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Review article

Hyperlexia: Systematic review, neurocognitive modelling, and outcome

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ABSTRACT

Hyperlexia is defined as the co-occurrence of advanced reading skills relative to comprehension skills or general intelligence, the early acquisition of reading skills without explicit teaching, and a strong orientation toward written material, generally in the context of a neurodevelopmental disorder. In this systematic review of cases (N = 82) and group studies (including 912 participants of which 315 are hyperlexic), we address: whether the hyperlexic profile is associated with autism and why, whether models of non-autistic reading can teach us about hyperlexia, and what additional information we can get from models specific to autistic cognitive functioning. We find that hyperlexia, or a hyperlexic-like profile, characterises a substantial portion of the autistic spectrum, in which the subcomponents of the typical reading architecture are altered and dissociated. Autistic children follow a chronologically inverted path when learning to read, and make extended use of the perceptual expertise system, specifically the visual word form recognition systems. We conclude by discussing the possible use of hyperlexic skills in intervention.

1. Introduction and definition

The term "hyperlexia" was coined in 1967 by Silberberg and Silberberg (1968), Silberberg and Silberberg (1967). Four features consistently describe hyperlexia: (1) the presence of an accompanying neurodevelopmental disorder; (2) advanced reading skills, relative to comprehension skills or general intelligence; (3) an early acquisition of reading skills without explicit teaching; and (4) a strong orientation toward reading material (Needleman, 1982). The same year, Rutter and Lockyer noticed that the same proportion of both their groups of "psychotic children" and control children could read at a normal level for their age, despite very little schooling for the "psychotic" group. They described the example of a boy who could read well above the average level, although he had been excluded from school at the age of six (Rutter and Lockyer, 1967). However, cases of hyperlexia were described as early as 1930 (Phillips), i.e. before the introduction of both hyperlexia and autism as a syndrome. Mentions of the hyperlexic profile can be found even earlier, in 1918, when Hollingworth and Winford wrote, "Cases where a generally stupid (*sic*) child is innately gifted with special ability to master the mechanics of reading, for example, are no doubt as frequent as cases where a generally capable child learns them with difficulty." (Hollingworth and Winford, 1918; cited in Thompson, 1966; p.24).

Fifty years after the term was first introduced, hyperlexia is often reported as one of the savant abilities in autism. However, its neurocognitive underpinning and how it relates to autistic cognition and typical reading acquisition, are yet to be established. Literature on hyperlexia consists of a large number of descriptive reports, combined with a small number of empirical studies testing specific assumptions about its prevalence and mechanisms. Beyond the established cross-sectional differences in decoding and understanding abilities, the distinct, developmental pathway of hyperlexics remains to be characterized. The last academic review on hyperlexia was published 14 years ago (Grigorenko et al., 2003). Multiple case and group studies have been published in the last decade, including the first imaging studies of hyperlexia, making a new review necessary.

Based on a systematic review, we first report the current state of knowledge on the prevalence of hyperlexia and its relation with autism. We present the cognitive processes underlying reading in non-hyperlexic, mature readers, and the sequence of typical reading acquisition.

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Table 1

Systematic review of hyperlexic case reports – *The number of criteria for hyperlexia refers to the four-feature definition given in section 1. **The oral language level is classified based on the indications used to determine the ADOS module: 1 refers to individuals with *no speech*, or who do not consistently use phrase speech; 2 refers to individuals with some *phrase speech* (defined as non-echoed three-word utterances that sometimes involve a verb and are spontaneous, meaningful word combinations), but not verbally fluent; 3 refers to fluent speech. *Verbal fluency* is broadly defined as having the expressive language of a typical four-year-old child: producing a range of sentence types and grammatical forms, using language to provide information about events, and producing logical connections within sentences (e.g., "but" or "though"). ***The autistic diagnosis is determined as explained in 3.2. Additional information on the scoring system can be obtained from the authors on request.

Publication	Case	Gender	Age at time of report (y = years, m = months)	Hyperlexic features mentionned* (1–4)	Reported onset of hyperlexia	Oral language level**	Autism-PDD mentionned	Autistic traits based on description***	Other condition mentionned-not autistic	Not enough information for autism diagnosis
Phillips (1930)	Gordon	M	10y	3	NA	2		X		
Scheerer et al. (1945)	L	M	11y	3	Before 5y	1		X		
Silberberg and Silberberg (1968)	Case 1	F	7y1m	4	NA	2				X
	Case 2	M	Grade 4	4	NA	2				X
	Case 3	F	3y	4	18 months	2	X			
	Case 4	M	Grade 2	4	NA	1				X
	Case 5	M	8y2m	4	NA	3				X
	Case 6	M	Grade 3	4	NA	3				X
Cain (1969)	Case 1: Millie	F	6y5m	4	NA	1	X			
	Case 2: Janey	F	7y	4	NA	2	X			
	Case 3: George	M	8y	4	NA	1	X			
Goodman (1972)	Case: Sam	M	7y11m	4	Before 4y	2	X			
Mehegan and Dreifuss (1972)	Case 1: 6 6.0	M	6y	4	NA	1	X			
	Case 2	M	7y	4	3y5m	3	X			
	Case 3	M	9y	3	6y	1	X			
	Case 4: 8.0	M	8y	3	5y	1	X			
Huttenlocher and Huttenlocher (1973)	Case 1: M.K.	M	7y	3	4y	1	X			
	Case 2: C.O.	M	4y11m	4	3y	1	X			
	Case 3: B.D.	M	5y	3	5y	1	X			
Cobrinik, 1974	Case 1: Anthony B.	M	12y6m	4	2y	1	X			
	Case 2: Robert C.	M	14y6m	4	Before 5y	2	X			
	Case 3: Terence D.	M	14y	4	Before 5y	2	X			
	Case 4: John M.	M	15y	4	Before 5y	1	X			
	Case 5: Malcolm P.	M	12y	4	Before 5y	2	X			
	Case 6: William W.	M	14y	4	Before 5y	3	X			
Elliott and Needleman (1976)	Case 1: V.	F	5y8m	3	1y3m	1		X		
Aram et al. (1984)	Case: MD	M	39y	4	4y5m	2	X			
Goldberg and Rothermel (1984)	Case 1: M.H.	F	17y8m	4	3y	2		X		
	Case 2: D.M.	M	6y8m	4	3y	2		X		
	Case 3: C.K.	M	8y8m	4	3y6m	1		X		
	Case 4: D.S.	M	5y3m	4	Before 4y	1		X		
	Case 5: T.M.	M	12y6m	3	4y6m	2	X			
Goldberg and Rothermel (1984) **(continued)**	Case 6: C.R.	M	10y1m	4	2y	2		X		
	Case 7: L.N.	M	5y2m	4	3y	2		X		
	Case 8: A.C.	M	12y4m	3	2y	2	X			
Siegel (1984)	Case: A.E.	F	7y9m	3	3y	1		X		
Burd et al. (1985)	Case: B.F.	M	7y	4	4y	2	X			
Burd and Kerbeshian (1985)	Case: W.	M	11y	4	NA	2	X			
Cossu Marshall (1986)	Case 1	F	12y5m	3	NA	2			X	
	Case 2	F	18y6m	3	NA	2			X	
Burd et al. (1987)	Case 1: A.	F	NA	4	Before 3y	1	X			
	Case 2: R.	M	NA	4	4y	2	X			
	Case 3: E.	M	28y	4	5y	2	X			
	Case 4: B.	M	NA	4	3y	2	X			
Lebrun et al.	Isabelle	F	9y	3	5y	3				X

(continued on next page)

Table 1 *(continued)*

Publication	Case	Gender	Age at time of report (y = years, m = months)	Hyperlexic features mentionned* (1–4)	Reported onset of hyperlexia	Oral language level**	Autism-PDD mentionned	Autistic traits based on description***	Other condition mentionned-not autistic	Not enough information for autism diagnosis
(1988)										
Smith and Bryson (1988)	Jon	M	7y2m	4	3y	1	X			
	Jay	M	7y2m	4	3y	1	X			
Burd and Kerbeshian (1988)	Sister	F	NA	4	3y6m	1	X			
	Brother	M	27y	4	3y	1	X			
Burd and Kerbeshian (1989)	1	M	NA	4	Before starting school	NA			X	
Ichiba (1990)	Case 1	M	11y	4	3y	2		X		
	Case 2	F	10y	4	3y	3		X		
Patti and Lupinetti (1993)	Case 1: Vera	F	22y	4	5y	2	X			
O'Connor and Hermelin (1994)	Case 1: Neil	M	8y	3	3y	3	X			
	Case 2: Christopher	M	5y	3	3y6m	3	X			
Worthy and Invernizzi (1995)	1	F	14y	3	"early"	NA		X		
Sparks (1995)	Case 1: D.Z.	M	9y	4	2y	2		X		
	Case 2: R.L.	M	8y	4	2 or 3y	2		X		
	Case 3: G.M.	F	10y	4	1y6m	3		X		
Glosser et al. (1996)	LA	M	6y	4	3y5m	3			X	
Kennedy (2003)	DS	M	15y	4	NA	2			X	
	HN	M	19y	4	NA	1				X
Turkeltaub et al. (2004)	Case: Ethan	M	9y	4	3y6m	2	X			
Craig and Telfer (2005)	Jason	M	5y	4	1y6m	1	X			
Jensen (2005)	Isaakk	M	NA	4	2y	2	X			
Talero-Gutierrez (2006)	Case1	M	2y	4	3y	2	X			
	Case 2	M	7y	4	4y	1	X			
Atkin and Lorch (2006)	Case 1: Paul	M	4y3m	4	3y	1	X			
Castles et al. (2010)	Case 1: JY	M	10y4m	4	Kindergarten	2	X			
	Case 2	M	8y2m	4	NA	2	X			
Joshi et al. (2010)	MS	M	16y	3	NA	3				X
Cardoso-Martins and da Silva (2010); study 2	Case 1	M	3y	4	2y	1	X			
	Case 2	M	3y	4	2y	1	X			
Lam & nica et al. (2013)	P1	M	4y4m	3	2y6m	3	X			
	P2	M	4y6m	3	3y	3	X			
	P3	M	5y	3	2y	3	X			
	P4	M	4y6m	3	Before 3y	3		X		
	P5	M	5y2m	3	Before 3y	3	X			
	P6	M	4y9m	3	2y6m	3		X		
Cardoso-Martins et al. (2013)	1	M	3y11m	4	NA	1	X			
Pacheva et al. (2014)	1	F	13y	3	NA	1	X			
Johnels and Miniscalco (2014)	1	M	7y	4	NA	2	X			
	Total cases 82	% M 79.27					Total 51	Total 18	Total 5	Total 8

Table 2
Systematic review of group studies on hyperlexia – *The number of criteria for hyperlexia refers to the four-feature definition given in Section 1.

Publication	N total	N with hyperlexia	Gender M	Gender F	Age range (years.months) or school grade level (year)	Reported diagnosis	Hyperlexic features mentionned* (1–4)
Silberberg and Silberberg (1971)	28	28	NA	NA	Preschool to grade 4.5	Varied (Autism, schizophrenia, cerebral dysfunction, hyperactivity, anxiety)	3
Mehegan and Dreifuss (1972) (4 detailed cases included in Table 1)	12	12	11	1	5.0 to 9.0	Mental retardation, psychotic and autistic features	4
Richman and Kitchell (1981)	10	10	8	2	5.9 to 9.7	Various (autism, language delay, hyperactivity, learning disability)	3
Healy et al. (1982)	12	12	11	1	5.0 to 11.0	Behavioral problems associated with autism	4
Cobrinik (1982)	9	9	9	0	9.8 to 13.2	Various (childhood schizophrenia, autism with mental retardation, psychosis at early onset, chronic brain syndrome with mental reta	4
Fontenelle and Alarcon (1982)	8	8	7	1	4.0 to 17.0	Primary autism, mental retardation, and language disorder	3
Whitehouse and Harris (1984)	20	20	20	0	13.0 to 25.0	Infantile autism	4
Snowling and Frith (1986);Exp 1	16	8	NA	NA	11.0 to 19.0	Autism	2
Snowling and Frith (1986); Exp 2, 3,4	40	NA	NA	NA	8.0 to 21.7	Various (autistic, mildly retarded, typical)	2
Welsh et al. (1987)	5	5	5	0	4.5 to 10.1	Autism or PDD	3
Tirosh and Canby (1993)	10	5	4	1	11.0 to 16.0	Autism	2
Aram (1997)	12	12	12	0	7.11 to 13.7	Delayed language, attentional deficits and/or psychosocial problems including autism	3
Cohen et al.(1997)	62	16	42	20	Mean 6.45	SLI and Hyperlexia, ruling out PDD	3
DeLong et al.(2002)	129	24	NA	NA	2.0 to 8.0	Autism	4
Grigorenko et al.(2002)	80	12	68	12	2.6 to 12.8	Developmental delay, PDD	2
Nation et al.(2006)	41	10	36	5	Mean 10.33	Autism	2
Newman et al.(2007)	40	20	36	4	3.0 to 19.75	Autism	3
Jones et al.(2009)	100	14	91	9	14.0 to 16.0	Autism	2
Saldaña et al.(2009)	36	14	25	1	Mean 13.9	Autism	2
Cardoso-Martins and da Silva (2010); study 1	18	6	17	1	6,2	Autism	4
Davidson and Weismer (2014)	94	58	82	12	2.0 to 6.0	Autism or PDD	2
Wei et al. (2015)	130	12	112	18	6.0 to 9.0	Autism	2

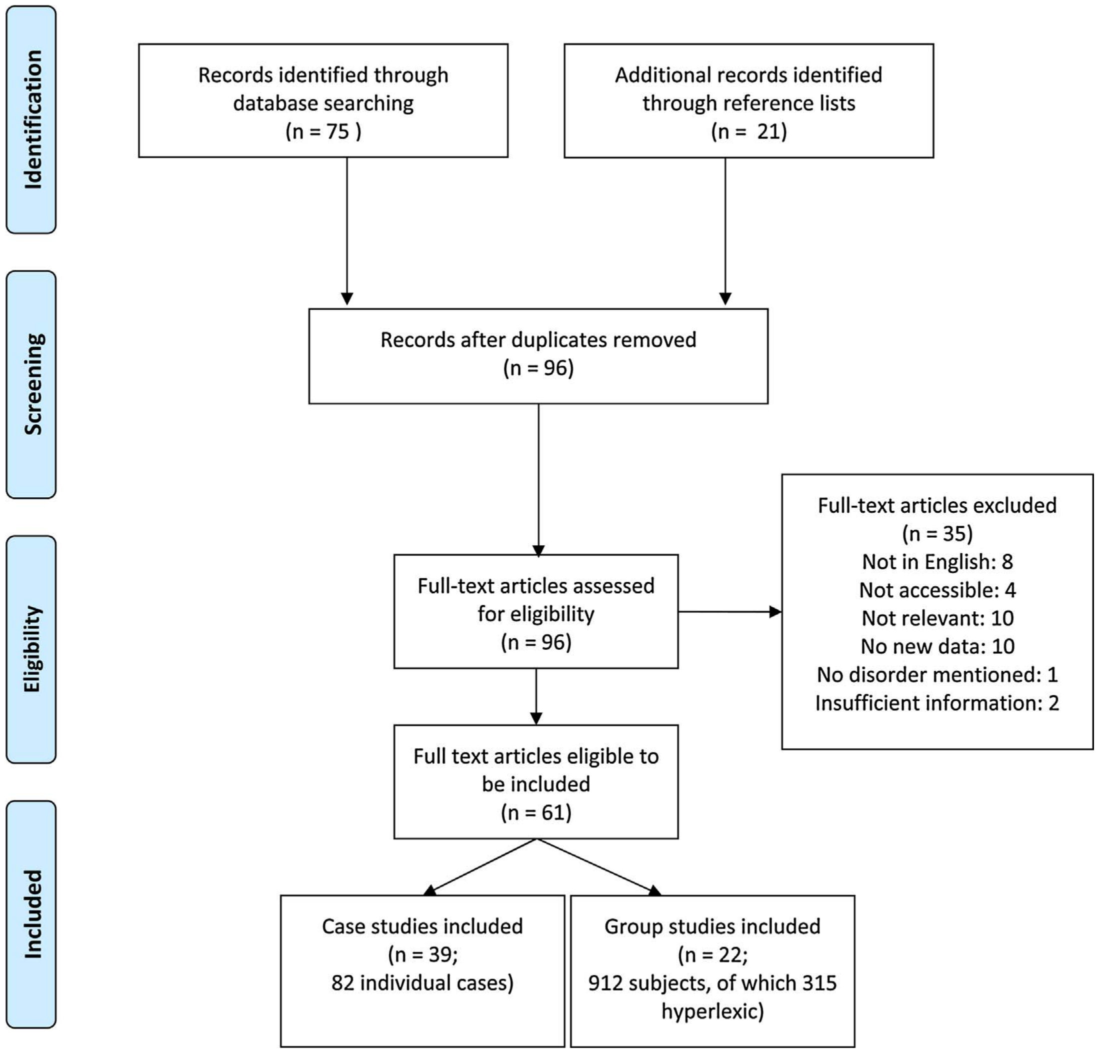

Fig. 1. Flow diagram based on PRISMA statement (www.prisma-statement.org).

We then describe how these processes differ in autism and hyperlexia. Last, we provide a summary of our current understanding of this phenomenon, its predictive value on future language abilities of hyperlexic children and how it could inform intervention decisions.

2. A systematic review: method

2.1. Literature search

We conducted a systematic review of single case descriptions (Table 1) and group studies (Table 2) reporting on hyperlexic children and adults. We used the PRISMA statement, a checklist for the reporting of systematic reviews, as a guideline to conduct the literature search (Moher et al., 2009). We followed the PRISMA 2009 Checklist while excluding the items that did not apply to the type of material we reviewed (i.e. items specific to intervention research).

The search terms (hyperlexia OR hyperlexic*) with no filter for the date were first used in January 2016 to find articles on Pubmed. The literature search and selection continued until May 2016. A few references were added after revisions in March 2017. Unpublished material was not explored. Publications were first selected based on the relevance of the title and abstract by A.O. and P.J., and then imported into Mendeley Desktop 1.16.1. for further reading and verification for inclusion criteria. The reference lists of included articles were cross-checked for potential additional articles. All studies included in the review were verified twice for inclusion criteria by A.O. and L.M.

2.2. Selection criteria

The papers that did not contain any new data (e.g. reviews), or at least a brief description of reading abilities and behavior were excluded, as well as papers that were not available in English. We only included articles that mentioned a neurodevelopmental disorder associated with the hyperlexic profile (see feature (1) in Section 1. and more about the definition in Section 6.1.) in accordance with the four-feature definition of hyperlexia.

Developmental and diagnostic information about hyperlexic individuals were extracted from the clinical observations contained in the articles. We examined the descriptions for mention of DSM-5 autism spectrum disorders criteria (American Psychiatric Association, 2013) (see 3.2) when no autism diagnosis was mentioned. The scarcity of information on individual phenotypes in group studies did not allow for this individual screening. Thus, case and group studies are reported separately in Table 1 and Table 2, respectively.

We extracted the demographic information (gender, age or age range, explicit diagnosis) for each case and group study. For group studies, we extracted the number of participants, the proportion of participants with hyperlexia in the group, the criteria used to detect it, and the oral language level when available. For case studies, we collected information relevant to the autism diagnosis, the number of hyperlexic features described in the text, the perceived age of reading onset if mentioned, as well as information on the intellectual and cognitive abilities, and oral language level of the participant. We also extracted developmental information and outcome, when available. The oral language level of the subjects was classified into one of three categories, similar to those used to select which ADOS (Autism Diagnosis Observation Schedule; Lord et al., 1989) module to use during diagnostic assessment (Table 1; details in the legend).

It was not possible to calculate a mean age in either table: in case studies, ages were expressed in different ways with different levels of precision, ranging from school grade to the precise number of years and months. Individuals were also sometimes described at different ages in the same paper. In most group studies, only an age range was given, and the mean age was often not reported.

We wanted to estimate the ratio of hyperlexic individuals with an autistic phenotype. The labels *autism*, *childhood schizophrenia,* or *early onset psychosis* or *infantile psychosis* were considered to be synonymous up to 1975 in our classification. When none of these were mentioned, two clinical experts (L.M. and B.F.A.) estimated whether the information provided on symptoms was sufficient to estimate a diagnosis according to DSM-5 criteria for autism (American Psychiatric Association, 2013).

2.3. Results

The initial search resulted in 75 papers. This search was complemented by scanning the references of the papers and applying our criteria to the additional potential articles. Four papers did not include the word hyperlexia but were cited in other papers about hyperlexia because the cases reported presented similar symptoms. This led to the screening and inclusion of an additional 21 articles and book chapters matching our criteria after full-text selection. Of these 96 articles, 61 remained after screening for inclusion criteria. A total of 58 papers and two book chapters were included in the review: 39 case reports that include detailed information on 82 individual cases, and 22 group studies covering 912 subjects, of which 315 were hyperlexic (Fig. 1).

3. Prevalence and relation with autism

3.1. Prevalence of hyperlexia in autism

There is no information available on the rates of hyperlexia in the general population. Data on the prevalence of hyperlexia is limited to its prevalence in pervasive developmental disorders (PDD) due to the antecedence of the description of hyperlexia to that of autism, the intertwinement of hyperlexia and neurodevelopmental disorders, and the increasing interest in hyperlexia in relation to autism. Estimates ranging from 6 to 20% were reported in the four studies that tried to estimate the prevalence of hyperlexia in PDD or autism using varying criteria to define hyperlexia. Based on a personal estimation and a stringent definition of hyperlexia, as well as of autism, as defined in DSM-3 (American Psychiatric Association, 1980), Burd and Kerbeshian (1985) estimated hyperlexia to be present in approximately **6%** of children on the autism spectrum. Wei et al. (2015) tested the reading profiles of 130 children with autism aged six to nine years old. They found a prevalence of **9.2%** for hyperlexia, defined by a somewhat broader definition: the discrepancy between word identification/rapid letter naming, and comprehension. Jones et al. (2009) adopted less stringent criteria (i.e. discrepancy between reading and IQ at the 10th percentile of the general population) and found that **14.1%** of their sample fit a hyperlexic profile. The highest prevalence (**20.7%**) was reported by Grigorenko et al. (2002). Their criteria were even more flexible, as children were considered hyperlexic if they met two out of the three following criteria: (1) standardized reading/decoding score at least two standard deviations above the level of intelligence; (2) age-equivalent reading/decoding score at least two years above the age-equivalent level of intelligence; (3) confirmation by clinical observations and evaluations. Information on the third criterion was very rarely available; hence, being qualified as hyperlexic mostly relied on the discrepancy between reading/decoding scores and intelligence levels. In summary, the reported prevalence of hyperlexia increases when the stringency of the criteria used diminishes (see Fig. 3).

The hyperlexic participants in the case studies we reviewed were 79% males (Table 1), a sex-ratio similar to that reported in autism throughout the 20th century. It is superior to the current reported sex-ratio in autism (approximately three males for one female, Zablotsky et al., 2015), which may be explained by improvements in the diagnosis of autism in females. A measurement of intelligence was reported in a subset of studies. We were not able to compare IQ between studies, nor compute an estimate of average intelligence, due to a high level of inconsistency between measurement methods. However, intelligence was most frequently in the normal range when measured by non-verbal tests – consistent with prototypical non-syndromic autism (Dawson et al., 2007).

We questioned whether hyperlexia was more frequently associated with a specific phenotype in the autism spectrum given its known heterogeneity. Surprisingly, although a delay in speech comprehension was present in most of the hyperlexic cases, which is inconsistent with the absence of speech-onset delay in Asperger Syndrome, six of 82 cases were diagnosed with Asperger syndrome, raising questions on the criteria that had been used to diagnose both Asperger Syndrome and hyperlexia. Asperger syndrome has been associated with high levels of reading skills across both decoding and comprehension (Huemer and Mann, 2010) which is not consistent with the definition for hyperlexia.

3.2. Prevalence of autism in hyperlexia

Hyperlexia is most commonly associated with the Autism Spectrum (AS), or at least autistic-like features (Grigorenko et al., 2003, 2002; Healy and Aram, 1986; Jones et al., 2009; Nation, 1999), although it has sometimes been reported in the absence of autism (Nation et al., 2002) or in association with other neurodevelopmental conditions. There are anecdotal reports of hyperlexia in Down syndrome (Cossu et al., 1993), Turner syndrome (Temple and Carney, 1996), and in various other neuro-developmental conditions (Fontenelle and Alarcon, 1982; Worthy and Invernizzi, 1995) not mutually exclusive with autism.

In our review, 51 of 82 (63.41%) case studies had an explicit autism diagnosis or explicitly mentioned autistic features. In the remaining 31 cases described without explicitly mentioning autism or autistic features, eight case studies did not contain sufficient information to score DSM-5 criteria, five presented a neurodevelopmental condition other than autism, but could not be qualified as autistic according to our criteria, and 18 suggested the presence of three or more out of seven DSM-5 criteria. This classification process resulted in a total of 69 of the 82 (84.15%) cases with either autism or several autistic features, confirming the strong association between autism and hyperlexia.

4. Typical reading

We first review the neurocognitive underpinning of reading in typical fluent readers and reading acquisition in typical development and then review the emerging evidence on neurocognition in autistic hyperlexia. Reading is defined here as getting meaning from print (Rayner et al., 2001). Although it is a complex process that calls on a wide range of brain areas, three main neural systems are thought to be involved in reading: (1) recognition of words, based on their visual characteristics, supported by the occipito-temporal cortex; (2) conversion from graphemes to phonemes, based on the parieto-temporal region; and finally (3) access to the lexicon and the meaning of words and sentences, supported by ventral and dorsal temporal neural systems of the left hemisphere (Dehaene and Cohen, 2007; Shaywitz and Shaywitz, 2008). These reading systems are not to be understood as a chronological sequence during reading (e.g. lexical access is involved from the beginning through word recognition), nor as a developmental

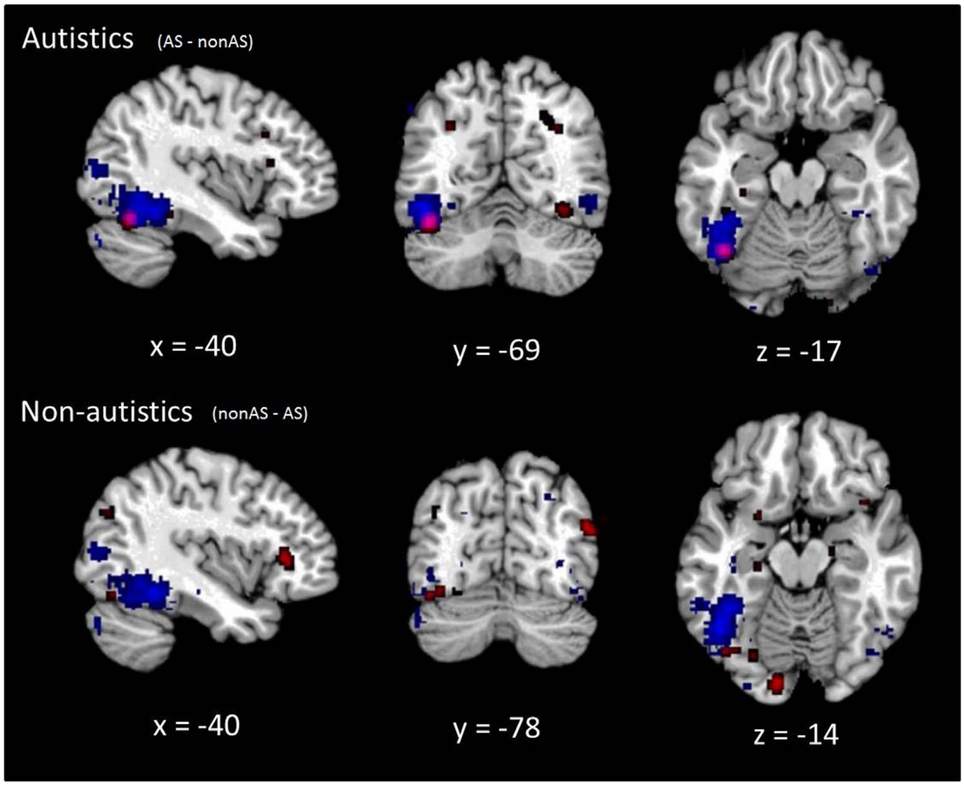

Fig. 2. Reading in the autistic brain – Overlap (pink) between results of an automated *meta*-analysis of 35 studies (N = 576) for 'visual word form area' (blue) generated using http://neurosynth.org, and regions showing differential activity between autistic and control groups (red) for word-related tasks in an ALE *meta*-analysis (81 autistics, 88 non-autistics) (Samson et al., 2012). (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article.)

sequence of acquisition (e.g. children speak before they read). We will present the main model explaining how these three systems typically function, followed by a summary of how these different steps mature in development.

4.1. Neurocognitive models of reading

4.1.1. Visual word form recognition

Visual word form recognition is a level of visual processing that extracts invariant information about the structure of visual words to shape a perceptual object (McCandliss et al., 2003). Expert readers need less than 250 ms to extract complex information from written words and access their grammatical features and meaning, despite large variations in font, size, colour, and location (McCandliss et al., 2003). The Visual Word Form Area (VWFA), located in the left occipito-temporal sulcus, is responsible for the rapid extraction of this abstract orthographic representation of ordered strings of letters (Dehaene and Cohen, 2011). This area is activated preferentially when written words or pseudo-words are presented, relative to other perceptually similar control stimuli (Gauthier et al., 2000; McCandliss et al., 2003; Szwed

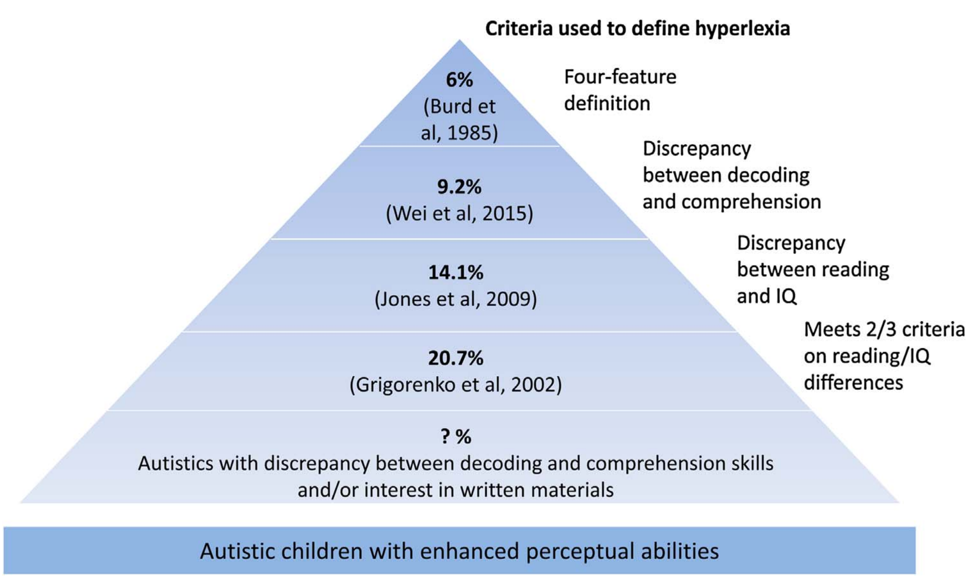

Fig. 3. Prevalence of hyperlexia and enhanced perceptual abilities – The reported prevalence changes with the stringency of the criteria employed. Less pronounced hyperlexic profiles may be much more frequent. Autistic children have strong perceptual skills from which hyperlexia could emerge.

et al., 2011), whether this presentation is conscious or unconscious (Dehaene et al., 2001). This activation is thought to be invariant for spatial location, with comparable activation patterns being observed for words presented in the left or right visual field (Cohen et al., 2002, 2000). The VWFA response is also case-insensitive: alternating-case words such as "tAbLe" activate the VWFA in the same way as visually familiar pure-case stimuli do (Polk and Farah, 2002). Selective disruption of the VWFA following surgery leads to pure alexia (an isolated reading deficit in the absence of other language impairments) (Cohen et al., 2004).

The VWFA is not only sensitive to the perceptual form of familiar words, but also to orthographic regularities. Real words or pronounceable pseudo-words activate the VWFA more than strings of consonants (Cohen et al., 2002; Price et al., 1996). Strings of letters violating the orthographic properties of the language activate the VWFA less, showing that it acquires its functional specialization according to arbitrary, culture-determined language properties (Dehaene et al., 2004, 2001). Real letters also activate the VWFA more than letter-like forms (Petersen et al., 1990), or letters of an alphabet that the subject cannot read (for example, Hebrew or Chinese characters for English readers) (Baker et al., 2007). In summary, the VWFA is an area shaped by reading experience (Baker et al., 2007), preferentially activated by pronounceable strings of letters, regardless of their size, position, CASE, or font.

4.1.2. Grapheme-to-phoneme conversion

The second step of reading is the conversion of visual linguistic information (graphemes) to oral linguistic information (phonemes), a process called grapheme-to-phoneme conversion. Two routes were initially proposed to explain how written text is transformed into pronounceable speech (Forster and Chambers, 1973; Marshall and Newcombe, 1973): a lexical route and a sublexical route. These two routes are controlled in the brain by two distinct, but interrelated, parieto-temporal neural systems: the more ventral lexical mechanism, associated with semantic access, and the more dorsal sublexical mechanism (Shaywitz and Shaywitz, 2008). The dual-route model has been elaborated and completed (Coltheart et al., 2001) based on the presence or absence of access to semantics.

The lexical route draws on the representations of printed words stored in the mental lexicon. When the reader sees a word, the visual form recognition system, presented above, extracts information about this perceptual object. If the word exists in the reader's orthographic 'library', the corresponding representation in the phonological lexicon is activated, and the word is read aloud. This route allows the reading of irregular words (such as *night* or *thought*) because their spelling does not follow the rules of the language; hence they require the intervention of the lexical route, i.e. previous knowledge of the words.

The sublexical route does not call upon the mental lexicon. Made-up pseudowords that the reader has never seen before have to be read out loud, employing the sublexical route. Using this path, the reader maps an unknown word serially from left to right and associates each letter in the letter string to its corresponding sound to read the word aloud. Expert readers have the ability to unconsciously switch between these two routes depending on the nature of the reading material.

4.1.3. Semantic access

Access to meaning is achieved when fluent decoding of printed material triggers previously acquired semantic knowledge. Semantic access involves a large network of interrelated systems in the left hemisphere, including the occipito-temporal, middle temporal, and premotor cortex (Shaywitz and Shaywitz, 2008). Semantic access may be limited in non-fluent readers by the effort put into decoding written materials. Comprehension may be limited to individual words, at the expense of higher level skills such as integration and inference, due to novice readers devoting most of their attention to their reading accuracy (Snyder et al., 2005). Hence, there is a strong correlation between reading accuracy and reading comprehension in children with typical language development (Nation and Snowling, 1997), even when they present learning problems (Shankweiler et al., 1999).

During typical reading, expectations related to phonology and meaning are involved from the early processing stages on (Wilson et al., 2011). Word reading skills have been linked to good oral vocabulary (Nation and Snowling, 2004), although not all reading skills are associated with vocabulary (Ricketts et al., 2007). High-imageability words (i.e. with rich semantic representations) are read more accurately than abstract words (Strain et al., 1995). Hence, perceptual, decoding, and semantic processes interact very early in the processing of written stimuli and cannot be easily distinguished.

Imaging studies have attempted to isolate the neural correlate of semantic access by contrasting visually presented words and pseudowords, although pseudowords may trigger semantic processing if they are similar to real words. A recent *meta*-analysis of these studies identified a semantic network encompassing the left middle temporal gyrus, angular gyrus, and inferior temporal gyrus (McNorgan et al., 2015).

4.2. Reading acquisition in typical development

Learning to read is defined here as the process of acquiring the skills that allow the child to identify and understand printed words. Competing theories of reading development all share the idea that many heterogeneous experiences lead to the acquisition of reading ability. Children typically progress through successive stages defined by different decoding strategies (Gough and Hillinger, 1980; Gough and Juel, 1991). At an early age, children start acquiring a wide variety of skills that will be used for reading later. Among these skills, the child's oral language abilities appear to be one of the most important factors for reading development in neurotypical children (Rayner et al., 2001).

Typical children are able to label and recognize letters at four years of age, when they already speak fluently. Precocious readers may learn to read words without external support starting from around three years of age (Fletcher-Flinn and Thompson, 2000; Jackson, 1988; Henderson et al., 1993), but most typically developing children have to be taught to read in an analytic, letters-to-words fashion. They mostly start with "baby writing" (writing what they hear) with spelling errors and incorrect pronunciation of irregular words (Cardoso-Martins et al., 2013; Cardoso-Martins and da Silva, 2010). Children first learn to recognize words by making associations between visual word forms and spoken words. During the 'selective association' stage, children acquire phonology, grammar, word meaning knowledge, and the social use of oral language. Children at this stage will often be found 'reading' a story while appropriately turning pages of a book and telling what they remember from previous readings, without any formal knowledge of decoding. In summary, children in the selective association stage associate the appearance of the printed pages of the book with the words they hear, but they do not decode the words before they learn the basics of decoding and letter-sound associations. This may explain why their 'decoding' is still font or context dependant (Dehaene, 2007).

Children adopt a new strategy based on the *alphabetic principle* around the age of five. They explicitly access the association between written symbols that compose the alphabet and speech sounds (Rayner et al., 2001) during the *cipher* or alphabetic stage (Frith, 1985). Novice readers usually know the most common letter combinations and frequent irregular words, although their pronunciation when reading is slow and not confident, and they understand the idea that print contains meaning. Their vocabulary increases very fast, approximately seven words per day in elementary school (Nagy and Herman, 1987), starting from 2500 to 5000 words when they are five years old (Beck and McKeown, 1991).

Children finally become *decoding readers* around the age of seven when they build fluency and confidence while reading. At this stage, they learn how to decode 3000 new words. Only then are typical children able to read more complicated irregular patterns of letters, such as vowel pairs. When developing their decoding skills, children first concentrate on processing individual words to decode the text, without focusing on the meaning of the sentences being read. As their decoding improves and becomes automatic, children increasingly understand what they read. When children shift from "learning to read" to "reading to learn" (Chall, 1983), they reach the *fluent comprehending reader stage*, where they acquire a substantial knowledge of spelling. They understand most of what they read and are able to interpret implicit elements in the text, such as metaphors, irony, or figurative language. They can switch strategies to maximize comprehension (e.g. read more complicated sentences more slowly, re-read what they do not understand the first time). The final step in learning to read, achieved in early adulthood, is the expert stage in which any word is read in less than 500 ms.

5. Cognitive processes in autistic hyperlexia

We now present the available literature on autistic hyperlexia in relation to the three steps of typical reading.

5.1. Autism, hyperlexia, and the three steps of reading

5.1.1. Visual perception

There are multiple sources of evidence for atypical visual perceptual processing in autism for both static and dynamic tasks. An extensive literature reports enhanced perceptual performances in autism. Autistics have a superior capacity to simultaneously process large arrays of visual information and larger visual receptive fields (Remington et al., 2012). They perform better than neurotypical people at tasks involving mid-level visual processes, such as pattern detection and manipulation, block design, visual search, mental rotation, and hidden figures (Kaldy et al., 2011; Mottron et al., 2013b). Letter-like symbols are consistently detected faster by autistics when used as stimuli in visual search tasks, even by those who are not known for their hyperlexic abilities, and at an early age (Gliga et al., 2015). Pattern detection and manipulation are generally enhanced in up to 50% of individuals (Caron, 2006). This may, however, be an overestimation due to the inclusion of autistic participants with milder phenotypes in some studies, in particular those without a speech onset delay. Barbeau et al. (2013) found that the inspection time on a simple visual task was 31% shorter for an autistic group with speech-onset delay than a typical control group matched for Wechsler IQ, whereas an autistic group without speech-onset delay did not show the same advantage.

Low-level visual skills are also enhanced. Kéïta and colleagues compared the contrast sensitivity of autistic and non-autistic participants using vertically-oriented gratings defined by luminance and texture of varying spatial frequencies (Kéïta et al., 2014). The autistic group demonstrated a higher sensitivity for luminance-defined, high spatial frequency gratings. The peak sensitivity was skewed towards higher spatial frequencies in the autistic group, which can be viewed as a selective bias towards detailed visual information. Perreault et al. (2011) subjected autistic and non-autistic individuals to a mirror symmetry detection task that necessarily involved spatial integration. Overall, the autistic group performed significantly better at detecting symmetry than the non-autistic group. The relationship between enhanced low-level visual skills and superior pattern manipulation is unclear, with some studies finding a relationship (Caron, 2006; Guy et al., 2015) and others finding an aggregation, but no direct link (Meilleur et al., 2014).

Few behavioral studies examined the performance of hyperlexic children at visual tasks. The early emergence of acute visual discrimination and pattern recognition skills in hyperlexic children with impaired cognitive abilities (N = 6) were first shown by Cobrinik (1974). The author suggested that these skills could extend to hyperlexic reading when children perceive words as complex visual patterns. Cobrinik later explored visual word recognition, and showed that hyperlexic children (N = 9) performed better than the control group (N = 10) in an incomplete word task (Cobrinik, 1982). The author explained this result by the hyperlexic children's use of a configurational approach, involving a large visual array, rather than serial single-letter analysis. De Hirsch also noted a facility for tasks involving visual perception in hyperlexic children (de Hirsch, 1971). There are reports of enhanced performance in perceptual tasks involving pattern processing in more recent case studies of hyperlexia, such as the WISC-III picture completion, picture arrangement, object assembly, and block design subtests (Johnels and Miniscalco, 2014), or more generally visually supported tasks such as the triangles subtest of the Kaufman Assessment Battery for Children but also the ability to solve puzzles when the picture is facing down, using only lines and shapes for reference(Craig and Telfer, 2005).

In a first attempt to look into the neural basis of hyperlexia using functional magnetic resonance imaging (fMRI), Turkeltaub et al. (2004) scanned the brain of Ethan, a nine-year-old hyperlexic boy whose reading skills were six years ahead of expectations for his age. They compared his fMRI scan with those of two non-autistic control groups, one matched for chronological age and the other for reading age. Ethan's neural activity in both hemispheres was greater than that of both control groups during a covert reading task. Ethan showed greater activity in the right posterior inferior temporal sulcus than reading age-matched controls. This region is homologous to the VWFA (also called the R-VWFA) and is involved in visual word form recognition. It is usually active in the early phases of reading development, when visual patterns are used to recognise words, and later disengaged when children become more proficient at reading and rely more on letter-to-sound correspondences (Turkeltaub et al., 2003). This suggests that Ethan, an expert reader, exhibited superior involvement of form-processing areas, usually activated in novice readers. In addition to the right hemisphere systems, Ethan's phonological systems were also hyper-activated, in parallel with his superior performance at a phonemic awareness task. This left hemisphere activation of phonological systems could be secondary to his language development. The authors hypothesise that the right hemisphere visual form recognition systems may have contributed to the development of his advanced reading skills prior to the development of language.

This is consistent with the results of an ALE (activation likelihood estimation) *meta*-analysis of 26 fMRI studies, where visual information was presented to autistic (N = 357) and non-autistic (N = 370) participants, that reported greater activity in the occipito-temporal and parietal regions in autistics (Samson et al., 2012). They found a pattern of higher activity in regions involved in visual processing and expertise, and lower activity in the frontal regions associated with planning, reasoning and decision making in the autistic than the non-autistic group, despite similar task performance. The autistic group also showed atypical functional spatial distribution of domain-specific responses. Higher activity in the fusiform gyrus, medial parietal cortex, middle posterior temporal gyrus, left inferior frontal gyrus, and bilateral lateral prefrontal cortex was observed in autism across the six studies (81 autistics, 88 non-autistics) in which letters and words were presented. The autistic group also showed no predominant left lateralization of task-related activity, consistent with previous studies of reading and language processing in this population. To summarize, the activity related to letter and word processing in autistics was more right-lateralized and higher in areas that subserve broader aspects of perceptual expertise (e.g. the fusiform gyrus). This was interpreted as enhanced resource allocation in areas associated with visual perception and expertise in autism, suggesting a greater reliance on mental imagery and visualization processes during reading in this population.

We further explored the link between temporo-occipital hyperactivation and reading by comparing the results extracted from this *meta*-analysis for reading tasks to the results of an automated *meta*-analysis using the website www.neurosynth.org. We used this tool to locate the

VWFA by entering 'visual word form area' as a search term. The differential activity in the left fusiform gyrus observed in the word processing domain in autistics relative to controls in the ALE *meta*-analysis (81 autistics, 88 non-autistics) and the VWFA, reported across 35 fMRI studies (N = 576), overlap (Fig. 2). However, the activity peaks for the controls relative to autistics were more posterior. The peak difference observed in the ALE *meta*-analysis may therefore reflect higher engagement of the VWFA in autism during reading tasks.

Consistent with these fMRI findings, Kikuchi et al. (2013) reported different neural pathways associated with reading ability in autism in a magnetoencephalography (MEG) study of 26 autistic children aged five to eight. In typical participants, the VWFA projects to language areas in the left hemisphere associated with phonological and lexico-semantic processes (Cohen, 2003), whereas autistic children showed mostly right-lateralized neurophysiological connectivity during visual recognition and phonological treatment of words. This rightward lateralisation of neurophysiological connectivity (as measured by signal synchrony) between the parietal and temporal regions was associated with higher decoding abilities, as measured by the Kaufman Assessment Battery for Children (K-ABC) Reading/Decoding subtest. This may indicate that early autistic reading ability is related to the processing of perceptual aspects of written material relying less on lexico-semantic access, i.e. involving a functional network restricted to the visual perception areas. Such lateralization has been observed with other savant skills (Corrigan et al., 2012). Kikushi's study had some limitations, as no measure of attention was collected to compare the possibly different levels of attention given to visual information and narrative sound information. There was also no brain structural information available for such young children, making it impossible to superimpose MEG-signals. Furthermore, a recent fMRI study found reduced functional connectivity between occipital and frontal regions during a word similarities task in an autistic group with low reading comprehension abilities, relative to healthy controls matched for age, IQ and decoding abilities (Bednarz et al., 2017), suggesting that visual areas could be more independent from areas involved in semantic processing in autism.

5.1.2. Grapheme-to-phoneme conversion

Studies on reading irregular words and non-words are particularly informative for understanding grapheme-to-phoneme conversion and lexical access. The ability to properly read irregular words indicates that, beyond letter-by-letter decoding, some memory of an entire word is stored and accessible. Conversely, reading non-words indicates that the reader may go back to a letter-by-letter strategy when facing an unknown word.

Castles et al. (2010) focused on the ability of reading irregular words exhibited by two hyperlexic children to investigate their lexical reading skills. They assessed their ability to read a set of 36 irregular words aloud. Both participants performed similarly to age-matched control groups on the reading task, but had significantly lower scores on both comprehension tasks, suggesting that lexical access is preserved in hyperlexia for the purpose of out-loud pronunciation.

Other studies have explored irregular word reading in hyperlexia, with several reports of good performance (Atkin and Lorch, 2006; Frith and Snowling, 1983; Welsh et al., 1987). Furthermore, on a non-word reading task, a group of six autistic children with hyperlexia performed similarly to a control group of typical children matched for reading ability, whereas a third group of autistic children without hyperlexia performed significantly worse (Cardoso-Cardoso-Martins and da Silva, 2010). This indicates that hyperlexic reading does not rely solely on lexical memory, but uses a letter-by-letter strategy to decode words when necessary.

There is also evidence that hyperlexia exists across many languages, whether the orthography is relatively regular, such as in Italian (Cossu and Marshall, 1986), Spanish (Talero-Gutierrez, 2006), Portuguese (Cardoso-Cardoso-Martins and da Silva, 2010), or Swedish (Johnels and Miniscalco, 2014), or highly irregular, such as in English (Castles et al., 2010; O'Connor and Hermelin, 1994) or French (Lebrun et al., 1988; Worthy and Invernizzi, 1995). Hyperlexia has even been reported in bilingual (Joshi et al., 2010) and trilingual (Lebrun et al., 1988) children who showed hyperlexic abilities in more than one language.

It is not known if the two mechanisms of the dual-route model (Introduced in 4.1.2) are equally used when autistics read relative to typical individuals, nor if hyperlexia relies more on one or the other route. That hyperlexic individuals decode better than they understand suggests that they mostly use a sublexical route in the absence of a mental lexicon, using their knowledge of the structural rules of the language to decode words letter by letter. Nevertheless, there is, at least, partial access to the lexicon in hyperlexia as proven by hyperlexic children's ability to read irregular words. Exceptional decoding skills also do not solely rely on memories of individual words, as hyperlexics can also decode non-words, which calls for a letter-by-letter strategy. Hyperlexia also exists in many different languages, showing that advanced decoding skills do not depend on the orthographic depth of the language (Zuccarello et al., 2015). When letter-sound correspondence rules are not reliable across the language's orthography, the reader has to be able to use both lexical and sublexical routes. Altogether, this evidence suggests that the two mechanisms of the dual-route can be alternatively used in hyperlexia depending on the material to be read (regular words, irregular words, pseudowords).

According to the Dual Route Cascaded model (Coltheart et al., 2001), the lexical route can be divided into two sub-routes: the direct-lexical route, that allows direct access to the orthographic representation of written words to translate them into spoken words with no semantic mediation, and the lexical-semantic route, that proceeds via semantics (Castles et al., 2010). Findings on hyperlexia support the existence of a direct lexical route from visual word recognition to spoken word production without semantic mediation.

5.1.3. Semantic access

Several studies report poor reading comprehension relative to decoding skills in autism in general (Dynia et al., 2016; Fernandes et al., 2016; Miller et al., 2016; Nation et al., 2006; Ricketts et al., 2013; Westerveld et al., 2016), but the discrepancy is even larger between decoding and comprehension in hyperlexia. In one study, hyperlexic children performed significantly worse than typical children matched for reading ability and autistic children without hyperlexia on a word comprehension task, whereas autistic children without hyperlexia did not differ from typical children (Cardoso-Cardoso-Martins and da Silva, 2010). Only a few studies have evaluated reading and comprehension *of the same words* in hyperlexia to investigate the relationship between both skills. Siegel (1984) found that A.E., a six-year-old hyperlexic girl, was able to read words and sentences that she could not understand. Conversely, Aram et al. (1984) reported a marked discrepancy between a hyperlexic man's ability to read meaningful and non-meaningful words, suggesting a facilitation effect from semantic access, similar to that found in non-autistic readers. However, these authors did not evaluate whether the participant understood the meaningful words. Hence, their result could be an effect of word frequency more than comprehension. Castles et al. (2010) addressed this issue by using the same 36 irregular words in their reading task and two comprehension tasks. They showed that there were no significant differences in reading accuracy for irregular words that the two hyperlexic participants could define, and those they could not. This indicates their ability to read aloud low-frequency irregular words in the absence of comprehension, whereas semantic representations of words have an impact on reading ability in typically developing children (Ricketts et al., 2007) and adult (Balota et al., 2004; Strain et al., 1995) readers.

Text level comprehension also appears to be altered in autism relative to typically developing individuals of similar reading level. In two studies, autistic participants were able to process semantic information just as well as controls when semantic processing was tested immediately after reading the relevant information, but their ability was impaired later in processing, for example when irrelevant sentences were included as distractors after the relevant semantic information (Henderson et al., 2011; Tirado and Saldana, 2015). Contextual proximity of relevant information has an impact on access to semantic information in autism, indicative of limited ability to grasp large-scale text meaning. Text level comprehension may be functional in autism under optimal conditions, but more fragile than in typical individuals. It might also vary depending on the nature of the reading material, as shown by a pilot study that found that reading comprehension is improved in autistic children when their interests are embedded in text (El Zein et al., 2014).

In an fMRI study (Harris et al., 2006), the autistic group (N = 14 adult males) showed less differences in the activation of language areas (left inferior frontal gyrus) when reading concrete versus abstract words, than a control group (N = 22), suggesting that the stimuli induced less semantic processing in the autistics. Autistics also had substantially less Broca's area activation during semantic processing than controls. The authors suggest that this area may develop atypically in autism, partially explaining the frequently observed difficulties related to semantics and language. An alternative interpretation is that autistics do not activate their semantic network when it is not explicitly required by the task.

The nature of the barriers to semantic access in autistics with hyperlexia remains largely unknown. The fact that hyperlexic children seem able to read words that they do not understand just as well as words that they can define may indicate that their decoding skills do not call on their semantic knowledge. This is in contrast to typical children who read words that they understand better than unknown words, suggesting the intervention of semantics at every stage of the reading process. The greater activation of networks in the right hemisphere for visual recognition and phonological treatment may make semantic access difficult, because the latter is located in the left hemisphere. Furthermore, information on semantic access in hyperlexia is collected at an early age when hyperlexia is generally identified. Hence, it reflects comprehension mechanisms at the early stages of hyperlexic reading. These processes may evolve with age. In summary, there may be semantic access in hyperlexia, although it does not represent the default mode of hyperlexic reading, at least before the complete development of the reading function. We will now sum up what we know of the developmental pathway of reading in autism and specifically in hyperlexic children.

5.2. *Reading acquisition in hyperlexic children and developmental pathway*

There are reports of hyperlexic autistic children reading as early as 18 months of age (Craig and Telfer, 2005; Sparks, 1995) and consistently before they speak, at least in a non-echolalic way. This represents an advance of approximately two years over neurotypical children (see Section 4.2.), even when gifted. Their reading abilities are self-taught, and hyperlexic children cannot be taught in the conventional manner that is used for typical children, suggesting different learning pathways. The sequence of abilities preceding oral speech in hyperlexic children is strikingly different from that of typical children, and the reading ability is usually discovered by parents when it is already fully developed, before fluent speech appears, which may not develop at all in some cases. This sudden emergence in the absence of intensive instruction was reported for all children in several of the group studies included in Table 2 (Richman and Kitchell, 1981; Healy et al., 1982; Whitehouse and Harris, 1984) and in most case studies (Huttenlocker and Huttenlocker, 1973; Goldberg and Rothermel, 1984; Burd et al., 1987; Smith and Bryson 1988; Turkeltaub, 2004). In a similar way, Kanner (1949,1951) noticed that nonverbal autistic children surprised their parents in emergency situations when they pronounced grammatically correct sentences, suggesting that they accumulated a large amount of language information before their first use of speech. Hans Asperger (1944) also described autistic children that could learn to read "particularly easily" while being "almost impossible" to teach.

Developmental information and its relation with present and future speech ability was scarce in the literature we reviewed. Several follow-up reports suggest the persistence of an interest in reading over time, whereas a few report a decreased and less compulsive interest (Sparks, 1995; Talero-Gutierrez, 2006). Some cases developed functional oral communication, whereas others did not. In some cases, speech started at the same period as reading: two studies (Cobrinik, 1974; O'Connor and Hermelin, 1994) reported a sudden emergence of fully formed speech after the apparition of reading skills in several subjects, whereas Burd et al. (1987) noted "Shortly after the onset of his intense interest in looking at printed materials, he began to use his first words and simultaneously began to read orally". More recently, Atkin and Lorch (2006, p.267) concluded on "the possibility of an atypical route to language acquisition" from their extensive observation of a 4-year-old autistic and hyperlexic boy. In summary, hyperlexia is more than a simple inversion of learning steps, but rather appears to be mediated by a different process for the acquisition of reading skills. It does not exclude speech, and may even be synchronized with its emergence.

Indeed, we found multiple reports on how hyperlexia has been used to encourage the emergence of oral and written communication, with positive results. Burd and Kerbeshian (1988), who first pictured hyperlexia as a predictor for a better outcome in PDD, presented the cases of two siblings with PDD and hyperlexia. Both exhibited a dramatic increase in their intellectual functioning, receptive and expressive language development, and adaptive behaviors after the onset of hyperlexia. The same researchers also reported examples of communication with autistic children through written commands that were obeyed by the children, and the case of a child with hyperlexia and hypergraphia whose primary method of communication was through typing words. They even mentioned a case where the child started uttering phrases written on cards, showing that his interest in reading led to an improvement in language and oral communication (Burd et al., 1987; Burd and Kerbeshian, 1985). In another longitudinal case report, oral communication was triggered by written messages (Jensen, 2005). Telero-Gutierrez (2006) also noticed a preference for written orders that their participant expressed by saying the word "write" when he was asked something during therapy. Craig and Telfer (2005) followed a hyperlexic boy from 5 to 12 years old and were able to use his decoding and writing ability to successfully scaffold language development. Furthermore, the presence of orthographic cues improves oral vocabulary acquisition in both controls and autistics, supporting intervention approaches that include written words (Ricketts et al., 2015). Burd's final note stating that "A small, portable personal computer may serve well in this regard." may not be as outdated as one might think (Burd and Kerbeshian, 1985; p.942).

6. Discussion

We examined the available descriptive and epidemiological data on hyperlexia through a systematic review of the literature. We also reviewed studies investigating the putative role of cognitive processes in the genesis and functioning of hyperlexia and the related cognitive processes in the typical literature. We summarized informative studies on the temporal course of this special ability. Three main questions emerge from the conclusions of this review: (1) Why does the hyperlexic profile characterize such a substantial portion of the autistic spectrum; (2) Can models of non-autistic reading teach us about hyperlexia, or do we need models specific to autistic cognitive functioning; and (3) Can we make use of the atypical path used by autistics when learning to read in intervention?

6.1. *Hyperlexia and autism*

Despite some uncertainty on the actual prevalence of hyperlexia in autism, this condition is the most commonly associated with hyperlexia. There are only a few reports of hyperlexia associated with other neurodevelopmental conditions, each of which show an elevated occurrence of autism (Howlin, 2008). This must be taken into account before concluding that the non-autistic conditions in which hyperlexia has occasionally been described have a direct, mechanistic link with its occurrence. In addition, the definition generally used in reports of non-autistic hyperlexia highlights the discrepancy between reading abilities and general intelligence or comprehension, but misses other criteria for hyperlexia, such as early onset and a compulsive interest in reading (Nation et al., 2002; Pennington et al., 1987). Similarly, Treffert defines hyperlexia as "*combination of precocious reading skills accompanied by significant problems with learning and language*" and describes three types of hyperlexia (Treffert, 2011). Hyperlexia type 1, are neurotypical children, who do not show any sign of developmental disorder. They do not qualify as hyperlexic according to our definition, and are called precocious readers in most articles. Beside precocious reading skills or poor comprehension in typical children, hyperlexia is defined by a very specific pattern of compulsive interest and exceptional skills, and we would like to "preserve the concept of hyperlexia" for this unique profile (Grigorenko et al., 2003; Healy et al., 1982).

It is currently impossible to give the precise prevalence of hyperlexia in autism because of the considerable variation in the prevalence reported for autism itself throughout the years, and the absence of information on the autism subtypes or clinical forms associated with hyperlexia.

Savant abilities and heterogeneous non-savant profiles of abilities may share mechanisms and contexts of occurrence (Dawson et al., 2008). The relation between hyperlexia and autism may raise the same issues as the relation between savant syndrome and autism, which is now beyond question (Heaton and Wallace, 2004; Mottron et al., 2013a,b), despite the fact that not all autistics are savants. The presence of hyperlexia, using the four-feature definition mentioned above, cannot be considered sufficiently sensitive to become a diagnostic criterion for autism. However, having a feature present in one tenth to one fifth of the autistic phenotype is still of considerable interest. For example, macrocephaly, found in less than 30% of autistic people (Libero et al., 2016), is still of major neurobiological interest.

The variability in the reported prevalence of hyperlexia, according to the stringency of its definition, may be informative as hyperlexia may be the extreme variant of a widespread cognitive profile among autistic people. Accordingly, a large portion of autistics show a dissociation between their decoding skills and reading comprehension (Jones et al., 2009; Miller et al., 2016; Nation et al., 2006; Ricketts et al., 2013; Westerveld et al., 2016). Hyperlexia may be a paradigmatic autistic savant ability to the same extent that savant abilities in general represent the clearest example of autistic expertise, in continuity with the restricted range of talents evident even in non-savant autism (Mottron et al., 2013a,b; Mottron et al., 2009). The very strong interest for words and letters that hyperlexic children show at a very early age may represent the most extreme manifestation of this frequent interest found in autistic children (Meilleur et al., 2015), and predispose them to develop savant abilities such as hyperlexia. In this context, hyperlexia can be represented as a pyramid where the most obvious examples are less frequent, but stand out on the basis of much more frequent but less spectacular hyperlexic profiles (Fig. 3).

6.2. Models of hyperlexia

6.2.1. Why do we need a new model for hyperlexia?

Through their extensive self-exposure to printed materials and thorough practice of reading, hyperlexic children acquire a vast catalogue of orthographic representations virtually mapped with phonological representations, bypassing the lexical-semantic route. In other words, hyperlexics are able to successfully couple written words with their corresponding sounds. Models of non-autistic reading inform us on the cognitive operations that are plausibly involved in hyperlexia. The first two logical steps of reading (word recognition and grapheme-to-phoneme conversion) are functional in hyperlexia, but the third (semantic access) does not seem to be achieved *prima facie*.

Although visual word recognition in hyperlexia is performed as fast as for any fluent reader, it is not known whether it is achieved in the same way, or goes through a different route. Visual word recognition in typical children is tightly associated with previous knowledge of the word and its meaning, as shown by the correlation between word reading accuracy and reading comprehension (Cain et al., 2004). There must be another system, or a different use of the same system in hyperlexia, to explain such fast decoding without access to meaning. The question remains as to whether the development of reading in hyperlexia is truncated (i.e. the third step is missing altogether while the first two are intact), inverted (i.e. the three steps do not come in the same chronological order), or altered (i.e. there are fundamental differences at every stage).

To address this question, we compared reading, learning, and development in hyperlexia and typical children, as well as how each step is performed in hyperlexia. However, the direct comparison of cognitive functions between typically developing and autistic individuals may be misleading (Cossu and Marshall, 1986; Karmiloff-Smith et al., 1997; Mottron et al., 2009). Approaching hyperlexia by comparing the reading skills of autistic children with those of typical adults is both 'adultomorphic' and 'normocentric', i.e. at risk of imposing an adult structure as well as non-autistic task division to autistic functioning during development (Mottron et al., 2008). This approach is useful for devising paradigms to test the components of hyperlexic reading, but may be misleading for the interpretation of their results. It is very unlikely that autistics with hyperlexia reach the fully developed decoding ability that comes at the end of the successful learning-to-read process in typical people, with a dysfunctional system with missing components. The evidence showing differences in behaviors, brain activity, and connectivity in autism in general, and hyperlexia in particular, suggests that hyperlexic reading is a substantially different process from typical reading. Hence, reading development in hyperlexia cannot be qualified as truncated or inverted, it is different at each and every stage.

We may need models specific to autistic cognitive functioning to account for the mechanistic aspect of hyperlexia, as well as its intrinsic relation with autism. The neural mechanisms involved in pattern detection are exceptionally active and may be more autonomous in autism (Mottron et al., 2009). Autistic strengths in visual tasks may be an asset in the processing of written strings of letters as the visual cortex is highly involved in visual word form recognition. The very first step of reading, visual word form recognition, may therefore be achieved faster and/or earlier than by typical children. These findings are consistent with the Enhanced Perceptual Functioning Model (Mottron et al., 2006; see Mottron et al., 2013a,b for an update) that proposes that autistic perception plays a greater role in complex cognitive operations such as those involved in reading. Moreover, superior decoding skills in autism are associated with more right-lateralized patterns of activation and more cerebral activity in areas subserving perceptual expertise, as well as stronger connectivity within the perceptual areas and altered connectivity between these areas and other brain regions, relative to typically developing individuals. In other words, autistic children may be attracted to words and letters at a young age because written material is particularly adapted to autistic neurocognitive abilities.

6.2.2. Towards a perception-oriented model for hyperlexia

For the second step of reading, grapheme-to-phoneme conversion, Mottron et al. (2013a,b) proposed the existence of a 'veridical mapping' (VM) mechanism that enables autistics to detect complex patterns through their recurrence, and map their composing elements with sets of items that have a similar (isomorphic) structure. Following this hypothesis, savant abilities require such matching between isomorphic sets of information found in human codes: this is the case for dates and day in calendar calculation, hue and color in synesthesia, and for notes and sounds in absolute pitch (Mottron et al., 2009). Decoding in hyperlexia requires the accurate coupling of isomorphic written and oral patterns at various scales of complexity: written material is rich in within-code (e.g. recurrence of syntactic and contextual patterns) and between-code (e.g. grapheme-phoneme, or oral-visual forms of words) isomorphisms. This hypothesis is consistent with intact associative learning in autism (Boucher and Warrington, 1976; Williams et al., 2006), and the fact that autistics are able to learn associations between paired stimuli more rapidly than non-autistics (Sears et al., 1994).

The use of intermodal correspondences required in hyperlexia implies the activity of associative regions and connection between brain areas. As stated before, cortical areas associated with visual perception and expertise are consistently more activated in autistic than non-autistic individuals (Samson et al., 2012), adding evidence for the enhanced role of perception in the autistic mind. In addition to this increased activation, there is evidence for increased connectivity, both locally within perceptual areas (Turner et al., 2006), and globally between these areas and other brain regions in autism (Mizuno et al., 2006), possibly forming a highly specialized network supporting the mapping between isomorphic elements necessary for the development of savant abilities. The combination between autistic perceptual strengths and their orientation toward the detection of structural similarities could explain why a substantial portion of autistic children develop an early interest for the visual patterns composing written material, which transforms into decoding expertise as they learn to map written words with their corresponding pronunciation. Hence, we can add a larger base to the pyramid mentioned in 6.1 to include the pool of autistic children with enhanced perceptual abilities from which hyperlexia could arise (Fig. 3).

6.3. Development

The main differences between typical and hyperlexic reading acquisition are that (1) the former relies on previously acquired language abilities, whereas the latter frequently emerges before any communicative oral language appears; (2) the age of onset of hyperlexia reported in the case studies we reviewed was consistently before the age of five years (with only one exception out of the 62 cases in which the onset was mentioned) and often much younger, starting at 18 months (Table 1), which is, to our knowledge, about 18 months earlier than the earliest reported precocious reading case in typical children (Henderson et al., 1993); and (3) hyperlexia manifests itself as an intense, transiently quasi-exclusive interest for all types of printed material, plausibly without an equivalent in non-autistic children at this age. It is not yet known how these three characteristics are mechanistically related. The advanced decoding skills and apparent independence between decoding and later speech ability are two elements that may or may not be causally related. The 'compensation' or 'inverse assumption' theory, stipulating that strengths are accompanied by a complementary deficit (for example, decoding would "compensate" for defective comprehension) may be misleading in the case of autism (Plaisted-Grant and Davis, 2009). It is also possible that early maturation of visual form recognition systems orients autistic activity and interest towards complex – and available – visual information, resulting in non-communicative access to language. Alternatively, speech, which typically develops for communication purposes and is intrinsically associated with comprehension, is delayed. Communication and comprehension are not necessary to decode words. Hence, this task can be performed at the 'lowest' possible level in the three-step hierarchy that characterizes typical reading, i.e. without the intervention of semantics. Additional steps may nevertheless appear further along development, benefitting from the knowledge of structural regularities acquired from the written code. Just as echolalia constitutes an initially non-communicative developmental step in the access to language specific to autism, hyperlexia may actually be more than non-communicative meaningless reading, but rather constitute a developmental step towards reading comprehension and possibly written and oral communication.

6.4. Intervention issues

The pattern of dissociation between decoding abilities and comprehension evident in hyperlexia results from the particular characteristics of autistic perception and learning modalities, the understanding of which is essential to adapt education and pedagogy to the special needs of autistic individuals. The main intervention methods currently in use in autism in North America (Applied Behaviour Analysis and Denver Model) involve the limitation or elimination of repetitive behaviours and restricted interests. Thirty-five years ago, Lovaas, the initiator of applied behavioral intervention for autistic children, claimed that repetitive behaviours and interests such as hyperlexia had to be suppressed in favor of more socially acceptable behaviours, because they hindered learning appropriate activities, such as playing and communication (Lovaas, 1981; p.350–351). As recently as 2013, the Denver model asserted that stereotyped behaviors and restricted interests did not favor learning and hindered the practice of new competences (Rogers and Dawson, 2010), despite the now well-known link between specific interests and special abilities in autism (Mottron et al., 2013a) and reports from autistic adults saying that their interests are beneficial and should be encouraged (Koenig and Williams, 2017). If hyperlexia is part of the language learning sequence of autistic children, attempting to replace it by typical instruction is likely to not succeed. We found multiple examples of how hyperlexia can be used to improve the life of autistic individuals and their peers (see 5.2.). The case-reports we reviewed support that hyperlexia cannot be considered as a developmental dead end. They provide proof of concept that hyperlexia can be used to develop a different form of communication, initially based on written words, but possibly evolving into oral communication later. This is made even more relevant by the fact that reading disturbance in autism is associated with language impairments (McIntyre et al., 2017). Legitimate large-scale studies are needed to confirm this hypothesis. A recent fMRI study showed that a strength-based reading intervention could lead to improved reading comprehension in autistic children through changes in their brain function (Murdaugh et al., 2015), which could potentially lead to increased communication.

Written language is widely available in the environment, draws the attention of most autistic children, has an obvious relation with oral language, and is used as a social link and cultural transmission tool in society. Moreover, in contrast to oral language, that is initially presented to the child in highly social situations, written language allows a non-social approach to language, making it more attractive for autistics. It can be accessed and practiced alone, without the help of another human, provided that written material is available. The role of intervention should be to ensure that the development of hyperlexia, emerging from the conjunction of a strong interest and remarkable abilities, ultimately results in adaptive benefits, which do not necessarily coincide with a decrease of autistic symptoms (Mottron, 2011).

Autistics frequently learn written language before oral language. This developmental sequence may not be disadvantageous, but simply different. Just as typical children cannot be taught how to write before they know how to speak, intervention may benefit from following the autistic developmental sequence. If further intervention studies confirm that it is feasible and effective, the child should be provided with information that he is spontaneously interested in and able to process. Strong interests, such as that observed with hyperlexic children and written material, may be considered as opportunities for development, as opposed to unwanted behaviours. Parents should thus be taught how to detect and encourage these interests, as well as how to make use of them to foster communication.

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