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Risk factors for hospital admission and length of stay for children with and without congenital anomalies: a EUROlinkCAT cohort study

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ABSTRACT

Objective To evaluate risk factors for hospital admission and length of stay (LOS) among children with and without congenital anomalies (CAs).

Design A population-based linkage cohort study including 50 353 children with major CAs and 1 259 925 children without CAs from four EUROCAT registry areas in three countries. Data on children born 1995–2014 were linked to hospital discharge databases 1995–2015. HRs and incidence rate ratios estimated risk of admission and LOS for children aged <1 and 1–4 years by birth cohort, gestational age, sex, maternal age, multiple births and maternal education. Estimates were pooled using random effects meta-analysis.

Results In children <1 year, twins/triplets with CAs were 34% more likely to be admitted and had over two times the LOS compared with singletons, while twins/triplets without CAs were over two and a half times as likely to be admitted and had six times longer stays. Despite this, a higher proportion of twins/triplets with CAs were admitted compared to those without CAs (91% vs 65%) and had longer LOS (20 days vs 10). Smaller increases in risk of admission or LOS were found in boys, young mothers and low maternal education. Preterm birth was a major risk factor for admission and LOS.

Conclusions While the impact of risk factors on hospital admission and LOS was generally greater in children without CAs, a higher proportion of children with CAs were admitted and had longer stays. These findings have implications for health care planning and for counselling parents regarding their child's future healthcare needs.

WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ Preterm birth increases the risk of admission and length of stay (LOS) in children with congenital anomalies (CAs). There is conflicting evidence for the effect of sex, limited information on maternal age and education and no evidence for multiple births in children with CAs.

WHAT THIS STUDY ADDS

⇒ Risk of admission and LOS were greatest for children aged <1 year who were born preterm and for twins/triplets. It was also increased in boys, children of young mothers (<20 years) and lower maternal education. While risks were higher for children without CAs, the proportions admitted and median LOS were higher for children with CAs.

HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ This information can help inform parents after a prenatal diagnosis or birth of a child with a CA, guiding them on what to expect regarding their child's future healthcare needs.

INTRODUCTION

Congenital anomalies (CAs) affected 2% of live births in Europe between 2013 and 2020.[1] While the survival of children with CAs has improved,[2–4] CAs remain a significant cause of childhood morbidity and long-term disability.[5] Children with CAs represented 9%–12% of non-birth hospital admissions in childhood (<18 years) in Australia (1980–1999),[6] California, South Carolina (1991)[7] and Texas (2001–2010).[8] In addition, children with CAs have longer stays in hospital than children without CAs.[6 9–11]

In Europe, the EUROlinkCAT project investigated the survival, morbidity and educational outcomes of children with CAs by linking CA data recorded in EUROCAT registries to regional or national mortality, hospital discharge, prescription and educational databases.[12] For children with CAs <1 year of age, 85% (95% CI 79% to 90%) were hospitalised compared with 31% (95% CI 26% to 37%) of children without CAs. This decreased to 56% (95% CI 51% to 61%) and 25% (95% CI 19% to 31%), respectively, at ages 1–4 years. The median length of stay (LOS) was 2–3 times longer for children with CAs in both age groups.[13]

As in the general population, preterm birth has been associated with an increased risk of admission[14 15] in those with CAs. Boys with CAs, such as Down syndrome (DS), were more likely to be admitted[6 14 15] and stay longer[16] than girls, but girls were more likely to be admitted if they had nervous system or musculoskeletal anomalies.[17] There is limited evidence for the effect of maternal age at birth on the risk of admission[14 18] and maternal education on LOS.[19] Reductions in hospital admissions and LOS over time for children with CAs have been reported in Australia[6] and in the United Kingdom.[17 20] The effect of multiple births on risk of admission, or LOS, has not been examined among children with CAs. Given the significant morbidity associated with CAs, it is essential to understand the risk factors for hospital admission and LOS in these children to inform health service planning, preventative care and to counsel families. This EUROlinkCAT study aimed to explore risk factors for hospital admission and LOS in hospital, among children with and without CAs in Europe.

METHODS

Cohort selection

A European, population-based linkage cohort study was conducted, including data on children with major CAs born 1995–2014 from four EUROCAT registries in three countries (Denmark, Finland and Italy).[12 13] Cases with minor CAs only were excluded. Children without CAs born during the same period and from the same population area were identified from birth registers and included as a reference population.[21] Three registries included the entire population of children without CAs, except Tuscany, which had a 10% random sample matched by year of birth and sex. The first birth year included in the study was Funen, Denmark (1995), Finland (1997), Tuscany, Italy (2005) and Emilia Romagna, Italy (2008).

Outcomes

Data on admission to hospital and LOS for children 0–4 years of age were obtained through linkage to hospital databases. Transfers of care, within or between hospitals, were combined into a single admission record if there was 1 day or less between a discharge and the next admission.[13] Obstetric stays, that is, the initial newborn stay following the birth, were identified as admissions on the date of birth (age=0) or the day after (age=1 day) and were excluded. Full details of the methodology used to exclude obstetric stays are described in previous publications.[13 21] Hospital admission data in each registry were standardised to a common data model, and each registry ran a centrally written analysis script.[21]

Risk factors

Risk factor information was obtained from maternity and birth records provided by national statistics or regional health authorities. The risk factors explored were birth cohort (1995–2004, 2005–2009 and 2010–2014); sex (male and female); multiple births (singleton and twins/triplets); gestational age (GA) at birth (<32, 32–36 and 37+ weeks); maternal age (<20, 20–34 and 35+ years) and maternal education based on the UNESCO International Standard Classification of Education[22] (Primary/preprimary/no education (low), any secondary (middle), tertiary/postsecondary (high)). Maternal education was used as a proxy for socioeconomic status (SES) as there were no area level measures of SES available in this study. Finnish maternal education data were excluded due to an error incorrectly classifying certain codes as missing. All comparisons are between the risk factor categories in children with CAs and separately in children without CAs; there is no direct comparison between children with and without CAs.

Statistical analysis

For each risk factor, the number and proportions of children with and without CAs admitted aged <1 year (<365 days) and 1–4 years (365–1825 days) were calculated using Kaplan-Meier survival analysis. This allowed for the censoring of children at age 5 years or occurring on 31 December 2015 (whichever came first), death, emigration or adoption. For each year, the child was in the study, the total number of hospital days for all admissions within that year was calculated; the median LOS in days per year and the corresponding lower and upper quartiles of LOS were then calculated for each age group among children with at least one hospital admission of ≥0.5 days for each age group. The total number of days in hospital in each of the 4 years was calculated and expressed as a rate per year in the study (not all children had the full 4 years, so we looked at years started). The average yearly rate for each child (ie, mean of years 1–4) was then calculated, followed by median rates for all children who were in hospital at any time over the 4 years. See Urhoj *et al*[13] for more information.

Cox proportional hazards models estimated HRs to quantify the associations of risk factors on the 'risk' of hospital admission. For the <1 year age group, time 0 for these models was birth; for the 1–4 year age group, it was day 365. Negative binomial regression models quantified the associations of risk factors on the LOS in hospital by estimating incidence rate ratios (IRRs), with the denominator being the length of time the child was in the study and the numerator being the number of days the child was in hospital. Univariable and multivariable models were fitted with the multivariable models, including birth cohort, sex, GA and maternal age. Maternal education and multiple births were not included in the multivariable models due to small numbers.

Random effects meta-analysis was used to pool the estimates of the proportions of children ever admitted to hospital, the HRs for hospital admission and the IRRs for LOS across each risk factor in children with and without CAs. The absolute excess risk of admission was estimated by subtracting the proportion admitted in the reference category from the proportion admitted in the category

of interest, giving the additional proportion admitted. Quantile estimation methods were used to obtain pooled estimates of the median LOS.[23] These methods use the median and quartiles for each registry to select an underlying parametric distribution based on the best fit of normal, log-normal, gamma and Weibull distributions. This allows the asymptotic variance of the median to be calculated and a random effects meta-analysis to be performed[24 25] using the 'metamedian' package in R. We provide the adjusted HRs and IRRs where available. Adjusting for multiple risk factors at the same time did not significantly change the estimates (online supplemental eTable 1).

Missing data

The high level of completeness of risk factor variables in the linked data is shown in online supplemental etable 2. Overall, up to 1% of data were missing for the risk factors, apart from maternal education, which was missing for 4% of children with CAs and 1% of children without CAs. By registry, the proportion of missing data was low, except for children with CAs in Tuscany, who had 6%–10% missing data, as the data were obtained from the Births registry rather than the anomaly registry. Due to low missing data levels, multiple imputation was not incorporated into the central analysis script.

All analyses used Stata V.16.0 (StataCorp LP, College Station, Texas) or R Statistical Software (V.4.1.0; R Core Team 2021).

Ethics approval

RESULTS

A total of 50 353 children with CAs and 1 259 925 children without CAs were included and followed up for 213 544.1 (mean 4.2) and 5 572 424.3 (mean 4.4) years, respectively. Table 1 shows the number of children with and without anomalies included in each registry and birth cohort. Most models had considerable heterogeneity, reflecting differences in admission and LOS across registries (online supplemental eTables 3 and 4). In total, 85.1% (95% CI 64.1 to 94.3) of children with CAs <1 year of age were admitted while 54.4% (95% CI 48.8 to 59.6) were admitted at 1–4 years of age. For children without CAs <1 year of age, 31.3% (95% CI 21.9 to 41.2) were admitted while 22.5% (95% CI 16.2 to 29.4) were admitted at 1–4 years of age. Children with CAs <1 year of age stayed in hospital a median of 7.9 days (95% CI 6.6 to 9.3) days, which dropped to 0.9 days (95% CI 0.7 to 1.1) at 1–4 years. Children without CAs <1 year of age stayed a median of 3.2 days (95% CI 2.8 to 3.7), which decreased to 0.4 days (0.2–0.6) at 1–4 years.

Preterm birth (<32 weeks) had the greatest impact on risk of admission and LOS for children with and without CAs in both age groups (table 2 and figures 1 and 2). Compared with term births, children under 1 with CAs born <32 weeks were over two times as likely to be admitted and had almost nine times longer stays, while those without CAs were almost seven times more likely to be admitted and had over 50 times longer stays. Similar associations of lower magnitude were seen for those born at 32–36 weeks GA and for children aged 1–4 years (figures 3 and 4).

Multiple births increased risk of admission and LOS in children <1 year, particularly in children without CAs. Twins/triplets with CAs were 34% more likely to be admitted and had more than two times the LOS than singletons, whereas twins/triplets without CAs were more than two and a half times as likely to be admitted and had more than six times longer stays (figures 1 and 2). Multiple births were associated with an additional 5.8% of children with CAs being admitted compared with an extra 35.0% of children without CAs (table 2). For children with and without CAs aged 1–4 years, multiple births were associated with a small increase in the risk of admission (figure 3) and two times the LOS (figure 4).

Table 1 Number of children with and without congenital anomalies for each registry by birth cohort

			Birth cohort			
			1995–2004	2005–2009	2010–2014	Total
Registry	Denmark, Funen (1995–2014)	Children with anomalies	1233	685	505	2423
		Children without anomalies	53 570	25 107	22 071	100 748
	Finland (1997–2014)	Children with anomalies	13 807	11 760	12 757	38 324
		Children without anomalies	395 140	257 184	259 355	911 679
	Italy, Emilia Romagna (2008–2014)	Children with anomalies	–	1179	4202	5381
		Children without anomalies	–	66 271	157 724	223 995
	Italy, Tuscany (2005–2014)	Children with anomalies	–	2048	2177	4225
		Children without anomalies	–	11 435	12 068	23 503
Total		Children with anomalies	15 040	15 672	19 641	50 353
		Children without anomalies	448 710	359 997	451 218	1 259 925

Table 2 Percent of children admitted, absolute excess risk (AER) and median LOS for children with and without CAs <1 and 1–4 years of age

Risk factor		Children with congenital anomalies			Children without congenital anomalies		
		Percent admitted*	Absolute excess risk (%)	Median LOS per year (95% CI)	Percent admitted*	Absolute excess risk (%)	Median LOS per year (95% CI)
<1 year of age							
Birth cohort†	1995–2004	70.8 (61.7–78.1)	5.3	9.9 (7.9 to 11.8)	22.9 (16.3–30.2)	–2.7	3.5 (2.5 to 4.5)
	2005–2009	81.4 (59.1–92.3)‡	Reference	7.2 (6.3 to 8.2)§	30.1 (23.6–36.8)¶	Reference	3.0 (2.2 to 3.8)**
	2010–2014	86.4 (54.8–96.5)	5.0	7.2 (6.2 to 8.2)	33.3 (22.7–44.2)	3.2	2.7 (1.8 to 3.7)
Sex	Male	86.2 (67.1–94.6)	Reference	7.4 (6.1 to 8.8)	33.3 (24.2–42.6)	Reference	3.2 (2.8 to 3.7)
	Female	84.2 (61.5–94.1)	–2.0	8.0 (6.5 to 9.4)	29.2 (19.5–39.6)	–4.1	3.2 (2.8 to 3.7)
Multiple birth	Singleton	85.1 (63.8–94.3)	Reference	7.3 (5.9 to 8.7)	30.3 (21.1–40.0)	Reference	3.1 (2.5 to 3.7)
	Multiple births	90.9 (80.1–96.0)	5.8	20.0 (17.9 to 22.1)	65.3 (51.0–76.4)	35.0	9.7 (7.3 to 12.2)
Maternal age (years)	<20	88.2 (68.5–95.9)	2.9	8.0 (6.8 to 9.1)	36.1 (25.3–47.0)	4.9	3.5 (2.9 to 4.1)
	20–34	85.3 (65.8–94.1)	Reference	7.6 (6.0 to 9.2)	31.2 (22.1–40.7)	Reference	3.2 (2.8 to 3.7)
	35+	85.4 (62.0–94.9)	0.1	7.9 (7.2 to 8.6)	31.4 (21.3–41.9)	0.2	3.3 (2.8 to 3.7)
Gestational age (weeks)	<32	99.3 (96.4–99.8)	16.0	61.7 (58.7 to 64.7)	94.8 (93.2–96.0)	66.7	45.2 (38.1 to 52.3)
	32–36	96.6 (89.4–98.9)	13.3	20.9 (17.5 to 24.2)	79.3 (66.9–87.5)	51.2	10.0 (6.7 to 13.3)
	37+	83.3 (60.4–93.6)	Reference	6.1 (5.1 to 7.0)	28.1 (19.5–37.4)	Reference	2.5 (1.9 to 3.1)
Maternal education	Low	89.1 (68.7–96.5)	0.0	9.2 (7.2 to 11.2)	41.6 (33.8–49.3)	8.3	3.6 (2.8 to 4.3)
	Middle	90.0 (75.1–96.2)	0.9	8.3 (6.6 to 9.9)	35.4 (29.3–41.5)	2.1	3.3 (2.7 to 4.0)
	High	89.1 (64.4–97.0)	Reference	7.2 (5.6 to 8.8)	33.3 (25.0–41.8)	Reference	3.3 (2.7 to 4.0)
1–4 years of age							
Birth cohort†	1995–2004	62.8 (62.0–63.6)	3.2	1.1 (0.4 to 1.7)	28.9 (25.1–32.9)	0	0.3 (0.2 to 0.3)
	2005–2009	54.2 (49.4-58.8)††	Reference	0.8 (0.6-1.1)‡‡	23.6 (17.5–30.3)§§	Reference	0.4 (0.2-0.5)¶¶
	2010–2014	49.9 (46.3–53.4)	–4.3	0.9 (0.6 to 1.3)	19.6 (14.9–24.8)	–4.0	0.5 (0.2 to 0.7)
Sex	Male	58.8 (53.8–63.4)	Reference	1.0 (0.7 to 1.3)	25.1 (18.0–32.8)	Reference	0.4 (0.2 to 0.6)
	Female	48.2 (41.1–54.9)	–10.6	0.9 (0.5 to 1.2)	19.7 (14.2–25.8)	–5.4	0.4 (0.2 to 0.7)
Multiple birth	Singleton	54.3 (48.9–59.3)	Reference	0.9 (0.7 to 1.1)	22.4 (16.2–29.3)	Reference	0.4 (0.2 to 0.6)
	Multiple births	56.0 (45.2–65.5)	1.7	0.8 (0.5 to 1.2)	24.1 (16.6–32.4)	1.7	0.4 (0.2 to 0.7)
Maternal age (years)	<20	53.6 (41.0–64.6)	–0.4	1.0 (0.8 to 1.2)	25.8 (18.6–33.7)	2.8	0.5 (0.2 to 0.7)
	20–34	54.0 (48.0–59.7)	Reference	0.9 (0.7 to 1.2)	23.0 (16.9–29.6)	Reference	0.4 (0.2 to 0.6)
	35+	54.4 (49.9–58.7)	0.4	0.9 (0.8 to 1.1)	21.0 (14.8–27.9)	–2.0	0.4 (0.2 to 0.6)

Continued

Table 2 Continued

Risk factor		Children with congenital anomalies			Children without congenital anomalies		
		Percent admitted*	Absolute excess risk (%)	Median LOS per year (95% CI)	Percent admitted*	Absolute excess risk (%)	Median LOS per year (95% CI)
Gestational age (weeks)	<32	70.1 (64.0–75.3)	17.2	1.5 (1.1 to 1.8)	38.2 (21.4–54.9)	16.0	0.6 (0.4 to 0.8)
	32–36	62.1 (53.3–69.8)	9.2	1.1 (0.8 to 1.3)	26.2 (17.4–35.8)	4.0	0.5 (0.3 to 0.8)
	37+	52.9 (47.6–57.9)	Reference	0.9 (0.6 to 1.1)	22.2 (16.1–29.0)	Reference	0.4 (0.2 to 0.6)
Maternal education	Low	51.8 (45.8–57.6)	−0.9	1.4 (0.6 to 2.2)	21.3 (12.6–31.6)	2.3	0.5 (0.3 to 0.8)
	Middle	54.7 (44.4–64.0)	2.0	1.1 (1.0 to 1.3)	21.4 (13.9–30.0)	2.4	0.5 (0.2 to 0.7)
	High	52.7 (42.2–62.2)	Reference	1.0 (0.9 to 1.0)	19.0 (12.5–26.5)	Reference	0.5 (0.2 to 0.7)

*Kaplan-Meier estimate of children ever admitted to hospital in age period from meta-analysis of all registries.
†Percent admitted 1995–2004 is based on Finland and Funen registries and cannot be compared directly to the percentage admitted in 2005–2009, which is based on all registries. The AER for 1995–2004 compared with 2005–2009 is calculated for Funen and Finland only.
‡65.5% (95% CI 76.9 to 50.6) for Funen and Finland only.
§7.9 (95% CI 4.9 to 10.8) for Funen and Finland only.
¶25.6% (95% CI 32.2 to 19.4) for Funen and Finland only.
**2.5 (95% CI 1.5 to 3.5) for Funen and Finland only.
††59.6% (95% CI 71.8 to 44.6) for Funen and Finland only.
‡‡0.7 (95% CI 0.3 to 1.0) for Funen and Finland only.
§§28.9% (95% CI 31.6 to 26.2) for Funen and Finland only.
¶¶0.3 (95% CI 0.2 to 0.2) for Funen and Finland only.
CAs, congenital anomalies; LOS, length of stay.

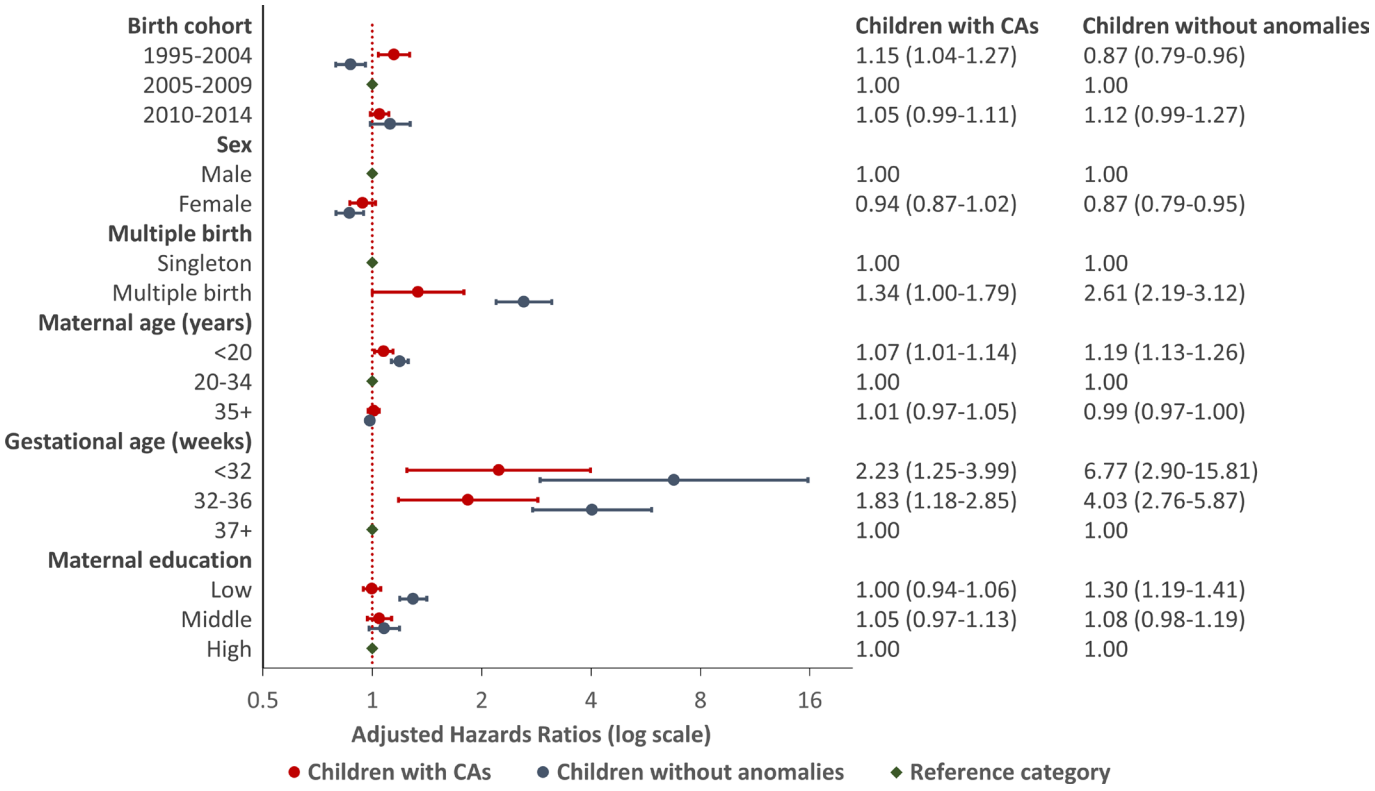

Figure 1 Cox proportional hazards ratios for risk of admission <1 year of age for children with and without anomalies. Birth cohort is adjusted for sex, gestational age and maternal age. Sex is adjusted for birth cohort, gestational age and maternal age. Maternal age is adjusted for birth cohort, sex and gestational age. Gestational age is adjusted for birth cohort, sex and maternal age. Multiple births and maternal education estimates are unadjusted in the models. CAs, congenital anomalies.

All girls (with and without CAs) were less likely to be admitted than boys, particularly those aged 1–4 years, who were 24% less likely to be admitted (figures 1 and 3). There was little difference in LOS between girls and boys (figures 2 and 4).

Risk of admission and LOS varied by birth cohort. For children born 2010–2014, those aged <1 year had no statistically significant risks of hospital admission or longer stays (figures 1 and 2), but those without CAs aged 1–4 years were significantly less likely (18%) to have an admission and had 50% longer hospital stays compared with children born 2005–2009 (figures 3 and 4). In contrast, children aged <1 year with CAs born 1995–2004 (based on Funen and Finland only) were 15% more likely to be admitted and had 46% longer stays compared with those born 2005–2009, while children without CAs were 13% less likely to have a hospital admission (figures 1 and 2).

For children in both age groups with CAs, young maternal age (<20 years) was associated with an increased risk of admission compared with mothers aged 20–34 years (figures 1 and 3). The effect was more marked in children without CAs as risk of admission increased by 19% in children aged <1 year and by 13% in those aged 1–4 years. Children born to young mothers also had longer LOS, particularly children without CAs, who had 34% longer stays aged <1 year and 29% longer stays at 1–4 years (figures 2 and 4).

Children aged <1 year without CAs born to mothers with low levels of education had a 30% increased risk of admission, and children with and without CAs had (40%–48%) longer stays than children born to highly educated mothers.

Comment

Principal finding

All the risk factors investigated in this large population-based study were generally associated with an increased risk of admission or LOS among children with and without CAs at 0–4 years of age. The association was typically greater in the first year of life and in children without CAs. Despite this, a higher proportion of children with CAs were admitted to hospital and had longer stays than children without CAs, reflecting the impact of the anomaly on their healthcare needs. Preterm birth and multiple births had the greatest association with risk of admission and LOS for children with and without CAs, and this persisted up to the child's fifth birthday. The association with preterm birth is unsurprising as all children born <32 weeks GA are admitted to hospital. These findings can be used to inform parents following a prenatal diagnosis or birth of a child with a CA.[26]

Strengths of the study

This study is based on data from high-quality population-based EUROCAT CA registries that standardised their

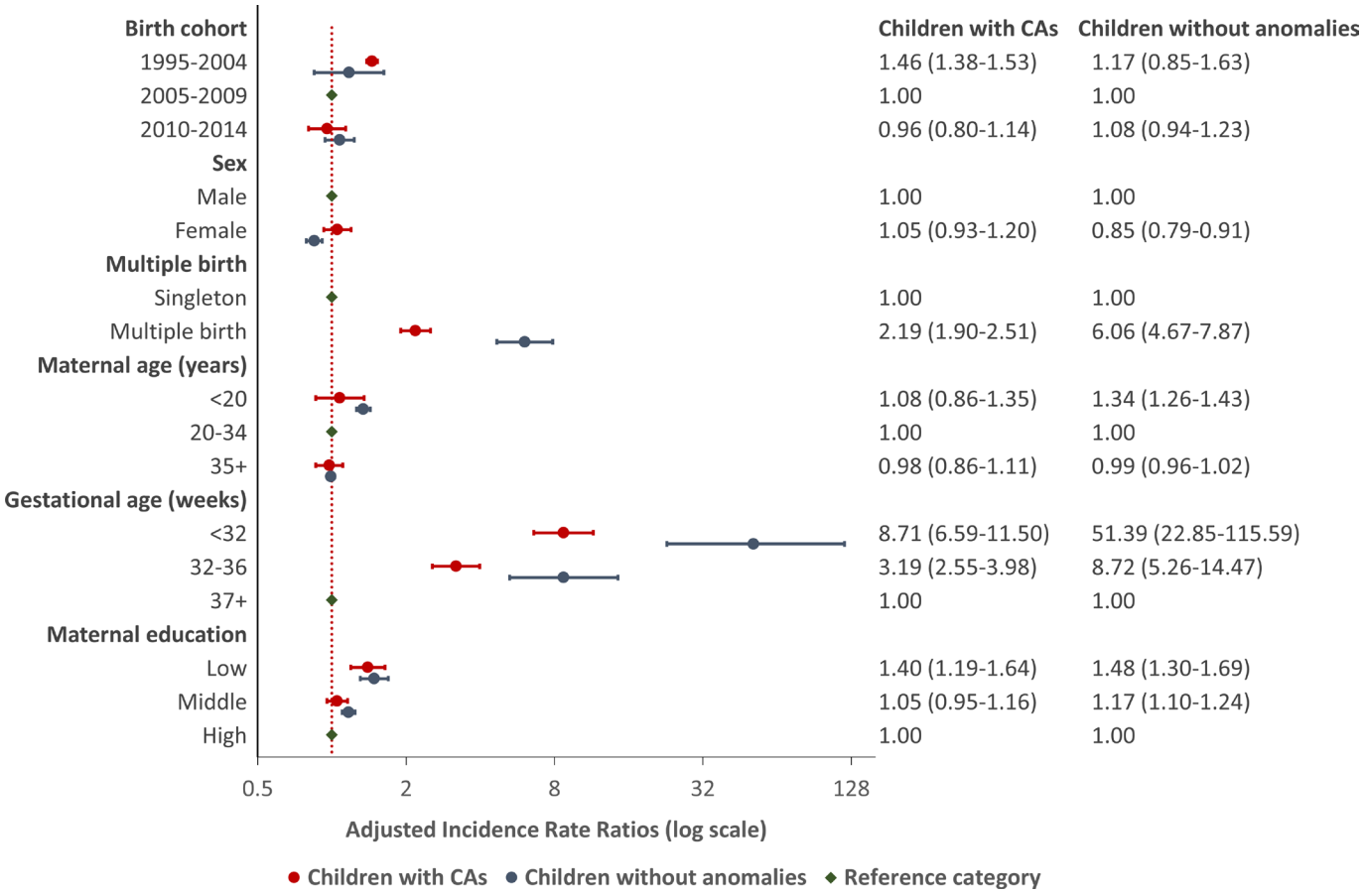

Figure 2 Incidence rate ratios for LOS <1 year of age for children with and without anomalies. Birth cohort is adjusted for sex, gestational age and maternal age. Sex is adjusted for birth cohort, gestational age and maternal age. Maternal age is adjusted for birth cohort, sex and gestational age. Gestational age is adjusted for birth cohort, sex and maternal age. Multiple births and maternal education estimates are unadjusted in the models. CAs, congenital anomalies; LOS, length of stay.

linked data according to the EUROlinkCAT common data model. Previous studies examining risk factors for admission or LOS in children with CAs included children with minor CAs[6] or relied on ICD-based hospital admission diagnoses without any verification of the anomaly.[17] Subsequently, they will have missed admissions among children with CAs and included admissions in children without CAs.[17 27] The high level of completeness of risk factor data will have minimised potential bias. The results are generalisable to western and northern Europe.

Limitations of the data

A limitation of this study is that all CAs were combined in the analysis, rather than investigating specific CAs. In an earlier study, the percentage of children with CAs <5 years who were hospitalised varied by type of anomaly,[13] so it is likely that risk factors for admission and LOS will also vary by type of anomaly. However, analysing the data for all children with CAs combined increased the power to detect the impact of these risk factors on the two outcomes.

Due to the number of multiple comparisons, four significant results would be expected by chance alone. However, the results were generally consistent with a greater association between risk factors and risk of admission or LOS seen in children <1 year than at 1–4 years.

Stratifying on GA may introduce collider bias as prenatal diagnosis of an anomaly may lead to early induction for planned birth and postnatal treatment. However, induction of birth due to an anomaly is usually performed at 35–36 weeks GA[28 29] due to risk of adverse outcomes associated with early preterm birth. Moreover, we found little difference in estimates between the unadjusted and adjusted results, which suggests that collider bias did not have a significant effect in this study as it was based on live births only, that is, babies with severe CAs may have been terminated following prenatal diagnosis of the anomaly or died in utero.

Interpretation

Given that, all children born <32 weeks GA need hospitalisation for survival, it is unsurprising that GA had the greatest impact on the risk of admission and LOS for children with and without CAs. Multiple births was the second most important risk factor for admission and LOS for both groups of children. Children from multiple births have a ninefold[30] increased risk of being born preterm compared with singletons, with 52%–54% of multiple births preterm.[30 31] GA will, therefore, have

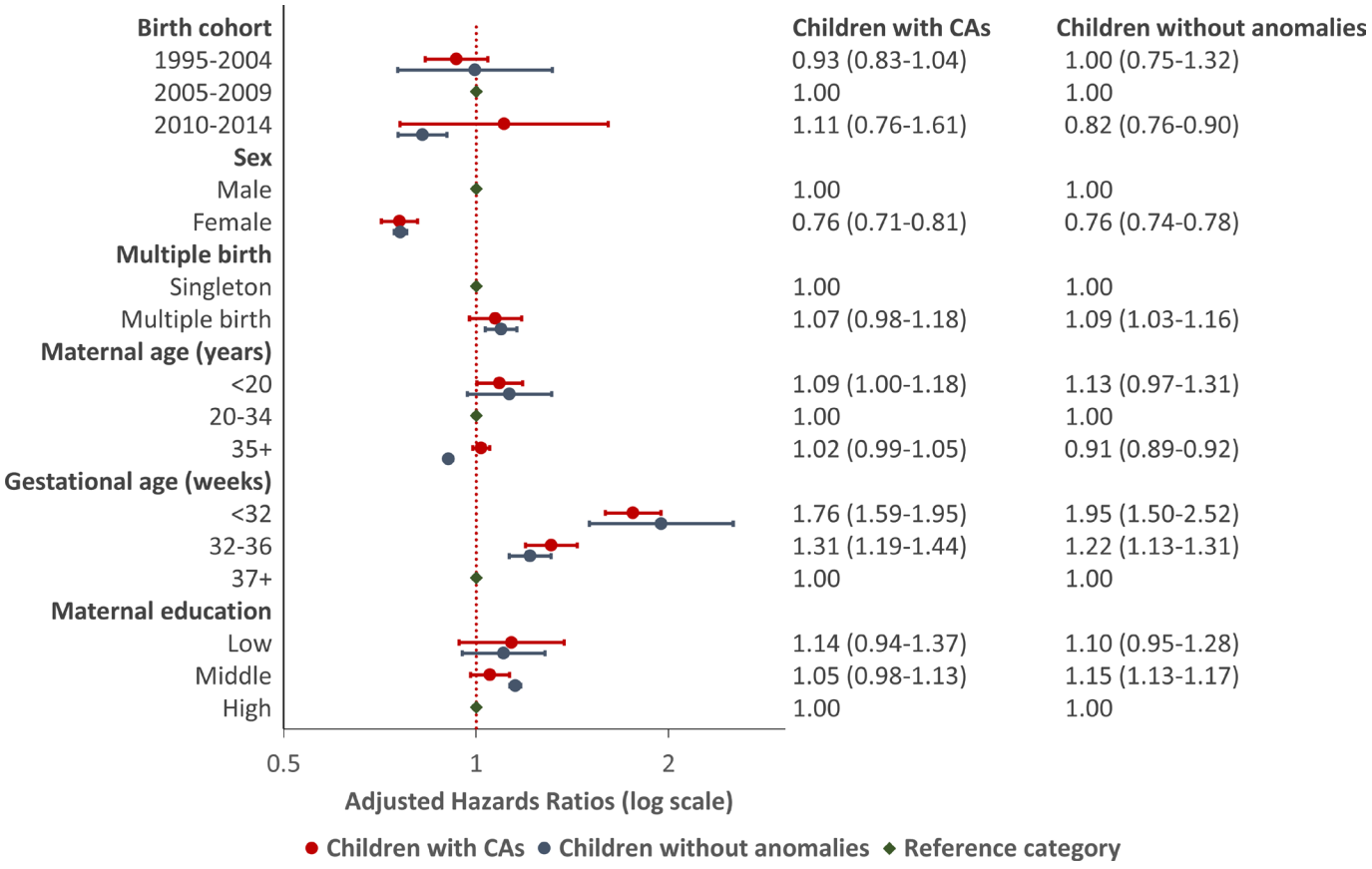

Figure 3 Cox proportional hazards ratios for risk of admission at 1–4 years of age for children with and without anomalies. Birth cohort is adjusted for sex, gestational age and maternal age. Sex is adjusted for birth cohort, gestational age and maternal age. Maternal age is adjusted for birth cohort, sex and gestational age. Gestational age is adjusted for birth cohort, sex and maternal age. Multiple births and maternal education estimates are unadjusted in the models. CAs, congenital anomalies.

contributed to the increased risk of admission and LOS seen in multiples. Unfortunately, in this study, multiple births could not be included in the adjusted models due to small numbers, so it is impossible to determine what impact multiple births, adjusted for GA, would have had on the risk of admission and LOS. Twins, particularly monochorionic twins, have a higher risk of CAs than singletons.[32] Despite this, the association between multiple births and risk of admission, or LOS in childhood, has not been examined previously in children with CAs. In the Oxford record linkage study (1970–1993), twins <5 years of age experienced two times the LOS and triplets almost eight times the LOS seen in singletons. No adjustment was made for GA or birth weight.[33] In Western Australia (1993–2008), twins <5 years were 30% more likely and higher order multiples were 210% more likely to be admitted to hospital than singletons. The mean LOS <5 years was 3.4 days for singletons, 7.2 days for twins and 14.2 days for higher order multiples.[34]

Male sex, young maternal age and middle or low education accounted for smaller increases in risk of admission or LOS for both children with and without CAs. Our findings are generally consistent with the literature, as previous studies reported that males with CAs,[6 17] DS[15] and congenital heart defects[35] had an increased risk of admission in childhood, and that boys with DS had longer hospital stays than girls.[36] Previous studies assessing the effect of maternal age and education on hospitalisation and LOS in children with specific CAs were limited to the first year of life (infancy). Young maternal age was associated with an increased risk of ≥2 admissions among children with CAs in infancy,[14] but it was not related to LOS among children with spina bifida[18] or gastroschisis.[37 38] In contrast, maternal education had no effect on the number of hospital admissions in infancy for children with spina bifida,[18] while less than high school graduate education was associated with increased LOS for children with late-detected critical congenital heart disease.[19]

While the risk factors explored tend to affect all children similarly, the impact of these on children without CAs was typically greater than that seen for children with CAs. Children without CAs have fewer hospital admissions than children with CAs, as the anomaly itself is a risk factor for admission and longer LOS in children with CAs.[13] These children may need surgery for their anomaly[39] and are more likely to need gastrostomy for tube feeding.[40] They are also more likely to be admitted to hospital with other illnesses, such as asthma or acute respiratory infections and stay longer in hospital for these than children without CAs.[6 9–11 41]

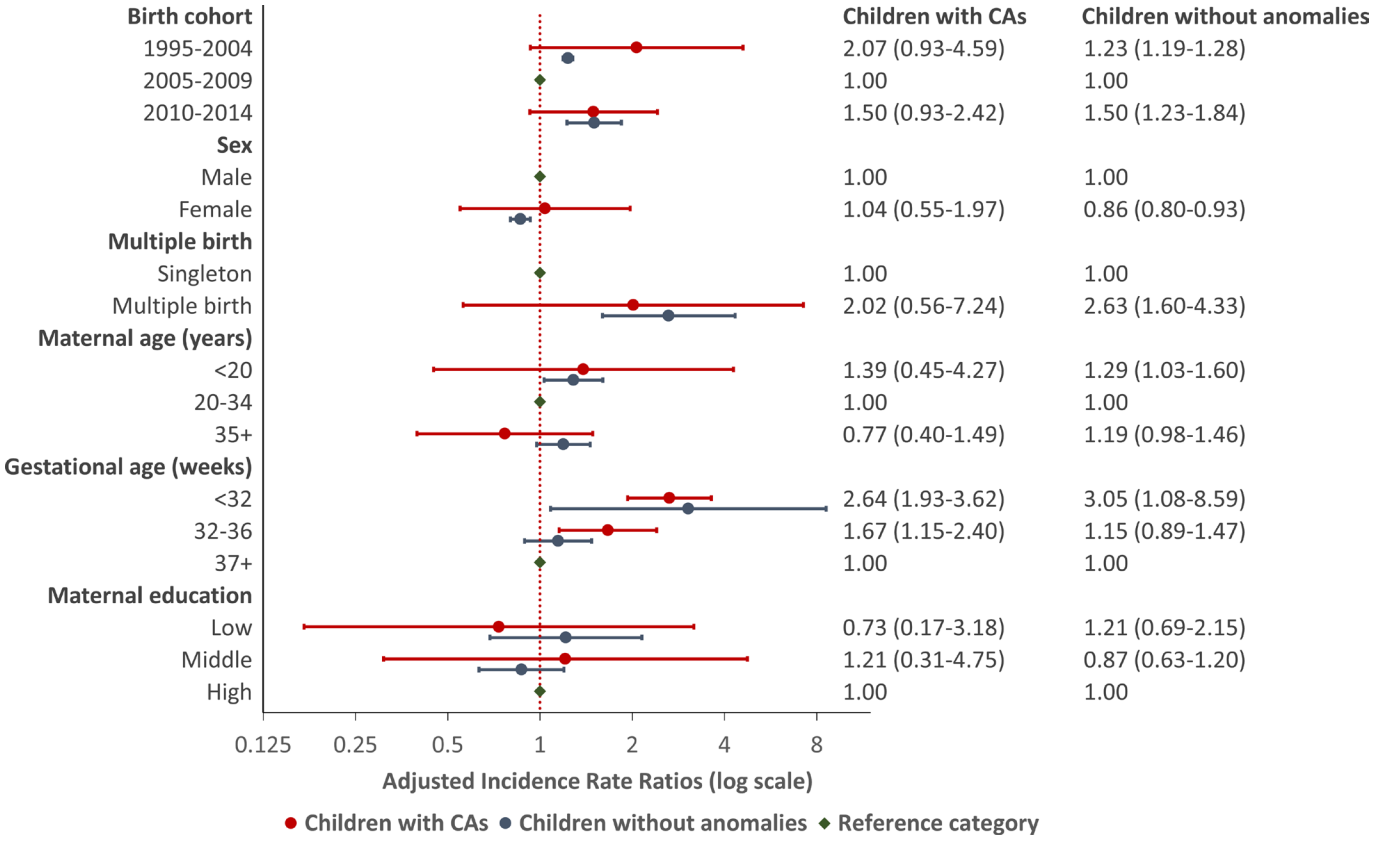

Figure 4 Incidence rate ratios for LOS at 1–4 years of age for children with and without anomalies. Birth cohort is adjusted for sex, gestational age and maternal age. Sex is adjusted for birth cohort, gestational age and maternal age. Maternal age is adjusted for birth cohort, sex and gestational age. Gestational age is adjusted for birth cohort, sex and maternal age. Multiple births and maternal education estimates are unadjusted in the models. CAs, congenital anomalies; LOS, length of stay.

CONCLUSIONS

While risk factors for admission and LOS are the same for children with and without anomalies, they have less impact on those with CAs as these children have more admissions and longer hospital stays reflecting their complex healthcare needs. Preterm and multiple births had the greatest association with the risk of admission and LOS persisting up to the child's fifth birthday. Our findings also demonstrate that male sex, young maternal age and maternal education should also be considered when advising parents on the future healthcare needs of their children. These findings are important for clinicians and other healthcare providers to counsel parents and help manage parental expectations around the care of their children.

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