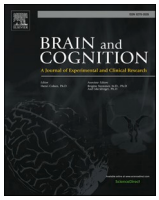
Brain and Cognition 173 (2023) 106099

Contents lists available at ScienceDirect

Brain and Cognition

journal homepage: www.elsevier.com/locate/b&c



Case Study

A longitudinal study on language acquisition in monozygotic twins concordant for autism and hyperlexia

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ARTICLE INFO

Keywords:
Autism
Hyperlexia
Case study
Longitudinal
Monozygotic twins
Language

ABSTRACT

Background: Hyperlexia, a strong orientation towards written materials, along with a discrepancy between the precocious acquisition of decoding skills and weaker comprehension abilities, characterizes up to 20% of autistic children. Sometimes perceived as an obstacle to oral language acquisition, hyperlexia may alternatively be the first step in a non-social pathway of language acquisition in autism.
Method: We describe two monozygotic twin brothers, both autistic and hyperlexic, from the ages of 4 to 8 years old. Following an in-depth diagnostic assessment, we investigated cross-sectionally and longitudinally their verbal and non-verbal cognitive abilities, language, reading and writing skills, interests, and strengths.
Results: The twins' features, including their high non-verbal level of intelligence, their special interests, and their skills in various domains, were highly similar. Their language consisted exclusively of letters and numbers until their fourth year. After that, their vocabulary broadened until they developed full sentences, and their perception-related interests expanded and merged over time to serve the development of other skills.
Conclusion: Our results show that hyperlexic skills can be harnessed to favor oral language development. Given the strong concordance between the twins' cognitive and behavioral phenotypes, we discuss the environmental and genetic influence that could explain their abilities.

1. Introduction

1.1. Background and rationale

Hyperlexia is generally defined as a strong orientation towards written materials associated with a discrepancy in reading skills: precociously acquired decoding skills and weaker comprehension abilities. It was first perceived as a rare ability, not immediately related to autism, but hyperlexia is now considered an autistic 'special ability' (Heaton & Wallace, 2004), and it could be the most frequent one. There is some disagreement on the definition of hyperlexia; some researchers use extreme precocious reading as their sole criteria, others debate over the level of discrepancy needed between decoding and comprehension skills, or decoding skills and general cognitive abilities, while others consider the early intense interest in written materials sufficient to be hyperlexic (Grigorenko et al., 2003). Hyperlexia is found in 6 to 21 % of autistic individuals depending on the definition used, and up to 84 % of hyperlexic individuals are on the autism spectrum (Ostrolenk et al., 2017).

The practical and theoretical importance of hyperlexia comes from the fact that it could represent an entry into the 'nonsocial' language characteristics of autism (see Kissine et al., 2023 for a *meta*-analysis on the independence of language development from socially mediated interactions), due to its high prevalence in most prototypical autism phenotypes (Ostrolenk et al., 2023, in prep.). Several cases of 'unexpected bilingualism', i.e., autistic children who acquire a language through non-interactional input such as the radio and television (Kissine et al., 2019; Vulchanova et al., 2012; Zhukova et al., 2021) or written material (Smith et al., 2011; Smith & Tsimpli, 1995), have been reported. Similarly in cases of hyperlexia, written input could provide children with the raw materials necessary to develop language without primarily requiring a socially mediated interaction. If correct, hyperlexia would predict a favorable prognosis in autistic children and should be harnessed in intervention strategies.

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https://doi.org/10.1016/j.bandc.2023.106099
Received 3 August 2023; Received in revised form 6 October 2023; Accepted 9 October 2023
Available online 13 October 2023

1.2. The neurobiological bases of hyperlexia

1.2.1. Neural aspects

Little is known about the brain substrates of hyperlexia. Early letter detection and recognition in the first year predicts future development of autism in at-risk children (Gliga et al., 2015) and is found in autism as early as 30 months (Kaldy et al., 2011). However, the neural bases of reading skills in autism cannot be studied with neuroimaging techniques until later in development. Two cases of hyperlexia in brain-damaged neurotypical adults have been published, one with transcortical motor aphasia, the other with a destruction of the left anterior cingulate cortex and corpus callosum (Glosser et al., 1996; Pacheva et al., 2014). However, the generalization of observations made in brain-injured adults to a population of non-cerebro-damaged children is risky. A single fMRI study directly investigated the cerebral bases of hyperlexia in a 9-year-old child (Turkeltaub et al., 2004) by comparing his brain activity during reading tasks to that of two non-autistic control groups, one matched for chronological age and the other for reading age. The child had increased activity in the right posterior inferior temporal sulcus compared to matched controls matched for reading age. This form processing area overlaps with the Visual Word Form Area (VWFA), which is specialized in the visual perception of written words. He also presented with atypical right-lateralized activity in an area analogous to the VWFA that is usually active in the early stage of reading acquisition and later disengaged in typical children. This cortical reallocation was also found using magnetoencephalography. One study found an association between a rightward lateralization of connectivity during word-related tasks and higher decoding abilities in autistic children from 5 to 8 years of age (Kikuchi et al., 2013), and another showed that an atypical right-lateralization in cortical activity during silent word reading is negatively correlated to measures of word comprehension and social abilities in autism (Ogawa et al., 2019).

In our previous work (Mottron et al., 2013; Ostrolenk et al., 2017), we formulated the hypothesis that hyperlexia emerges from a superior ability for visual recognition and complex pattern processing applied to written text, supported by heightened activity in cortical areas related to visual perception, and increased connectivity with other brain regions. Additionally, we showed that areas related to visual perception and expertise are more strongly activated in autism during word-related tasks, and that the peak of activation for autistic compared to non-autistic people found in a *meta*-analysis of fMRI studies overlaps with the VWFA (Ostrolenk et al., 2017; Samson et al., 2012). Recent work suggests the integration of hyperlexia into the more general framework of imbalances in information network hierarchy, between sensory and higher-order default mode regions (Hong et al., 2019).

1.2.2. Genetic aspects

There is no information on the genetic underpinnings of hyperlexia. Since hyperlexia is unquestionably associated with autism (Ostrolenk et al., 2017; Solazzo et al., 2021), it is likely that the genetics of hyperlexia overlap, at least partially, with that of autism. To our knowledge, a single case of hyperlexia with an identified neurogenetic condition (tuberous sclerosis; Pacheva et al., 2014) has been reported, for which a mere random association cannot be excluded. Therefore, hyperlexia is most likely linked with non-syndromic familial autism, and has a similar sex ratio: 79 % male in our systematic review of hyperlexia (Ostrolenk et al., 2017), slightly higher than that of non-syndromic autism (Dougherty et al., 2022; Jacquemont et al., 2014).

The heritability of autism is an established fact, with heritability rates ranging from 64 % to 91 % (Tick et al., 2016). However, the nature and limits regarding the genetic versus environmental influence remain unanswered inasmuch as the determination of the inherited part of the autism phenotype depends on our understanding of the heterogeneity of the 'autism spectrum' (Mottron & Bzdok, 2020). There is high family concordance in autism that extends well beyond the autism diagnosis, and where high concordance is found for the broader autistic phenotype and for non-autism-specific language impairments (Castelbaum et al., 2020). This familial concordance and higher concordance in homozygotic than in heterozygotic twins was one of the earliest pieces of evidence for the genetic influence in autism (Colvert et al., 2015). Critically, more recent work emphasizes the absence of a relationship in symptom severity between autistic individuals from a concordant family showing that "the factors responsible for variation in severity within the clinical range diverge from those that are responsible for the heritability of the condition itself" (Castelbaum et al., 2020). These recent studies have also highlighted the existence of non-concordant cases even in homozygotic twins (Castelbaum et al., 2020; Constantino, 2021). These discordant monozygotic twin pairs have theoretical importance for the understanding of the heritability of autism. However, these discordant results should be balanced with the existence of extreme concordance between twin pairs, i.e., cases involving a similarity that goes largely beyond that of a shared diagnostic category.

Hyperlexia has mainly been reported at its most noticeable stage, the early onset of decoding skills. There are few accounts of how this skill develops prior to becoming obvious, and how it evolves as the child ages and enters school. The longitudinal investigation of the language development of a pair of autistic monozygotic twins who are both highly prototypical (i.e., representative of the core of the autism category, see Mottron & Gagnon, 2023 for more on prototypical autism) and with a highly concordant phenotype can be particularly informative. The reporting of a single other pair of monozygotic twins concordant for autism and hyperlexia as well as for multiple other phenotypical features (Smith & Bryson, 1988) allows for comparative study between these exceptional occurrences. The present case study documents the developmental history, behavioral phenotype, oral language development, interests, reading abilities, and other exceptional skills in two monozygotic twin brothers, Paul and Luc (pseudonyms used for privacy reasons), who are prototypically autistic and hyperlexic in the strictest sense (i.e., meeting all the features of the definition), investigated from their diagnosis at age four to school age at eight years old.

1.3. Case history

1.3.1. Familial background and developmental history

Paul and Luc's parents were born in Spanish-speaking countries and were 42 and 38 years old at the beginning of this study. The mother had five children, of which one died *in utero* due to placental thrombosis. The twins live with two typically developing siblings: a brother who is 3 years older and a sister from a different father who is 9 years older than them. Their brother is academically advanced with typical socio-communicative development. There is no history of psychiatric or neurological conditions in any of the first- or second-degree relatives. Both parents are university level educated and employed. They communicate in French and Spanish together and with their children, with occasional use of English.

Paul and Luc were born to their 34-year-old mother at the end of an uneventful 34-week pregnancy in Montreal, Canada. Paul weighed 2400 g and Luc weighed 2600 g at birth. They both spent a week in the neonatal intensive care unit after their birth due to an episode of bradycardia. They were described as difficult babies who cried a lot and slept little. Both children took their first steps around 12 months and were potty trained at three years old. Their first words, as defined in the ADI-R (Rutter et al., 2003) as the meaningful use of single words other than 'mama' or 'dada', appeared at 15 months for both children, and they had not acquired the use of phrases at the time of the first assessment when the children were four years and three months old (4y3m).

1.3.2. Diagnostic assessment

Paul and Luc were 4y3m when they underwent an autism diagnostic assessment after their family doctor raised concerns over their speech delay. The assessments were performed by a psychiatrist (LM) and an educational psychologist, and consisted of direct interactions with the

child, an in-depth interview with the mother, and an Autism Diagnostic Observation Schedule (ADOS-2) module 1 assessment (see scores in Table 1; Lord et al., 2012). Paul and Luc were both diagnosed with Autism Spectrum Disorder using DSM-5 criteria (American Psychiatric Association, 2013) and severe speech delay. An ADHD diagnosis was added a few months later for both children.

1.3.2.1. Paul. Communication and social interactions. In Paul, the first warning sign for autism occurred relatively abruptly when the child was 16 months old where he stopped answering to his name, and his language skills stalled as tantrums became more frequent. Retrospective questioning on the pre-clinical period did not reveal any warning signs. At the time of diagnosis (4y3m), Paul mostly interacted with objects and had very minimal spontaneous social interactions that were always stereotyped and atypical in nature. He did not participate in turn-taking in communicative or playful interactions and he pushed away any hand extended to interact with him. Additionally, his social behaviors were not adapted to different people in different situations, e. g., he would hug and kiss any adult that he was told to greet, whether the individual was familiar or unknown. He had no interactions with children his own age besides his brother. Regarding language, he was able to express basic needs using simple words, pointing, and hand leading. His spoken vocabulary was composed of individual words that he would not use in sentences. These words included naming letters, numbers, and a few words from his favorite shows, in addition to jargon (i.e., unintelligible gibberish). The parents estimated that he knew over 500 words but could not use them to communicate. He also exhibited immediate and delayed echolalia, often demonstrated with songs. He had multiple daily tantrums with violence directed towards himself and others that could last up to 45 min and were associated with transitions and interruptions in his usual routine.

Repetitive behaviors and intense interests. At the time of diagnosis, Paul had a quasi-exclusive interest in letters that had started in his first year of life. For instance, he would move towards toys with letters as early as 6 months old, and he could write words at 18 months old starting with the word 'Mozart' that he had learnt from the children's TV show Baby Einstein. He also lined up toy cars, displayed prolonged visual inspections of 3D objects, liked to move his fingers in front of his eyes and would cover his ears as a reaction to loud noises. He had precocious abilities in letter identification: he was able to correct inverted letters in the alphabet and he could write long words such as 'Beethoven' forwards and backwards on paper. He also had precocious abilities in accurately reproducing complex 2D figures. Paul also particularly enjoyed classical music and sang and hummed a lot. He liked to categorize objects based on their visual features, such as shapes and colors. Combining two of his interests, he was seen writing the word 'Beethoven' while humming Beethoven's 9th Symphony. He was very fond of Baby Einstein and liked to write words from the show, draw its characters, and listen to classical music, hence satisfying most of his interests.

1.3.2.2. Luc. Communication and social interactions. Luc's early development was very similar to his brother's. He also presented with an abrupt interruption of his language development around 16 months old. He did not use communicative speech for apparent communicative purposes until the age of 3 years and 10 months, and prior to this, his language consisted exclusively of naming letters. At the time of diagnosis and in contrast with his twin, Luc could make simple word combinations and perform some pragmatic functions. For example, he was able to imitate gestures on demand or play pretend. Similarly, he could initiate direct gaze but in an unmodulated way. However, until right before the assessment, he had no overt social interactions with his peers. He showed uninhibited familiarity and used atypical social openings, such as nodding his head, or saying "hello" and leaving immediately. His mother had never observed an interaction between Luc and another child that she could qualify as reciprocal. He would sometimes initiate interactions with his twin by joining Paul's activities of interest, and he would name letters and numbers whenever they were present in his environment. Luc's communicative language in the presence of other children was limited to a few common words that were intermingled with a jargon sounding like a pseudo-language. He exhibited immediate echolalia of sentences even when they were not addressed to him, and sometimes reversed pronouns and used common words inappropriately (e.g., said 'sorry' when someone else dropped something, or 'thank you' when he gave a cookie to someone else). He also communicated through hand leading, and like his brother, he exhibited frequent long tantrums where he threw objects.

Repetitive behaviors and intense interests. In his second year of life, Luc started to inspect car wheels at length, followed by an intense fixation on letters and numbers from 12 months on, and writing words shortly after at 18 months old. He was able to name letters in three languages (English, French, and Spanish), correct the spelling of words that he had never used orally, and fix inversions in alphabetical order both forwards and backwards. He could also memorize complex classical melodies and sing them accurately. He rapidly solved 48-pieces puzzles and built Lego structures with over 100 pieces. He had a lot of creativity for these structures and could build realistic cars or airplanes without models, and Luc was generally very good with any game that required matching shapes together.

Table 1
ADOS-2 Module 1 scores for Paul and Luc at 4 years and 3 months old.

ADOS SCORES	Paul	Luc
Social Affect (max. 20)	15	13
Restricted and Repetitive Behaviors (max. 8)	7	6
Total score (max. 28, cut-off for autism = 12)	22	19
Comparison score (max. 10)	9	8

2. Methods

The twins first took part in a group study around 4y6m where they underwent five sessions of detailed cognitive evaluation and complete assessment of their expressive and receptive language. The family was seen an additional 11 times between the children's ages of five and eight years old where questionnaires and interviews with the mother (three sessions), the children's language assessments (four sessions), and assessments of reading and writing skills and documentation of interests (four sessions) were collected. Each child was tested separately by a trained graduate student or speech therapist at a local research center. We followed an iterative process where each session was planned according to observations and information collected in the previous sessions. The investigation was guided by the information that was missing to answer our research questions. Following the mother's advice, we introduced a visual representation (i.e., icons and written words) of the testing schedule at the beginning of each session with a box to color marking the completion of a task. This addition improved the children's compliance in the research assessments. This study was approved by the CIUSSS-NIM Research Ethics Committee. The twins' mother gave informed written consent. Testing had to be interrupted unexpectedly in March 2020 due to onset of the COVID-19 pandemic and could not resume. This prevented us from completing further planned assessments.

2.1. Neuropsychological assessment

The twins' cognitive abilities were assessed between ages 4y8m and 5y using three standardized neuropsychological assessments. The first assessment was the *Wechsler Preschool & Primary Scale of Intelligence - Fourth Edition* (WPPSI-IV; Wechsler, 2012). It is normed for ages 2y6m through 7y7m. For children older than 4y, it includes six mandatory subtests for the calculation of a full-scale IQ (FSIQ): Information,

Similarities, Block Design, Matrix Reasoning, Picture Memory, and Bug Search. The second measure used was the *Mullen Scales of Early Learning* (MSEL; Mullen, 1995). It is normed from birth to 5y8m and shows good convergent and divergent validity for autistic children (Swineford et al., 2015). It is composed of five subtests: Gross Motor Skills (only normed up to 33 months), Visual Reception, Fine Motor, Expressive Language, and Receptive Language. Lastly, the *Raven's Coloured Progressive Matrices - Board Form* (RCPM; Raven et al., 1998) was used as a non-verbal measure of fluid intelligence that is normed for children 3y9m to 10y2m showing good testability with minimally verbal children (Courchesne et al., 2015). It is composed of 36 matrices of increasing difficulty, where the child must complete a given matrix with one of six options by manipulating pieces to fill the hole in the matrix.

2.2. Oral language assessment

Paul and Luc's language skills were assessed at three different ages (4y4m, 5y6m, and 7y4m) over four video-recorded testing sessions that were performed in French by a speech therapist who had extensive experience with autistic children (Table 2). Flexible testing methods were used such as switching between tests, rescheduling a portion of the assessment to a later time if needed, allowing the child to walk around, and accepting English answers on French tests (Courchesne et al., 2019). Choices regarding the tests used were based on the child's abilities that were previously observed by the research team or reported by the mother. The objective was to start from what each child could do and gradually increase the difficulty to find the boundaries of their current abilities. The tests were all selected individually by the speech therapist. A tantrum interrupted Paul's language testing at T3, and the onset of the COVID-19 pandemic prevented us from scheduling a new session.

Note. EOWPVT-2000: Expressive One-word Picture Vocabulary Test – 2000 Edition; EVIP: French version of the Peabody Picture Vocabulary Test; IMBDC: MacArthur-Bates Communicative Development Inventories in French; CELF cnd-f: Clinical Evaluation of Language Fundamentals for francophone Canada.

2.3. Reading assessment

We created custom reading assessments adapted to each child's skills. Magnetic letters were used at first based on the twins' spontaneous interest in them, but they were abandoned due to the children playing with the letters in their own way instead of focusing on the task. Reading assessment worksheets of increasing difficulty were then used at 6y5m and 6y10m. The six worksheets comprised of 12 items each using upper- and lower-case letters, starting with single letter naming, then syllables of increasing complexity, followed by simple words. The words made of three to eight letters with a frequency higher than 30 per million were taken from the Lexique 3.55 database (New et al., 2001) and selected based on their probable familiarity for a young child and their orthographic complexity.

Table 2
List of language tests and subtests administered at the three testing times.

T1 (4y4m)	T2 (5y6m)	T3 (7y4m)
EOWPVT-2000	IMBDC (parent-report)	EVIP Form A
Expressive	Words produced	CELF cnd-f (Luc only)
EVIP Form A	Grammatical complexity	Following directions
Expressive	Mean of the 3 longest	Recalling sentences
Receptive	Utterances	Expressive vocabulary (both)
IMBDC (parent-report)	Sentences	Word classes – expressive and receptive
Words produced	EVIP Form A	Formulated sentences (both)
Words understood	Receptive	Understanding spoken paragraphs
Sentences (Luc only)	CELF cnd-f	
	Following directions	Word structure
	Formulated sentences	Sentence assembly

2.4. Other strengths and interests

Information on the twins' strengths and interests was extracted from parent-report interviews, a standardized questionnaire, and direct observation. An in-depth semi-structured interview with the mother when the children were 5y8m documented the development of their interest in letters and numbers, and their hyperlexic skills (adapted from Ostrolenk et al, 2023, in prep.). A parent survey was administered when the children were 6y4m and 8y4m using The Autism Preschoolers Strengths and Interests Questionnaire (APSIQ; Larose et al., 2021) to document the children's strengths and interests. Direct observation was performed when the children were 6y4m and 6y5m during the Montreal Stimulating Play Situation that is a standardized play situation assessing children's interests through spontaneous behaviors (Jacques et al., 2018). In this situation, children are placed in a room containing 40 toys that were selected because of their appealing properties for autistic preschoolers. The observation was video recorded and lasted approximately 30 min, divided into three 10-minute periods: free play, followed by 'lateral tutorship' (Mottron, 2017) where the evaluator used toys that the child showed interest in while next to the child without interacting, and semi-directed play where the evaluator showed toys one-by-one and how to use them while engaging with the child. We later adapted the play situation to focus on objects related to written materials (e.g., books, dictionary, magnetic letters, chalk board, etc.) that we tested when the children were 6y10m.

3. Results

3.1. Neuropsychological assessment

The results of the twins cognitive testing performed between ages 4y8m and 5y are shown in Table 3. Paul's IQ could not be evaluated using the WPSSI-IV and Luc was distracted by the letters in this task, although he was able to obtain a score. Both children scored in the lower percentiles on the MSEL, but in the range of superior intelligence on the RCPM resulting in a discrepant profile consistent with prototypical autism (Dawson et al., 2007; Mottron & Gagnon, 2023).

3.2. Language assessment

Feasibility issues: The language assessments were challenging due to issues with comprehension and compliance. Despite the use of flexible testing, qualitative observations proved to be more valuable than standardized testing scores. Moreover, the children spontaneously used French, English and Spanish, of which only French was formally assessed. Standardized scores did not represent the children's language progression since they were compared to norms for typically developing children in their age range.

Qualitative aspect of oral language performance: Paul's answers to test questions were highly driven by his interests. For example, when asked to point to the image corresponding to a verbal label, he would point to

Table 3
Paul and Luc's performance on neuropsychological tests.

Test	Paul			Luc		
	Age (m)	Score	Percentile	Age (m)	Score	Percentile
WPPSI-IV	60	N.A.	–	58	77	6
MSEL	59	55	1	56	79	8
RCPM	60	25	92.5	57	25	95.5

Note. N.A. = Test not administered; m = months. WPPSI-IV: Wechsler Preschool & Primary Scale of Intelligence - Fourth Edition; MSEL: Mullen Scales of Early Learning; RCPM: Raven's Coloured Progressive Matrices - Board Form.

both the correct answer, and an image of interest, or he would flip the pages of the manual to find one. He became less impulsive over time: at T1 and T2, but not at T3, where he would point to an image before hearing the instructions. His phonology and syntax had noticeably improved as well. By T3, he was able to use simple sentences without errors in French and English. The unintelligible jargon, very present at T1 and T2, had almost disappeared at T3. However, his expressive vocabulary progressed only minimally: at T3, he was able to name familiar items from a picture (e.g., cat, apple) but would sometimes give wrong, but related, answers (e.g., saying "fire, the water" when he was shown a firefighter). His responses were sometimes echolalic, not overtly addressed, and of idiosyncratic/obscure relation to the tasks (e.g., answering "My name is Richard Brown" when asked to point to an image). Delayed echolalia was present at all three times, but his respect of speaking turns improved at T3. Testing was interrupted by a temper tantrum at T3 and could not be resumed.

Luc generally collaborated well during testing sessions. He would often point to things while saying "hey, look" but without coordinated eye contact. At T3, his interactive skills had greatly improved: he spontaneously asked several questions to the evaluator and mentioned when he did not know the answer to a question. He was also able to have a simple short conversation on a single topic and respect speaking turns. Luc's phonology and syntax had noticeably improved, and the occasional transformation of sounds (e.g., vowel substitution, nasal assimilation) observed at T1 and T2 had disappeared at T3. He used complete sentences of increasing complexity, but with morphosyntactic imperfections, and echolalia reduced over time. He spontaneously recited many sequences (e.g., letters and numbers) and could manipulate them (e.g., count backwards, count every other number, etc.). However, his expressive vocabulary and receptive language as measured by the tests progressed only slightly over time. On a subtest where he was asked to formulate a sentence using a word, Luc would instead repeat the word or ask questions about an image showing his verbal skills, but not scoring on the test.

Quantitative oral language performance: The results of the standardized language tests at T1, T2, and T3 are presented in Supplementary Table A.1, A.2 and A.3, respectively. At T1 (age 4y4m), direct testing of expressive language either placed the twins at the 1st or 2nd percentile (i.e., EOWPVT-2000 expressive for both twins, and EVIP Receptive for Luc) or they could not be administered (i.e., EVIP expressive for both children, and receptive for Paul). Parent-reported (IMBDC) words produced and words understood gave both twins an age equivalent around 16 months when they were 52 months at the time, and questions about sentences uttered gave XP an age equivalent of 12–13 months old. Questions about sentences uttered were not asked for Paul since he did not produce more than single words at the time. At T2, both children had an age equivalent of 35 months on the EVIP receptive language assessment and 48 months on the CELF when they were 66 months old. The IMDBC placed Paul at an age equivalent of 24 months on average, besides a score above 30 months for *Sentences*, while Luc reached the ceiling score (30 months) on all categories except *Words produced* (27 months). At T3, Paul's language could not be evaluated due to a tantrum. Luc's age equivalents ranged between 31 and 69 months and displayed better performance for receptive than expressive language.

3.3. Written language assessment

On the reading worksheets, both children had perfect scores for single letter naming in upper- and lower-cases at ages 6y5m and 6y10m. Paul was particularly quick in his answers, but he got easily frustrated when difficulty increased. At 6y5m, he had a perfect score for reading simple syllables in upper- and lower-cases but refused to answer any further after four consecutive errors when presented with a more complex worksheet. At 6y10m, he refused to continue past the single letters, although he was able to read words at home. Luc was more collaborative, but less advanced in his reading. At 6y5m, he was able to read half of the upper- and lower-case simple syllables. He scored 6/36 on worksheets with more complex syllables including French letter combinations (ou, ton) and short words. He read certain common whole words (e.g., his name, papa, bébé, pipi), but sometimes inverted letters (either inverting and mirroring letters when he read 'ou' as 'no', or just inverting the order reading 'ul' for 'lu'). He tended to spell the words that he could not read and made links with other words that he knew ("mou comme mouton!"). At 6y10m, Luc read all lower-case simple syllables correctly, scored 7/12 on upper-case simple syllables, and 10/36 on complex syllables or short words. When trying to assess their writing skills, we could successfully ask them to write a few letters, but they would soon start drawing and writing their current favorite characters' names making formal assessment impossible.

3.4. Other strengths and interests

3.4.1. Qualitative notes on other interests

The semi-structured interview with the mother at the children's age of 5y8m revealed that both twins were highly skilled with construction toys and could align objects with extreme precision as soon as they started crawling. Paul would sometimes build structures with toys to voluntarily produce an image with the structure's shadow. For both twins, any toy could be used to compose letters such as placing little cars in a letter shape or using plasticine to form letters. At 5y8m, the mother estimated that this interest occupied 90 % of both children's free time. The mother reported that most new spoken words were pronounced after being read. Their interest in letters eventually opened to other interests. For example, Luc became passionate about classical music and would ask his mother to write the name of the composer he was listening to. Later, when the children became interested in Sonic the Hedgehog, they would hum the theme song, write words related to the cartoon, draw the main character, mold it with clay, etc. While Paul made elaborate drawings incorporating written text, Luc was more interested in singing and building 3D structures (with Lego bricks or clay). The twins could spend over an hour on an activity involving one of their intense interests, while conversely, attracting their attention for learning purposes was challenging if the material proposed was unrelated. Their interest in letters was used to communicate at home: the children could write a word to design a toy that they wanted their mother to get. The family also used pictograms with images and written words around their house to organize the twins' time. Books were freely available in their bedroom, and they always had access to paper and pencils. The use of an electronic tablet was limited to 10–15 min per day after school. Both children liked to watch videos in English with subtitles, although they mainly spoke French and Spanish at home. Paul frequently used an application where he traced over written words with his fingers. Finally, their interests had soothing abilities: singing the alphabet song could help calm the children when needed. The twins spent most of their free time together dedicated to parallel or shared activities related to their interests.

3.4.2. Systematic investigation of interests

The Autism Preschoolers Strengths and Interests Questionnaire (Larose et al., 2021) administered at ages 6y4m and 8y4m revealed that Paul and Luc both presented a multitude of varied, but highly interconnected intense interests. The twins' interests expanded through their partial similarity with a previous interest (e.g., they cared for a stuffed Sonic the Hedgehog, but not stuffed animals in general). Over time, their interests tended to become more complex; for example, an interest in books was added to their interest in letters. Additionally, new interests related to social activities appeared such as playing with friends, role playing, and playing cards. The mother also highlighted their interest in drawing, geometric shapes, music, and singing although they were not included in the questionnaire. The twins had the same level of interest in 21 out of 23 items at 6y4m and 20 out of 23 items at 8y4m. Their objects of interest remained mostly stable over these two years with 16 out of 23

items appearing at both times (Fig. 1).

When placed in the Montreal Stimulating Play Situation (Jacques et al., 2018), Paul and Luc walked directly to a magnetic board with letters (i.e., one of 40 items in the room) and played with it for most of the free time they were allowed. On a second visit to the room, Paul could not wait until the evaluators were finished installing the room; he entered forcefully, immediately headed to the magnetic board, removed all the letters already on it and wrote all the names of Baby Einstein's

Item	Age 6y4m	Age 8y4m	
Trains	P (I), L (I)	P (I), L (H)	=, ⇑
Encyclopedias	P (I), L (I)	P (I), L (I)	=
Logos	P (I), L (I)	P (H), L (H)	⇓
Stuffed toys	P (I), L (I)	P (H), L (H)	⇓
Animated character figurines	P (I), L (I)	P (I), L (I)	=
Cars	P (I), L (I)	P (I), L (I)	=
Legos	P (I), L (I)	P (I), L (I)	=
Numbers	P (I), L (I)	P (I), L (I)	=
Letters	P (I), L (I)	P (I), L (I)	=
Metro/Bus routes	P (I), L (I)	P (M), L (I)	⇓, =
Electric lines	P (I), L (I)	No interest	⇓
Computers/Tablets	P (I), L (I)	P (I), L (I)	=
Toys with visual/sound effects	P (I), L (I)	P (I), L (I)	=
TV shows and videos	P (I), L (I)	P (I), L (I)	=
Clocks	P (H), L (M)	No interest	⇓
Dinosaurs	P (H), L (M)	P (H), L (H)	=, ⇑
Books	P (H), L (H)	P (I), L (I)	⇑
Pipes	P (M), L (M)	No interest	⇓
Traffic lights	P (M), L (M)	P (M), L (I)	=, ⇑
Weather forecast	P (M), L (M)	No interest	⇓
Playing cards	No interest	P (I), L (I)	⇑
Playing with friends	No interest	P (H), L (H)	⇑
Role playing	No interest	P (H), L (H)	⇑

Fig. 1. Cross-sectional and longitudinal evolution of Paul and Luc's interests between ages 6y4m and 8y4m as reported by their mother on the Autism Preschoolers Strengths and Interests Questionnaire (Larose et al., 2021) Note. P = Paul; L = Luc; I = Intense interest; H = High interest; M = Moderate interest; Items for which the children had little, or no interest, are not reported. The last column indicates the evolution between the two testing times for Paul and Luc, respectively.

characters. Paul tended to play next to the evaluator with no interaction other than occasional glances showing his interest. Luc initiated interactions when he needed help operating a toy. Both children had very strong intentions on what they wanted to do with the letters and did not react well to external interventions (e.g., pushing the evaluator's hand, throwing their magnetic letters to the floor). Lateral tutorship (i.e., doing things next to the child as opposed to with the child; Mottron, 2017) proved to be a more efficient strategy, for which a second board and set of letters were purchased and installed in the room in the literacy-oriented play situation.

4. Discussion

This longitudinal study of autistic hyperlexic monozygotic twins followed between the ages of four and eight years old adds to the literature on hyperlexia, specifically on the chronology of the interest in letters and other intense interests, and their relationship with oral language development. Due to hyperlexia's occurrence in monozygotic twins, it enriches the current hypotheses pertaining to the relative weight of genes and environment in the orientation of prototypical autistic children toward written material.

From letters to language - relevance for models of language acquisition in prototypical autism: Letters and numbers represented the majority of these hyperlexic toddlers' language around 3 years old. Their development combined a severe language delay with a focused precocity in their interest in written material, which is a marked difference from typical development. In a parallel group study, we have found that this represents a common pattern in autistic preschoolers (36 % of a representative population; Ostrolenk et al., 2023, in prep). The interest in written material, despite its restricted, pervasive, and apparently non-communicative nature at first, demonstrated its capacity to expand, complexify, and merge with other interests over time. Focus and task motivation drastically improved when the children's interests were integrated in the testing or learning material, which is consistent with other studies showing that embedding autistic children's interests in intervention is effective (Davey, 2020; El Zein et al., 2016; Harrop et al., 2019; Solis et al., 2022). The twins' interests triggered the acquisition of new skills and fostered peer-relationships where they could be used to help with everyday communication and played a comforting role as a source of pleasure and soothing. Although the interest in letters seemed independent from social interaction and communication at first, it played a large role in the relationship between the twins who showed enjoyment of social sharing around their joint interests. Additionally, the family used written words to improve at-home communication and learning. Both children were able to use sentences in a conversation by the end of the study suggesting that hyperlexic skills can be harnessed to support other skills and communication in general. Social bonding between siblings through their special interests and skills has been reported in other pairs of autistic twins (Horwitz et al., 1965; Smith & Bryson, 1988).

Overall, hyperlexia did not prevent the twin's further development of language which, although it remained delayed at the end of the study, followed a prototypical 'bayonet-shape'. This developmental profile comprises a large plateau without overt communicative oral language followed by a relative catch-up in the school years, allowing communicative functions, while still retaining autistic atypicalities and limitations (Charman et al., 2003; Gagnon et al., 2021; Luyster et al., 2007; Pickles et al., 2014). This pattern is the most common in prototypical autism (Gagnon et al., 2021; Mottron & Gagnon, 2023; Wodka et al., 2013). The development of language in this pattern is delayed and independent of the precursors of communication associated with the acquisition of oral language in typical development (Kissine et al., 2023).

We have suggested that the early phases of the plateau period in the 'bayonet-shape' model of language development is the manifestation of a developmental bifurcation towards the treatment of non-socially biased information in autism (Mottron & Gagnon, 2023). Autistic children's interest in complex perceptual, mostly visual isomorphic structures available in their environment, such as written text, would trigger implicit learning of the rules that govern these structures which is helped by their enhanced perceptual skills (Mottron et al., 2013). Hyperlexia would therefore represent one of the possible consequences, in autism, of the widely replicated over-efficiency of mid-level visual processes. They benefit other visual-perceptual processes ranging from simple shape recognition (Caron et al., 2006), shape segmentation (Thérien, Degre-Pelletier, Barbeau, Samson, & Soulières, 2023), mental rotation (Thérien et al., 2022), to fluid intelligence reasoning (Simard et al., 2015). Children's interest in perceptual material could contribute to the future development of special abilities, such as hyperlexia, itself nurturing oral language (Heaton & Wallace, 2004; Mottron et al., 2021). This idea is supported by studies showing that fully-formed speech can emerge shortly after the onset of reading in hyperlexia (Burd et al., 1987; Cobrinik, 1974; O'Connor & Hermelin, 1994). Like in Smith and Bryson's (1988) case report, Paul and Luc's other intense interests were related to visual or auditory perception which is consistent with the development of special abilities in autism through the attraction for structured perceptual material (Heaton & Wallace, 2004; Mottron et al., 2009, 2021; Mottron & Gagnon, 2023). Of note, the majority of the 15 hyperlexics studied by Lin (2014) had enhanced orientation for visuo-spatial material, and some understanding of language (mostly written). Most also presented immediate or delayed echolalia, which, as noted by Howlin (1981, p. 98) predicts future progress in speech. Perceptual strengths and a preference for complex visual patterns, combined with an ability to make intermodal correspondences and higher resource allocation to cortical areas involved in visual perception, could set the table for the early acquisition of decoding skills independently from oral language in hyperlexia.

Multiple exceptional abilities often co-exist in a single autistic individual (Bal et al., 2022; Meilleur et al., 2015). Indeed, hyperlexic traits are mentioned in case studies on other abilities, specifically calendar calculation. For example, DBC, an autistic calendar calculator with an IQ in the low-average range (Mottron et al., 2006) presented a "spontaneous interest in reading and writing" at four years old and a self-taught ability to read and write by age five, whereas his practice of calendar calculation only started at age 12 and he had an interest in numbers in general. It is possible that DBC's interest in written symbols led to his later interest in calendars, triggering the development of his exceptional calendar calculation skills. Another hypothesis is that his interests in reading and writing and in calendars share a common underlying cause, and that the self-taught development of these skills from his interests share a common mechanism, possibly the enhanced perceptual functioning described above. Although inconsistencies exist in the research, neuroimaging studies point to various abnormal patterns of activation in the autistic brains while reading at all ages. Most studies conclude to an increased reliance on areas involved in visual processing and reduced recruitment of areas involved in lexico-semantic processing during reading tasks. Autistics are characterized by a lack of left-lateralization and an atypical right-lateralization of cortical activation patterns compared to TD peers, a pattern correlated with higher decoding skills (Kikuchi et al., 2013) and lower comprehension (Ogawa et al., 2019). The right-lateralization of cortical activations during reading observed in hyperlexia has been reported in other exceptional abilities such as artistic skills (Corrigan et al., 2012), suggesting that common neural features may support the development of different exceptional abilities in autism.

Intelligence: In spite of their low scores on the WPPSI-IV and MSEL, the RCPM revealed that both twins had excellent cognitive abilities as long as verbal skills were not needed. Similarly, the language assessments, even though they were specifically chosen for the children and adapted to reach their potential, yielded few results above floor level. This was not informative of the children's progress in spoken language over time, which could only be reported with qualitative observations.

Generally, the twins showed irregular performance and their daily moods greatly affected our test results. Had we not explored their skills in detail over multiple sessions, we could have concluded that some of these skills did not exist, when in fact the child stopped answering because he did not want to continue testing.

The general concept of flexible testing for strength-informed assessments, and taking children's interests into account when designing strength-based interventions, can be applied more widely and may allow for the cooperation of children usually considered untestable (Courchesne et al., 2015, 2019; Mottron, 2017). In a study comparing a standard testing condition with a motivation/attention condition, designed for each individual, Koegel and colleagues (1997) found that three autistic children who could not be given a score under the standard condition scored in the average range in the motivation/attention condition. One case was particularly relevant to our topic: child 5 was 3:4, could read at first-grade level, and had an intense interest in books. In the motivation/attention condition, he was permitted to look through books during breaks, which greatly improved his performance on the test. This suggests that reduced motivation may be limiting the scores of autistic children, and that we sometimes assess attention and compliance when the target is language or intelligence.

Harnessing intense interests for oral language acquisition: Stereotyped behaviors and intense interests have been generally considered noxious in the autism literature in the past and targeted by intervention models arguing that they get in the way of learning new skills (Rogers & Dawson, 2010). Precocity of the interest in written material and letter identification is not usually considered by parents as indicating the emergence of oral language. There is a current trend towards reconsidering intense interests and repetitive behaviors as potentially beneficial (Koenig & Williams, 2017), contributing to the development of skills, a better quality of life (Grove et al., 2018; Epstein et al., 2017), and improved educational and social outcomes in school (Wood, 2019). The simultaneous appearance of speech and reading (Burd et al., 1987), and the surprising appearance of fluent speech in children who had previously developed reading skills (Cobrinik, 1974; O'Connor & Hermelin, 1994) have indeed been reported. More recently, a longitudinal intervention case study showed that reading and writing abilities could be used to support language development in a hyperlexic boy followed from the age of 5 to 12 (Craig & Telfer, 2005). Another strength-based intervention for autistic children, targeting visual processing as an area of intact functioning, showed changes in brain function associated with improvements in reading comprehension (Murdaugh et al., 2016). A follow-up study focused on the specific processes that improved reading comprehension compared two groups of autistic children, half of which were randomly assigned to receive the intensive reading intervention (Murdaugh et al., 2017). The participants were scanned before and after the 10-week intervention program using fMRI. Post-intervention scans revealed greater activation in visual processing and frontal regions in the group who had received the intervention compared to the group who did not, as well a different activation patterns in core language areas during sentence tasks. Activations in the left thalamus and right angular gyrus during a word task were only observed in the intervention group; these areas are involved in the integration of visual, auditory, and spatial information, which is essential to semantic processing.

Is hyperlexia as genetically constrained as autism itself? Our study offers some insight on the level to which genetics constrain the selection of a special ability in autism. The two cases reported here, although they have not been the subject of a genetic study, very probably belong to non-syndromic autism, because of their very high non-verbal IQ, normal walking age, and the absence of any warning sign for an identified mutation. However, in the absence of genetic investigation, this cannot be demonstrated beyond clinical, cognitive, and develelopmental arguments. To our knowledge, hyperlexia has only been associated in a single case with an identified neurogenetic condition (tuberous sclerosis; Pacheva et al., 2014) where hyperlexia was defined as the spontaneous acquisition of reading skills with appropriate comprehension, thus not matching our definition. The twins' interests and strengths were highly similar, including the developmental precedence of early specific interests over overt socio-communicative atypicalities. The autistic and hyperlexic monozygotic twin brothers described by Smith & Bryson (1988) were similar to Paul and Luc on many aspects including their average walking age and age of first words, the brutal interruption of typical language progression in the middle of the second year, and their early autism diagnosis (25 months). Several perception-oriented behaviors in the visual modality were mentioned (e.g., "waving a pencil or similar object in the periphery of their vision") and the rote recitation of a favorite book while turning its pages was done in the exact same way by both twins. Both had decoding skills, acquired autonomously at age 3, with a comprehension delay. They were both considered intellectually disabled with scores in the 50–60 range using the non-verbal scale of the Merrill-Palmer. This is probably an underestimation of their intellectual abilities given that the Merrill-Palmer scales result in lower scores than the Wechsler Intelligence Scale for Children–Fourth Edition, which is itself known for underestimating autistic children (Dempsey et al., 2020; Nader et al., 2016).

Another case of a shared savant ability between autistic monozygotic twins who also had an interest in printed material has been published. Horwitz and colleagues (Horwitz et al., 1965, 1969) published a retrospective case study of monozygotic twin brothers who were both calendar calculators. Autism is not explicitly mentioned, but the twins' description strongly suggests an autistic profile, and they were referred to as autistic in later accounts (Sacks, 1985). Their IQ scores were low, and their oral language and walking age were delayed, but they had a strong memory for dates and for the weather on specific dates. They displayed impressive self-taught calendar calculation skills but could not perform simple math operations. Like the twins in the present study, they had a very short attention span and low ability to focus unless they were interrogated about the calendar, in which case they showed focus and patience. The first twin to develop calendar calculation skills, George, acquired them at age 6 after spending hours starring at a calendar and made no errors as soon as the skill became apparent. George could also reproduce melodies on a piano, but could not be taught to read music, and spontaneously learned the Greek alphabet. The other twin, Charles, also developed an intense interest in dates and calendars a little later which transformed the relationship between the brothers who became inseparable. The parental role in the development of this interest was reduced to giving the twins access to a perpetual calendar and providing praise for their performance.

A higher concordance of autism in monozygotic than dizygotic twins is an undisputed argument in favor of the influence of a genetic mechanism in autism. The genetic similarity and the major role of the shared environment in monozygotic twins, which is even greater than in dizygotic twins (Hallmayer et al., 2011; Horwitz et al., 2003), predicts high phenotypical similarity. If autistic individuals are particularly drawn to certain types of stimuli and more likely to develop exceptional abilities in response to this exposure, we can expect to observe even higher levels of concordance in autistic monozygotic twins. The existence of another pair of highly concordant autistic monozygotic twins who are both hyperlexic (Smith & Bryson, 1988), but also cases with more divergent exceptional abilities (Horwitz et al., 1965), support this argument.

Non-concordance of autism severity in 10 % of monozygotic twins has been attributed to random, post-natal non-shared environmental influence (Constantino, 2021). Conversely, extreme concordance in monozygotic twins suggests that the genetic contribution to autism may extend beyond diagnosis, up to and including phenotypic cross-sectional (e.g., interest in printed material) and longitudinal (e.g., age of speech regression) linguistic features. While the possibility of contagion among twins cannot be excluded, this hypothesis is hardly tenable to explain such similarity including the age of developmental milestones. The very high level of similarity between two homozygotic twins sharing the

same environment cannot be directly attributed to their homozygosity. However, the extreme concordance of the transversal and developmental characteristics of hyperlexia and the multiple other interests in the context of autism in the two published pairs of autistic monozygotic twins pleads in favor of hyperlexia being under genetic influence.

Funding

This work was supported by a scholarship from Fonds de Recherche du Québec – Santé (AO), a scholarship from the Transforming Autism Care Consortium (AO), and the Marcel and Rolande Gosselin Research Chair on the cognitive neuroscience of autism (LM). The funding sources were not directly involved in this study.

CRediT authorship contribution statement

Alexia Ostrolenk: Conceptualization, Methodology, Investigation, Data curation, Visualization, Project administration, Writing – original draft, Writing – review & editing. **Valérie Courchesne:** Methodology, Investigation, Writing – review & editing. **Laurent Mottron:** Conceptualization, Methodology, Resources, Writing – original draft, Writing – review & editing, Supervision, Funding acquisition.

Declaration of Competing Interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Data availability

The authors do not have permission to share data.

Acknowledgement

The authors would like to thank the twins and their parents for their involvement in this study and extreme generosity with their time. We are also grateful to Catherine Cantin, Annie-Claude Côté, and Alina Vocila for their important contribution to the assessments and testing. We also thank Samantha Wunderlich for her work on editing.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.bandc.2023.106099.

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