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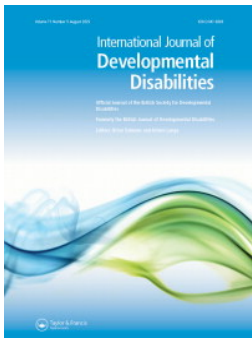
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Likelihood status and classification issues to be considered in familial research on communication disorders

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ABSTRACT

Objectives: Research investigating early identification of developmental conditions often uses an 'increased likelihood' methodology that recruits undiagnosed infants with affected family members. These studies are often cross-sectional or short-term follow-ups. Using grouping data from a wider longitudinal study of increased likelihood for developmental language disorder and autism, we aimed to investigate whether (i) likelihood status changes over time as family members acquire additional diagnoses and younger siblings are born (ii) Criteria used to determine likelihood affects status change and (iii) likelihood status in infancy associates with diagnostic outcome at 7 years.

Methods: 67 infants categorised into 3 likelihood groups based on family history of communication difficulties: Average likelihood of communication difficulties (AL); increased likelihood of language impairment (ILLI); increased likelihood of social communication difficulties (ILSCD). Caregivers completed an interview about the presence of communication difficulties when infants were around 12 months old. At 7 years old likelihood status was re-assessed, along with diagnostic outcome information.

Results: At 7, 25.4% of children changed likelihood status based on newly available family information. Using stricter criteria to group children only reclassified 4 children but lowered this moment to 16.4%. Neither broad nor strict likelihood groups predicted diagnostic outcome at 7.

Conclusions: Longer-term follow-up revealed issues with increased likelihood methods that need considering when conducting this type of research. An early assessment-based approach is likely to lead to greater progress in establishing successful prediction of later diagnosis and support for children with communication difficulties.

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Introduction

Autism and Developmental Language Disorder (DLD) are two relatively common conditions which are identified during childhood. Both affect social and structural communication skills which often require recognition, support and management through school life and beyond (Botting 2020; Dubois et al. 2020). There is now a body of work indicating that these conditions show familial aggregation, whereby siblings and first-degree relatives are more likely to show an increased likelihood of also being described as either autistic (Hansen et al. 2019; Ozonoff et al. 2024) or having a language impairment (Tallal, Ross, and Curtiss 1989; Capelli et al. 2024). This increased likelihood line of investigation may be useful in early assessment and intervention. Clinicians routinely screen and monitor children against developmental communication milestones. The ability to prioritise

certain groups of increased-likelihood children is therefore potentially an advantage in practice.

Thus, in parallel with this research, there has been a promising line of study investigating more subtle outcomes relating to communication and social behaviours in babies that have affected family members. Although some behaviours associated with autism (Hatch et al. 2021) as well as differing EEG patterns (Lau et al. 2023) may be identifiable in very young children, the average age of diagnosis for communication disorders is much later. For autism median age at diagnosis is 51 months of age in the US (Baio et al. 2018), and 55 months in the UK (Brett et al. 2016). For DLD there is much less information about age at diagnosis, with only one published article to our knowledge (de Bree, Wiefferink, and Gerrits 2024). This study reports that 25% of diagnoses are made after the age of 8 years, and less than half before children are 3 years old.

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Rannard, Lyons, and Glenn (2004, 2005) also note that 20% of their parent sample reported receiving no help until their child with DLD was five years old. Increased likelihood methodology may therefore have important potential for a better understanding of early signs of these conditions, and ultimately for providing recommendations for early assessment and intervention.

For autism in particular, where a body of work already exists, increased-likelihood samples show some identifiable differences in infancy, however for DLD the evidence is very sparse with only a few notable exceptions (e.g. Spicer-Cain et al. 2023). The following section briefly summarise the current evidence regarding outcomes for children identified in infancy as having increased likelihood of social communication disorders.

Increased likelihood of communication disorders

Autism prevalence is reported globally to average around 1% of all children (Zeidan et al. 2022). Over the past decade studies have reported that children at increased likelihood of autism show differences in their language (Spicer-Cain et al. 2023), social development (Walton 2016) and early behaviour (Charman et al. 2015). Jones et al. (2014) review indicates multiple factors that might be important in early identification including social interaction, gesture, language and motor development although less convincing evidence currently exists for repetitive behaviours and executive function. There has also been a recent interest in early trajectories associating with likelihood and diagnostic status (Longard et al. 2017).

Developmental Language Disorder affects just over 7% of children (Norbury et al. 2016) and like autism has life-long implications (Dubois et al. 2020). Yet, in contrast to autism, there is much less research about DLD in general (McGregor 2020) and almost none examining very early differences in children with increased likelihood of this communication difficulty. One exception is research by Spicer-Cain et al. (2023) which indicated that few differences were identifiable at 12 months for this group, despite clear patterns of difference in a group at increased likelihood of autism. There has also been some recent work reviewing the robustness of language screens for slightly older children (Bao, Komesidou, and Hogan 2024). A review of the limited DLD evidence base suggests that factors such as gesture, vocabulary, sentence comprehension and absence of two-word combinations at 30 months might be useful places to start (Sansavini et al. 2021). In children at average likelihood of developing autism or DLD, infant communication skills have been shown

to predict language at 7 (Määttä et al. 2016). However, indicators from very young ages and for populations at increased likelihood of communication disorder have not yet reliably been identified for DLD.

Methodological issues

Although there is some evidence that increased likelihood methodology may be useful, there are several complexities surrounding explorations of this kind which have not been fully addressed in the literature, largely because the developmental trajectories reported are relatively short term. Researchers have begun to highlight the fact that increased-likelihood research is complex and raises many complex challenges. For example, Zwaigenbaum et al. (2009) begin to address some of the methodological issues that are associated with increased likelihood infants. They emphasise the importance of looking at the individual level, sample size issues and the need for careful selection of assessments. Similarly issues around heterogeneity, classification of outcomes (especially for non-autism diagnoses) and the need for multiple measurements of each construct have also been raised (Jones et al. 2014). This literature also briefly touches on some other key issues which have not been much discussed elsewhere. These include the length of time to follow up assessments, the initial inclusion criteria and classification framework for 'higher likelihood' participants, change in likelihood status and the need for comparison groups from families with other clinical profiles. Despite these methodological challenges being raised, there have been no papers to the authors' knowledge which have documented these issues empirically, or which have focussed on the stability and robustness of likelihood status.

Present study

This article addresses some of these methodological gaps by reporting on a group of infants who were categorised as being at either average likelihood of developing communication difficulties (AL); at increased risk of structural language impairment (but not social communication disorder) (ILLI); or at increased risk of social communication difficulties (with or without language concerns) (ILSCD). The sample were then followed up to age 7 years which is relatively unusual in the increased-likelihood literature. The following questions were asked:

1. Does likelihood status change or remain the same for children over time?

2. Is movement over time affected using broad versus strict Time 1 classification criteria?
3. How does likelihood status relate to preliminary diagnostic outcomes?

Note that this article does not consider whether early predictors may be useful in showing more dimensional differences between language scores across likelihood groups which is not the focus here, and is reported elsewhere in infancy (Spicer-Cain, Hasson, and Botting 2024) and at later ages.

Methods

Recruitment

The families who took part in the study were recruited via the use of social media. A project website was established, and social media channels including Facebook and Twitter were used to share information about the project. Parents were able to access the website to view relevant information about the study, and could submit a contact form if they wished to receive further information. It is important to note that the wider study was aiming to recruit groups of increased likelihood children and was actively targeting these groups, rather than recruiting a fully representative sample. The limitations of this strategy are discussed later. Adverts to take part therefore specifically invited families who had a one-year old either with or without older children who had autism or DLD, and used related hashtags to draw attention to these conditions.

Ethical approval was obtained from the City St George's University of London SHPS ethics committee prior to the commencement of the study. Consent was taken from parents in writing at the start of the research visit. The children taking part were too young to give verbal assent, but a willingness to engage with the researcher was taken to indicate implied assent to the session.

Participants

Participants initially comprised 111 children who were part of a wider study of language and communication skills which was not initially designed to track likelihood status over time, but to test a new battery of dynamic assessment for infants. The responses to dynamic assessment of early communication skills have been previously reported for 92 of this group (Spicer-Cain, Hasson, and Botting 2024). Families were recruited through schools, social media and

clinical contacts and represent a volunteer sample. Infants recruited to the study met the following inclusion criteria:

- Age of 23 months or younger at the time of first assessment.
- No known developmental, physical or sensory difficulties at the time of enrolment in the study.
- Exposure to English in the home for at least 40 h per week.

For the purposes of this longitudinal report, participants also needed to have likelihood and diagnostic outcome data available at a second time point when they were 7 years of age (Time 2). There were 67 children who were eligible and whose data were used in the current analyses. There were no significant differences between those who are included in this article and those were not, regarding ethnicity, IDACI (Income Deprivation Affecting Children Index, <https://www.gov.uk/government/statistics/english-indices-of-deprivation-2019>), sex, age, or time 1 likelihood status (all p values > 0.15).

Demographic data related to participants was collected using some questions from the Parent Report Questionnaire of the UK Communicative Development Inventory (Alcock et al. 2020), namely the questions on parental age, parental education, family income and exposure to additional languages. Two additional questions were also included on a separate form asking about history of developmental difficulties in the immediate family, and difficulties during pregnancy or birth. Additionally, a verbal family history was taken from all parents by the first author, who is an experienced speech and language therapist. Details of the participants are presented below in Table 1 for the whole sample.

Measures and analysis

On the basis of the demographic data collected, children were divided into three groups based on their genetic likelihood of language and social communication difficulties at 2 time points – when they were infants (between 2013 and 2015) and again when they were 7 years of age (between 2000 and 2022).

At both time points the groups were initially defined in the same way using broad criteria as follows:

- Average Likelihood Group (AL): The inclusion criteria for this group were that the infants were considered to be typically developing (i.e. there were

Table 1. Demographic characteristics of whole sample ($n = 67$).

Sex	34 (50.7%) female / 33 (49.3%) male
Age months	12.7 (SD = 3.4)
Birth order	26 (38.8%) 1st born 25 (37.3%) 2nd born 11 (16.4%) 3rd born 5 (7.5%) 4th born
Ethnicity ($n = 59$)	49 White British 8 Mixed ethnicity 2 Other
Maternal education ($n = 66$)	60 (91%) Graduate or postgraduate education 6 (9%) High school education or below 16497.15 (SD = 7752.87)
IDACI rank ($n = 66$) (postcode area score between 1 and 32,844, 1 being the most deprived)	
IDACI = Income Deprivation Affecting Children Index.	

no diagnoses of genetic disorder, physical disability or sensory impairment during the first year of life); and they had no immediate family members (parents or siblings) with evidence of language, literacy or social communication difficulties.

- **Increased Likelihood of Language Impairment (ILLI):** The inclusion criteria for this group were that infants were considered typically developing but had siblings and/or parents with a diagnosis of language disorder, dyslexia or literacy difficulties; or a history of late talking; or who had received speech and language therapy for language and/or speech; and had no immediate family with autism or concerns around social communication.
- **Increased Likelihood of Social Communication Difficulties (ILSCD)** (with or without additional language issues): The inclusion criteria for this group were that the infants were considered typically developing but an older sibling, half-sibling or and/or parent was diagnosed with autism; was under assessment for autism diagnosis; or that these family members scored in the clinical range on the Social Interaction Deviance Composite Score of the Children's Communication Checklist, 2nd Edition (CCC-2; Bishop 2003).

As detailed above, in this broad approach identifying increased likelihood groups involved using a number of indicators as well as clinical diagnoses, and included half siblings. This is different to some of the previous literature, where parent or sibling diagnosis has been required, but mirrors some recent studies where children whose parents or siblings are suspected of having a condition, or who score highly on screening measures have also been included in increased likelihood groups (e.g. Charman et al. 2023; Bazelmans et al. 2024). On the one hand this means that our likelihood groups are relatively heterogeneous, but on the other hand this reflects our

knowledge of familial patterns in communication disorders and the profile of clinical caseloads.

In a second analysis stage, the initial likelihood groups were reclassified using stricter criteria. In this categorisation the analysis only included children whose immediate relatives had confirmed diagnoses or assessment pathways as having increased likelihood status.

Note that this analysis is not reporting on change in infant profile such that increased signs of communication disorder have appeared later in the proband. Instead, it sought to document change in likelihood status based on affected family member status. Although this may be informally documented in sibling studies, to our knowledge this has not been formally reported previously.

The analysis did however explore proband diagnostic outcomes at 7 alongside both likelihood methods described above. It considered outcomes mainly a broad way, considering any source of concern as valid because it is known that formal diagnoses of communication disorders are often made later in childhood (Davidovitch et al. 2023; de Bree, Wiefferink, and Gerrits 2024) even in children who have affected siblings (Bazelmans et al. 2024), and because very few children ($n = 8$) have formal assessment, diagnostic and treatment outcomes. However, in order to examine the best fit predictions for later outcomes relationships between outcome and both broad and stricter Time 1 groupings are reported, and the tables indicate where those with formal outcomes sit in terms of the likelihood groups.

Data was stored and managed using SPSS v.30 (IBM, 2024).

Results

Likelihood status reclassification across time

Using broad criteria

Initially all likelihood status was determined by using the broader criteria set out in methods. In this

Table 2. Demographic information for the 3 groups.

	AL	ILLI	ILSCD
Proportion of sample	$n = 39/67$ (58.2% of the whole sample)	$n = 17/67$ (25.4% of the whole sample)	$n = 11/67$ (16.4% of the whole sample)
Sex (m/f)	22 / 17	7 / 10	4 / 7
Age at first assessment (mean/SD)	11.8 months (2.8)	12.5 months (2.5)	17.1 months (3.4)
Exposed to other languages	5 (12.8%) (Swedish = 1, Finnish = 1, German = 2, Italian = 1)	0 (0%)	1 (5.9%) (French)
Weekly hours exposed to other languages (mean/SD)	24.8 (8.6)	N/A	24 (N/A)
First borns (n; %)	19 (48.7%)	7 (41.2%)	0 (0%)

AL = Average Likelihood; ILLI = Increased Likelihood of Language Impairment; ILSCD = Increased Likelihood of Social Communication Difficulties.

section, the characteristics of children in each of the likelihood groups are reported. Next, information about reclassification of status based on 7-year family information is presented. Note that change in likelihood status is not due to proband children showing more clinical behaviours, but to new family history information revealing increased numbers of affected family members.

Time 1 likelihood status. At time 1 when infants were 12 months of age, most of the children were in the average likelihood group. Demographic and family information about the children in each group is given in Table 2.

For the average likelihood group there were no concerns about the speech, language, literacy or social communication of siblings or parents. In total, 6 of the ILLI children had elder siblings who were currently accessing speech and language therapy for speech and/or language, where there were no concerns about social communication, and two of these elder siblings presented with severe language difficulties as part of a profile of global developmental delay ($n = 1$) or Down Syndrome ($n = 1$). A further 2 children had a parent with a history of speech and language therapy or concerns around late talking; 2 children had siblings with dyslexia; and 7 had parents who reported a diagnosis of dyslexia. In the ILSCD group, 9 children had elder siblings or half-siblings with a diagnosis of autism or who had been referred for a diagnostic assessment for autism. For the remaining 2 children, the CCC-2 completed by parents indicated social communication difficulties in at least one elder sibling, *via* the Social Interaction Deviance Composite scores falling at or below -14 , and parents had concerns about social interaction. Three elder siblings in this group had additional diagnoses: one of Attention Deficit Disorder, one of Down Syndrome and one of Cri-Du-Chat Syndrome.

Time 2 likelihood status. When the children were 7 years of age the caregivers gave a repeated family

history which was used to reclassify the children again with this information. At this point, regrouping revealed that 17/67 (25.4%) of the children with follow up data had moved groups as follows:

26/67 (38.8%) children were classified at time 2 as AL (change of -13 children)

21/67 (31.3%) now met criteria for ILLI (change of $+4$ children)

20/67 (29.9%) could now be classified as ILSCD (change of $+9$ children)

Eight of the 39 AL children (20.5%) could now be classified as ILLI, and 5/39 (12.8%) as ILSCD. Four of the 17 ILLI children (23.5%) could now be classified as ILSCD based on new information. All of the 11 ILSCD children remained in this group. Thus, the proportions of children in each group were now much more evenly spread across likelihood groups. Figure 1 illustrates the changes. The number of children in the combined increased likelihood groups (ILLI + ILSCD) increased from 41.8 to 61.2%.

Reasons for likelihood status changes. Of the 8 children who were originally classified as AL, and later classified as ILLI, 3 had parents who had received a dyslexia diagnosis between T1 and T2. 2 further children had elder siblings who had been assumed to be typically developing at T1, but were showing literacy difficulties at T2. Four children were reclassified on the basis of having younger siblings with speech and language difficulties, one of whom also had a parent who had received a dyslexia diagnosis.

Of the five children who were originally classified as AL, and later classified as ILSCD, 1 had an older sibling who was undergoing autism assessment, and the four others had older or younger siblings who scored in the clinical range on the Social Interaction Deviance Composite Score of the CCC-2 (Bishop 2003) at T2 assessment.

Of the four children who were originally classified ILLI, and later classified as ILSCD, two had older

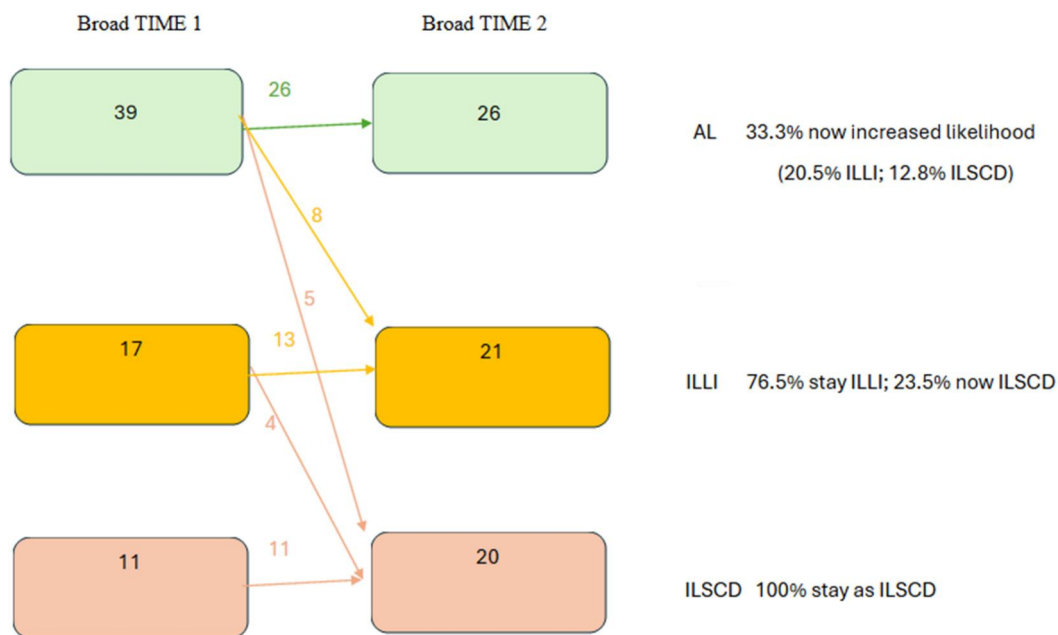


Figure 1. Change in likelihood status using broad and strict criteria ($n = 67$).

siblings who had received an autism diagnosis between T1 and T2, one had an older sibling who was undergoing autism assessment, and one had an older sibling who scored in the clinical range on the Social Interaction Deviance Composite Score of the CCC-2 (Bishop 2003) at T2 assessment. In two of these cases, these older siblings had been noted at T1 to have speech and language difficulties, but not concerns related to autism.

Using stricter criteria

Because the wider study used relatively broad criteria to indicate increased likelihood, the analysis above was repeated using the stricter criteria outlined in methods. First the differences that stricter criteria made to each group at Time 1 are reported, before moving on to present any changes in likelihood status over time. Importantly, only 4 children moved groups as a result of stricter criteria, therefore the detailed demographic information is not repeated here.

Time 1 likelihood status. When stricter criteria were applied, the AL group expanded slightly to include 41/67 children including 20 first-borns (61.2% of the sample of 67), the two additional children being reclassified from the ILLI group. This was because they had a parent or sibling with a history of late talking but no formal assessment or diagnosis of language difficulties.

However, there were still 17 children in the ILLI group (7 first-borns; 25.4%). This was because 2

children were reclassified *out* into the AL group, but 2 children were now included *within* this group who were previously classified in the ILSCD group below. Both of these previously ILSCD children had half-siblings with diagnosed dyslexia and concerns had been raised about sibling autism but were not formalised. They therefore no longer met ILSCD criteria using stricter classification. In total, 10/17 had family members with dyslexia (often also with secondary speech/language concerns) and 7 had relatives who had received a diagnosis or treatment for speech and language difficulties.

Nine of the ILSCD group from the broader approach met our stricter criteria groupings (13.4% of the sample of 67), none of whom were first-borns. Two of the 11 children in our broader classification for this group were moved into the ILLI group when using stricter criteria. Four children had relatives with a diagnosis of autism, and five others had family members who were undergoing diagnostic assessment for autism.

Thus, at Time 1, using stricter criteria did not markedly change the composition of the groups, with only 4 children reclassified into different likelihood groups.

Time 2 likelihood status. When re-reclassified using stricter criteria at 7 years of age, 11/67 (16.4%) children had moved likelihood groups at Time 2:

- 33/67 (49.2%) were classified as AL (change of -8)
- 21/67 (31.3%) as ILLI (change of $+4$)
- 13/67 (19.4%) as ILSCD (change of $+4$)

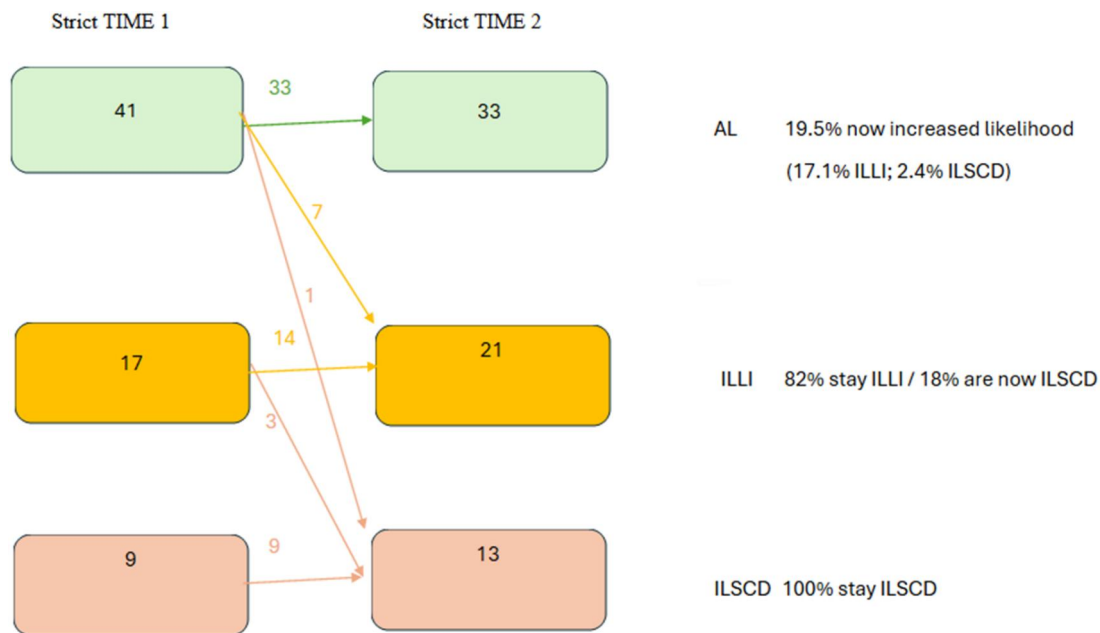


Figure 2. Change in likelihood status using broad and strict criteria ($n = 67$).

The movement patterns were represented by 7/41 AL children (17%) who were now considered ILLI, and 1/41 (2.4%) as ILSCD. Three of the 17 ILLI children (23.5%) could now be classified as ILSCD, and again all 9 of the ILSCD children stayed in the same classification. See Figure 2 for details.

Using the stricter criteria, the number of children in the combined increased likelihood groups (ILLI + ILSCD) increased from 38.8% to 50.7%.

Reasons for likelihood status changes. Of the 7 children who were originally classified AL and now classified ILLI, 2 had parents who received a dyslexia diagnosis between T1 and T2, and 2 had older siblings who were showing literacy difficulties at T2 who had been presumed typically developing at T1, but was undergoing autism assessment at T2. 3 children had younger siblings who had received or were receiving support from an SLT for speech and/or language difficulties.

Only one child moved from AL classification at T1 to ILSCD classification at T2 using stricter criteria. This child had an older sibling who was assumed typically developing at T1, but was undergoing autism assessment at T2.

Of the 3 children who were originally classified ILLI and now classified ILSCD, two had older siblings who had received an autism diagnosis between T1 and T2, and one had an older sibling who was undergoing autism assessment at T2. Two of these older siblings had been identified with speech language difficulties but not concerns related to autism at T1.

First born children

Note that a substantial proportion of the 67 children were first-borns (26/67; 38.8%) mainly in the AL group, and therefore likelihood status may have been difficult to ascertain. Therefore, movement analysis was re-run with these first-born children removed. Using broad criteria, 11/41 children (26.8%) moved likelihood group and using strict criteria 10/41 (24.3%) moved classification representing similar results to the entire cohort.

Summary of likelihood status results

A substantial minority of children moved likelihood classifications. This was driven mainly by changes from the AL category into the ILLI group. Using stricter criteria did not affect overall proportions of change a great deal. However, the overall proportion of children who were reclassified did fall from 25.4% to 16.4% and this was mainly due to fewer AL and ILLI children being re-identified as ILSCD at Time 2.

Birth order did not appear to make a meaningful contribution to reclassification effects, although movement proportions were more similar using broad vs. strict criteria when the first-borns were excluded from the analysis.

Time 1 likelihood status and 7 year outcome status

Table 3 shows the broad outcomes for each Time 1 risk group using broad criteria and Table 4

Table 3. Broad likelihood status and outcome at 7 years of age ($n = 67$).

		No LI or SCD concerns	LI	SCD
T1 likelihood broad	AL	22 56.4%	7 (4) 17.9%	10 (2) 25.6%
	ILLI	11 64.7%	1 5.9%	5 (1) 29.4%
	ILSCD	4 36.4%	1 (1) 9.1%	6 (1) 54.5%

Children in brackets are the small subset of 8/67 children with formal AX, DX or TX.

LI = Language Impairment; SCD = Social Communication Difficulties.

Table 4. Stricter likelihood status and outcome at 7 years of age ($n = 67$).

		No LI or SCD concerns	LI	SCD
T1 likelihood strict	AL	24 58.5%	7 (4) 17.0%	10 (2) 24.3%
	ILLI	10 64.7%	1 5.9%	6 (1) 29.4%
	ILSCD	3 33.3%	1 11.1%	5 (1) 55.6%

Children in brackets are the small subset of 8/67 children with formal AX, DX or TX.

LI = Language Impairment; SCD = Social Communication Difficulties.

shows outcomes using strict criteria to classify at Time 1.

In total, nearly half all the children in this study (30/67; 44.7%) had been identified at Time 2 (aged 7) as having some broad concerns around either language or social communication which is much higher than expected even when oversampling increased likelihood groups. For most of these children however, these concerns were not formalised and some were parent worries. For 8 children (in brackets on the tables), formal assessment procedures, diagnoses and intervention for a communication disorder had taken place.

As can be seen, there was little predictive value in likelihood status alone, with high numbers of children in all groups having been flagged as experiencing issues with social communication. The rate of late identification of issues is unexpectedly high, especially for the AL group. Possible reasons for this and the considerations needed for future research will now be discussed.

Discussion

This article sought to explore the longer-term likelihood status of a group of children at increased risk of communication disorders. In relation to our first research question, changes in family diagnosis information led to considerable movement in status from Time 1 (1–2 years of age) to Time 2 (6–8 years of age). This was largely driven by children initially in an average likelihood group being reclassified as concerns emerged about family members. Interestingly, however, the trend was not particularly linked to first-borns included in this subgroup. Regarding

research question 2, our data also suggests that using broader criteria than often applied (i.e. taking parental concern into account) does not much change overall proportions in likelihood groups, the movement patterns between them over time, or the associations between family likelihood and 7-year outcome. Thus, a broader approach, may help recruitment to sibling studies and improve understanding of subclinical and broader phenotypes. Finally, for the final research question, this dataset suggests that likelihood classification established in infancy does not provide useful predictions for diagnostic outcomes at age 7, regardless of whether strict or broad criteria is used. These results are now discussed in detail.

Although at one level, it may not seem surprising that likelihood status changes over time, to our knowledge this has never been formally reported or discussed in the literature as a methodological challenge. Note that this change in likelihood status has been driven by changes in sibling or parent profiles, not by increased communication concerns regarding the probands themselves. One reason that this is not well documented may be that most studies do not follow up increased likelihood infants beyond early childhood. It is therefore interesting to note the different pathways of familial likelihood that this longer-term follow up has afforded, revealing that a number of different factors contributed to the moveable status observed. These included the fact that some older siblings were diagnosed with communication difficulties relatively late in middle childhood in line with existing literature (Brett et al. 2016; de Bree, Wiefferink, and Gerrits 2024). Furthermore, because developmental diagnoses are somewhat fluid over time, a few older siblings also shifted profiles, especially from concerns about language difficulties to being identified as also being autistic. Because the proband children were only 12 months in this study, many of their older siblings were not yet at school at Time 1 assessment. Thus, the sibling literacy status was not established until follow up when dyslexia was more apparent. In a similar vein, some parents also gained diagnoses in the intervening period or had reflected on their own communication history more accurately. Finally, this is the only study to our knowledge to note the diagnoses of younger siblings. While this factor is perhaps less usefully clinically, this information is of interest theoretically when tracking familial aggregation of communication difficulties. In sum, the changes in likelihood status reported here may have implications for the interpretation of previous literature which has suggested that the likelihood of

being autistic (Hansen et al. 2019; Ozonoff et al. 2024) or having a language impairment (Capelli et al. 2024) is higher in families with recognised forms of these conditions.

Overall, the data from this study suggest that likelihood status in infancy is not a particularly useful predictor of later communication disorders, even within the most stable subgroup (ILSCD). Instead, research and practice need to focus on establishing feasible early assessments that can reliably screen children who may go on to have additional needs. Large cohort studies with general population samples have identified reliable associations between infant language and pre-school language development (Peyre et al. 2014); broad language and educational outcomes (Hohm et al. 2007); as well as between parent reported questionnaires and later communication (Gasparini et al. 2024). However other recent studies have shown only weak associations between infant language measures and later language status at the individual level (Reilly et al. 2010; Lowe et al. 2023). Thus, more work is needed to provide sensitive direct assessments of communication skills in infancy. One promising avenue is that of Dynamic Assessment, which measures the amount of scaffolding needed for a child to succeed at a task. Our own recent work in this area indicates that good task reliability and differences between likelihood groups can be achieved using such measures, which is a first step towards developing a clinical tool. Nevertheless, even with this approach the picture remains complex: siblings of children with social communication difficulties showed differing patterns of development (rather than lower scores per se), and children at higher likelihood of language and literacy difficulties (ILLI) did not show convincing differences from AL peers (Spicer-Cain, Hasson, and Botting 2024). Studies indicate that parents can often identify concerns with communication before 24 months (Rannard, Lyons, and Glenn 2004) but formal identification and support may only be available much later. Thus, research to improve early diagnosis based on preschool information is essential (Gascoigne and Gross 2017). The current study suggests that a combination of factors may need to be considered to give the best chance of optimum early support to families, including likelihood status (Hansen et al. 2019; Capelli et al. 2024), early communicative behaviour (Spicer-Cain, Hasson, and Botting 2024; Gasparini et al. 2024) and potentially other associated factors (Lebeña et al. 2024).

It is important to note that the knowledge base is confounded somewhat by the wide variation in

assessment and diagnostic pathways even within a country or region (Zavaleta-Ramírez et al. 2020). The professional background of practitioners, the geographical location, manner of referral, socio-economic status and family perseverance are all factors in determining access to a diagnosis or treatment, the type of assessments and the age at which formalised support is given (Thomas, Schulz, and Ryder 2019; Denman et al. 2021). The limited research regarding diagnostic age, particularly for DLD also needs expanding, especially since existing evidence suggests there may be different profiles related to age at diagnosis (de Bree, Wiefferink, and Gerrits 2024). Nevertheless, the rate of infants with autistic siblings who later receive an autism diagnosis appears to be relatively stable over different cohorts at least for autism (Ozonoff et al. 2024).

Thus, in parallel with assessment development, research with extended longitudinal periods is also needed to investigate whether infant likelihood status has better predictive value once children are older (and therefore likely to have a diagnosis if one is warranted). Recent work by Bazelmans et al. (2024) has begun to address this for families with autistic children. The substantial minority of children who move from AL to increased likelihood groups, and who have informal or formal support in place at 7 is of particular interest here, both theoretically and methodologically. The sample of parents in this report had unusually high levels of education, and many were trained as speech and language therapists, teachers or psychologists. The possibility that the participating parents had well informed low-level concerns about their children and were especially motivated to sign up to the study cannot be discounted. These findings also cannot rule out the fact that some of these caregivers may be particularly sensitive to certain behaviours associated with communication difficulties and may be more able to access formal assessment and treatment pathways. The increased public awareness of autism and the disruptive social effects of Covid might also contribute to social concerns being noted more readily. This would in part explain the lower proportion of children being classified as ILSCD at Time 2 when stricter criteria are used.

Limitations and implications for future research and practice

It is important to acknowledge that our inclusion of parents and half siblings in our analysis, as well as features associated with communication disorder

(such as dyslexia and late talking) means that our groups may be more heterogenous than some of the literature. However, these related concerns are often more easily recognisable for families, since awareness of DLD is particularly poor compared to prevalence (McGregor 2020). Given that research into increased likelihood of DLD is not yet very evident in the literature, it seemed important to capture increased likelihood more completely by including these highly related difficulties.

It is important to note that this article does not intend to view changes in likelihood as outcomes in themselves. Instead, the findings highlight that the age at which likelihood is considered (especially in practice) might affect the status of a given individual. In addition, if 'true' likelihood is sometimes not detectable in infancy (i.e. false negatives where the infant is deemed average likelihood, but at a later family members gain diagnoses) then this might be a useful contextual issue within with to consider existing sibling research into developmental disorders. The challenges of moveable likelihood status are likely to be particularly important for studies of familial trends in DLD and other developmental disorders where this evidence base is not as well-established as it is for autism.

Nevertheless, it is important to note that our sample was neither large nor very diverse. The social-media and word-of-mouth based recruitment strategy used here led to our sample not being representative of all families. Thus, while this article aims to raise a discussion about this potential methodological challenge rather than provide definitive conclusions about likelihood status, in future research it is essential that a wider mix of families and larger samples are recruited. In particular this will help to determine whether this high proportion of clinically identified AL children stems from specific sample demographics. Importantly, if there are biases towards parents with high levels of observation or towards those with existing concerns about their infant, for much of the previous literature this would not be visible due to the short follow up periods involved.

Conclusions

This study's findings suggest that likelihood status may be somewhat moveable as children develop, due to additional family diagnoses. Furthermore, this approach may not be a particularly useful classification strategy for recruiting and predicting children who will later develop communication difficulties.

This is evidenced by the fact there were no obvious differences across our AL, ILLI and ILSCD groups in terms of 7-year diagnostic pathways. Nevertheless, it is highly plausible that these groups may show more subtle differences in their later language and social outcomes, which warrants further investigation. These data are currently being collected and analysed for this cohort.

Regardless of which type of outcome is the focus, the findings presented here indicate that the picture may be complicated by the fact that infant likelihood status seems somewhat moveable. This adds to the methodological conversation begun by Zwaigenbaum et al. (2009). It is important therefore for clinicians and researchers to revisit family history conversations to note newly emerging concerns about direct family members. Our findings also suggest that the inclusion of more informal concerns by practitioners may lead to slightly less stability in likelihood groupings, but in contrast these broad criteria are more likely to capture useful information for diagnosis and support at an individual level.

This longer-term data raises issues when using increased likelihood methods that need considering when conducting research and needs further discussion in the literature. A more nuanced, dimensional approach involving infant assessment is likely to lead to greater progress in establishing successful early identification and intervention programmes for children with communication difficulties.



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No potential conflict of interest was reported by the authors.

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Data availability statement

The dataset on which this paper is based includes personal information. Data is therefore not available publicly. Reasonable requests for data that can be sufficiently anonymised should be sent to the authors directly.

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