphemes (letters or groups of letters) and phonemes (speech sounds). In order to learn those matches, repeated training on grapheme-phoneme correspondances is indispensable. In addition, other activities may be proposed to stimulate the motivation to read.
- No study of the overall French population with respect to the prevalence of dyslexia is available. Such a study needs to be set up. Various studies have estimated the prevalence of dyslexia (moderate to severe) to be a little less than 5% from the primary

school grade CP (prevalences are usually determined for children aged 10 years in the international studies). Certain authors have reported that dyslexia accounts for about a quarter of the cases of reading difficulty in children. The data generated by studies on dyslexia therefore cannot be generalized to all children experiencing difficulty in learning to read.

- Dyslexia becomes patent in a child when he/she has begun learn to read in primary school grade CP (6-7 years). Dyslexia presents as a lack of mastery of grapheme-phoneme correspondances. The distinction between below-average readers and dyslexic children cannot be clearly established at that stage. However, factors (probable but not certain) may argue in favor of dyslexia: persistence of a spoken language disorder; family members presenting with dyslexia.
- Dysorthography has to date mainly be studied in children presenting with dyslexia. Does dysorthography not related to a specific reading disorder exist? The literature does not enable a response to that question. Studies of the cognitive mechanisms and determinants of isolated dysorthography should therefore be promoted. In that type of study, writing and reading performances are to be evaluated concomitantly.
- It would appear that dyscalculia is less frequent than dyslexia but prevalence data are lacking. Children presenting with dyscalculia have a poor understanding of the principles governing the activities of counting which constitute the basis on which all subsequent arithmetical skills are built. The children also have atypical difficulties in memorization and learning addition and multiplication tables.
- Dyslexia, dysorthography and dyscalculia are persistent disorders which may be encountered in junior and high school pupils despite prior rehabilitation. The disorders constitute a handicap. It is important for teachers to be informed and trained in order to promote setup of the indispensable adaptations enabling maintenance of affected pupils in the ordinary school environment.
- Combination of the three disorders is not rare. This finding has consequences in terms of the teacher's pedagogic actions, the clinician's management and the researcher's study orientations.
- Dyslexia, dysorthography and dyscalculia, together or in isolation, may also be associated with coordination disorders (dyspraxia) or handwriting disorders (connected or unconnected to the dyspraxia) or attention deficit/hyperactivity disorder. In over half of the cases, dyslexia is subsequent to a specific spoken language disorder known as 'dysphasia'. This finding has led researchers to investigate for common underlying mechanisms involving, in a variable manner, the main sensory-motor systems.
- Dyslexia, dysorthography and dyscalculia may also be associated with affective disorders (anxiety, depression) and behavioral disorders secondary to the scholastic difficulties encountered or existing as a veritable concomitant disease. The second possibility leaves open the question of the plurality and inter-relationship of the determinisms. In practical terms, for the expert group, each disorder is to be tackled specifically and the child managed overall. The analysis of the psychological development of the child and the child's interactions with his/her environment constitute natural parts of that management which combines a pedagogic (at school) and care (rehabilitation and psychotherapy) approach.
- The research on disorder explanatory mechanisms has mainly focused on dyslexia. The presence of a dysfunction in the development of the brain areas normally involved in speech sound representation and processing (phonology) is the accepted hypothesis for dyslexia. However, since the year 2000, numerous publications have suggested other

hypotheses to explain the associations between dyslexia and other developmental disorders. With regard to developmental disorders of the pervasive development disorder (PDD) type, the current literature does not enable a conclusion to be formulated with respect to whether the mechanisms underlying learning disabilities associated with PDD are of the same nature as those underlying specific learning disabilities.

- The hypothesis that dyslexia has a familial component is not new. International studies of twins have shown that when a monozygotic twin has dyslexia, the probability of the other twin having it is 70%. Research on the genes that may be involved is recent but has generated concordant results: the genes listed are involved in migration (in the early stages of brain development) of the neurons situated in the brain areas recruited much later in learning to read. Have the 'genes for dyslexia' been discovered? At most, alleles which increase the risk of developing dyslexia as a function of interactions with numerous other factors (biochemical, traumatic, linguistic, socio-educational, psychological, etc.) have been discovered. This does not mean that the primary etiology is always genetic. Other factors may be involved in isolation or together. An exclusively biological determinism paradigm is thus unjustified.
- By definition, dyslexia can only be diagnosed when the disorder has as its primary cause an overall retardation, sensory handicap, educational deficit, language difficulty or patent mental disorders. This does not mean that affective, familial, socioeconomic and cultural factors play no role in the emergence and severity of dyslexia. In particular, it has been established that dyslexia expression by a child results from cerebral and cognitive dysfunction (which may have multiple causes) and the influence of numerous environmental factors, including the linguistic environment, the degree of regularity of the writing system, the teaching methods used and many other factors. It is indispensable for studies to be conducted to determine the role of the interactions between those factors in the emergence of dyslexia.
- Children with learning difficulties in school are detected in class by the teachers and by the parents. But the nature of the difficulties (retardation or specific disorder) has to be evaluated. Teachers (particularly those specializing in children with difficulties) want information and training with respect to the instruments they can use.
- The systematic screening during the obligatory examination in the child's 6th year of life (article L.541-1 of the Education code), conducted by National Education physicians and nurses, cannot address dyslexia since learning to read has not yet started. However, the examination can detect children presenting with a specific spoken language disorder (possibly already evidenced by the examination at age 4 years). A specific spoken language disorder diagnosed in nursery school is a risk factor for dyslexia in 50% of cases. Screening instruments have been compiled and several are used in the context of systematic screening<sup>9</sup>.
- When a learning difficulty has been reported by a teacher, the child may undergo first-line individual screening. The individual screening may be conducted in school by the National Education physicians or specialized psychologists and teachers in the network for assistance to pupils in difficulty (RASED), if those people have been trained to use the instruments. The examination contributes to defining the complementary investigations necessary in order to formulate a diagnosis.
- Diagnosis is conducted using several specific instruments which consist in test batteries. The test battery may require the skills of several professionals forming a pluri-

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<sup>9</sup> An expert commission has conducted a considerable study of the subject. The report is available from the Ministry of Health's website: [www.gouv.sante.fr](http://www.gouv.sante.fr)

disciplinary team. For that reason, some forty reference centers have been set up in the teaching hospitals (CHU).

- After the diagnosis of dyslexia, dysorthography or dyscalculia, the precise evaluation of the deficient functions and investigation for associated disorders, individualized management is generally proposed. The latter consists in: remediation conducted by specialized professionals (several times per week) sometimes in the context of pluri-disciplinary networks and precisely targeting the deficit functions; pedagogic management to enable the child to pursue learning in the preserved fields. The effects of management are to be regularly assessed and management is to be reoriented if required. The associated disorders are not to be overlooked. Each requires specific work but management always addresses the whole child. This raises the question of coordinating the various parties.
- It is difficult to formulate an opinion on the methods of rehabilitation and training currently proposed for dyslexia since most have not undergone scientific validation studies. The most frequent methods are of the speech therapy type and most frequently address training the child's phonological capabilities. Other rehabilitation measures are designed to enable the child to develop compensatory strategies to circumvent the handicap. The fact that the rehabilitation program is based on a precise assessment of the child's skills and weaknesses and is implemented interactively with a trained professional argues in favor of its efficacy. Nonetheless, rigorous studies on the many methods that are currently being developed and which do not always have a theoretical basis are to be recommended.
- Prevention is a little explored field. Recent rigorous studies on English-speaking children have evaluated the effects of pedagogic training as a first-line response in school for children at risk of dyslexia (familial history of dyslexia) and children who experience difficulty learning to read. Prevention is implemented before any diagnosis. The results of the research show that positive effects are obtained with short-duration training sessions, repeated daily, with small groups with similar needs. Early intervention (as of the first sign of reading difficulty) is most effective. For children not showing improvement, complementary individual management is then to be implemented on the basis of a diagnosis. It would appear judicious to promote empirical studies in France to test the advantages of a preventive strategy.
- Prevention must also address the teaching arrangements and adaptations in order to enable children presenting with dyslexia, dysorthography or dyscalculia to continue to be taught in all subjects and throughout school in an ordinary environment without suffering any consequences of their handicap. In particular, the decree relating to the provisions for school, secondary school and higher education examinations and competitive examinations should be applied to pupils presenting with severe specific learning disabilities who, as candidates in a competitive examination, are handicapped. Implementation of the decree, very uneven in France, requires correct information and preparation of the children's families.

## Recommendations

The present scientific literature review and synthesis conducted by the expert group convened under the auspices of Inserm was designed to take stock of the progress with respect to specific scholastic learning disabilities which affect about one quarter of children with learning difficulties.

Following the analysis and synthesis, the expert group formulated a number of essential findings.

In the scientific community, there is currently an almost unanimous consensus on the nature of the specific disorders which prevent children suffering from them from learning, particularly learning to read. It is now recognized, in particular, that the disorders essentially consist in deficits (probably very early in onset and for some of them with a genetic component) in certain language processes (particularly phonological processing), which give rise to the specific learning disabilities for reading.

The most widespread circulation of the scientific advances possible, particularly to all the medical, paramedical and National Education professionals who are responsible for children presenting with specific learning disabilities is important in order to ensure both the earliest screening possible for children at risk and the setup, without delay, of measures designed to reduce the deficiency and minimize the consequences on the children's scholastic performances.

The implementation of measures needs to be graduated as a function of the severity of the disorders but must obligatorily include an evaluation stage using validated instruments calibrated for the child's age. Adaptation of teaching methods to the types of difficulties experienced by each child must be systematic.

Only fragmentary data on the conditions for efficacy of a number of rehabilitation and specific training methods applied in the event of disturbed cognitive functions are available. A large number of the methods have still to undergo validation studies. The great variety of the methods calls for great vigilance on behalf of those prescribing them and using them. The coordination between the various partners (in school and outside of school) is indispensable and should enable an individualized approach to each case.

Fields of learning other than reading may also be subject to specific disorders that, just as much as reading disorders, require a scientific approach to the study of their mechanisms and management appropriate to each individual case. The coexistence of those disorders with the reading disorder increases the severity of the clinical presentation and calls for recourse to a pluri-disciplinary team (reference center) possibly structured to form regional networks. The access of the greatest number to all the competent professionals is in fact an objective to be achieved.

Affective disorders are frequent in children presenting with specific learning disabilities. Their presence may raise questions with respect to management priorities. The disorders may appear to be the consequence of a specific learning disorder or a concomitant disorder that will exacerbate the manifestations and, in that case, provide the grounds for not neglecting specific rehabilitation with respect to the learning disabilities.

Among the numerous fields still requiring expert investigation, the frequency of the various types of specific disorders and their combinations on the population scale remains a priority

if only to more accurately measure the impact of the specific disorders on learning in terms of public health.

The National plan of action for children presenting with a specific language disorder defined in 2001<sup>10</sup> formulated five priority approaches that in part overlap those presented below. The expert group's proposals are to be situated in a scientific approach shedding light, on the basis of recent data, on certain aspects of the operations to be deployed, already deployed or to be evaluated. When the proposals for action address a new field such as that suggested for prevention, the actions are to be tested, evaluated and compared with other approaches before large-scale deployment. For the recommendations already implemented at national level, the objective of the present collective expert review was not to evaluate their application.

In the course of the expert review process, meetings with patient and parent associations, and educational, medical and paramedical (speech therapists, neuropsychologists, psychologists, etc.) professionals showed a commitment to sharing and ensuring the convergence of knowledge, experience and know-how, if possible in the form of institutionalized exchanges.

## **Enhancing detection, screening and prevention**

The knowledge acquired on learning disabilities should be made available to the public, particularly the parents and professionals concerned. The knowledge will contribute to facilitating detection of children experiencing learning difficulties.

In school, teachers are the first and best placed people to identify children presenting with learning difficulties. An understanding of the nature of specific learning disabilities and their presentations should be incorporated in initial teacher training as should new knowledge with respect to educational practices.

### **INFORMING THE PROFESSIONALS AND PUBLIC ABOUT SPECIFIC LEARNING DISABILITIES AND THEIR MANAGEMENT**

By definition, a specific learning disability cannot be attributed to intellectual retardation, a sensory handicap or a patent psychiatric disorder. The disorders are thus unexpected given the other aspects of development. Most frequently, they persist into adulthood. For instance, dyslexia is a lasting and persistent disorder in the acquisition of reading that may be patent even in intelligent pupils with good visual and auditory perception and for whom no socioeconomic, medical or educational factor can explain the difficulties experienced. Parents and the general public should be made aware of specific learning disabilities in order to prevent the worry and inaccurate diagnosis. The information given should enable them to understand the process of scholastic acquisition (reading, writing, arithmetic) and to be more alert to the first signs of difficulties.

A number of early signs of specific learning disabilities are now known. Although investigation for some of those signs may be conducted for before the child begins to read, no sign that can be evidenced in nursery school is a sure indicator of a future specific deficit in learning to read, write or do arithmetic. The accumulation and persistence of the various indices are to be taken into account and will enable a diagnosis to be formulated. Thus, the

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<sup>10</sup> [www.sante.gouv.fr](http://www.sante.gouv.fr)

absence of a disorder before primary school does not mean that the child will not experience learning difficulties.

#### **TRAINING TEACHERS TO BETTER UNDERSTAND AND RECOGNIZE SPECIFIC LEARNING DISABILITIES**

Training in the chronology of acquisition would afford teachers the ability to pay special attention to children who, in kindergarten, present with risk factors for specific disorders in learning to read (perceptual confusion between related sounds, deformation of words, difficulty repeating rhymes, difficulty remembering invented words, not knowing the names of letters, etc.) and arithmetic (retarded acquisition of numbers and counting).

Early deficits in phoneme segmentation and discrimination (elementary sounds of the spoken language) are among the most reliable indicators for future reading difficulties as are deficient phonological short-term memory (evaluated, for instance, by repetition of invented words) and deficient letter recognition. After the start of primary school, the main manifestation of difficulties learning to read is the absence of mastery of the correspondances between letters or groups of letters (graphemes) and sounds (phonemes).

Training in the functions involved in learning to read (phoneme segmentation and discrimination, phonological short-term memory, letter recognition, etc.) would enable teachers to detect, at the start of or during primary school, children presenting with difficulties and thus enable rapid setup of in-class training.

The various numerical activities beginning in kindergarten enable children to master the procedures of counting by pointing and counting on the fingers. Children generally acquire those abilities before the end of kindergarten. With primary school (CP, 6-7 years) comes the systematic teaching of a new code, the Indo-Arabic code, and the algorithms associated with it, yielding a power for operation resolution that the verbal code cannot impart. The transition from the spoken code to the Indo-Arabic code or vice versa is initially based on verbal knowledge. This explains why, for instance, in French, the transcription of quantities such as seventy five ('soixante-quinze') gives rise to errors such as 6015, observed in the second part of the CP.

Training in the functions solicited in learning arithmetic would enable teachers to recognize the first signs of dyscalculia in children as a poor understanding of the principles of counting, the use of primitive counting strategies on the fingers, and later abnormal and persistent difficulty in memorizing the results of the simplest sums and multiplications.

The need to train secondary school teachers should also be stressed. Numerous children have disorders that persist through junior and high school despite the rehabilitation dispensed. Without training on the subject, teachers may consider such pupils to suffer from a mental retardation or disorder and come to the conclusion that the pupils are not in the right educational environment.

#### **PROMOTING APPROPRIATE USE OF INSTRUMENTS IN THE CONTEXT OF SCREENING**

The screening instruments for specific learning can only be used when the child has begun scholastic learning (reading, writing, arithmetic), i.e. after age 6 years. However, instruments are available that enable screening for risk factors for learning disabilities before the age of 6 years. The instruments address spoken language, non-verbal capabilities, attention, memory, etc.

In the context of individual screening after age 6 years, several instruments and tests are available to investigate whether a child presenting with, for instance, difficulty learning to read is liable to have a specific disorder (dyslexia). The instruments calibrated in France have specific objectives for which their sensitivities and specificities have been determined. Investigating different functions and capabilities, the instruments may be used in a complementary manner during the initial assessment. They are able to identify children requiring a differentiated pedagogic approach, those requiring follow-up and those requiring referral to a specialized professional for diagnosis.

In the context of the obligatory health assessment at age 6 years, when the child enters primary school (article L.2325.1 of the Code of Public Health), the instruments screening for risk factors based on the results of longitudinal studies are to be used. However, early detection is only of value if preventive actions having demonstrated their efficacy in the context of rigorous evaluations can be set up.

It is noteworthy that an expert commission set up (decree dated February 8, 2002) to formulate national recommendations on the instruments for use on children by professionals in the context of the plan of action for children presenting with a specific language disorder issued a report that was made public in 2006<sup>11</sup>.

#### **DEVELOPING AND EVALUATING PEDAGOGIC TRAINING BY ADAPTING MODELS HAVING DEMONSTRATED THEIR EFFICACY ABROAD**

Studies, mainly conducted on English-speaking children, have demonstrated the efficacy of certain forms of pedagogic training (actions on deficient cognitive function) in elementary school children (CP, 6-7 years, or CE1, 7-8 years) presenting with decoding disorders. On the basis of the studies, training is to consist in a specific, intensive and explicit activity. The activity is to address, first, the grapheme-phoneme matches both in synthetic (from the grapheme-phoneme match to the word) and analytical (from the word to the grapheme-phoneme match) tasks and, secondly, the capability for phoneme analysis, discrimination and fusion. Training is to be pursued until reading is mastered and is to enable the children to recognize, discriminate and write words increasingly rapidly. A daily session lasting half an hour to an hour is recommended for individual children or small groups with similar requirements. Assessment of the benefits of training may be conducted after a few months.

The results of the studies available show that the type of training in elementary school (CP, 6-7 years) has positive effects on automation of recognition of written words, reading comprehension and, to a lesser degree, writing. The effect on reading speed nonetheless requires confirmation.

Using those studies as a model, pedagogic training should be tried as of the start of primary school (CP: 6-7 years) for children presenting with a spoken language disorder and as of the second part of CP for weak decoders. Similarly, the potential benefit of preventive training, as of kindergarten (GS: 5-6 years) for children at risk of dyslexia (with a spoken language disorder, weak phonological skills or a familial history of dyslexia) should be studied.

Such experiments in France would enable evaluation of whether pedagogic training in school that is limited in duration, does not marginalize the children and is accessible to all, would be beneficial for some of the children enabling them to recover, in a stable and lasting manner, a reading level similar to the normal level without other management. The studies would also show whether transient 'slow readers' can be differentiated from children with

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<sup>11</sup> [www.sante.gouv.fr](http://www.sante.gouv.fr)

dyslexia requiring complementary care and individual management. If the preventive strategy were validated in France, it would enable faster access to reference centers for children in real need of a diagnosis.

In the context of those studies of pedagogic training in the school environment, teacher training would be necessary for preventive management in kindergarten (GS: 5-6 years) of children presenting with risks for learning to read in the same way as for management of children experiencing reading difficulties in the first year of primary school (CP: 6-7 years).

#### **PROMOTING THE PEDAGOGIC ARRANGEMENTS AND ADAPTATIONS NECESSARY FOR PREVENTING OTHER LEARNING DIFFICULTIES**

Several published studies address the positive effects of pedagogic arrangements and adaptations which enable a child presenting with a specific disorder in a field to acquire the knowledge required for the child's class level in other subjects (mathematics, history, life and earth sciences, etc.) without being handicapped by his/her disability. Methods include, for example, reading the problems in mathematics out loud or giving additional time for reading them in the event of dyslexia, or decreasing the writing load in the event of an orthographic or handwriting disorder using a computer (word-processor with spell check or vocal dictation in the most severe cases).

These pedagogic arrangements and adaptations are to be set up and evaluated through school (primary, secondary and higher education) in order for the child not to be penalized by the written language disorder (reading, writing) and in order for the child to benefit from other learning experiences. Such studies could contribute to defining the conditions under which children presenting with a specific learning disorder and subject to individual care elsewhere can be integrated into normal school.

Decree No. 2005-1617 dated December 21, 2005, relating to the adaptation of examinations and competitive examinations in school and higher education must be applied to pupils presenting with specific disorders, who, as candidates for an examination or competitive examination, are handicapped as provided for by the new international nomenclature on deficiencies, disabilities and handicaps. However, the family associations report very uneven application of the law, depending on the area of residence ('*département*'). In addition, given the diversity and seriousness of specific learning disabilities, the concept of handicap is sometimes poorly perceived by parents, who thus hesitate to have recourse to the local handicapped persons' facility in order to procure those adaptations.

### **Enhanced management**

#### **SETTING UP AND EVALUATING VARIOUS INDIVIDUAL CARE MODALITIES FOR DYSLEXIA**

The individual care modalities for dyslexia are to be based on the scientific knowledge acquired in recent years. The modalities have to take into account the diversity of each case: the precise nature of the cognitive disorder presented by the child and the child's environment.

Increasingly numerous published findings on written language development and the predictive factors for that development together with the effects of specific training are associated with indications, orientations and practical conditions with respect to disorder

management. The studies evidence that intensive training programs (generally half an hour daily, 4 days per week) over relatively short durations are beneficial providing that they are precisely and specifically targeted on a deficient cognitive function with respect to improving the deficient function and with generalization to reading and writing.

Individualized speech therapist's management of a specific written language acquisition disorder is justified as of the start of primary school (CP: 6-7 years) if a spoken language disorder persists, during CP if the pedagogic training in class has proved insufficient or in the event of signs of severity such as total non-acquisition of the grapheme-phoneme code during learning. The effects of management are to be regularly quantitatively and qualitatively evaluated using calibrated tests.

It would be of interest to experiment on, evaluate and compare several care modalities as a function of age and impaired function, and under various conditions (with daily repetition in class and/or at home of the program defined by the professional managing the child's case; harmonization with pedagogic interventions, etc.) and with various methods of compensation (visual and kinesthetic aids, for instance). The objective is to optimize care modalities as a function of the child's need.

The use of standardized instruments (video games, recreational audiovisual recordings) appears necessary in research and clinical practice in order to control the quality and quantity of the information targeting the child during the training sessions. The advantage of computerized instruments (digitization of speech, for instance), in addition to the fact that they provide specific exercise on auditory acquisition, is that they enable graduation of the difficulty of the exercises and potential adaptation of the exercises to each case as a function of age or to the severity of the phonological deficit. If the results of the studies are positive, the findings could subsequently be generalized to clinical practice.

However, the proliferation of instruments, particularly computerized instruments, whose objectives and contents have not been controlled and whose efficacy has not been evaluated provide the grounds for creating an independent scientific body to validate and accredit the remedial instruments for learning disabilities. Comparative studies (in the form of a clinical trial) of the various instruments would enable definition of their effects and limitations.

The evaluation of the effects of training using those instruments is to be based on a methodology enabling confirmation of a specific effect on the trained function. The efficacy criteria of training are: the effect on the specific cognitive function trained; generalization to the identification procedures for written words (precision and time); generalization to the understanding and spelling of isolated words and words in context.

#### **PROMOTING AND EVALUATING THE VARIOUS MODALITIES FOR INDIVIDUAL MANAGEMENT FOR LEARNING DISABILITIES OTHER THAN DYSLEXIA**

The methods of management of disorders associated with written language acquisition are to be the subject of evaluations.

The studies show that spoken language disorders have repercussions on written language learning and that early rehabilitation (at the latest at age 5 years) with respect to the spoken language enables written language learning to be tackled under better conditions.

With regard to handwriting disorders, the quantitative and qualitative results of the specific tests enable determination of whether the disorder affects gesture coordination, perception and/or visual production and visual and spatial production. Criteria could be determined with respect to the ages and modalities of managing handwriting disorders: at the end of

kindergarten or at the start of primary school if the disorders are severe; before the end of the first year of primary school (CP: 6-7 years) in the event of a persistent disorder in order to prevent deviant strategies in letter sequencing becoming established. It is indispensable to interface the management implemented by the psychomotrician and ergotherapist and the teaching interventions.

Disorders of numeric code acquisition are frequently associated with written language acquisition disorders. Few studies have been conducted on the instruments and modalities of management. Management as of the start of the primary school is to be envisaged and evaluated insofar as those associated disorders frequently exacerbate the child's failing status.

#### **PROMOTING AND EVALUATING MULTIMODAL MANAGEMENT FOR FREQUENTLY ASSOCIATED DISORDERS**

In the context of specific learning disabilities, the literature reports frequent association of affective and behavioral disorders, which require psychotherapeutic management without, however, neglecting the specific rehabilitation for the specific learning disabilities. Management of the associated behavioral and affective disorders must be initiated early in the event of disorders reported prior to primary school and if the disorders impact on the child's cooperation with the pedagogic and rehabilitation project.

Neuropsychological and psychomotor disorders associated with learning disabilities currently constitute a wide field of research. The disorders raise the question of offer in terms of rehabilitation techniques that may vary depending on the associated signs such as perceptual-motor disorders, visual-spatial disorders or attentional disorders and in terms of trained recognized professionals (such as neuropsychologists).

Those more severe cases are exposed to a serious threat to scholastic outcome and social adaptation. The cases should benefit, irrespective of the children's geographic or socioeconomic situation, in the familial and school environment, from the most pertinent resources with respect to the current state of knowledge. The development of reference centers<sup>12</sup> is a very important step forward and the setup of health networks consisting in multidisciplinary teams coordinated on a regional scale also appears an interesting alternative.

#### **PROMOTING AND EXPERIMENTING WITH SETUP OF COORDINATED DIAGNOSTIC AND CARE NETWORKS**

The diagnosis of a specific learning disorder, which is indispensable in order to adapt management, frequently involves a pluri-disciplinary team (e.g. reference center) which needs to be coordinated by a referring professional: the clinical examination enables identification of a patent disorder and verification of its specific nature, severity and persistence; precise evaluation of the impaired functions is implemented by the professional involved (speech therapist, psychomotrician, ergotherapist, psychologist, neuropsychologist). The various cognitive functions involved in learning are to be evaluated using validated instruments. The tests address, for instance, the specific capabilities for reading, which are deficient in the child (identification of written words, precision and rapidity) and the related capabilities (phoneme analysis capability, phonological short-term

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<sup>12</sup> About forty now exist.

memory, visual analysis capability, etc.). Comparing and contrasting the results generated by batteries evaluating cognitive profile and by the specific tests enable confirmation of the specificity of the disorder. The evaluations are to be conducted using tests standardized on the child's age. The results are to be qualitative and quantitative and state the test name and the results in terms of standard deviation or percentile relative to the reference population standards.

Management of learning disabilities requires regular evaluation of the child's course. The effects of rehabilitation are to be evaluated at least every 6 months with a new assessment using tests similar to those for the initial assessment. The assessment is to enable quantitative and qualitative evaluation of the child's course and review of management in order to take the pertinent decisions (pursuit of training, redefinition of the objectives, reorientation, alternation of pauses, discontinuation of rehabilitation). Recourse to a professional other than the one implementing the rehabilitation program is useful if the child's progress and progress generalization in class are not sufficient.

Formalized networking of all those contributing, in close cooperation (National Education specialized personnel, health professionals, etc.) is under study in a few centers and is to be evaluated with a view to generalization to the country as a whole. The coordination of care and family counseling may be ensured by a professional with appropriate training. It is also important to promote regular meetings between the rehabilitators and teachers in order to harmonize their reciprocal actions, set up a personalized scholastic program, and provide for the pedagogic adaptations and arrangements necessary in order to integrate the child in the class and ensure access to the various types of learning. The setup of such networks appears, a priori, as a particularly appropriate response to severe forms of learning disorder whose complexity clearly necessitates comparing and contrasting the opinion of several professionals. Those cases can only be effectively managed providing close and repeated contacts with the child's teacher are established.

In addition, it is appropriate to reduce geographic inequalities with respect to management offer and to equip the areas without coverage, in particular with regard to proximity resources on which teachers can draw (Rased, Sessad, National Education physicians). Those provisions, if they existed in sufficiently large numbers, would enable the reference centers to be dedicated to diagnostic evaluations.

#### **PROPOSING COMMON TRAINING TO ALL RESOURCE PEOPLE IN ADDITION TO SPECIFIC TRAINING BY DISCIPLINE**

In schools, there are professionals (specialized teachers, psychologists, National Education physicians) able to help detect pupil learning difficulties and propose and implement an appropriate response. Other professionals outside the school (speech therapists and other rehabilitators, psychologists, neuropsychologists, physicians and other specialists) are also frequently called in for diagnosis and remediation.

Initial and in-service training of all those professionals is to be envisaged in relation to scientific progress. The training must enable all the professionals to become familiar with the use of validated identification instruments and critical analysis of the proposed management methods.

In addition to precise training specific to each professional's role, a training course common to the various professions would enable the professionals to work in close cooperation in order to facilitate the management necessary for pupils with learning difficulties or presenting with specific learning disabilities.

## **Widening our knowledge of scholastic learning and learning disabilities**

The research orientations proposed are designed to develop an enhanced understanding of scholastic learning (reading, writing, arithmetic, etc.) and learning disabilities, in particular enhanced understanding of the causes of dyslexia and estimation of the frequency of the various specific learning disabilities in France.

### **WIDENING OUR UNDERSTANDING OF THE MECHANISMS OF DYSLEXIA**

Numerous theoretical models to explain dyslexia currently exist: phonological deficiency theory, temporal auditory processing theory, visual theories, cerebellar theory, magnocellular theory, etc. The diversity is due to the fact that there are doubtless several explanatory factors for dyslexic disorders that can be applied to various subgroups of the dyslexic population. Dyslexia remains a complex disorder including numerous symptoms other than reading impairment and is frequently associated with other learning disabilities.

The phonological theory has given rise to the greatest number of studies and is currently the most widely validated. Almost all the studies conducted on children with dyslexia, irrespective of their language, have evidenced associated phonological deficiencies. The deficits objectively demonstrated in the three fields of skills related to reading (phoneme analysis, phonological short-term memory, image and object naming) contribute to explaining the child's reading level. The children's performances in the various fields remain deficient even when compared with those of younger children with the same reading level. This shows, first, that the dyslexic child does not have the same developmental trajectory as the normal reader; secondly, the phonological disorder is causally related to reading learning difficulties. This has been confirmed by longitudinal studies: the phonological skills evaluated before learning to read are indicative of the subsequent reading level of the child.

However, except in rare studies, the phonological theory has not been compared with other alternative or associated theories. Longitudinal studies in which the children would be followed up from the start of the main section (or even intermediate section) of kindergarten (5-6 or 4-5 years) up to the end of primary school cycle 2 (8 years) or 3 (11 years) would be very informative. Those studies should evaluate the involvement of phonological, visual and motor skills in learning to read using the most similar methodologies possible (taking into account accuracy scores and processing speed in tasks with or without time limits, etc.). Research on cognitive mechanisms which specifically influence reading speed and the speed-precision interactions should also be encouraged.

### **DEVELOPING RESEARCH ON THE MECHANISMS INVOLVED IN LEARNING TO SPELL**

The specificities of French spelling, particularly in written language production, are such that the knowledge generated by research on other writing systems is difficult to transpose. Thus, in the absence of precise data on the errors made by all children or adults, it is difficult to determine the extent to which the number and type of errors are a normal performance or, on the contrary, provide grounds for predicting a disorder necessitating specific management.

Studies on the mechanisms involved in learning to spell and implementation of the various components of writing should be developed. The studies should take into account the type of teaching given. The studies should tackle the question of lexical spelling acquisition and the determinants of success and failure. The studies should also address the learning and

implementation of derivational morphologies ('*chat*'; '*chatte*'; '*grand*'; '*grande*') and flexional morphologies (past participle vs. infinitive; gender and number agreements of nouns and adjectives, etc.).

#### DEVELOPING RESEARCH ON THE MECHANISMS INVOLVED IN LEARNING ARITHMETIC

The studies on dyscalculia and mathematical difficulties are much less frequent and less advanced than those on dyslexia while the frequency and etiology of dyscalculia remain poorly elucidated. While numerous hypotheses have been advanced, the data are currently not sufficient to enable orientation of pedagogic and rehabilitation responses. While the hypothesis of selective impairment of the brain structures responsible for numerical processing has been suggested, the hypothesis requires stronger support. The deficiency in the research field is all the more surprising in that learning arithmetic and mathematics constitutes one of the major objectives of schooling in all technologically advanced societies.

Research on the relationships between early skills in infancy with respect to quantities and subsequent numerical acquisitions, on the nature, rhythm and between-individual differences in those acquisitions, their evolution and their impact on learning mathematics should be developed. The results would shed light on the principles for remediation.

#### DEVELOPING EPIDEMIOLOGICAL STUDIES IN FRANCE

In France, there are no epidemiological data on specific learning disabilities based on samples of children representative of the overall population.

Cross-sectional studies on representative samples are thus indispensable in order to elucidate the prevalence of the various specific learning disabilities. The studies could evaluate the influence of the various criteria used to classify the disorders on their frequency and define instruments and a standardized methodology. Epidemiological studies would also enable elucidation of the role of the family's social status and educational level.

Similarly, longitudinal studies of child cohorts initiated very early in the infant's life could enable study of the determinants potentially involved early in the child's cognitive development (e.g. in the context of the Eden cohort and Elfe cohort). The factors most closely linked to specific learning disabilities should be identified.

Epidemiological and preventive (research-action) studies involving schools and mixed educational and scientific teams would enable information sharing between the two fields and should also be developed.

## Meeting-debate held on January 16, 2007 <sup>13</sup>

Opening the meeting, Christian BRECHOT, Director General of Inserm, greeted the participants and thanked all those having contributed to the collective expert review directed by Jeanne ETIEMBLE. The meeting illustrated the manner in which Inserm now wishes to work, at least for certain collective expert reviews. Christian BRECHOT considered it important to obtain the point of view of the professionals in the field with respect to the conclusions drawn by the expert review.

### **Introduction: the Inserm collective expert review approach**

Jeanne ETIEMBLE explained that the collective expert review approach consists in reviewing the international literature on a given subject. The review is a response to a request formulated by a sponsor, in the present case, the RSI (social regimen for free-lance workers, formerly CANAM), in the context of a program focusing on child health. After the expert reviews on obesity and mental disorders, the third collective expert review, considered herein, addressed the specific learning disabilities, namely dyslexia, dysorthography and dyscalculia. The group of experts - whose composition was validated by the Directorate General of Inserm - consisted of 11 specialists. The group issued a report consisting in three sections: analysis of the international literature; synthesis of the experts' work; and the experts' recommendations. Jeanne ETIEMBLE stressed the distinction between the analysis of the international literature implemented by the experts and a status report on French practices in the field considered. The latter was not included in the expert review's scope and is rather a matter for the High Authority of Health. The collective expert review on specific learning disabilities in children is now at the stage when it is rendered public and the present meeting is intended to enable all parties to express their feelings, in particular to prevent any misunderstandings.

The subject: 'Dyslexia, dysorthography and dyscalculia' was selected in order to define a field of expert review less broad than scholastic failure for which considerably greater resources would have been necessary. While dyslexia is a frequent concern, there are numerous erroneous ideas with regard to the subject. It would therefore seem of value to review the question. Dysorthography and dyscalculia have been the subject of markedly fewer articles. Jeanne ETIEMBLE agreed that dysphasia, a specific spoken language disorder, constitutes such an important subject that it could in itself have been the subject of an expert review. The same observation applies to dyspraxia. However, the experts decided to consider those two disorders in the context of 'disorders associated' with dyslexia, dysorthography and dyscalculia.

Claire MELJAC (psychologist, Hôpital Sainte-Anne) noted that dysorthography, which is associated with dyslexia in most cases, was addressed indirectly and was thus present in the expert review. However, no significant findings have emerged with respect to dyscalculia and certain results on the subject are far from certain. Claire MELJAC therefore wondered why dyscalculia was included in the title of the expert review.

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<sup>13</sup> Report compiled in real time by the company Ubiquus ([www.ubiquus.fr](http://www.ubiquus.fr))

Ariel CONTE (CORIDYS) considered that the framework of the expert review, restricted to dyslexia, dysorthography and dyscalculia, was liable to restrict its public health interest when the teaching hospitals (CHU) statistics showed that few cases of homogeneous dyslexia existed. Won't decision-makers have a narrow vision of specific learning disabilities?

## **Principal findings of the collective expert review**

### **Reading**

Liliane SPRENGER-CHAROLLES, (member of the expert group) noted that the aim of reading is comprehension. This requires a satisfactory level of spoken language understanding and a good level of automatism in the identification of written words (accuracy and speed). In alphabetical writing, two procedures may be used for identification: a lexical procedure (the processing unit is the word) and a sub-lexical procedure (the processing unit is the grapheme in relation with the phoneme).

Studies have shown that children strongly use the sub-lexical procedure at the start of learning. The success of learning depends on the regularity of the grapheme-phoneme correspondances which are more transparent in Spanish than in French and more transparent in French than in English. Thus, Spanish-speaking children automate written word identification procedures for written words faster and better than French-speaking children who in turn are faster and better than English-speaking children. Dyslexia is mainly due to deficient automation of those procedures which is exacerbated when the writing is opaque. Moreover, systematic and early teaching as of the start of primary school of grapheme-phoneme correspondances is what helps pupils most. Lastly, it would appear that the most reliable predictors of learning to read are the ability to conduct phoneme analysis and the degree of knowledge of letters together with phonological short-term memory capability and rapid naming capability. Those predictors enable early identification (as of kindergarten age 5-6 years) of children liable to experience reading learning difficulties with a high reliability.

### **Writing, arithmetic**

Michel FAYOL (member of the expert group) explains that an essential point consists in determining whether, for writing, there are disorders which are not related to reading and, for arithmetic, disorders that are not specific to arithmetic. The French language is an 'inconsistent' system in terms of production: writing it is much more difficult than reading it. Phonological training again has a very favorable impact on learning the bases of writing. A large part of lexical writing acquisition seems to be related to the practice of reading.

Michel FAYOL noted that only few studies were available on arithmetic disorders and that there was no reliable classification of errors. It is therefore difficult to distinguish a normal course from a problematic course and misunderstandings with regard to associations of disorders may result. In any event, sensitivity to changes in quantity emerges very early. The acquisition of the native language symbol system enables setup of counting behaviors. Even though there is an international definition of dyscalculia, the criteria are not universally recognized. Dyscalculia may be as frequent as dyslexia but is rarely observed in isolation. There is no difference between boys and girls with respect to the frequency of dyscalculia.

### **Why are disorders so frequently associated?**

Jean-François DEMONET (member of the expert group) stressed a difficulty confronting researchers. The latter are uneasy in epistemological terms due to a paradox: skills highly dependent on culture and scholastic learning prove, in the light of the scientific studies, to be closely related to particularities of the organization of the human brain and particularities of the genome (or even gene mutations). Given that surprising finding, Jean-François DEMONET pleaded in favor of a multidisciplinary approach, which is currently difficult due to the diversity of the epistemological viewpoints.

With regard to reading considered from the viewpoint of cognitive functions, Jean-François DEMONET explained that in the absence of procedure automation, individuals who were dyslexic in childhood need 150 milliseconds more than individuals who were not dyslexic to decode a word (i.e. a minute more to read a page).

Writing is associated with additional difficulties compared to reading, particularly due to the exhaustive nature of written expression: everything must be written down! In addition, a given sound may have very numerous written forms. This is referred to as the 'phono-orthographic non-transparency of the French language'. Because of those characteristics, learning a lexicon, particularly in the French language, constitutes a major workload for the long-term memory.

Arithmetic makes use of a more or less large stock of memory knowledge (multiplication tables) or the application of computation rules enabling memorized knowledge to be combined. The degree of working memory load is greater when none of the basic stages has become automatic.

Jean-François DEMONET formulated the hypothesis that dyslexia, dysorthography and dyscalculia share common mechanisms, namely:

- defective perception of the basic units,
- defective automation of between-format conversion,
- an excessive working memory load.

Jean-François DEMONET then reviewed the cerebral substrates for those linguistic functions and mentioned the importance of the frontal cortex for working memory. He presented the topography of the regions activated during reading, writing and arithmetic. The topography of the microscopic anomalies reported for the brains of a few dyslexic persons show that the regions involved cover those that seem to be important for the functions related to written language and arithmetic.

### **Biological approaches vs. social approaches**

Franck RAMUS (member of the expert group) addressed the opposition that has sometimes been suggested between the biological approaches and social approaches to the three disorders under study. In his opinion, the Inserm report does not reflect an exclusively biological approach: the report does not pass over the social and environmental (familial, psychological, pedagogic) factors that may play a role in the emergence of learning disabilities. However, even children with no sociocultural handicap may present with disorders, for example in learning to read, for which no primary social cause can be evidenced. The researcher must thus consider a cognitive deficit, i.e. a problem in the processing of certain information by the brain. As the report describes in detail, in reality,

there may be a major deficit, at phonological level, in the processing and mental representation of sounds and speech.

This finding presupposes the existence of a cerebral lesion and the research conducted on dyslexic individuals' brains has confirmed the existence of specificities in those individuals. A number of indices, including the familial character of disorders such as dyslexia, argue in favor of the existence of genetic factors (but not exclusively genetic factors) that explain the cerebral particularities. Researchers have evidenced the variant form of certain genes resulting in a slightly different setup of certain brain areas playing a role in speech and language and recruited later for learning reading, writing and arithmetic. However, Franck RAMUS stressed that the genetic factor is not to be considered synonymous with determinism: the logic is probabilistic and depends in part on interactions with very diverse environmental factors. For that reason, strictly biological or strictly social paradigms are to be ruled out.

### **Prevention, screening and management**

Catherine BILLARD (member of the expert group) stressed the value of prevention in the management of all children presenting with reading disorders. The international literature has shown that early management, in school, of children presenting with decoding difficulties enables 50 to 75% of those children to catch up with their peers in a few months. With regard to prevention, the concept of 'child with similar needs' seems operative in order to define management meeting the criteria defined by the scientific literature. The expert group argues in favor of evaluating such an approach in France. This necessitates defining severity criteria on the basis of which, for children not having benefited from the first stage, the decision on whether or not to add additional care to assistance in school will be taken. Spoken language disorders define a population at risk but not all the members of that population will present with the disorders considered (dyslexia, dysorthography and dyscalculia). Preventive management of spoken language disorders is not problematic and is defined by the ANAES recommendations. However, on the basis of the reference meta-analysis in the field, a preventive program is only effective if associated with work on the phonological skills in spoken and written language.

With regard to individual management (in terms of care) post-diagnosis, it must precisely define:

- the orientations of rehabilitation (to be worked on intensively and evaluated),
- the conditions for access to care for deprived people,
- articulation with the teaching programs in school in the form of a coordinator who will have, in particular, the crucial role of explaining to the parents.

### **Debate**

After having congratulated the expert group on its work, Michel ZORMAN (Grenoble Teaching Hospital (CHU)) returned to certain formulations in the text: the question of the independence of the disorders with respect to sociocultural factors compared to Franck Ramus' remarks on the existence of biological and environmental interactions. With regard to 'unfavorable environmental conditions', Michel ZORMAN preferred the term educational or pedagogic deficiency in which fundamental components of education are not present. That may occur in any sociocultural environment. Lastly, Michel ZORMAN noted the presence of redundant components, for example in the case and group studies, impairing the

legibility of the report. Michel ZORMAN also contested the distinction between phonological dyslexia and surface dyslexia. All the cases of phonological dyslexia in his care become, at a certain age, surface dyslexia. Michel ZORMAN considers it indispensable to dissociate the measurements of the efficacy of the two procedures (lexical and sub-lexical) from the causes which may reduce their efficacy. Liliane SPRENGER-CHAROLLES stressed that the study of the causes of disorders necessitates longitudinal follow-up of children, in the beginning of the follow-up, phonological disorders were very often observed in children who later became dyslexics.

Georges DELLATOLAS (Inserm) considered that the diagnostic criteria for scholastic learning disorders were not sufficiently discussed in the synthesis: the experts did not state whether it was necessary to use them. The criterion of interference with scholastic performance may lead to learning disabilities being defined as a function of the entourage's requirements, particularly with elitist aims. The prevalences stated show, in any event, that the criteria are not used. Jeanne ETIEMBLE observed that the disorder definition criteria, when applied, have at least the value of providing a basis for comparison of epidemiological studies. Franck RAMUS agreed that the definitions of the criteria are not satisfactory to anybody. However, they constitute the only international standard available.

Bernard GOSSOT (Inspector General, National Education, honorary) noted that the synthesis states that 'the prevalence of dyslexia is a little less than 5%' but that the reference population was not stated. The age at which children's 'reading difficulties' were detected were not stated either. Because of those imprecisions, 'we see dyslexia everywhere'. Catherine BILLARD agreed that it was necessary to take the remark into account to avoid confusion with regard to the term 'dyslexic'. Michel ZORMAN considered that it was possible to state that dyslexia affected 5% of the overall population from the first year of primary school (CP: 6-7 years). He also suggested indicating (with no further details of the proportions) that all children experiencing reading difficulty were not dyslexic. Referring to a report that he wrote in 2005 for the Inspectorate General of Social Affairs and the Inspectorate General of National Education on the complex disorders of spoken and written language, Bernard GOSSOT noted that the number of children presenting with a true complex disorder of spoken and written language was extremely low. In that context, the prevalence of 5% was very high in his opinion. Jean-François DEMONET asked what criteria were used to estimate the prevalence. Bernard GOSSOT stated that no precise criterion had been defined. Physicians, psychologists and parent associations were nonetheless interviewed and described their analysis of the situation to the authors. The authors also observed that the number of children expressed relative to the population was fairly low although at the outset the number was stated to be high. Jean-Marie DANION stressed the difficulty in obtaining an accurate figure on that point.

## **Recommendations**

### **Training**

Jeanne ETIEMBLE indicated that a first recommendation, with respect to training, was considered particularly important by the expert group. Training must enable all the players to speak the same language and understand each other.

Gérard TOUPIOL (National Federation of Special-Needs Teachers, specialized teachers working in the context of specialized help networks) noted that the synthesis cited numerous instruments which are not known by National Education professionals. The network of

specialized assistance would appear, in any case, best situated for identifying pupils and recommending, if necessary, further screening for learning disabilities. Jeanne ETIEMBLE pointed out that a commission meeting under the auspices of the Directorate General for Health (DGS) had published a report available to all on the Ministry of Health's website.

Jeanne-Marie URCUN (national physician, National Education) considered it of value to state the instruments under discussion: detection instruments, diagnostic instruments and remediation instruments are available. They are not interchangeable. In any event, 'no one can act alone' and pluri-disciplinary work constitutes a requirement. However, it is not for one to act in another's place. Claire MELJAC considered that an instrument without training for the user was devoid of sense. Calibration and instructions for use are not sufficient: there must be an exchange with a person aware of the limitations and sensitivity of an instrument. Ariel CONTE confirmed that the use of instruments greatly depends on training, culture and individual representations. Moreover, certain recommendations have long existed. They need to be applied.

Scania de SCHONEN observed that difficulties often begin with pediatricians' unfamiliarity with cognitive neuroscience: pediatricians rely on psychiatrists to identify children's cognitive disorders while psychiatrists are not the best trained professionals for that purpose. Scania de SCHONEN wished that a unit could be set up with the mission of studying the channels enabling physician (pediatricians, psychiatrists) and speech therapist training in the cognitive disorders of children. She also recommended implementation of an international technology watch with a view to updating the conditions for use of the existing instruments.

Anne TERLEZ (Pupil Parent Federation, PEEP) indicated that she was particularly aware of the quality of the expert review. She considered that the experts 'could have gone much further' in their recommendations. PEEP militates for equal opportunity which supposes, in particular, accentuating screening (which goes further than detection). Teacher awareness of learning disabilities and child remedial work and follow-up requires (initial and in-service) training for healthcare professionals and National Education teachers. More broadly speaking, pragmatism is urgent. The RASED are frequently precarious due to lack of resources. The same applies to school medicine and the protocols set up cannot be applied. Opening schools to health professionals is indispensable and partnerships must be set up and oriented towards finding original solutions. Anne TERLEZ also reported an original experiment set up by the Issy-les-Moulineaux PEEP: the latter launched dyslexia screening on the commune scale. National Education professionals were specifically trained. PEEP wishes that the action, whose cost is assumed by the Issy-les-Moulineaux *commune*, may be generalized to all of the Hauts-de-Seine *département*.

Sylviane LEWICK-DERAISON (speech therapist and UNADREO representative) was pleased to see the position of the networks recognized in the experts' recommendations. However, she considered the proposed formulation too vague. The assistance network (RASED) constitutes a network in schools while the health networks are outside the schools. The referral professionals are also numerous. Hence, those concepts need to be clarified without confusing disease and teaching. This led Sylviane LEWICK-DERAISON to point out what the previous speaker described was in fact only an experiment.

### **Preventive strategies**

Jeanne ETIEMBLE wished to tackle the preventive strategy that could be deployed as soon as a difficulty was identified in a child. In France, the trend remains characterized by individual

management. Jeanne ETIEMBLE wished the participants to describe how they envisage working as a network on preventive objectives.

Referring to the 14 studies on reading difficulties in primary school published outside of France, Michel ZORMAN suggested that the synthesis referred to those studies on the basis of which a controlled and evaluated study could be requested from National Education. Jean-Marie DANION considered that that type of recommendation fully reflected the type of recommendation for which the collective expert review approach may be legitimate. Catherine BILLARD insisted that the study methodology should determine whether the observed effect is a specific effect or placebo effect. The results must not be only expressed in terms of percentage significant improvement but also in terms of the results for the children.

Laurence VAIVRE-DOURET considered that one dimension related to the associated disorders is currently missing from the preventive context. Psychomotricity may constitute a means of prevention in the National Education system, for instance by evidencing an absence of laterality setup. The National Education system may thus have an interesting role to play.

Jeanne ETIEMBLE observed, particularly on the basis of the experiment reported by the PEEP representative, that it was possible to implement the preventive recommendation involved. The experiments should be allowed to multiply, particularly as they respond to several problems at the same time without remaining confined to the field of dyslexia. However, such a strategy does not obviate the need for individual management for certain children. Accordingly, the question resides in the interface between individual management (care) and the pedagogic management to be implemented in school.

### **Individual management/coordination and articulation between education and care**

Bernard GOSSOT reminded those present of the distinctions between the concepts of detection, screening and diagnosis. Detection may be implemented by teachers but not alone. RASED can contribute useful help on that point, particularly through management in class of specific small groups. Outside personnel could be called in: speech therapists, SESSAD personnel, etc. The school physicians and psychologists are not sufficiently present. Screening is to be the responsibility of the Maternal and Infantile Protection (PMI) physician or school health physician. Lastly, diagnosis is the responsibility of the reference centers. Catherine BILLARD stated that between the reference centers and teachers were resource persons, thus avoiding a teacher directly addressing a family to a reference center. The reference center has a second-line vocation in terms of severity or recalcitrance of the problem.

Frédérique LEFEVRE (Ministry of National Education) stated that schools are only authorized to treat scholastic difficulties at pedagogic level. When the teacher does not know which pedagogic response to use, he/she can turn to other players such as RASED. Frédérique LEFEVRE also noted that schools are not care establishments. Schools can contribute to prevention through a pedagogic response as the 2005 act states. Michel FAYOL indicated that the Ministry of National Education recently considered introducing, for primary school children (CM1: 9-10 years), an instrument enabling teachers to conduct detection which could be followed, if appropriate, by implementation of activities such as those discussed during the debate. Frédérique LEFEVRE also stated that there were non-mandatory evaluations enabling teachers to implement pedagogic responses.

Bernard GOSSOT indicated his disagreement with the idea that the National Education system would rule out any intervention with a care aim: while the school is not a care establishment, care professionals can intervene to facilitate education. In addition, Bernard

GOSSOT indicated that the term pedagogic training might be badly perceived by teachers who might consider it to reflect a behaviorist approach. Catherine BILLARD stated that the concept of 'training' did not in any way constitute the principal pedagogic action.

Ariel CONTE noted that, in Canada, schools include teacher - speech therapists with dual skills. He expressed the wish that the cleavage between health professionals and educational professionals become less marked in France. He also indicated that the networks' budgets did not enable the latter to manage the children who needed it most.

Isabelle DABOVILLE (referral teacher) stated that a dialog with the parents could result in setup of a personalized project providing for intervention of the care services in schools and arrangements in the children's school timetable. A question persisted, however, with respect to the way in which the requests for resources emerging in that context would be received by the Commission for Rights and Autonomy (CDA).

Jean-François DEMONET observed that the National Education system was wary of being put in the position of diagnosing a 'serious brain disease'. Nonetheless, National Education system physicians exist. If they existed in sufficient number, they could play an indispensable interface and orientation role for a disorder that is first expressed at school. The care response is to be interfaced with the pedagogic response. The cleavages must be replaced by an interconnection between pedagogic concerns and health concerns. In order to do so, means and structures are required. The number of school physicians and psychologists, for example, would have to increase. Jean-François DEMONET argued in favor of mixed management, irrespective of whether individual or collective.

### **Pedagogic arrangements and adaptations**

Ariel CONTE noted that all the arrangements granted to dyslexic subjects are granted because of the handicap. However, the practical organization of those arrangements is the problem. Also, practices vary widely from one department to another. As a neurologist, Jean-François DEMONET stressed that application of decree No. 2005-1617 dated December 21, 2005, relating to adaptation of examinations and competitive examinations in secondary school and higher education constituted a daily difficulty and an extremely frequent request from families. Alas, there is great confusion on the subject, including among academic personnel, in particular given the varied seriousness of the disorders.

Valérie CAPUANO-DELESTRE (ASH Bureau, scholastic adaptation and schooling of handicapped pupils, Ministry of National Education) stated that the adaptation of examinations is independent of the percentage handicap and its recognition by the MDPH (departmental facilities for handicapped persons). But a difficulty persisted with respect to specific learning disabilities due to the 50% cutoff defined as the necessary condition for recognition of a handicap. In contrast, the decision with respect to adaptation of examinations depends on the National Education system, independently of the percentage handicap. Michel ZORMAN hoped that the compensation granted for pupil examinations did not compensate for a diagnosis but for a social disadvantage with regard to subsequent studies. Without such a principle, the compensation would benefit too many pupils and in that case lose all pertinence. Jean-François DEMONET stated that a certified diagnosis of dyslexia at a given time in a pupil's education did not necessarily in itself constitute a sufficient argument for adaptations with respect to examinations. Compensation for deficiencies enabled pupils to pursue high-level studies in many cases. The rationale for adaptation of exam conditions was based on evidencing, by an appropriate neuropsychological examination, persistent deficiency in the ability to read and understand a text in a time interval compliant with the standard.

Jeanne-Marie URCUN noted that the circular cited covers all children presenting with a health disorder or handicap. The measures that can be envisaged under the circular are related to the consequence of a disorder in schooling and a higher level of individual performance should not deprive the pupil of the adaptations to which his/her handicap gives him/her a right. Franck RAMUS suggested that the slope with respect to compensating for a 'social disadvantage' is slippery. He suggested that the handicap should be compensated for and the handicap recognized as liable to evolve: the right to compensation may be justified at a given time and not justified later in the pupil's studies.

Catherine BILLARD wondered whether a child suffering from a handicap liable to evolve could have a right to adaptations without being recognized as having a handicap by the departmental establishment for handicapped people. Jeanne-Marie URCUN said that any request in that context was to be forwarded to the MDPH. The request is then reviewed by a physician designated by the MDPH. The physician meets with the child and his/her family in order to envisage the adaptations that could be implemented. The request for adaptation is then forwarded to the Education authority, which, in the very large majority of cases, responds positively. Valérie CAPUANO-DELESTRE considered that the parents could be advised to contact the National Education physicians of the school attended by the child. Submission to the MDPH would take place since it is scheduled in the procedure. However, that may vary depending on the department.

Bernard TOPUZ (Directorate of Childhood and the Family, Seine-Saint-Denis) reported that certain parents refuse referral to the departmental establishment for handicapped persons because they consider the disorder transitory. Bernard TOPUZ also stressed the inequalities related to the first psychological relationships in infancy and noted that the question raised earlier for the social aspects also needs to be raised for factors of a psychological nature. Jean-Marie DANION considered that there is a risk of 'dramatizing' the disorder in the parents' eyes. In his opinion, that situation calls for cautious wording of the recommendation. Ariel CONTE confirmed that handicap recognition constitutes a recurrent debate in all the associations. But the SESSAD cannot be set up without a clear definition of the handicap.

Christine GETIN (TDAH association) considered that the inequalities experienced in the field were particularly difficult. She argued in favor of a special effort with regard to the 'psychological education' of the parents, a concept that she prefers to that of 'guidance' for parents given that a good understanding of a disorder constitutes a first step towards its resolution.

**Those attending:** Maria de AGOSTINI (Expert, Epidemiology, Inserm), Rémy BAILLY (Centre Référent du Langage et troubles d'apprentissage, Pitié-Salpêtrière), Xavier BLANC (Association Les Lavandes, Orpierre), Fabienne BONNIN (Centre expertise collective Inserm), Christian BRECHOT (Director General, Inserm), Sandrine BROUSSOULOUX (INPES), Catherine BUTIKOFER (Association APEDA/FLA), Valérie CAPUANO-DELESTRE (Ministry of National Education), Patrick CHANSON (Deputy Director, Disc Inserm), Michèle CHARNAY (Association APEDYS), Anne-Marie CHARTIER (INRP), Dominique CHAUVIN (Centre Référent Pitié-Salpêtrière), Ariel CONTE (Association CORIDYS), Isabelle DABOVILLE (Syndicat enseignant-Union nationale des syndicats autonomes, SE UNSA), Jean-Marie-DANION (Professor of Psychiatry, Advisor to the Director General, Inserm), Georges DELLATOLAS (Inserm), Jean-François DEMONET (Expert, Neurology, Inserm), Dominique DONNET-KAMEL (Associate, Inserm-association, Disc), Marie-Christine DROCHON (SNMSU-UNSA, syndicat national des médecins scolaires et universitaires), Anne-Christine DUPONT (FNO/UNADREO), Jeanne ETIEMBLE (Director, Collective expert review center, Inserm), Michel FAYOL (Expert, Laboratoire de Psychologie sociale et cognitive, Université Clermont-Ferrand), Pascale GERBOUIN (Inserm), Christine GETIN (Association TDAH), Bernard GOSSOT (Académie Créteil), Barbara KNOCKAERT (SNUIPP), Anne LAUNOIS (Expert review assistance, Disc), Frédérique LEFEVRE (DGESCO), Marie-Thérèse LE NORMAND (Expert, Inserm), Sylviane LEWICK-DERAISON (UNADREO), Claire MELJAC (CHS Sainte-Anne-UPPEA), Anne MIGNOT (Press office, Inserm, Disc), Anne-Marie MONTARVAL (Association, APEDA/FLA), Isabelle NOCUS (Université Nantes UFR Psychologie), Anne-Laure PELLIER (Collective expert review center, Inserm), Christine PLOUZENNEC (AFPS), Franck RAMUS (Expert, Laboratoire sciences cognitives et psycholinguistique, ENS Paris), Scania de SCHONEN (Université Paris 5), Taraneh SHOJAEI-BROSSEAU (MGEN), Liliane SPRENGER-CHAROLLES (Expert, Laboratoire psychologie expérimentale, Boulogne), Anne TERLEZ (PEEP), Gérard TOUPIOL (FNAME, Fédération nationale associations maîtres E), Monique TOUZIN (CHU Bicêtre), Bernard TOPUZ (PMI), Jeanne-Marie URCUN (Physician, Education Nationale DGESCO),

Laurence VAIVRE-DOURET (Hopital Necker), Céline VRIDAUD, (FNAME, Fédération nationale associations maîtres E), Michel ZORMAN (CHU Grenoble).

**Apologies for absences:** Jean-Claude AMEISEN (Ethical Committee, Inserm), Pierre BAROUILLET (Expert, Université Genève), Marc BRODIN (Université Paris), Martine CARAGLIO (Académie de Paris), Claude CHEVRIE-MULLER (Inserm), Philippe CLAUS (Inspection EN), Stanislas DEHAENE (Inserm, CEA), Ghislaine DEHAENE-LAMBERTZ (CNRS), Michel DELEAU (Université Rennes), Agnès FLORIN (Université Nantes), Nicole GENEIX (Observatoire de l'enfance), Nicolas GEORGIEFF (Institut des Sciences Cognitives, Bron), Nathalie GUIGNON (DGS, DREES), Jean-Emile GOMBERT (Expert, Université Rennes), Michel HABIB (Expert, CHU Marseille), Gisèle JEAN (SNES-FSU), Irène KAHN-BENSAUDE (Ordre des médecins), Brigitte LEFEUVRE (DGS), Jean-Jacques LEMIRE (School psychologist), Philippe MEIRIEU (Université Lyon), Marie-Noëlle METZ-LUTZ (CNRS), Agnes MITTON (CMPP Corbeil Essonnes), Gille MOINDROT (SNUIPP), Dominique MOTHES (SNMSU), Philippe NIEMEC (SE-UNSA), Monique PLAZA (Université Paris), Hélène RABATE (SNPDEN), Jean-Charles RINGARD (Académie Nantes), Patrick ROUMAGNAC (SNIEN-UNSA), Martine SAFRA (Education nationale), Gérard SCHMIT (CHU Reims), Anne TURSZ (CNRS), Sylviane VALDOIS (Expert, Université Grenoble), Louis VALLÉE (CHRU Lille), Florence VEBER (Santé Mairie de Paris), Jean-Luc VILLENEUVE (SGEN-CFDT).

Inserm and the expert group would like to thank all the participants for the quality of the debates and would also like to thank the numerous representatives of the Education and Health sector who forwarded comments on the synthesis and recommendations. Their contributions have been taken into account in the final version of the document.

## Inserm collective expert review

### Methodology

An Inserm collective expert review<sup>14</sup> sheds scientific light on a given subject in the field of health on the basis of a critical analysis and synthesis of the international scientific literature. The expert reviews are implemented at the request of institutions wishing for access to recent research data pertinent to their decision-making process with respect to public policy. An Inserm collective expert review is to be considered as an initial stage that is necessary but most frequently not sufficient to result in decision-making. The conclusions of the collective expert review contribute to, but cannot replace, debate between the professionals involved or society debate if the questions addressed are particularly complex and sensitive.

At the request of a sponsor, the Inserm collective expert review may be accompanied by an 'operational' expert review addressing application of the knowledge and recommendations and taking into account contextual factors (existing programs, structures, players, training, etc.). The latter type of expert review elicits contributions from the players in the field able to respond to the feasibility aspects, representatives of the administrations or institutions responsible for promoting applications in the field involved, experts having contributed to the reviews, and representatives of patient associations. The sharing of varied cultures and experience enables a complementary approach to the collective expert review in an operational framework. Moreover, a variety of work (recommendations for good practices, public hearings, etc.) implemented under the auspices of the High Authority for Health (HAS) may follow an Inserm collective expert review.

Collective expert review has been an Inserm mission since 1994. Some sixty collective expert reviews have been implemented in numerous health fields. The Institute guarantees the conditions under which the expert review is implemented (exhaustiveness of the document sources, qualification and independence of the experts, transparency of the process).

The Inserm Center for Collective Expert Reviews organizes the various stages of collective expert review from the initial problem statement through to communication of the report, with the assistance of Inserm departments. The Center team, consisting of engineers, researchers and a secretariat, implements the document searches, logistics and chairing of the expert review meetings. The team contributes to the scientific writing and to compiling the expert review products. Regular exchanges with other public organizations (EPST) implementing the same type of collective expert review have enabled similar procedures to be set up.

#### **Problem statement**

The problem statement phase enables definition of the sponsor's request, checking that accessible scientific literature on the issue raised is available and drawing up specifications which state the framework of the expert review (status report on the perimeter and main themes of the subject), its duration and budget, documented by a convention signed by the sponsor and Inserm.

During the problem statement phase, Inserm also organizes meetings with patient associations in order to ascertain the questions those associations wish to have addressed and the data sources available to them. The information is incorporated in the scientific program of the expert review. For certain subjects, exchanges with industrial partners are indispensable in order to obtain access to complementary data not available in the databases.

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<sup>14</sup> Inserm accredited label

### **Expert review monitoring committee and assistance unit setup**

A monitoring committee consisting of sponsor and Inserm representatives is set up. The committee meets several times during the expert review to monitor the progress of the review, discuss any difficulties encountered in addressing the issues, ensure compliance with the specifications and examine any new factors in the regulatory and political context pertinent to the ongoing review. The committee also meets at the end of the expert review for presentation of the conclusions and prior to compilation of the final version of the report.

For expert reviews addressing sensitive issues, an assistance unit is also set up and consists in representatives of the Directorate General of Inserm, scientific board, ethical committee of Inserm, communication department, human and social science researchers and specialists in the history of science. The role of that unit is to identify, at the start of the expert review, the issues liable to have strong resonance for the professionals involved and civil society, and to suggest hearings of professionals in related fields, representatives of civil society and patient associations. In short, the unit is responsible for measuring the perception that the various recipients may have of the expert review. Before publication of the expert review report, the assistance unit pays special attention to the wording of the synthesis and recommendations, including, if necessary, the expression of the various points of view. Downstream of the expert review, the unit is responsible for strengthening and enhancing the circulation of the results of the expert review, for instance by holding colloquia or seminars with the professionals of the field and players involved or holding public debates with representatives of civil society. Those exchanges are to ensure enhanced understanding and adoption of the knowledge generated by the expert review.

### **Literature searching**

The specifications drawn up with the sponsor are translated into an exhaustive list of scientific questions reflecting the perimeter of the expert review with the assistance of referral scientists in the field and members of Inserm. The scientific questions enable identification of the disciplines involved and construction of a key-word arborescence employed in the systematic searching of international biomedical databases. The articles and documents selected on the basis of their pertinence with respect to answering the scientific questions constitute the document base, which is forwarded to the experts. Each member of the group is asked to add to the document base over the course of the expert review.

Institutional reports (parliamentary, European, international, etc.), raw statistical data, associations' publications and other documents from the gray literature are also inventoried (non-exhaustive) in order to complement the academic publications provided to the experts. The experts are responsible for taking or not taking into account those sources depending on the interest and the quality of the information supplied. Lastly, a review of the main articles in the French press is supplied to the experts during the expert review in order to enable them to follow developments on the theme and the social repercussions.

### **Constitution of the expert group**

The expert group is formed on the basis of the scientific skills necessary for analysis of the bibliography collected and on the basis of the complementarity of the group members' approaches. Since an Inserm collective expert review is defined as a critical analysis of the academic knowledge available, the choice of the experts is based on their scientific skills certified by publications in peer-review journals and their recognition by their peers. The expert recruitment logic, based on scientific skills and not on knowledge in the field, is to be stressed in that it is a frequent source of misunderstandings when the expert reviews are published.

The experts are selected from the French and international scientific community. They are to be independent of the partner sponsoring the expert review and recognized pressure groups. The composition of the expert group is validated by the Directorate General of Inserm.

Several scientists outside of the group may be requested to contribute occasionally to a particular theme during the expert review.

Expert review implementation lasts between 12 and 18 months, depending on the volume of literature to be reviewed and analyzed and the complexity of the subject.

### **Initial expert group meeting**

Before the first meeting, the experts receive a document explaining their mission, the scientific program (issues to be addressed), schedule, the expert review bibliographic database to date and articles more specifically addressing certain experts on the basis of the skills.

During the first meeting, the expert group discusses the list of issues to be reviewed and completes or modifies it. The group also examines the document base and proposes supplementary searches with a view to enriching that base.

### **Expert critical analysis of the literature**

During the meetings, each expert orally presents a critical analysis of the literature with respect to the aspect allocated to the expert in his/her field of expertise and communicates the accepted facts, uncertainties and controversies with respect to current knowledge. The questions, remarks and points of convergence or divergence elicited by the group analysis are taken into consideration in the section that each of the experts compiles. The analysis report, consisting of various sections, thus constitutes the state of the art for the various disciplines pertinent to the issue under review. The bibliographic references used by the expert are cited in and at the end of each section.

### **Synthesis and recommendations**

The synthesis summarizes the broad lines of the literature analysis and identifies the main findings and principles. Contributions from contributors outside the group may be summarized in the synthesis.

The synthesis is more specifically intended for the sponsor and decision-makers with a view to use of the knowledge presented therein. The wording of the synthesis is to take into account the fact that it will be read by non-scientists.

As of report publication, the synthesis is posted on Inserm's website. The synthesis is translated into English and posted on the NCBI/NLM site (National Center for Biotechnology Information of the National Library of Medicine) and Sinapse site (Scientific INformAtion for Policy Support in Europe, European Commission site).

If requested by the sponsor, certain collective expert reviews include 'recommendations'. Two types of 'recommendations' are formulated by the expert groups. 'Principles for action' based on a validated scientific reference system with a view to defining future public health action (mainly in screening, prevention and management) but which are not under any circumstances to be considered 'operational' recommendations insofar as no economic or political components have been taken into account in the scientific analysis. 'Research orientations' are also proposed by the expert group with a view to filling in the gaps in scientific knowledge observed during the analysis. Once again, these proposals cannot be considered 'priority' research without their being put into perspective. That is the task of the pertinent authorities.

### **Critical review of the report and synthesis by prominent 'readers'**

For certain expert reviews addressing sensitive subjects, a critical reading memorandum is requested from several prominent 'readers' selected on the basis of the scientific or medical knowledge and managing or evaluating French or European research programs or having contributed to ministerial working groups. Similarly, the report and synthesis (and recommendations) may be submitted to figures with good knowledge of the 'field' and able to grasp the socioeconomic and political issues associated with the knowledge (and proposals) presented in the expert review.

### **Presentation of the conclusions of the expert review and debate**

A seminar open to the various sectors involved in the subject of the expert review (patient associations, professional associations, unions, institutions, etc.) enables an initial debate on the conclusions of the expert review. On the basis of that exchange, the final version of the synthesis document incorporating the various viewpoints expressed is compiled.