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Importance of accounting for sibling age when examining the association between family size and early childhood cognition, language and emotional behaviour: a birth cohort study

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ABSTRACT

Objectives Larger sibships are associated with poorer cognitive and language outcomes but have different impacts on child emotional development. Previous studies have not taken into account sibling age, nor have impacts across multiple neurodevelopmental domains been considered in the same participant group. This study investigated the influence of family size indicators on early childhood cognitive, language and emotional-behavioural development. The effect of sibling age was considered by evaluating these relationships separately for different sibling age categories.

Design Prospective birth cohort study.

Setting Participants in the Barwon Infant Study were recruited from two major hospitals in the Barwon region of Victoria, Australia, between 2010 and 2013 (n=1074 children).

Participants The 755 children with any neurodevelopmental data at age 2–3 years excluding twins and those with an acquired neurodisability.

Outcome measures Cognitive and language development was assessed using the Bayley Scales of Infant and Toddler Development, Third Edition, and emotional-behavioural development was measured with the Child Behaviour Checklist for Ages 1½–5.

Results Greater household size was associated with a reduced cognitive development score (adjusted mean difference (AMD) −0.66 per extra household member; 95% CI −0.96 to −0.37; p<0.001) without age-specific differences. However, poorer expressive language was only observed for exposure to siblings between 2–6 and 6–10 years older. Having siblings 2–6 years older was associated with less internalising behaviour (AMD −2.1 per sibling; 95% CI −3.1 to −1.0; p<0.001). These associations persisted after multiple comparison adjustment.

Conclusions The influence of siblings on early childhood development varies substantially by sibling age and the neurodevelopmental outcome under study. Although family size alone appears important for cognitive development, age-specific findings emphasise the importance of sibling interaction in early childhood expressive language development and emotional behaviour.

Strengths and limitations of this study

- ► The first birth cohort study to examine the association between family size and early childhood neurodevelopment while accounting for sibling age.
- ► Cognitive, language and emotional-behavioural outcomes were assessed in parallel, allowing for better discrimination of differential patterns of association across these outcomes.
- ► A comprehensive consideration of other determinants for each outcome was made, resulting in robust confounding adjustment.
- ► Large sibships were uncommon in the study sample which limits the evaluation of dose–response patterns and the generalisation of the results to very large sibships and households.
- ► The study only considers older siblings given the young age of the index child and did not have enough power to examine sex-specific effects.

INTRODUCTION

The early environment is critical in promoting early child neurodevelopment and lays the foundation for long-term outcomes and future productivity at a societal level.[1] Social environment, including family size indicators, is an important developmental determinant, with the focus of prior studies being on overall sibship size and, to a lesser degree, birth order.[2 3]

Both larger sibship size and later birth have a well-established association with poorer cognitive, language and educational outcomes extending into adulthood.[4–7] The most influential theory to explain these findings—the Resource Dilution model[5–7]—proposes that as sibship size increases, parental resources available to individual offspring decrease.[5–7] The concept of shared parental resources

extends beyond socioeconomic resources. There is evidence from the Avon Longitudinal Study of Parents and Children (ALSPAC), a large UK birth cohort, that children in larger sibships receive less of their parents' time and that within sibships latter-born children are at the greatest disadvantage both in terms of parental investment and neurodevelopmental outcomes.[8]

Intriguingly, different patterns are seen in the association of sibship size or birth order with mental health and emotional and behavioural development. Latter-born children exhibit less internalising problems such as anxiety and depression.[9–14] This appears to differ by age of sibling or birth order. In the ALSPAC birth cohort, the 4-year-old children with older siblings displayed less internalising emotional behaviour,[12] consistent with other international studies,[9–11 13 14] but those with younger siblings displayed more internalising behaviour.[12] Possible explanations are that relationships with older siblings provide a buffer to stress or that latter-born children benefit from greater parental experience and a household already adapted to children.[12] No consistent pattern of association has been observed between having older siblings and child externalising emotional behaviour across these same studies.[9–12]

These differences in findings by outcome suggest different impacts of having siblings for different domains of child development. Findings for child internalising, in particular, suggest that sibling age may be a factor but this has never been directly examined. That is, age-specific sibling effects have not been investigated for either child cognition or emotional and behavioural development. Previously, we have demonstrated that sibling age has an important impact on infant immune development.[15 16] We reported in detail on household composition and sibling patterns in association with immune disorders, taking into account age of index child and siblings as a source of exposure and dose response.[15 16] Here, we investigated the influence of various family size indicators on cognitive, language and emotional development by 2-3 years of age. In particular, we aimed to assess the effect of sibling age by evaluating these relationships separately for different age categories. It was hypothesised that accounting for sibling age would clarify the role of siblings in neurodevelopment.

METHODS

Sample

The aims and methodology of the Barwon Infant Study (BIS) have been described previously.[17] In brief, a birth cohort of 1074 infants was assembled in the Barwon region of Victoria, southwest of Melbourne, Australia, using an unselected antenatal sampling frame. Women were recruited prior to 28 weeks' gestation between 2010 and 2013. Infant exclusion criteria were: (1) delivery before 32 weeks, (2) serious neonatal illness, (3) major congenital malformation or genetic disease and (4) family having moved out of the Barwon Statistical Division by the time of birth. Mother–infant pairs were reviewed at regular intervals. Comprehensive questionnaire, clinical and biological measures were collected antenatally and at birth; 4 weeks; 3, 6, 9, 12 and 18 months; and 2 years.[17] Questionnaire data include over 1500 measures on the infant, mother and father, including: multiple indicators of socioeconomic status, sociocultural and linguistic factors, maternal diet and lifestyle during the pregnancy, breast feeding and infant diet, infant behaviour and temperament, maternal stress and mental health through pregnancy and subsequent follow-up, parenting practices and childcare arrangements. Clinical measures include clinical perinatal and neonatal history, birth anthropometrics, and a range of detailed phenotype indicators across multiple study domains including child socioemotional, behavioural, cognitive and language development (neurodevelopment).[17]

This analysis includes those children with relevant neurodevelopmental data at age 2–3 years and excludes twins (n=20 in inception cohort) and children with known acquired neurodevelopmental disorder (n=1).

Measures

Child neurodevelopment at 2–3 years of age

Cognitive and language development were assessed by a trained research assistant at age 2–3 years with the Cognitive Development, Expressive Communication and Receptive Communication subscales of the Bayley Scales of Infant and Toddler Development, Third Edition (BAYLEY-III).[18] This measure has high test–retest reliability with corrected Pearson coefficients ranging from 0.85 to 0.87 for the relevant subscales at ages 19–26 months.[19] Emotional behaviour was assessed by the Internalising and Externalising subscales of a standardised parent-reported measure, the Child Behaviour Checklist for Ages 1½–5 (CBCL).[20]

Data analysis

BAYLEY-III raw scores (adjusted for postconceptional age at test, child sex, assessor and assessor's test experience) and CBCL T-scores (adjusted for child age and sex) were used in analyses.

Factors associated with child development were first evaluated in linear regression models adjusting for process and measurement factors only (such as child age at assessment or administering research assistant). Possible non-linear associations were assessed using fractional polynomial models, and separately by stratifying by quintiles and/or clinically relevant cut-offs. Nominal categorical variables were evaluated by each stratum versus baseline and by a likelihood ratio test of models with and without inclusion of the categorical variable. Tests for trend were additionally undertaken for ordinal categorical covariates by using a single predictor taking category rank scores.

In our further evaluation of sibling effects and family size factors, birth dates of infants and their siblings were used to determine age of siblings at birth of the index

child. The siblings were then placed into age-specific categories (0 to <2 years, 2 to <6 years, 6 to <10 years and 10 to <18 years). Siblings aged 18 years and over were classified as adults. The effect of non-parental adults in the household was evaluated based on the number of additional adults residing in the family home beyond the first two. A separate indicator variable was included in the model to account for lone parent status. Total household size was defined as the number of children and adults residing in the home excluding the index child and first two adults. Again, adjustment was made for lone parent status. As sibling variables were not independent, the association between number of siblings in each age-specific category and neurodevelopmental outcome was evaluated in separate multivariable linear regression models. Additional analyses included mutual adjustment for all other age-specific categories, and adjustment for total number of household residents. The latter served as a test of age-specific effects that differed from an age-independent effect of additional household members. All models were adjusted for process and measurement factors (such as child age at assessment or administering research assistant) and for potential confounders.[21] Additional potential confounders were included if they changed the estimate for the main effect by at least 15%.[22 23] Adjustments for multiple comparisons were also made using the method of Benjamini-Hochberg with a false discovery rate of 5%.[24]

In a sensitivity analysis, inverse probability weighting was used to account for initial non-participation and attrition by assessing whether differences between responders and non-responders likely influenced estimates.[25] We used Stata V.14.2 software (StataCorp) for all analyses.

Patient and public involvement

Extensive one-on-one consultations with the families of paediatric patients and other members of the public were used to inform the design of BIS. A survey which had identified Australian paediatricians' top research priorities[26] was also referred to. Potential participants were informed of the time commitment and biospecimen collection at each wave of the cohort. A consumer group is actively involved in disseminating study results to participants, for example, through newsletters and a website.

Parents or guardians provided written informed consent at prenatal recruitment and again when the child was 2 years of age.

RESULTS

Study sample

At the 2-year review, 837 (78% of the inception cohort) participated (online supplemental figure 1). Of these, 675 (81%) completed the CBCL. Of the 830 approached to take part in the BAYLEY-III, 703 (85%) participated. One participant was outside of the age criteria at the time of assessment and so this child's BAYLEY-III scores were excluded. Twins (n=10) and children with a known acquired neurodevelopmental disorder (n=1) were further excluded. Overall, 755 children had any relevant neurodevelopmental data at age 2–3 years, with final models investigating family size indicators including n=635–637 for CBCL outcomes, n=663–665 for Bayley III Cognitive Development, and n=556–558 for Bayley III Expressive Communication. The majority of participants were full-term infants with Australian-born parents (table 1). Median number of older siblings in the households was 1 (IQR 0–1; further details table 1) and 5.6% of families had more than two adult family members. Mean child age at BAYLEY-III assessment was 29.4 months (SD 1.7) and mean age at completion of CBCL was 29.5 months (SD 1.8; table 1).

Neurodevelopmental outcomes

Mean performance on all BAYLEY-III measures (table 1) was above that expected from US normative data, consistent with prior Australian research.[27 28] Performance across the BAYLEY-III measures was highly correlated (Pearson coefficient r=0.66–0.73; online supplemental table 1). Due to this collinearity we focus on findings for Cognitive Development and Expressive Language, which had the greatest divergence (Pearson correlation coefficient 0.66). There was little correlation between the BAYLEY-III and CBCL (r=−0.06 to 0.04; online supplemental table 1).

Factors associated with child neurodevelopment

We identified a comprehensive range of factors associated with each of the neurodevelopmental outcomes evaluated. Where there was evidence of non-linear patterns of association between continuous factors and neurodevelopmental outcome, this was addressed by modelling the factor as a categorical variable, with the exception of household income where we observed a ceiling effect with each outcome at $A100 000–$A125 000 and an otherwise linear association below this, and addressed this by censoring household incomes above $A100 000 at $A100 000. Greater sibling number and total household size were each associated with lower cognitive and language scores and with less internalising behaviour (table 2). Additional key factors associated with cognitive and language scores included higher parental education, maternal country of birth, lone parent status, preterm delivery before 34 weeks and breastfeeding duration (table 2; online supplemental table 2); and these were included as potential confounding factors in further analyses of family size indicators with cognitive and language outcomes. Key factors associated with internalising or externalising behaviour included household income, maternal country of birth, maternal age, lone parent status, maternal smoking in pregnancy, maternal marijuana use in pregnancy, mode of birth, birth condition (Apgar score at 5 min), breastfeeding duration and maternal perceived stress from pregnancy to 6 months (table 2; online supplemental table 2); and these were included as potential confounding factors in further analyses of family size indicators with emotional and behavioural development.

Table 1 Characteristics of the project sample (n=755)

	N	Mean (SD) or % (n)*
Socioeconomic and demographic factors		
Sex (male)	755	53.6% (405)
Age at CBCL test (months)	691	29.5 (1.8)
Age at BAYLEY-III test (months)	668	29.4 (1.7)
Maternal age at conception:	755	
<25 years		6.0% (45)
25–40 years		90.9% (686)
>40 years		3.2% (24)
Paternal age at conception:	719	
<25 years		4.6% (33)
25–40 years		83.2% (598)
>40 years		12.2% (88)
Median parental education category†	754	4.3 (1.0)
Mean annual household income antenatal and first year (10 000 AUD units)‡	749	9.6 (3.2)
Family size indicators		
Number of adults in household at birth (including parents):	752	
1 adult		2.0% (15)
2 adults		92.4% (697)
≥3 adults		5.6% (42)
Number of siblings in household at birth§:	753	
0 siblings		44.2% (333)
1 sibling		35.9% (270)
2 siblings		16.5% (124)
3 siblings		2.9% (22)
≥4 siblings		0.5% (4)
Any siblings aged 0 to <2 years in household at birth§	753	12.2% (92)
Any siblings aged 2 to <6 years in household at birth§	753	42.4% (319)
Any siblings aged 6 to <10 years in household at birth§	753	6.8% (51)
Any siblings aged 10 to <18 years in household at birth§	753	4.5% (34)
Cultural-linguistic factors		
Any grandparents identifying as Aboriginal or Torres Strait Islander	752	1.9% (14)
Mother born in Australia	755	90.2% (681)
Antenatal health, diet and exposures		
Maternal total fish intake (g/day)	728	27.3 (26.2)
Maternal fish oil supplementation in pregnancy	735	54.4% (400)
Average alcohol intake in trimester 1 (standard drinks):	755	
Nil		64.1% (484)
<1 a week		27.4% 207)
1–6 a week		8.1% (61)
1–3 a day		0.4% (3)
No of occasions >5 standard drinks at a time over trimester 1:	740	
Nil		92.0% (681)
1–2		5.8% (43)
>2		2.2% (16)
Maternal smoking (timing over this pregnancy):	751	

Continued

Table 1 Continued

	N	Mean (SD) or % (n)*
Nil		88.4% (664)
Early (preconception and/or T1) but stopped		7.9% (59)
Early and late		3.7% (28)
Maternal recreational drug use in this pregnancy (marijuana)	749	1.3% (10)
Birth outcomes		
Premature birth (<37 weeks)	755	3.8% (29)
Mode of delivery:	755	
Unassisted vaginal		48.3% (365)
Assisted vaginal		20.5% (155)
Unscheduled caesarean		14.7% (111)
Elective caesarean		16.4% (124)
Apgar score at 5 min	745	9.0 (0.9)
Birth weight:	755	
<2500 g (<5 lb 8oz)		1.7% (13)
2500–4200 g		89.7% (677)
>4200 g (>9 lb 4oz)		8.6% (65)
Weight by sex and gestational age at birth (Z-score):	755	
<–1.5 (low)		2.4% (18)
–1.5 to 1.5		86.5% (653)
>1.5 (high)		11.1% (84)
Postnatal		
Breastmilk feeding (duration):	752	
Nil or <1 week		3.6% (27)
1 week to <6 months		31.8% (239)
≥6 months		64.6% (486)
Maternal stress and mental health		
Edinburgh Depression Scale (raw score in pregnancy)	568	5.4 (3.9)
Mean early Perceived Stress Scale score (antenatal, 4 weeks, 6 months)	752	18.1 (6.2)
Parenting practices		
HOME-SF Emotional Support subscale (9 months)	339	8.1 (0.9)
HOME-SF Cognitive Stimulation subscale (9 months)	339	6.6 (1.3)
Neurodevelopment outcomes		
BAYLEY-III Cognitive Scale (scaled score)	667	10.8 (2.0)
BAYLEY-III Receptive Communication Scale (scaled score)	569	12.0 (2.0)
BAYLEY-III Expressive Communication Scale (scaled score)	560	12.1 (2.6)
CBCL Externalising Scale (T-score)	666	44.8 (9.2)
CBCL Internalising Scale (T-score)	666	41.7 (9.1)

*Exclusions: twins, child with known acquired neurodevelopmental disorders, BAYLEY-III scores for child outside of age criteria.
†Highest level of education attained (six categories: <10 years of school, 10–11 years of school, completed year 12, trade certificate/apprenticeship or diploma, undergraduate degree, postgraduate degree).
‡Mean household income reported in pregnancy and at 12-month review with income above $A100 000 censored at a maximum of $A100 000.
§Regardless of whether they are a blood relation.
BAYLEY-III, Bayley Scales of Infant and Toddler Development-Third Edition; CBCL, Child Behaviour Checklist for Ages 1 ½–5; HOME-SF, Home Observation for Measurement of the Environment-Short Form.

Table 2 Selected participant characteristics and associations with neurodevelopmental outcome in four key domains

	Cognitive Development[a]			Expressive Language[a]			Externalising[b]			Internalising[b]		
	Mean Difference	95% CI	P value	Mean Difference	95% CI	P value	Mean Difference	95% CI	P value	Mean Difference	95% CI	P value
Socio-economic and demographic factors												
Maternal age:												
<25 years	−1.7	−2.8, −0.52	0.005	−1.7	−3.5, −0.020	0.05	6.5	3.3, 9.7	<0.001	4.6	1.4, 7.9	0.005
25–40 years	Reference			Reference			Reference			Reference		
>40 years	−1.4	−2.9, 0.15	0.08	−0.63	−2.8, 1.6	0.57	−4.3	−8.1, −0.42	0.03	-3.0	−6.9, 0.83	0.12
Median parental education category[c]	0.84	0.55, 1.1	<0.001	1.0	0.62, 1.5	<0.001	−0.66	−1.4, 0.077	0.08	−0.43	−1.2, 0.30	0.25
Mean annual household income antenatal & first year (10 000 AUD units)	0.19	0.051, 0.33	0.008	0.24	0.042, 0.45	0.02	−0.91	−1.3, −0.55	<0.001	−0.84	−1.2, −0.48	<0.001
Household composition factors												
Total household size at birth	−0.66	−0.96, −0.36	<0.001	-1.0	−1.4, −0.61	<0.001	−0.063	−0.83, 0.71	0.87	−1.3	−2.1, −0.57	0.001
Total number of siblings at birth	−0.61	−0.94, −0.29	<0.001	−1.1	−1.6, −0.69	<0.001	0.063	−0.77, 0.89	0.88	−1.5	−2.4, −0.73	<0.001
Lone parent household	−0.57	−2.2, 1.0	0.49	−0.58	−2.8, 1.7	0.61	1.8	−2.5, 6.2	0.40	4.7	0.37, 9.0	0.03
Cultural-linguistic factors												
Number of grandparents identifying as Aboriginal or Torres Strait Islander	−1.1	−2.7, 0.45	0.16	−0.96	−3.1, 1.2	0.37	0.76	−3.4, 4.9	0.72	0.36	−3.8, 4.5	0.86
Maternal country of birth:												
Australia	Reference			Reference			Reference			Reference		
Majority English-speaking country	−0.39	−1.6, 0.81	0.52	−1.7	3.5, 0.0030	0.05	4.4	1.5, 7.3	0.003	2.1	−0.84, 5.0	0.16
Other country	−1.1	−2.5, 0.30	0.13	−0.94	−3.0, 1.1	0.36	−1.1	−4.7, 2.6	0.56	1.8	−1.8, 5.5	0.33
Antenatal health, diet & exposures												
Maternal diet – total fish (g/day)	0.0013	−0.0094, 0.012	0.81	0.0041	−0.010, 0.020	0.58	0.0022	−0.025, 0.029	0.87	0.017	−0.010, 0.043	0.22
Any fish oil supplements in pregnancy	0.63	0.074, 1.2	0.03	0.74	−0.046, 1.5	0.06	0.49	−0.91, 1.9	0.49	−0.55	−2.0, 0.87	0.45
Average alcohol intake trimester 1 (standard drinks):												
Nil	Reference			Reference			Reference			Reference		
<1 a week	0.099	−0.54, 0.74	0.76	−0.68	−1.6, 0.22	0.14	0.48	−1.1, 2.1	0.56	0.19	−1.4, 1.8	0.82
1–6 a week	0.13	−0.91, 1.2	0.81	−1.8	−3.2, −0.29	0.02	0.78	−1.9, 3.5	0.57	1.5	−1.2, 4.2	0.27
1–3 a day	−1.9	−6.1, 2.3	0.37	−2.6	−9.2, 4.0	0.44	−0.45	−11, 10	0.93	3.2	−7.3, 14	0.55
(test of trend)			0.92			0.008			0.49			0.29
Number of occasions>5 standard drinks at a time over trimester 1	0.097	−0.31, 0.51	0.64	−0.55	−1.2, 0.08	0.09	0.59	−0.49, 1.7	0.29	1.1	0.019, 2.2	0.05
Maternal smoking (timing over this pregnancy):												
Nil	Reference			Reference			Reference			Reference		
Early but stopped	0.61	−0.44, 1.7	0.26	0.064	−1.4, 1.5	0.93	2.9	0.26, 5.5	0.03	4.1	1.5, 6.7	0.002
Early and late	−0.64	−2.2, 0.87	0.40	−2.7	−5.0, −0.35	0.02	2.8	−0.89, 6.4	0.14	2.2	−1.4, 5.9	0.23
Any maternal recreational drug use in this pregnancy (marijuana)	1.4	−1.4, 4.1	0.33	0.24	−3.6, 4.1	0.90	7.5	1.8, 13	0.01	3.9	−1.8, 9.6	0.18

Continued

Table 2 Continued

	Cognitive Development[a]			Expressive Language[a]			Externalising[b]			Internalising[b]		
	Mean Difference	95% CI	P value	Mean Difference	95% CI	P value	Mean Difference	95% CI	P value	Mean Difference	95% CI	P value
Birth outcomes												
Prematurity[d]:												
Term	Reference			Reference			Reference			Reference		
Late preterm	−0.39	−2.0, 1.2	0.63	0.82	−1.7, 3.4	0.53	−0.18	−4.1, 3.7	0.93	1.2	−2.8, 5.1	0.56
Moderate preterm	−2.9	−6.5, 0.77	0.12	−7.1	−14, −0.51	0.03	1.5	−7.6, 11	0.75	5.4	−3.6, 14	0.24
Mode of delivery:												
Unassisted vaginal	Reference			Reference			Reference			Reference		
Assisted vaginal	−0.20	−0.94, 0.53	0.59	0.029	−1.0, 1.1	0.96	0.12	−1.7, 1.9	0.90	1.6	−0.22, 3.4	0.09
Unscheduled caesarean	0.31	−0.53, 1.1	0.47	0.87	−0.33, 2.1	0.15	−0.26	−2.4, 1.8	0.81	1.6	−0.52, 3.6	0.14
Elective caesarean	−0.17	−0.99, 0.65	0.68	−0.22	−1.3, 0.91	0.70	−1.4	−3.4, 0.61	0.17	−2.2	−4.1, −0.20	0.03
Apgar score at 5 min	−0.011	−0.38, 0.36	0.95	0.25	−0.31, 0.81	0.38	−1.2	−2.1, −0.42	0.003	−0.23	−1.0, 0.59	0.59
Postnatal												
Breastmilk feeding (duration):												
Nil or <1 week	-1.0	−2.7, 0.60	0.21	−3.4	−5.6, −1.2	0.003	2.6	−1.0, 6.3	0.16	1.1	−2.5, 4.7	0.56
>=1 week but <6 months	−0.85	−1.5, −0.25	0.006	−1.3	−2.1, −0.41	0.004	0.025	−1.5, 1.6	0.98	−0.9	−2.4, 0.64	0.25
>=6 months	Reference			Reference			Reference			Reference		
Maternal stress & mental health												
Edinburgh Depression Scale (pregnancy):												
Low Risk (<10)	Reference			Reference			Reference			Reference		
Moderate Risk	−0.52	−1.5, 0.49	0.31	0.029	−1.5, 1.5	0.97	4.1	1.3, 7.0	0.005	5.8	3.1, 8.6	<0.001
High Risk (>12)	−0.34	−1.8, 1.1	0.64	−0.93	−2.8, 1.0	0.34	0.66	−3.4, 4.7	0.75	4.5	0.56, 8.4	0.03
Mean Perceived Stress Scale score (pregnancy, 4 weeks, 6 months)	−0.013	−0.059, 0.033	0.57	−0.02	−0.084, 0.043	0.53	0.35	0.24, 0.47	<0.001	0.42	0.31, 0.53	<0.001
Parenting practices												
HOME-SF Emotional Support subscale (9 months, n=275–334)	0.024	−0.42, 0.47	0.92	0.3	−0.36, 0.97	0.37	0.54	−0.65, 1.7	0.37	0.59	−0.52, 1.7	0.30
HOME-SF Cognitive Stimulation subscale (9 months, n=275–334)	0.18	−0.12, 0.48	0.23	0.33	−0.090, 0.76	0.13	0.12	−0.68, 0.92	0.77	−0.18	−0.93, 0.56	0.63

*Estimated change in raw score on the BAYLEY-III adjusting for sex, postconceptional age, researcher, researcher experience (number of prior assessments within study) and excluding twins, those with known acquired neurodevelopmental disorders, and those outside of age criteria of 2–3 years.
†Estimated change in T-score for problem behaviours reported on the Child Behaviour Checklist for Ages 1½–5 adjusting for sex and chronological age and excluding twins, known acquired neurodevelopmental disorders.
‡Highest level of education attained (six categories: <10 years of school, 10–11 years of school, completed year 12, trade certificate/apprenticeship or diploma, undergraduate degree, postgraduate degree).
§Note that the associations reported here between prematurity and BAYLEY-III measures (cognitive development and expressive language) are after adjustment for postconceptional age.
BAYLEY-III, Bayley Scales of Infant and Toddler Development, Third Edition; HOME-SF, Home Observation for Measurement of the Environment-Short Form.

Family size indicators and child cognitive and language development

Family size indicators were investigated further, taking into account the age of siblings (table 3). Different patterns of association were observed for cognitive performance vs expressive language. Greater number of siblings was associated with poorer cognitive scores across multiple age-specific categories with a comparable magnitude of effect (table 3). However, these age-specific sibling effects were not independent of total household size (table 3). Thus, total household size rather than age-specific sibling effects appeared more important for cognitive development, with an estimated 0.66 point decrease in the mean raw score for cognitive development for each additional household member regardless of age (95% CI −0.96 to −0.37; p<0.001). Dose response was evident (figure 1). A similar pattern of results was observed for receptive language scores (online supplemental table 3).

By contrast, an association between a greater number of siblings and poorer expressive language scores was only observed among siblings aged 2–10 years older and these associations tended to persist after adjustment for household size, particularly for siblings in the 6–10 year category (table 3). The highest magnitude of association was observed for siblings 6–10 years older with an estimated 2.8 point decrease in the mean raw score for expressive language with each additional sibling (95% CI −4.1 to −1.5; p<0.001; q<0.001; table 3). There was also an estimated 1.2 point decrease in the mean raw score for expressive language with each additional sibling in the 2–6 years category (95% CI −1.8 to −0.67; p<0.001; q<0.001; table 3). Thus, the association between greater number of siblings and better expressive language development was dependent on the age of the older siblings.

Family size indicators and emotional and behavioural development

Age-specific effects were also observed for emotional behaviour. Each additional sibling aged 2–6 years older was associated with an estimated 2.1 point decrease in mean Internalising T-score (95% CI -3.1 to -1.0; p<0.001; q<0.001), and this persisted after adjusting for total household size (table 3). Having older siblings less than 2 years older was associated with an estimated 2.6 point increase in mean Externalising T-score per sibling although evidence for this was attenuated after accounting for the false discovery rate (95% CI 0.52 to 4.7; p=0.01; q=0.07; table 3). Again, this persisted after further adjustment for total household size (table 3). Thus, associations between siblings and emotional behaviour were dependent on the age of the older siblings. More siblings between 2 and 6 years older was protective against internalising behaviour while having siblings less than 2 years older appeared adverse for externalising behaviour.

Additional adjustments, non-linear modelling and sensitivity analyses

The main findings reported above persisted after adjustment for additional potential confounders (online supplemental table 4) and we did not find evidence for non-linear patterns of association using fractional polynomial modelling. The magnitudes of these associations were also not materially changed after accounting for initial non-participation and attrition in analyses using inverse probability weighting. For example, per additional sibling aged 2-6 years older, the estimated mean decrease in expressive language score and in internalising behaviour score changed from 1.2 (95% CI 0.67 to 1.8) to 1.2 (95% CI 0.50 to 1.9) and from 2.1 (95% CI 1.0 to 3.1) to 2.1 (95% CI 0.97 to 3.2), respectively. Similar changes were observed for the other main findings. The associations reported in table 3 between age-specific sibling numbers and neurodevelopmental outcome were additionally evaluated with mutual adjustment for all other age-specific categories, with no material change in estimates (data not presented).

Proportion of variance in cognition, language, internalising and externalising explained

For cognitive development, the fully adjusted model in table 3 explained 21.2% of the variance in scores. Larger household size alone explained 3.2%. For expressive language, 23.6% of variance was explained by the full model, with 4.2% of variance accounted for by number of siblings aged 2–6 and 6–10 years older. For internalising behaviour, the corresponding proportions were 18.5% and 2.5% (considering number of siblings aged 2–6 years older) and, for externalising behaviour, 15.7% and 0.7% (siblings aged 0–2 years older). Thus, the overall estimated contribution of the early life factors measured was substantial, and larger total household size (cognitive development) and age-specific sibling influences (expressive language, emotional behaviour) were important components within these early life factors.

DISCUSSION

Greater family size was associated with poorer child cognitive and receptive language performance at age 2 years, consistent with past research. However, contrasting patterns were observed for the other three neurodevelopmental outcomes. For expressive language, greater sibling number was associated with lower scores, but only for siblings 2–10 years older. Different patterns were observed when comparing the two emotional behaviour outcomes. For internalising behaviour, having siblings between 2 and 6 years older was associated with a better outcome. However, exposure to a sibling under 2 years older appeared to be associated with more externalising behaviour, although our findings with externalising behaviour should be interpreted with caution as evidence was weak after accounting for the false discovery rate due to the multiple comparisons tested.

Table 3 Family size indicators and child neurodevelopmental outcomes at 2–3 years of age

		Fully adjusted*			Additional adjustment for total household size		
		β	95% CI	P value	β	95% CI	P value†
Cognitive development							
By total household size:	**Per household member‡**	**−0.66**	**−0.96 to −0.37**	**<0.001**			
By age of household members:	Per sibling aged under 2 years	0.39	−0.46 to 1.2	0.37	0.74	−0.11 to 1.6	0.09
	Per sibling aged 2 to under 6 years	−0.76	−1.2 to −0.36	<0.001	−0.29	−0.84 to 0.25	0.28
	Per sibling aged 6 to under 10 years	−1.5	−2.4 to −0.53	0.002	−0.78	−1.8 to 0.25	0.14
	Per sibling age 10 to under 18 years	−0.20	−0.95 to 0.55	0.60	0.55	−0.26 to 1.4	0.18
	Per non-parental adult (18+ years)§	−0.64	−1.3 to 0.040	0.07	−0.057	−0.79 to 0.68	0.88
Expressive language							
By total household size:	Per household member‡	−0.98	−1.4 to −0.57	<0.001			
By age of household members:	Per sibling aged under 2 years	0.19	−0.96 to 1.3	0.74	0.69	−0.46 to 1.8	0.24
	Per sibling aged 2 to under 6 years	**−1.2**	**−1.8 to −0.67**	**<0.001**	**−0.64**	**−1.4 to 0.097**	**0.09**
	Per sibling aged 6 to under 10 years	**−2.8**	**−4.1 to −1.5**	**<0.001**	**−1.9**	**−3.3 to −0.52**	**0.007**
	Per sibling age 10 to under 18 years	−0.036	−1.1 to 1.0	0.95	1.1	−0.022 to 2.3	0.05
	Per non-parental adult (18+ years)§	−0.23	−1.2 to 0.70	0.63	0.76	−0.24 to 1.7	0.14
Internalising							
By total household size:	Per household member‡	−1.3	−2.1 to −0.56	<0.001			
By age of household members:	Per sibling aged under 2 years	−0.49	−2.6 to 1.6	0.64	0.11	−2.0 to 2.2	0.92
	Per sibling aged 2 to under 6 years	**−2.1**	**−3.1 to −1.0**	**<0.001**	**−1.5**	**−2.9 to −0.12**	**0.03**
	Per sibling aged 6 to under 10 years	0.25	−2.2 to 2.7	0.84	2.5	−0.15 to 5.2	0.06
	Per sibling age 10 to under 18 years	−0.50	−2.6 to 1.6	0.64	1.0	−1.2 to 3.3	0.37
	Per non-parental adult (18+ years)§	−0.57	−2.4 to 1.2	0.53	0.73	−1.2 to 2.7	0.46
Externalising							
By total household size:	Per household member‡	−0.0086	−0.81 to 0.79	0.98			
By age of household members:	**Per sibling aged under 2 years**	**2.6**	**0.52 to 4.7**	**0.01**	**2.7**	**0.59 to 4.9**	**0.01**
	Per sibling aged 2 to under 6 years	−0.45	−1.5 to 0.63	0.42	−0.84	−2.3 to 0.59	0.25
	Per sibling aged 6 to under 10 years	0.82	−1.6 to 3.3	0.51	1.0	−1.7 to 3.8	0.45

Continued

Table 3 Continued

		Fully adjusted*			Additional adjustment for total household size		
		β	95% CI	P value	β	95% CI	P value†
	Per sibling age 10 to under 18 years	0.016	−2.1 to 2.2	0.99	0.029	−2.3 to 2.4	0.98
	Per non-parental adult (18+ years)§	−1.1	−2.9 to 0.72	0.24	−1.3	−3.3 to 0.69	0.20

Bolded text (and corresponding estimates) highlights the household compositional factors most relevant to each neurodevelopmental domain.
*Cognitive Development and Expressive Language: Estimated change in raw score on the Bayley Scales of Infant and Toddler Development, Third Edition, adjusting for postconceptional age, sex, administering researcher, experience of researcher, mean parental education category, mother born in Australia, lone parent status, preterm delivery <34 weeks, breastfeeding duration category to 6 months. Internalising Behaviour and Externalising Behaviour: Estimated change in T-score for problem behaviours reported on the Child Behaviour Checklist for Ages 1½–5 adjusting for sex, chronological age at assessment, mean household income, maternal country of birth category, maternal age category, lone parent status, maternal smoking in pregnancy, maternal marijuana use in pregnancy, mode of birth, birth condition (Apgar score at 5 min), breast feeding duration category to 6 months, and mean maternal perceived stress from pregnancy to 6 months.
†Test of trend for each age category independent of total number of additional children and adults in household per household member beyond the index child and first two adults.
‡Per household member beyond the index child and first two adults.
§Defined as the number of adult (18+ years) household members beyond the first 2.

The patterns of association between household composition and cognitive development replicate substantial research that has observed poorer outcomes for children related to larger sibship size, later birth order or larger household size,[2–7] and extend this by demonstrating a similar effect regardless of the age of the sibling(s) or for additional adult household members. This is consistent with predictions from the Resource Dilution model and additionally provides an insight that parental resources do not appear to be disproportionately taxed by children under a particular age but by the overall number of household members.

In contrast, expressive language appeared to be specifically impacted by sharing a household with siblings aged 2–10 years. This finding would not be expected from the Resource Dilution model. Our findings were more consistent with the Confluence model,[4] an alternative model that had been proposed to explain association between family size indicators and educational attainment in the 1970s. The Confluence model considers the contribution of all household members—including siblings—to the overall developmental experiences of a child; with a central assumption that the developmental contribution of sibling interactions depends on the developmental maturity of those siblings. In the context of language development, the Confluence model predicts that language development is driven by the overall developmental maturity of the language exposure within a household, and that interactions with older siblings in the household can compensate for reduced interactions with

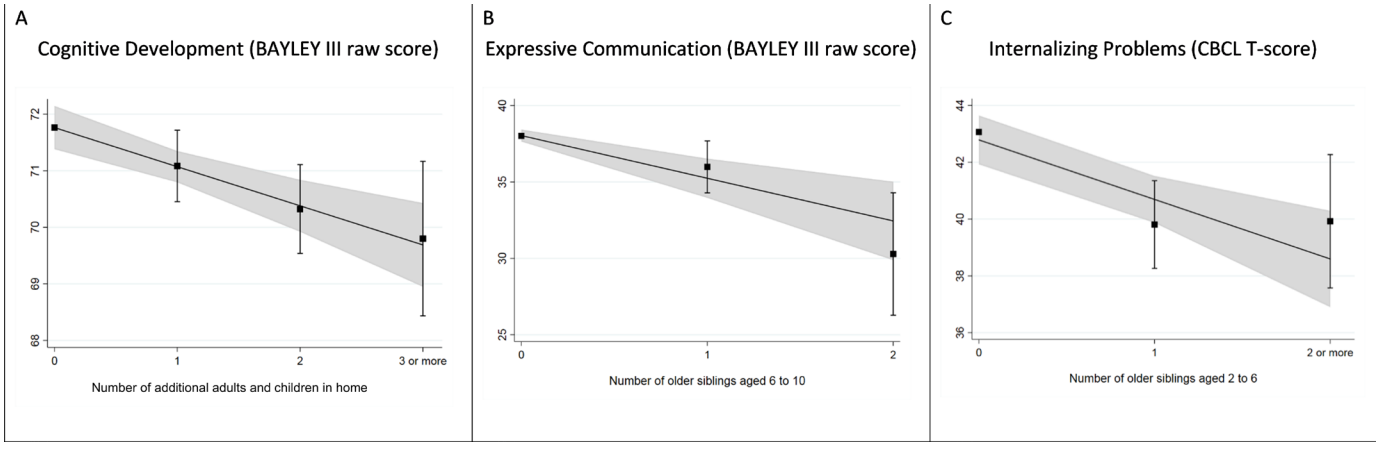

Figure 1 Dose response in selected association between family size indicators and child neurodevelopmental outcomes at 2–3 years of age NB. Estimated mean (square) and 95% CI (tails) for each estimate and predictive margins (line) with 95% CI (shaded area) for linear trend with outcome (in units). We additionally modelled each of the above associations in fractional polynomial models and did not find evidence of non-linear patterns of association within the distribution of family size indicators above. BAYLEY-III, Bayley scales of infant and toddler development third edition; CBCL, Child Behaviour Checklist for ages 1½–5.

adults, but that this depends on the age of the sibling. Consistent with this, prior studies have found that the presence of siblings is associated with decreased and altered child-directed speech from parents,[29] and that siblings potentially make a substantial contribution to the spoken environment of the index child. Indeed, by 1–2 years of age, children spend similar time with older siblings as either parent[30] and recent work demonstrates that the quality of interactions with older siblings is associated with child language development with similar dimensions of intersibling interaction observed to be as important as those established for parent–child interaction.[31] Significantly, the age of that older sibling appears to be a major determinant of the quality of such intersibling interactions.[32 33] In this context, our overall findings indicate that beyond 10 years of age, the net contribution of older siblings to overall quality of language exposure may compensate for reduced child-directed speech from parents.

Considering emotional and behavioural outcomes, less internalising emotional behaviour was evident among children with older siblings which is highly consistent with prior research.[9–14] Importantly, we extended previous work by identifying that this positive effect seems to be restricted to siblings with an inter-birth interval of 2–6 years. The underlying mechanisms by which siblings are protective for childhood internalising, and by which siblings with narrower inter-birth interval may increase externalising, are not understood and these age-specific findings indicate intersibling interaction needs to take account of the developmental stage of each sibling. Previous studies examining the relationship between sibship and externalising behaviour are conflicting[9–13] and our findings give a possible explanation for this in that evaluation of total sibling number may overlook age-specific effects. Our age-specific findings for both internalising and externalising indicate that further investigation of differences in early intersibling interaction by age of the older sibling may help identify positive and negative aspects of intersibling interaction. Existing qualitative studies of sibling interaction have described more positive responses to older sibling teaching with greater intersibling interval but more negative feedback and controlling behaviours for smaller intersibling intervals (less than 2 years),[33] and these latter qualities may have contributed to the observation here that only older siblings close in age were associated with externalising by 2 years.

The major strength of this study is that it is the first birth cohort to examine both age-specific sibling effects on early childhood neurodevelopment and household composition contemporaneously across diverse neurodevelopmental outcomes at age 2 years. The evaluation of multiple neurodevelopmental outcomes in parallel with the same exposure methodology and within the same cohort provided an opportunity for the better discrimination of differential patterns of association across those outcomes. A comprehensive consideration of other determinants for each outcome was made, allowing for robust confounding adjustment. This was demonstrated by the high variance proportion explained for various aspects of child development. For example, the early life factors we examined, including larger household size, explained 21% of the variation in cognition at age 2 years.

There are, however, some limitations of this study. Large sibships were uncommon in our sample which limits the evaluation of dose–response patterns and the generalisation of our results to very large sibships and households. Also, given the young age of the index children, the analysis here focused on older siblings; the effect of younger siblings will be examined as this cohort matures. While direct measures of parental cognitive performance and language were not available to us, we did adjust for parental education as a proxy measure for each. Information on the quality of intersibling interactions was not available so we were, therefore, unable to additionally investigate these.[32 33] As with any longitudinal cohort study there is risk of bias due to initial selection bias, attrition or missing data. The sample of children in our study was population derived, and we applied propensity weighting in an additional sensitivity analysis of all main findings to evaluate any bias due to initial recruitment or attrition, which gave reassurance against any material bias associated with this. Missing data were minimal (<5%) for all main findings presented. Finally, this study did not have enough power to examine whether the reported associations changed depending on the sex of the index child and that of the sibling(s). Future studies should consider sex-specific effects.

There are a number of key implications. First, our findings provide ongoing evidence of poorer child cognitive and language development in larger families, and indicate a need for further enhancing supports for larger families. Importantly, our findings also extend prior research by finding differential sibling patterns with cognitive and language development. Expressive language development was only adversely influenced by siblings between 2 and 10 years older, indicating a differing underlying mechanism. This should lead to better awareness of risk and targeting of language screening and supports. Finally, the age-specific findings emphasise the importance of developmental contributions of sibling interaction and taking the age of each sibling into account in both language and child emotional outcomes. This has been neglected in much previous epidemiological research on early child neurodevelopment.

CONCLUSIONS

This study confirms family size indicators as important factors associated with early childhood neurodevelopment across multiple domains, and brings out important differences by considering these outcomes in parallel. Our findings provide further evidence for association between household size and poorer child cognitive and language development, with implications for further targeted supports. Our findings also support beneficial aspects of having siblings, such as protection against symptoms of anxiety and depression. For the first time, the age-specific nature of these associations has been

described. We recommend further research to identify the specific underlying factors that contribute to better outcomes when exposed to siblings of specific ages and to promote these healthy sibling interactions across the first 2 years of life.

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