# Hospital care in the first ten years of life of children with congenital anomalies in six European countries: Data from the EUROlinkCAT Cohort linkage study

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# Hospital care in the first 10 years of life of children with congenital anomalies in six European countries: data from the EUROlinkCAT cohort linkage study

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## ABSTRACT

**Objective** To quantify the hospital care for children born with a major congenital anomaly up to 10 years of age compared with children without a congenital anomaly.

**Design, setting and patients** 79 591 children with congenital anomalies and 2 021 772 children without congenital anomalies born 1995–2014 in six European countries in seven regions covered by congenital anomaly registries were linked to inpatient electronic health records up to their 10th birthday.

**Main outcome measures** Number of days in hospital and number of surgeries.

**Results** During the first year of life among the seven regions, a median of 2.4% (IQR: 2.3, 3.2) of children with a congenital anomaly accounted for 18% (14, 24) of days in hospital and 63% (62, 76) of surgeries. Over the first 10 years of life, the percentages were 17% (15, 20) of days in hospital and 20% (19, 22) of surgeries. Children with congenital anomalies spent 8.8 (7.5, 9.9) times longer in hospital during their first year of life than children without anomalies (18 days compared with 2 days) and 5 (4.1–6.1) times longer aged, 5–9 (0.5 vs 0.1 days). In the first year of life, children with gastrointestinal anomalies spent 40 times longer and those with severe heart anomalies 20 times longer in hospital reducing to over 5 times longer when aged 5–9.

**Conclusions** Children with a congenital anomaly consume a significant proportion of hospital care resources. Priority should be given to public health primary prevention measures to reduce the risk of congenital anomalies.

### WHAT IS ALREADY KNOWN ON THIS TOPIC

⇒ Congenital anomalies are a major cause of mortality and morbidity in childhood.
⇒ Studies from the USA and Australia have evaluated how much greater hospital care needs are for children with anomalies.
⇒ Studies have reported on cardiac anomalies and Down syndrome, but there is sparse literature on gastrointestinal and other anomalies.

### WHAT THIS STUDY ADDS

⇒ During the first year of life, 2.4% of European children with a congenital anomaly accounted for 18% of days in hospital and 63% of surgeries.
⇒ Over the first 10 years of life, the percentages were 17% of days in hospital and 20% of surgeries.
In the first year of life, children with gastrointestinal anomalies spent 40 times longer in hospital than children without a congenital anomaly.

### HOW THIS STUDY MIGHT AFFECT RESEARCH, PRACTICE OR POLICY

⇒ This study shows that although congenital anomalies are infrequent, they account for a significant proportion of hospital care resources.
⇒ This highlights that priority should be given to public health primary prevention measures to reduce the risk of congenital anomalies.

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## BACKGROUND

In Europe, 2%–3% of all babies are born with a major congenital anomaly (birth defect).[1] Congenital anomalies are a major cause of mortality and morbidity in childhood. In Australia, children under 18 with congenital anomalies account for around 22% of all days spent in hospital by children (derived from the data in table 1).[2] Three American studies have evaluated in-patient hospital care needs of children with congenital anomalies after infancy,[3–5] but the availability of data from Europe is much scarcer.[6–8] Studies are also often based in centres of excellence and children attending may not be representative of those attending local hospitals. Studies have reported on cardiac anomalies and Down syndrome,[4 7 9 10] but there is sparse literature on gastrointestinal[8] and other anomalies.

A European study of children with congenital anomalies up to the age of 10 years showed that these children were more likely to be admitted to hospital with longer median lengths of stay.[11] The aim of the present study is to analyse these European data to estimate the total hospital associated

**Table 1** The number and percentage (95% CI) of children with congenital anomalies (CAs) and reference children (without CAs) admitted to hospital according to each registry and age

| | | | Children with CAs | | Reference children | |
|---|---|---|---|---|---|---|
| Registry | Birth years | Age | Total number | Percentage admitted to hospital (95% CI) | Total number | Percentage admitted to hospital (95% CI) |
| Italy, Tuscany* | 2005–2014 | <1 year | 4225 | 93.2 (92.4 to 94.0) | 23 503 | 39.6 (39.0 to 40.3) |
| | | 1–4 years | 4121 | 49.8 (48.1 to 51.5) | 23 503 | 18.8 (18.2 to 19.3) |
| | | 5–7 years | 2484 | 33.7 (31.2 to 36.3) | 13 793 | 16.0 (15.2 to 16.9) |
| Italy, Emilia Romagna | 2008–2014 | <1 year | 5381 | 93.9 (93.2 to 94.5) | 223 995 | 37.3 (37.1 to 37.5) |
| | | 1–4 years | 5210 | 47.2 (45.6 to 48.8) | 223 958 | 16.4 (16.3 to 16.6) |
| | | 5–7 years | 1911 | 25.9 (20.2 to 32.7) | 98 401 | 9.4 (9.0 to 9.7) |
| Denmark, Funen | 1995–2014 | <1 year | 2423 | 73.7 (71.9 to 75.4) | 100 748 | 27.9 (27.6 to 28.1) |
| | | 1–4 years | 2285 | 64.8 (62.8 to 66.9) | 99 945 | 27.4 (27.1 to 27.6) |
| | | 5–9 years | 1862 | 44.4 (41.9 to 46.9) | 81 352 | 16.3 (16.0 to 16.6) |
| The Netherlands, North Netherlands*† | LBZ 2013–2014 | <1 year | 555 | 79.9 (76.5 to 83.2) | | |
| | | 1–4 years | 530 | 56.4 (51.0 to 62.0) | 5730 | 29.3 (27.4 to 31.3) |
| | LMR 1995–2010 | <1 year | 6975 | 66.5 (65.4 to 67.6) | 55 770 | 34.8 (34.4 to 35.2) |
| | | 1–4 years | 6520 | 56.4 (55.2 to 57.7) | 54 770 | 28.6 (28.2 to 29.1) |
| | | 5–9 years | 4660 | 38.2 (36.8 to 39.7) | 39 245 | 20.7 (20.3 to 21.1) |
| Spain, Valencian Region | 2010–2014 | <1 year | 4260 | 96.5 (95.9 to 97.0) | 168 563 | 25.6 (25.4 to 25.8) |
| | | 1–4 years | 4093 | 40.9 (39.1 to 42.9) | 168 495 | 13.3 (13.1 to 13.6) |
| United Kingdom, Wales | 1998–2014 | <1 year | 17 448 | 71.9 (71.3 to 72.6) | 531 784 | 31.4 (31.2 to 31.5) |
| | | 1–4 years | 16 558 | 68.5 (67.7 to 69.2) | 509 565 | 38.0 (37.8 to 38.1) |
| | | 5–9 years | 12 313 | 46.5 (45.5 to 47.5) | 357 934 | 25.7 (25.5 to 25.9) |
| Finland | 1997–2014 | <1 year | 38 324 | 60.7 (60.3 to 61.2) | 911 679 | 21.2 (21.1 to 21.3) |
| | | 1–4 years | 37 213 | 54.7 (54.2 to 55.3) | 909 733 | 28.2 (28.1 to 28.3) |
| | | 5–9 years | 27 121 | 38.8 (38.2 to 39.5) | 701 127 | 18.0 (17.9 to 18.1) |

All data in this table are identical to that presented in table 1 in Urhoj *et al*.[11]
*Reference children were from a 10% random sample of all children excluding EUROCAT children in Tuscany and a 20% random sample in Northern Netherlands.
†All numbers are rounded to the nearest 5 for the Northern Netherlands. Two datasets, LMR and LBZ, covering the registry area, were used, LMR for birth years 1995–2010 and LBZ for 2013–2014. LBZ data for reference children were only included for 1–4 years as outpatient contacts in 2013 were recorded as admissions and<1 year data were therefore excluded.
LBZ, Dutch hospital data; LMR, Dutch hospital data.

healthcare of children with congenital anomalies as a proportion of the total hospital-associated healthcare for all children up to age 10 years and to quantify the relative care needed for children with specific congenital anomalies. Congenital anomaly registries that were unable to provide information on children without congenital anomalies were excluded from the present study but were included in the earlier European study by Urhoj *et al*.[11]

## METHODS

### Study populations

This study is a population-based data linkage cohort study. The European network of population-based registries for the epidemiological surveillance of congenital anomalies (EUROCAT) includes high-quality multiple source registries that ascertain all major congenital anomalies in terminations of pregnancy as well as births. This study includes all children with major congenital anomalies born between 1995 and 2014 in six full-member EUROCAT registries and one associate member (Finland). All children without congenital anomalies born during the same time period and from the same population area covered by a registry were the reference population. Five registries included all reference children. Tuscany included a 10% sample and the Northern Netherlands a 20% sample, with both samples being randomly selected with frequency matching on year of birth and sex.

### Congenital anomalies

Congenital anomalies are coded using the WHO International Classification of Diseases (ICD) 9th or 10th Revision with the British Paediatric Association code extension. For full member registries, cases are automatically assigned by the EUROCAT Data Management Program to define major congenital anomaly subgroups in accordance with the EUROCAT Guide V.1.4.[12] Finland independently assigned the congenital anomalies to the EUROCAT subgroups. Cases with minor anomalies only are excluded.

Analyses were performed for (1) all children with any major congenital anomaly, including chromosomal or genetic conditions, (2) all children with a congenital heart defect (CHD), including any with chromosomal or genetic conditions, (3) children with specific isolated structural anomalies and (4) children with Down syndrome. Isolated anomalies are defined as a non-genetic congenital anomaly in one organ system only or with a known sequence, where multiple congenital anomalies cascade as a consequence of a single primary anomaly.[13]

### Linkage to hospital databases

Each participating registry linked their birth data on children with and without congenital anomalies to their hospital in-patient databases to identify hospital admissions before the child's 10th birthday, or up to 31 December

**Table 2** Children with a congenital anomaly and children with a congenital heart defect (CHD) and their days in hospital and surgeries expressed as a proportion of all days and surgeries in the whole population according to their age at admission

| Age of child | | Registry | | | | | | | Median (IQR) of all registries |
|---|---|---|---|---|---|---|---|---|---|
| | | IT, Tuscany | IT, E Romagna | DK, Funen | Netherlands, Northern† | SP, Valencian Region‡ | Wales | Finland | |
| | ***% live births with any major congenital anomaly**** | ***1.8*** | ***2.3*** | ***2.3*** | ***2.4*** | ***2.5*** | ***3.2*** | ***4.0*** | ***2.4 (2.3, 3.2)*** |
| < 1 year | % days in hospital | 11.8 | 17.6 | 16.5 | 14.3 | 23.6 | 24.5 | 27.0 | 17.6 (14.3, 24.5) |
| | % surgeries | 62.2 | 62.1 | 57.0 | | 75.7 | 75.5 | 63.4 | 62.8 (62.1, 75.5) |
| 1–4 years | % days in hospital | 12.4 | 18.3 | 14.9 | 17.3 | 19.2 | 18.0 | 23.4 | 18.0 (14.9, 19.2) |
| | % surgeries | 16.4 | 18.1 | 22.2 | | 26.7 | 23.9 | 15.1 | 20.2 (16.4, 23.9) |
| 5–9 years | % days in hospital | 8.7 | 10.6 | 13.0 | 8.8 | | 14.1 | 12.3 | 11.5 (8.8, 13.0) |
| | % surgeries | 6.4 | 6.7 | 15.8 | | | 12.1 | 12.0 | 12.0 (6.7, 12.1) |
| < 9 years | % days in hospital | 11.6 | 17.5 | 15.5 | 14.6 | | 19.8 | 22.8 | 16.5 (14.6, 19.8) |
| | % surgeries | 15.3 | 19.2 | 24.4 | | | 21.8 | 20.2 | 20.2 (19.2, 21.8) |
| | ***% live births with a CHD*** | ***0.7*** | ***0.8*** | ***0.9*** | ***0.7*** | ***1.1*** | ***1.0*** | ***1.8*** | ***0.9 (0.7, 1.1)*** |
| < 1 year | % days in hospital | 5.3 | 7.6 | 7.9 | 5.7 | 12.8 | 13.5 | 15.5 | 7.9 (5.7, 13.5) |
| | % surgeries | 34.9 | 38.0 | 33.1 | | 57.3 | 56.7 | 42.2 | 40.1 (34.9, 56.7) |
| 1–4 years | % days in hospital | 4.5 | 6.0 | 6.6 | 7.0 | 9.9 | 8.1 | 10.6 | 7.0 (6.0, 9.9) |
| | % surgeries | 4.0 | 5.1 | 8.3 | | 10.3 | 7.9 | 6.1 | 7.0 (5.1, 8.3) |
| 5–9 years | % days in hospital | 2.8 | 3.0 | 4.1 | 2.4 | | 5.6 | 4.4 | 3.5 (2.8, 4.4) |
| | % surgeries | 1.6 | 2.0 | 5.2 | | | 3.9 | 4.5 | 3.9 (2.0, 4.5) |
| < 9 years | % days in hospital | 4.8 | 6.8 | 6.9 | 5.7 | | 9.8 | 11.4 | 6.9 (5.7, 9.8) |
| | % surgeries | 4.5 | 7.1 | 9.6 | | | 8.5 | 8.9 | 8.5 (7.1, 8.9) |

*Live birth prevalence calculated from numerators and denominators in table 1 allowing for 10% and 20% sampling for Tuscany and Northern Netherlands, respectively.
†Data on surgeries were not available for the Northern Netherlands registry linkage.
‡Follow-up data from ages 5–9 were not available for the Valencian Region registry linkage.

2015. The Northern Netherlands registry linked their data to two different hospital databases that covered different birth years. Children with discharge codes for congenital anomalies in the hospital databases that were not found in the EUROCAT registries were excluded from the study. Linkage success was high for all registries, with 97% of children with anomalies and 95% of children without anomalies being linked to either a hospital database or another healthcare or population database (for those children who did not have a hospital admission). The Italian registries

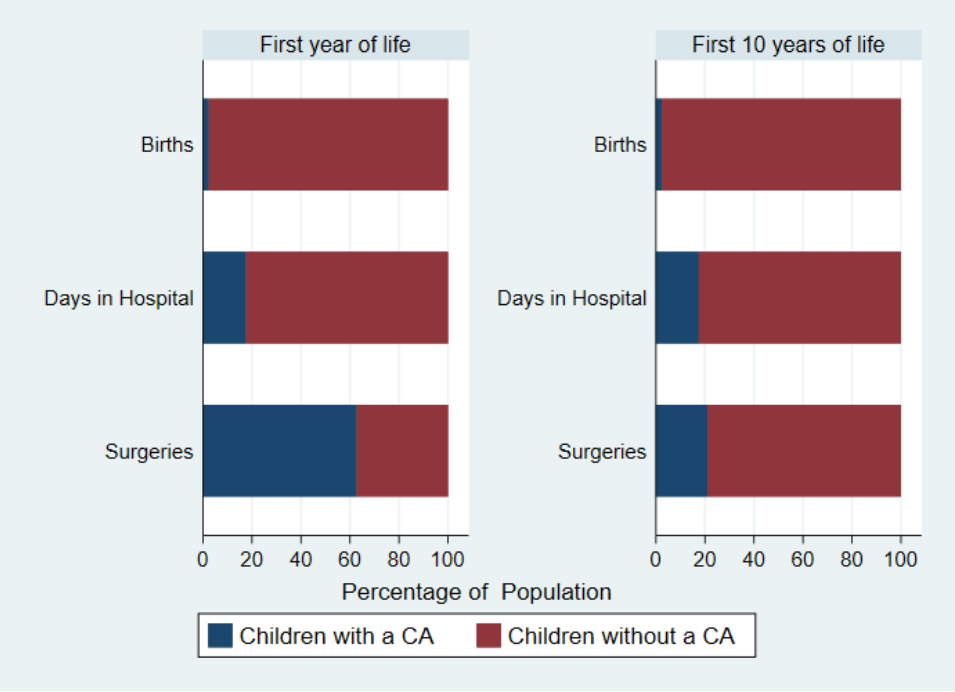


**Figure 1** The proportions of births, days spent in hospital and number of surgeries for children with congenital anomalies during (i) their first year of life and (ii) their first 10 years of life. CA, congenital anomaly

**Table 3** The number of days in hospital for a child without an anomaly and the multipliers (How many times longer children with anomalies spend in hospital compared with children without anomalies) for all children with congenital anomalies, children with selected isolated anomalies and children with Down syndrome: median and IQR of the registries

| | **Average number of days in hospital per year for a child without an anomaly** (expressed as median and IQR of all registries) | | | |
|---|---|---|---|---|
| | <1 year | 1–4 years | 5–9 years | 0–9 years |
| | **1.98 (1.37, 2.11)** | **0.26 (0.25, 0.35)** | **0.10 (0.10, 0.11)** | **0.36 (0.34, 0.39)** |
| | **Multipliers (median and IQR of all registries)** (number of days in hospital for child with anomaly divided by number of days for child without an anomaly (top row of table)) | | | |
| All anomalies including chromosomal and genetic | 8.8 (7.5, 9.9) | 8.1 (7.5, 9.6) | 5.0 (4.1, 6.1) | 7.5 (7.2, 7.8) |
| Isolated anomalies: | | | | |
| Spina Bifida | 13.2 (9.2, 13.9) | 6.3 (5.6, 28.0) | 29.1 (10.8, 66.8) | 17.3 (9.6, 25.5) |
| Hydrocephalus | 16.9 (12.9, 23.3) | 13.6 (7.4, 29.8) | 10.4 (6.7, 13.4) | 15.7 (10.7, 23.8) |
| Severe microcephaly | 7.8 (7.2, 14.3) | 6.4 (5.2, 21.3) | 6.9 (0.0, 10.9) | 13.2 (5.6, 24.4) |
| Congenital cataract | 3.5 (3.2, 5.6) | 4.6 (3.9, 7.2) | 2.6 (1.9, 3.7) | 3.6 (3.4, 3.8) |
| Congenital heart defects (CHD) | 6.9 (5.9, 10.6) | 5.7 (4.5, 7.4) | 3.3 (3.0, 3.4) | 5.9 (5.8, 6.6) |
| Transposition of great arteries | 21.0 (13.1, 23.8) | 12.8 (9.9, 16.6) | 5.2 (4.7, 6.2) | 13.0 (11.8, 14.6) |
| Ventricular septal defect | 5.5 (4.4, 9.3) | 3.3 (3.0, 6.7) | 2.1 (1.5, 2.2) | 4.0 (3.8, 5.8) |
| Atrial septal defect | 9.3 (8.0, 11.5) | 7.7 (5.8, 14.2) | 3.6 (2.7, 4.0) | 7.3 (6.6, 9.6) |
| Atrioventricular septal defect | 18.7 (11.7, 19.6) | 24.6 (14.6, 29.6) | 4.9 (3.6, 10.4) | 16.1 (11.7, 20.2) |
| Tetralogy of Fallot | 17.5 (14.4, 19.5) | 15.7 (11.5, 16.4) | 6.1 (3.3, 12.7) | 14.8 (14.6, 16.0) |
| Pulmonary valve stenosis | 8.9 (6.7, 11.3) | 8.7 (5.2, 23.0) | 3.3 (2.7, 5.0) | 5.8 (5.6, 9.2) |
| Aortic valve atresia/stenosis | 9.9 (7.4, 14.8) | 3.7 (2.1, 9.6) | 6.1 (5.7, 6.3) | 7.4 (5.9, 10.3) |
| Mitral valve anomalies | 11.4 (5.9, 18.3) | 11.6 (5.1, 33.0) | 6.7 (3.8, 9.3) | 12.1 (10.1, 16.2) |
| Hypoplastic left heart | 20.3 (6.8, 37.9) | 29.2 (18.2, 42.9) | 23.9 (9.5, 30.1)* | 29.6 (12.6, 36.5) |
| Coarctation of aorta | 14.2 (13.7, 17.2) | 10.0 (6.9, 13.2) | 4.3 (3.9, 5.9) | 11.9 (10.1, 12.7) |
| Patent ductus arteriosus as only CHD in term infants (≥37 weeks) | 4.1 (2.1, 5.4) | 2.7 (1.6, 4.0) | 0.6 (0.2, 1.3) | 2.9 (2.3, 4.5) |
| Cystic adenomatous malformation of lung | 10.1 (6.8, 12.3) | 1.8 (1.5, 5.1) | 0.6 (0.0, 1.2)* | 3.8 (3.7, 4.5) |
| Cleft lip with or without cleft palate | 6.2 (5.9, 7.0) | 5.3 (3.1, 7.2) | 4.6 (3.6, 5.4) | 5.5 (5.3, 5.7) |
| Cleft palate | 5.8 (4.9, 6.5) | 5.4 (3.9, 6.9) | 2.8 (2.5, 3.0) | 5.4 (4.6, 7.5) |
| Oesophageal atresia with or without tracheo-oesophageal fistula | 28.4 (25.5, 35.9) | 20.6 (13.2, 36.5) | 7.4 (3.7, 9.1) | 22.0 (17.7, 24.2) |
| Duodenal atresia or stenosis | 15.8 (15.3, 20.8) | 3.1 (1.2, 4.9) | 2.3 (1.6, 3.9) | 9.3 (8.5, 9.9) |
| Atresia or stenosis of other parts of small intestine | 46.0 (27.4, 53.2) | 22.0 (7.8, 54.3) | 3.9 (2.4, 8.8) | 31.0 (20.0, 43.5) |
| Ano-rectal atresia and stenosis | 9.1 (7.8, 11.3) | 6.1 (3.1, 8.6) | 4.5 (2.5, 5.6) | 7.6 (5.4, 10.5) |
| Diaphragmatic hernia | 17.8 (9.7, 22.1) | 4.3 (1.9, 6.2) | 2.7 (0.8, 4.6) | 12.9 (9.3, 16.7) |
| Gastroschisis | 32.7 (26.4, 35.9) | 4.3 (2.3, 11.1) | 1.2 (0.0, 1.4) | 15.3 (12.2, 17.1) |
| Omphalocele | 17.9 (10.8, 22.5) | 4.3 (2.1, 12.6) | 1.2 (0.6, 2.6) | 11.1 (7.3, 14.3) |
| Multicystic renal dysplasia | 4.8 (3.8, 9.5) | 2.8 (1.8, 3.6) | 1.5 (1.0, 5.3) | 3.6 (3.0, 4.2) |
| Congenital hydronephrosis | 5.3 (4.6, 7.4) | 5.3 (5.0, 11.4) | 2.7 (1.5, 2.9) | 4.9 (4.0, 6.6) |
| Hypospadias | 3.2 (2.9, 6.1) | 7.2 (4.4, 10.3) | 2.6 (2.3, 3.6) | 4.6 (3.2, 5.7) |
| Limb reduction defects | 2.9 (2.8, 3.6) | 2.7 (1.8, 12.6) | 3.0 (1.4, 5.0) | 3.9 (2.2, 6.4) |
| Clubfoot—talipes equinovarus | 3.4 (3.2, 4.1) | 1.9 (1.5, 3.0) | 2.7 (2.4, 5.2) | 3.3 (3.3, 3.5) |
| Hip dislocation and/or dysplasia | 2.4 (1.6, 4.7) | 2.0 (1.0, 7.3) | 0.9 (0.6, 1.6) | 1.8 (1.3, 4.9) |
| Polydactyly | 2.0 (1.2, 2.4) | 2.1 (1.4, 2.8) | 1.3 (0.9, 1.7) | 1.8 (1.5, 2.3) |
| Syndactyly | 2.1 (1.5, 2.7) | 2.1 (1.4, 3.0) | 1.3 (1.0, 1.4) | 1.9 (1.5, 2.3) |
| Craniosynostosis | 6.1 (5.0, 6.7) | 4.6 (3.0, 10.5) | 1.3 (0.8, 2.2) | 4.6 (4.5, 4.7) |
| Genetic syndromes | | | | |
| Down syndrome | 14.1 (12.8, 15.9) | 14.2 (10.0, 15.7) | 5.5 (2.9, 6.5) | 12.5 (10.3, 14.2) |
| Down syndrome with CHD | 20.1 (16.9, 23.9) | 17.8 (13.8, 21.3) | 6.0 (3.5, 7.3) | 17.6 (15.1, 19.0) |
| Down syndrome without CHD | 8.4 (7.9, 11.0) | 7.2 (5.4, 8.9) | 3.7 (3.1, 5.0) | 7.1 (6.6, 8.6) |

*Estimates are less reliable as they are based on less than 50 children and/or on data from less than four registries.

followed children up to 7 years and the Spanish registry up to 5 years. For the Northern Netherlands, only those children born 1995–2010 were followed up to 9 years. Data for reference children aged <1 year, born 2013–2014 in the Northern Netherlands, were excluded as outpatient contacts in 2013 had been incorrectly recorded as admissions. Detailed information on linkage and standardisation is given elsewhere.[14–16]

### Length of stay and surgeries

Length of stay (LOS) was calculated as the number of days between the date of admission to hospital and the date of discharge. For hospital stays, where the date of admission and discharge occurred on the same day, the LOS was considered to be 0.5 days. Admissions associated with birth only (ie, obstetric stays immediately after birth with no additional procedures) were excluded.

Surgeries were coded according to the coding systems used in the national health systems. Italy and Spain used the ICD, Ninth Revision, Clinical Modification, England and Wales used the Office of Population Censuses and Surveys Classification of Interventions and Procedures and Finland and Denmark used national adaptions of NCSP (NOMESCO Classification of Surgical Procedures). Information on surgeries for the Northern Netherlands was incomplete and not analysed. Two paediatricians independently determined if a code was for a surgical procedure and a consensus between the two clinicians was reached over the final list of surgery codes. Online supplemental appendix table 1 summarises the broad decisions made over which procedures were considered surgeries.

### Statistical analysis

The total number of days spent in hospital and the total number of surgeries for children with specific congenital anomalies and for reference children were calculated for children in their first year of life, from 1 to 4 years and from 5 to 9 years of age. An estimate of these numbers for children aged 0–9 years was obtained by weighting the number of days and surgeries within each age group by the ratio of the child-years of exposure that would have been observed if full follow-up of all children had occurred in that age group divided by the years of exposure that were observed in the age group. This weighting was necessary as, if in one registry only around 20% of children had full follow-up after the age of 5, then the numbers of days and surgeries were multiplied by a factor of 5 (1/20%) to enable the totals from 0 to 9 to be calculated per live birth.

The whole population of live births was assumed to be the children with anomalies plus the reference children for five registries. For the two registries with 10% and 20% samples of reference children, the whole population of live births was assumed to be the children with anomalies plus the number of reference children divided by the sampled fractions.

Within each registry, the average length of stay per child was calculated by dividing the total number of hospital days by the number of children known to be alive at the start of each specific age group (<1 year, 1–4 years, 5–9 years), adjusted for their length of follow-up. The average length of stay for children with an anomaly was divided by the average length of stay for reference children, to estimate how many times longer a child with an anomaly was in hospital compared with a reference child.

The median values and IQRs of all the registry estimates were calculated to provide European estimates. The relative lengths of stay were also compared according to the birth cohort of the children: those born 1995–2004, 2005–2009 and 2010–2014.

### Ethics approval

All EUROCAT registries obtained ethical, governance and other permissions for the data linkage according to their national legislation and arrangements. University of Ulster obtained ethics permission for the Central Results Repository on 15 September 2017 (Institute of Nursing and Health Research Ethics Filter Committee, number FCNUR-17–000).

## RESULTS

Data on 79 591 children with anomalies and 2 021 772 reference children were available for analysis (table 1) from seven registries.[11] In all registries, a higher proportion of children were admitted during their first year of life than during the following 4 years, with children with anomalies being much more likely to be admitted.

Table 2 and figure 1 show the percentage of children with congenital anomalies and their percentage share of hospital stays and surgeries. The live birth prevalence of children with congenital anomalies varied according to registry with Finland having the highest prevalence (4.0%). The high prevalence in Finland has been partly explained by inclusion of more minor heart anomalies in the Finnish registry.[11] During the first year of life in the seven regions, a median of 2.4% (IQR: 2.3, 3.2) of children with a congenital anomaly accounted for around 18% (14, 24) of days in hospital and 63% (62, 76) of surgeries. During the first 10 years of life, the percentages were 17% (15, 20) of days in hospital and 20% (19, 22) of surgeries. The percentage share of surgeries fell as the children grew older, while the percentage share of days in hospital only decreased after age 5 years.

Table 2 shows that around one-third of children with a congenital anomaly had a CHD and that 40% of surgeries under age 1 year and 8% of all surgeries under age 10 years were performed in these children. The proportions of days in hospital reduced considerably after the first year of life but remained raised up to the age of 9 years.

Table 3 shows that children with congenital anomalies spent around nine times longer in hospital during their first year of life than reference children. On average, reference children spent almost 2 days in hospital in their first year (row 1 of table 3); therefore, children with anomalies were likely to spend around 18 days in hospital (9×2). Children with congenital anomalies spent around five times longer in hospital than reference children from ages 5–9 years (0.5 days per year compared with 0.1 day per year). Children with gastrointestinal anomalies were in hospital the longest; children with atresia or stenosis of other parts of small intestine being in hospital over 40 times longer in their first year of life and 20 times longer in ages 1–4 years. Children with abdominal wall defects such as gastroschisis had extremely long stays only in their first year of life. Children with Down syndrome and CHD spent longer in hospital than children with only Down syndrome.

We observed no significant trends of the relative lengths of stay for three different birth cohorts (1995–2004, 2005–2009 and 2010–2014 (data not shown).

## DISCUSSION

This study illustrates the high hospital healthcare needs of children with congenital anomalies, with a relatively small proportion of children (2.4%) accounting for a large proportion of hospital care, particularly in their first year of life (18%). The relative needs decreased as the children grew older, but still remained much greater for children with congenital anomalies than reference children up to age 10 years (17%). These results differed by registry with Finland having the highest prevalence (4.0%) and Italy, Tuscany (1.8%) the lowest which was probably due to the inclusion by Finland of more minor anomalies. However, the pattern of a large proportion of hospital care occurring in the first year of life which decreased as the children grew older occurred in all registries. The results from Finland were very similar to those observed in Australia, where the 4.6% of children born with a major congenital anomaly accounted for

25% of all days spent in hospital in the first year of life and 22% of days up to 17 years of age (derived from the data in table 1)[2] compared with 4.0%, 27% and 23% (up to age 10) in Finland.

Children with CHD accounted for 40% of surgeries in the first year. In addition to cardiac surgery, surgeries for other anomalies, surgeries for feeding tubes, or minor surgeries performed in hospitals rather than out-patients due to the increased risk of anaesthesia,[17] are included. Our results are consistent with those from two American studies finding that for the first year of life around 10% of all costs are incurred by infants with a CHD[3] and that the median costs for these children was about ten-fold higher than for children without a CHD.[4]

Two hospital-based studies in America found that children with Down syndrome spent 25 times longer in hospital in their first year of life and eight times longer when aged 1 to 4 years compared with children without Down syndrome.[18 19] These findings are similar to our study estimates that children with Down syndrome aged <1 year and 1–4 years spent 14 times longer in hospital compared with reference children.

Two population-based studies in America found the financial costs for children with clefts were 5.4 times higher in the first year of life than for other children and 10 times higher in the first 2 years of life,[20 21] which is consistent with our study findings for children aged <1 year.

A hospital-based study in America by Hook-Dufresne *et al* reported that the lengths of hospital stay in the first year of life of infants with gastroschisis were between 17–24 times greater than other infants in hospital without gastroschisis.[22] This is reasonably consistent with our finding that the lengths of stay for such infants were 33 times greater (IQR: 26, 36) compared with all children without any congenital anomaly.

This study did not find differences in the relative length of time in hospital over birth cohort despite both the rate of terminations of pregnancy for fetal anomaly (TOPFA) for severe anomalies increasing and the survival of liveborn children with congenital anomalies increasing.[23] These changes appear to be of similar magnitude, effectively cancelling each other out.

The strength of this study is that it is based on standardised data from high quality population-based congenital anomaly registries who are all members of EUROCAT and who standardise their data according to the EUROlinkCAT common data model. Population-based congenital anomaly data linked to hospital discharge records will identify all hospital admissions to children with congenital anomalies (if linked), whereas using hospital data only may miss some admissions if a child with an anomaly is admitted due to an unrelated condition, such as an accident/infection, and the anomaly is not reported. Calculating the comparisons of hospital stays and surgeries for children with congenital anomalies relative to the total population within each registry enables valid comparisons to be made across Europe.

A limitation of the study is that it relies on the successful linkage of children with congenital anomalies to healthcare data. Overall, 97% of children with a congenital anomaly and 95% of reference children were linked. We do not believe that the proportion of children not linked to the hospital discharge records would unduly affect the calculations performed as the proportions missed are similar for the children with anomalies and reference children, and our results are consistent with the published literature based on population-based data.

A further limitation is that the overall hospital healthcare needs associated with congenital anomalies are underestimated as out-patient visits, the need of additional care during the pregnancy and the hospital care for pregnancies that result in a termination or a stillbirth are not included.

## CONCLUSIONS

The hospital care needs of children with congenital anomalies account for a significant proportion of hospital care provision to children and priority should be given to public health preventive measures such as the reduction in teenage pregnancies, reduction of cigarette smoking, alcohol consumption, substance misuse, occupational exposures, viral infections and the use of teratogenic medications during pregnancy.[24] In addition, the fortification of flour with folic acid should be adopted to reduce the occurrence of neural tube defects.[24] Strategies for preventing infections and hospitalisations should also be implemented for children with congenital anomalies including vaccinations (COVID, influenza, rota virus, RS virus) and the reduction in exposure to infections in the most vulnerable periods in infancy and after surgery.

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**Data availability statement** Data may be obtained from a third party and are not publicly available. We are legally not allowed to share the third-party administrative data used in this study as it belongs to the data providers in each of the regions i.e. the regional or national statistical organisations. The study team had access to aggregate data only from each region i.e. the linked patient level data remained in the local region. We encourage any interested parties to apply to the EUROlinkCAT management team to assist them in obtaining approval from the data providers in each region/country to use the aggregated data for an approved study (http://www.EUROlinkCAT.eu/contactinformationanddatarequests).

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