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School-recorded special educational needs provision in children with major congenital anomalies: A linked administrative records study of births in England, 2003-2013

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Abstract

Background
Children with major congenital anomalies (MCAs) disproportionately experience complex health problems requiring additional health and educational support.

Objectives
To describe survival to the start of school and recorded special educational needs (SEN) provision among children with and without administrative record-identified MCAs in England. We present results for 12 system-specific MCA subgroups and 25 conditions. We also describe the change of prevalence in recorded SEN provision before and after SEN reforms in 2014, which were implemented to improve and streamline SEN provision.

Methods
We created a birth cohort of 6,180,400 singleton children born in England between 1 September 2003 and 31 August 2013 using linked administrative records from the ECHILD database. MCAs were identified using hospital admission and mortality records during infancy. SEN provision in primary school was defined by one or more recording of SEN provision in state-school records during years 1 to 6 (ages 5/6 years to 10/11 years).

Results
Children with any MCA had a 5-year survival rate of 95.1% (95% confidence interval (CI) 95.0, 95.2) compared with 99.7% (95% CI 99.7, 99.7) among children without an MCA. 41.6% (75,381/181,324) of children with an MCA had any recorded SEN provision in primary school compared with 25.7% (1,285,572/5,008,598) of unaffected children. Of the 12 system-specific MCA subgroups, children with chromosomal, nervous system and eye anomalies had the highest prevalence of recorded SEN provision. The prevalence of recorded SEN provision decreased by 4.8% (99% CI -5.4, -4.3) for children with any MCA compared with a reduction of 4.2% (99% CI -4.3, -4.2) for unaffected children, when comparing pupils in year 1 before and after 2014.

Conclusion
We observed that approximately two fifths of children with MCAs have some type of SEN provision recorded during primary school, but this proportion varied according to condition and declined following the 2014 SEN reforms, similar to children unaffected by MCAs.

Keywords
birth defects; cohort study; congenital abnormalities; ECHILD; special educational needs

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Highlights

- We used linked hospital and school records to identify children with different MCAs and follow them up to the end of primary school, when they are 11 years old
- We found that two in five children with an MCA had any recorded SEN provision in primary school – 1.6 times the proportion in children without an MCA
- Our findings also suggest that SEN provision reduced for all children after government reforms of the SEN system in 2014, including those with MCAs
- Our findings provide information about survival to primary school and the likelihood of specialist support in primary school for children with hospital-identified MCAs in England, which may be useful for families, clinicians and service providers

Introduction

Major congenital anomalies (MCAs) include a variety of structural and functional abnormalities of prenatal origin that are present are birth [1, 2]. Whilst individual anomalies are rare, in England, MCAs are estimated to affect between 2-3% of live births [3, 4]. They are termed "major" because of their significant impact on health, survival, or the individual's physical or social functioning [1]. MCAs can occur in isolation or as a group of anomalies and, for up to half of MCAs, there is no known aetiology [5]. Known causes of MCAs can be stratified into genetic and chromosomal abnormalities occurring pre-conception and environmental exposures post-conception, including teratogenic agents, mechanical forces and vascular disruptions. It is estimated that up to 30% of MCAs are caused by a combination of genetic and environmental factors. Clinical recognition of MCAs is contingent on access to health care, but typically occurs within the first six months of life, and usually before age two years [1, 2].

MCAs are a leading cause of infant mortality [4]. However, over the last few decades, with advances in neonatal care and surgical interventions, the survival of infants born with MCAs has been improving [6, 7]. Ten-year survival estimates for many MCAs are now over 90%, resulting in growing numbers of children with these conditions completing primary school [8]. Studies of selected MCAs, including orofacial clefts and congenital heart disease, show a higher risk of educational achievement below the national average and a greater need of specialist educational support at school for children with these conditions compared to their unaffected peers [9, 10], known as special educational needs (SEN) provision in England. This will contribute important prognostic information for families/carers, as well as help with planning future provision to support this growing population of children.

To the best of our knowledge, evidence on these longer term outcomes is restricted to selected MCAs mostly captured in registry data and, aside from isolated orofacial clefts [11], is not available at a national level in England. Evidence on temporal trends in SEN provision amongst children with similar health conditions can also build an understanding of how external factors, specifically policy changes, have impacted the likelihood of eligibility for SEN provision in school. For example, in 2014, the SEN system in England underwent reforms following the Children and Families Act 2014 and subsequent code of practice [12, 13]. Key principles for the reformed system included: earlier identification; increased participatory decision making; a delegated SEN budget for schools; and changes to the categories of SEN provision offered to children. These reforms were implemented to "offer simpler, improved and consistent help for children and young people with special educational needs" [14]. Yet, as far as we are aware, the precise impact of these changes on children with specific health conditions remains unknown.

Using a national cohort of births in England between 2003 and 2013, we aimed to describe patterns of survival to the start of compulsory education and frequencies of recorded SEN provision across children with and without MCAs identified in hospital and mortality records. We present results for 12 system-specific MCA subgroups and 25 conditions. Specifically, we estimated: rates of survival up to the age 5 years; the prevalence of recorded SEN provision of those attending a state-funded primary school; and differences in the prevalence of recorded SEN provision in year 1 (age 5/6 years) between children attending state-funded school before and after the 2014 SEN reforms.

Methods

Data sources

We used the 'Education and Child Health Insights from Linked Data' (ECHILD) database which comprises linked de-identified data from Hospital Episode Statistics (HES), Office for National Statistics (ONS) mortality records and the National Pupil Database (NPD) [15–17]. HES and NPD contain information recorded on state-funded hospital attendances and school activity in England. HES Admitted Patient Care (APC) contains clinical information on inpatient stays (such as diagnoses and procedures) and captures 97% of all birth admissions in England, meaning that these data can be used to define population-based birth cohorts [15, 18]. Linkage to subsequent hospital admissions in HES APC allows for longitudinal follow-up. ONS mortality data, which is routinely linked to HES, provides information on the cause and date of deaths registered in England and Wales (including those that occur outside of hospital) [19]. The NPD contains several school censuses (Supplementary Table 1, Supplementary Appendix 1), which provide information on school enrolments, as well as pupil-level data, including recorded SEN provision, attainment, absences, and exclusions [16]. We used the Department for Education's opensource 'Get Information about Schools' (GIAS) register to add school type to ECHILD using each school's unique reference number [20]. We also downloaded congenital anomaly prevalence data submitted by England-based registries to the 'European Network of Population-based Registries for Congenital Malformations' (EUROCAT) for external cohort validation [3].

Birth and school cohorts

Our study population (the "birth cohort") included all live singleton births recorded in NHS-funded hospitals in England between 1 September 2003 and 31 August 2013. Birth admissions were identified using a combination of diagnostic and procedure codes, healthcare resource group codes and administrative variables (as outlined in Zylbersztejn et al. [18]). We excluded infants from multiple births, to minimise the risk of linkage error, and infants who were not resident in England, to minimise the risk of loss to follow up [18]. To create the sub-cohort of children attending state-school (the "school cohort"), we then excluded children who: did not link to the NPD; were not present in any NPD census in school years 1 to 6 (ages 5/6 to 10/11 years); or were only present in NPD censuses $\geq$2 years outside the expected age (see Supplementary Figure 1, Supplementary Appendix 1). Pupils not following the national curriculum do not have a school year listed in the NPD, so were instead assigned a year based on their age in each academic year. The maximum date of follow up was 31 August 2019 (the last full academic year before the COVID-19 pandemic).

Major congenital anomalies

We used International Classification of Diseases 10th Revision (ICD-10) codes based on EUROCAT guide 1.5 to define MCAs in this study (Supplementary Table 2, Supplementary Appendix 1) [21]. A child was defined as having "any MCA" if they had one or more relevant ICD-10 code(s) in hospital admission or mortality records during the first year of life (i.e. infancy). The same criteria were applied across the whole cohort, i.e. for all birth years. MCAs were classified by 12 system-specific subgroups. Infants could belong to multiple subgroups (e.g. a child with a nervous system and eye anomaly is counted in both groups), but for each of the 12 subgroups we also identified children who did not have MCAs in any of the other systems ('isolated' anomalies). Within the 12 subgroups we also defined 25 conditions (including isolated and non-isolated cases), which were selected based on a group size of $\geq$200 at the end of follow up to avoid problems with statistical disclosure. Conditions were defined using EUROCAT guidelines, apart from congenital diaphragmatic hernias and anorectal malformations, where we used definitions developed previously [22, 23]. We were unable to apply EUROCAT minor anomaly exclusion rules where 5-digit ICD-10 codes were required (see Supplementary Table 2, Supplementary Appendix 1).

Outcome: Mortality

We reported death from any cause before the age of 5 years to coincide with the expected age at entry into year 1, the first full year of compulsory education and the start of key stage 1 in state-funded schools in England. We also present survival rates to age 7 years, corresponding to the expected age at entry into year 3 (and the start of key stage 2). Occurrence and date of death were derived from HES-ONS linked mortality records. Follow-up began at birth and ended at the earliest of: death, the end of the study period (31 August 2019), or each child's 5^{th} birthday (or 7^{th} birthday for survival rates to age 7 years).

Outcome: recorded SEN provision

In state-funded schools in England, children are entitled to receive SEN provision if they have "a significantly greater difficultly in learning than the majority of others of the same age, or have a disability which prevents them from making use of facilities generally provided by mainstream schools" [24]. There are two broad categories of SEN provision [24]: SEN support (previously, 'School Action'/'School Action Plus'), which is arranged by the school and may include different educational materials or small group support; and Education, Health and Care Plans (EHCP; previously, 'statement of SEN'), which is arranged by local authorities for children whose needs cannot be met by the lower level of provision and may include one-to-one support in the classroom and therapies outside school. To this, we added a third category, 'specialist provision', to differentiate between children attending non-mainstream schools where support for all children differs, including smaller classroom sizes and an adapted curriculum (Supplementary Table 1, Supplementary Appendix 1).

We used the NPD alternative provision, pupil referral unit and termly school censuses linked to the GIAS register to define four categories of recorded SEN provision in each school year: (1) none, where there was no recorded SEN nor evidence of attendance at a special school or alternative provision in any NPD census; (2) SEN support in mainstream school, where 'School Action', 'School Action Plus' or 'SEN support' (following the 2014 legislative changes) was recorded in at least one census and there was no record of an EHCP nor a record of attending a special school or alternative provision in any NPD census; (3) EHCP in mainstream school, where a 'statement of SEN' or 'EHCP' (following the 2014 legislative changes) was recorded in at least one census and there was no record of attending a special school or alternative provision in any NPD census; (4) specialist provision, where there is a record of attendance at a special school or alternative provision in any census.

Covariates

We derived the following variables from each child's HES APC record at birth to compare socio-demographic characteristics of the birth and school cohorts: year of birth; phenotypic sex (female or male); region of residence; five-group index of multiple deprivation, based on the lower super output area of residential address recorded at birth [25]; and racial-ethnic group (six major groups aggregated from 16 categories). We use the term racial-ethnic group purposefully to emphasise that this covariate includes both race- and ethnicity-based identifiers (e.g. "White" and "British", respectively). This paper is descriptive and therefore we do not include adjustment for any covariates in the main analyses.

Analysis

We firstly compared the prevalence of MCAs in our birth cohort with the average prevalence reported across England-based registries between 2004 and 2014. To estimate the proportion of children who survived to ages 5 and 7 years, we used the Kaplan-Meier estimator with 95% confidence interval (CI). Next, we identified the children who appeared in any of

the NPD censuses between year 1 and year 6, to construct the school cohort. We described socio-demographic characteristics (in numbers and percentages) of children in the birth cohort who were alive at school entry compared with those in the school cohort, to assess the extent to which the final cohort was representative of the birth cohort. We then calculated the prevalence of recorded SEN provision at any census between years 1 and 6, by MCA status. We replicated these analyses restricting the definition of recorded SEN to school years 1 to 2 to assess the stability of the main results, given that children born after 31 August 2008 did not have complete follow up to year 6 (Supplementary Figure 1, Supplementary Appendix 1).

To examine whether the prevalence of SEN provision differed for children with and without MCAs before and after the 2014 reforms, we defined SEN provision using NPD censuses in year 1 only because children born in 2012/13 did not have follow-up beyond year 1. We firstly plotted a time series of the prevalence of SEN provision recorded in year 1 for children with and without an MCA. We then divided the school cohort into two (five-year) periods: births from 1 September 2003 to 31 August 2008 and births from 1 September 2008 to 31 August 2013. Those born in the earlier period finished year 1 before 2014/15, when legislative changes were implemented. We described the number and proportion of children with recorded SEN provision in any NPD census in year 1 over these two periods, by MCA. We calculated the absolute difference (in percentage points) in the prevalence of recorded SEN provision between the two periods, by MCA status. We report 99% confidence intervals to conservatively estimate standard errors, acknowledging the multiplicity of related calculations.

All analyses were carried out in Stata v17 within the ONS secure research service. MP, KML, BDS and RG had access to the raw data in this study, and analyses was carried out between 02/12/2022 and 11/09/2023. None of the authors had access to information that could identify individual participants. The code used to create key study variables and implement analyses is available at https://github.com/UCL-CHIG/HOPE_study_MCA_SEN/. The analyses for this study followed directly from the aims, that is to study survival patterns and prevalence of SEN provision by MCA status (no protocol is available). We added time series figures, characteristics of linked/unlinked children and a comparison to prevalence of MCAs reported in registry data in response to peer review comments. This study is reported as per the REporting of studies Conducted using Observational Routinely-collected Data (RECORD) guideline (S1 Checklist).

Results

Birth cohort description and survival rates

After excluding multiple births and non-England residents, our birth cohort comprised 6,180,400 singleton live births in NHS-funded hospitals in England from 1st September 2003 to 31st August 2013 (Figure 1). Of these, 3.5% (219,249) had evidence of at least one MCA identified in hospital or death records in the first year of life. The most common anomalies were those of the cardiac (53,741; 0.9% of the birth cohort), limb (34,366; 0.6%), and urinary systems (29,158; 0.5%; Table 1). The estimated prevalence of MCAs in this study was about twice the average prevalence reported by regional registries for available MCAs over the same period, with some exceptions (Supplementary Table 3, Supplementary Appendix 1). Similar prevalence rates were reported for the orofacial and abdominal wall MCA subgroups and for severe cardiac anomalies, congenital diaphragmatic hernia, limb reduction defect and Down syndrome. The prevalence of children with an identified MCA increased from 3.3% amongst births in 2003/04 to 3.8% in 2012/13, with the largest system specific sub-group increase among children with any cardiac anomaly (from 0.7% to 1.1%; Supplementary Figure 2, Supplementary Appendix 1).

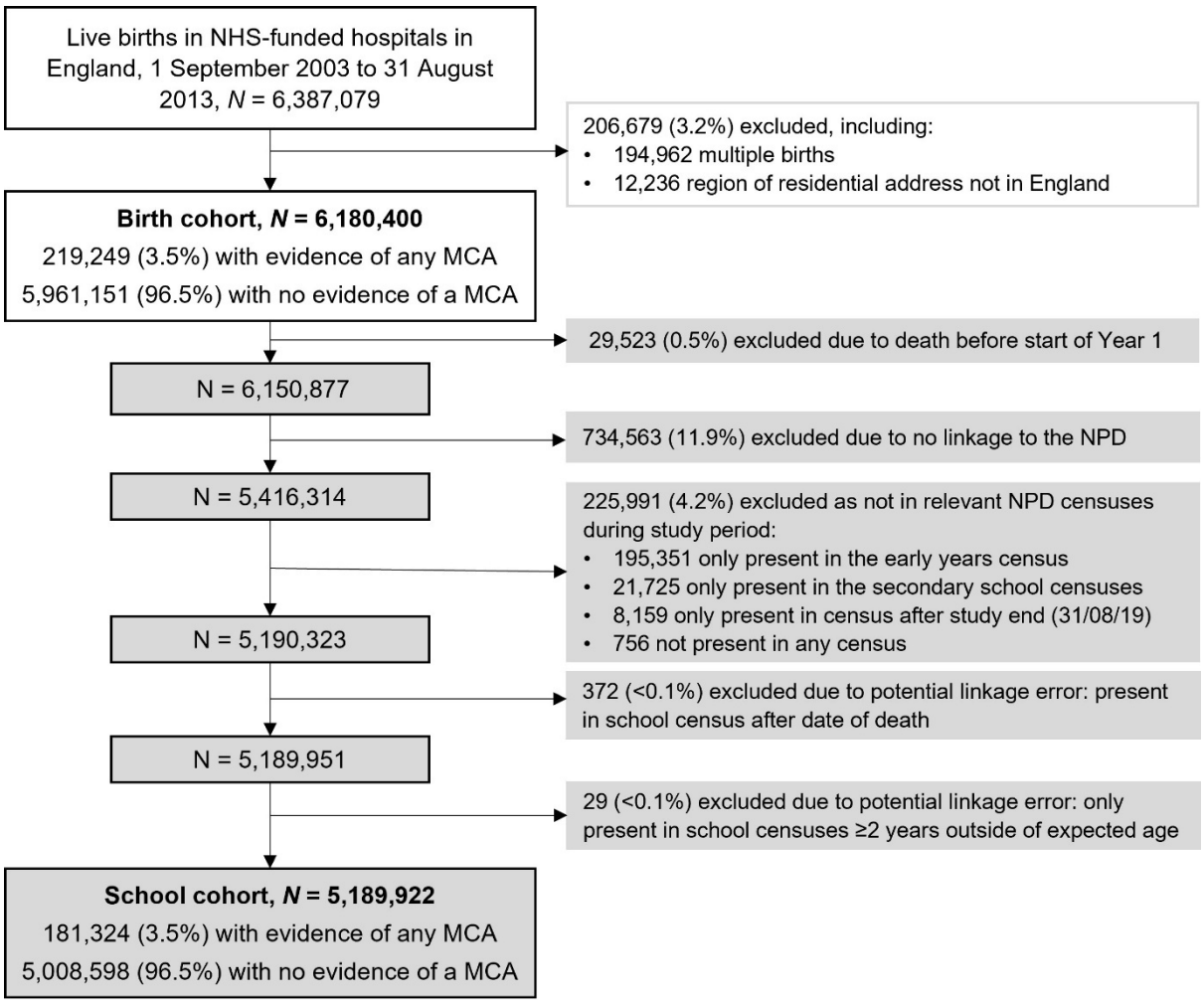
For children without an MCA, survival to age 5 and 7 years was stable at 99.7% (95% CI, 99.7-99.7 for each age). Children with at least one MCA had survival rates of 95.1% (95% CI, 95.0-95.2) at 5 years and 95.0% (95% CI, 94.9-95.1) at 7 years. Among the 12 system-specific subgroups, average 5-year survival was under 90% for those with any nervous system, cardiac, respiratory, digestive or chromosomal anomaly. Children with isolated anomalies had higher survival rates than on average for children in their anomaly subgroup. Conversely, survival rates were lower for the isolated chromosomal anomaly group, compared with specific conditions (Down syndrome and Turner syndrome). The condition with the lowest survival rate was congenital diaphragmatic hernia (66.9% to age 5 years, 95% CI 64.4-69.2).

School cohort description and SEN provision

Of the 6,180,400 children in the birth cohort, 16.0% (990,478) were excluded from the school cohort, including: 29,253 children who died before the expected age of entry into year 1; 734,563 children without a linked NPD record; 225,991 children (with a linked NPD record) who were not present in any NPD census during years 1 to 6 within the study period; and 372 present in NPD after date of death (evidence of possible linkage error; Figure 1). The final school cohort consisted of 5,189,922 children, including 3.5% (181,324) with evidence of at least one MCA. Children were more likely to be included in the school cohort if they were born in later study years, lived outside London and were from a White racial-ethnic group (Supplementary Table 4, Supplementary Appendix 1). The proportion of children in the birth cohort who were included in the school cohort was 84.0% among children without an MCA compared with 82.7% of children with any MCA (rising to 84.3% and 86.9%, respectively, when accounting for deaths before age 5 years; Supplementary Table 5, Supplementary Appendix 1). Of all studied MCA sub-groups and conditions only one (indeterminate sex) had inclusion rates under 84% after accounting for deaths before age 5 years.

Of the 5,008,598 children in the school cohort without an MCA, 25.7% (1,285,572) had any recorded SEN provision during years 1 to 6 (22.5% with SEN support in mainstream school, 1.8% with an EHCP in mainstream school and 1.4% in specialist provision; Table 2; Figure 2). In comparison, of the 181,324 children with at least one MCA, 41.6% (75,381) had any recorded SEN (27.6% with SEN support in mainstream school, 6.6% with an EHCP in mainstream school and 7.3% in specialist provision). Within system-specific subgroups, any recorded SEN provision was highest among children with any chromosomal (93.1%; 6,712/7,208), nervous system

Figure 1: Derivation of the birth and school cohorts



MCA = major congenital anomaly, NPD = national pupil database.

(73.3%; 6,358/8,676) or eye anomalies (66.0%; 2373/3,599). 8.7% (624/7,208) of children with any chromosomal anomaly had SEN support in mainstream school recorded, compared with 25.8% to 39.1% among other system-specific anomaly subgroups.

EHCPs in mainstream school were most frequently recorded among children with any chromosomal (35.3%; 3,548/7,208), nervous system (16.6%; 1,440/8,676) and eye anomalies (15.4%; 555/3,599). Those with abdominal wall, urinary system, genital or limb anomalies were the least likely to have such provision (<5%). Broadly, the proportion of each MCA group with recorded EHCPs in mainstream school was similar to the proportion attending specialist provision, except for children with nervous system and chromosomal anomalies. Among children with microcephaly, 57.4% (952/1,659) attended specialist provision compared with 13.0% (215/1,659) with a recorded EHCP in a mainstream setting. Within the system-specific subgroups, children with isolated anomalies had lower proportions of recorded SEN than on average for the subgroup. The overall pattern of results is comparable when restricting the results to key stage 1 only (years 1 and/or 2), with a slight decrease in the prevalence of recorded SEN across all MCA subgroups and conditions (Supplementary Table 6 and Supplementary Figure 3, Supplementary Appendix 1).

Differences in recorded SEN provision, by reform period

Overall, the prevalence of children with SEN provision recorded in year 1 decreased from 21.1% in 2009/10 to 15.0% in 2018/19 (Figure 3), with the same decreasing pattern observed for children with and without an MCA. As shown in Supplementary Figure 4 (Supplementary Appendix 1), when split by category of recorded SEN provision, this decrease is only observable for SEN support (the largest group; 88.3% of all recorded SEN provision in year 1).

The proportion of children with any recorded SEN provision in year 1 decreased by 4.2% (99% CI −4.3,−4.2) after the 2014 SEN reforms for children without an MCA, compared with 4.8% (99% CI −5.4,−4.3) for those with any MCA (Figure 4; Supplementary Table 7, Supplementary Appendix 1). Of the MCA subgroups, the proportion of children with any recorded SEN provision decreased most for abdominal wall (−8.2%, 99% CI −12.2,−4.1), respiratory system (−6.3%, 99% CI −11.0,−1.6) and cardiac (−6.1%, 99% CI −7.3,−4.8) anomalies. Of the categories of SEN provision, these decreases are only observed in SEN support in mainstream school for children without an MCA (−4.6%, 99% CI −4.6,−4.5) (Figure 5; Supplementary Table 7, Supplementary Appendix 1). Children with any MCA had a similar decrease in SEN

Table 1: Birth cohort description and survival rates, by MCA, system-specific subgroups and selected conditions[a]

	Birth cohort	Up to age 5 years		Up to age 7 years	
	N (%)	Deaths	5-year survival % (95% CI)	Deaths	7-year survival % (95% CI)
No MCA	5961151 (96.5)	18565	99.7 (99.7, 99.7)	19265	99.7 (99.7, 99.7)
Any MCA	219249 (3.5)	10691	95.1 (95.0, 95.2)	10953	95.0 (94.9, 95.1)
Nervous system anomalies					
Any nervous system anomaly	11971 (0.19)	2094	82.5 (81.8, 83.2)	2205	81.6 (80.8, 82.2)
Isolated nervous system	7322 (0.12)	870	88.1 (87.4, 88.8)	927	87.3 (86.5, 88.1)
Microcephaly	2349 (0.04)	478	79.7 (78.0, 81.2)	525	77.6 (75.8, 79.2)
Hydrocephaly	2574 (0.04)	463	82.0 (80.5, 83.4)	486	81.1 (79.5, 82.6)
Spina Bifida	1343 (0.02)	174	87.0 (85.1, 88.7)	[b]	[b]
Eye anomalies					
Any eye anomaly	4404 (0.07)	376	91.5 (90.6, 92.3)	402	90.9 (90.0, 91.7)
Isolated eye anomaly	2849 (0.05)	23	99.2 (98.8, 99.5)	[b]	[b]
Anophthalmos/ Microphthalmos	672 (0.01)	119	82.3 (79.2, 85.0)	[b]	[b]
Congenital cataract	1271 (0.02)	88	93.1 (91.5, 94.3)	[b]	[b]
Congenital Glaucoma	448 (0.01)	20	95.5 (93.2, 97.1)	[b]	[b]
Ear, face, and neck anomalies					
Any ear, face, and neck	1663 (0.03)	91	94.5 (93.3, 95.5)	[b]	[b]
Isolated ear, face, and neck	993 (0.02)	[b]	[b]	[b]	[b]
Cardiac anomalies					
Any cardiac anomaly	53741 (0.87)	6050	88.7 (88.5, 89.0)	6161	88.5 (88.3, 88.8)
Isolated cardiac anomaly	37080 (0.60)	3015	91.9 (91.6, 92.1)	3057	91.8 (91.5, 92.0)
Severe cardiac	14260 (0.23)	2584	81.9 (81.2, 82.5)	2626	81.6 (80.9, 82.2)
Respiratory anomalies					
Any respiratory anomaly	3941 (0.06)	493	87.5 (86.4, 88.5)	[b]	[b]
Isolated respiratory anomaly	2121 (0.03)	83	96.1 (95.2, 96.8)	[b]	[b]
Choanal atresia	804 (0.01)	82	89.8 (87.5, 91.7)	[b]	[b]
Orofacial anomalies					
Any orofacial anomaly	9611 (0.16)	480	95.0 (94.6, 95.4)	492	94.9 (94.4, 95.3)
Isolated orofacial anomaly	6503 (0.11)	29	99.6 (99.4, 99.7)	[b]	[b]
Cleft lip	2589 (0.04)	51	98.0 (97.4, 98.5)	[b]	[b]
Cleft palate	4891 (0.08)	300	93.9 (93.2, 94.5)	310	93.7 (92.9, 94.3)
Cleft lip and palate	3656 (0.06)	183	95.0 (94.2, 95.7)	[b]	[b]
Digestive system anomalies					
Any digestive system anomaly	16098 (0.26)	1731	89.2 (88.8, 89.7)	1763	89.0 (88.5, 89.5)
Isolated digestive system anomaly	9343 (0.15)	518	94.5 (94.0, 94.9)	[b]	[b]
Oesophageal atresia	1664 (0.03)	200	88.0 (86.3, 89.5)	[b]	[b]
Small intestine atresia	1934 (0.03)	167	91.4 (90.0, 92.5)	[b]	[b]
Hirschsprung's disease	1508 (0.02)	60	96.0 (94.9, 96.9)	[b]	[b]
Biliary atresia	440 (0.01)	45	89.8 (86.5, 92.3)	[b]	[b]
Anorectal malformation	1713 (0.03)	173	89.9 (88.4, 91.2)	[b]	[b]
Congenital diaphragmatic hernia	1524 (0.02)	505	66.9 (64.4, 69.2)	[b]	[b]
Abdominal wall anomalies					
Any abdominal wall anomaly	4110 (0.07)	292	92.9 (92.1, 93.6)	[b]	[b]
Isolated abdominal wall anomaly	2854 (0.05)	97	96.6 (95.9, 97.2)	[b]	[b]
Omphalocele	1286 (0.02)	155	87.9 (86.0, 89.6)	[b]	[b]
Gastroschisis	2539 (0.04)	139	94.5 (93.6, 95.3)	[b]	[b]
Urinary system anomalies					
Any urinary system anomaly	29158 (0.47)	1200	95.9 (95.7, 96.1)	1228	95.8 (95.5, 96.0)
Isolated urinary system anomaly	24703 (0.40)	469	98.1 (97.9, 98.3)	[b]	[b]
Bladder exstrophy	704 (0.01)	[b]	[b]	[b]	[b]
Genital anomalies					
Any genital anomaly	25914 (0.42)	488	98.1 (97.9, 98.3)	[b]	[b]

Continued

Table 1: Continued

	Birth cohort	Up to age 5 years		Up to age 7 years	
	N (%)	Deaths	5-year survival % (95% CI)	Deaths	7-year survival % (95% CI)
Isolated genital anomaly	22252 (0.36)	100	99.6 (99.5, 99.6)	[b]	[b]
Hypospadias	17727 (0.29)	235	98.7 (98.5, 98.8)	[b]	[b]
Indeterminate sex	1044 (0.02)	119	88.6 (86.5, 90.4)	[b]	[b]
Limb anomalies					
Any limb anomaly	34366 (0.56)	985	97.1 (97.0, 97.3)	1010	97.1 (96.9, 97.2)
Isolated limb anomaly	28911 (0.47)	159	99.5 (99.4, 99.5)	[b]	[b]
Limb reduction defect	2180 (0.04)	142	93.5 (92.4, 94.4)	[b]	[b]
Chromosomal anomalies					
Any chromosomal anomaly	9734 (0.16)	1612	83.4 (82.7, 84.2)	1646	83.1 (82.3, 83.8)
Isolated chromosomal anomaly	3330 (0.05)	347	89.6 (88.5, 90.6)	[b]	[b]
Down syndrome	6260 (0.10)	492	92.1 (91.4, 92.8)	503	92.0 (91.3, 92.6)
Turner syndrome	381 (0.01)	32	91.6 (88.3, 94.0)	[b]	[b]

[a]any groups contain children with and without additional anomalies in other system-specific subgroups (% survival is group average), isolated groups do not contain children with anomalies in other system-specific subgroups; [b]small numbers (<8) suppressed to prevent disclosure of identities (including deductive disclosure when subtracting deaths by age 7 years from deaths by age 5 years). CI = confidence interval, MCA = major congenital anomaly.

support (−4.4%, 99% CI −4.9,−3.9), together with slight decrease (−0.4%, 99% CI −0.7,−0.1) in the prevalence of EHCPs in mainstream school.

Discussion

We found 5-year survival rates of 95.1% for children with MCAs born between 1 September 2003 and 31 August 2013, compared with 99.7% of children without these conditions. 41.5% of children with any MCA attending state school had any recorded SEN provision compared with 25.6% of children without an MCA. Differences in any recorded SEN provision between those with and without MCAs were greater for EHCPs and specialist school provision. Among children with MCAs, there was substantial variation in recorded SEN provision, with those with chromosomal, nervous system and eye anomalies having the highest prevalence of any recorded SEN provision. Nearly 1 in 20 fewer children had recorded SEN provision in year 1 after the 2014 reforms, with the decline mainly observed in support at the lower level in mainstream schools.

Interpretation and comparison with other studies

Patterns of survival for congenital anomalies in our study are similar to pooled estimates presented in a 2020 meta-analysis, highlighting increasing survival for children with many of these conditions over time [6]. Estimates of SEN provision for children with MCAs are similar to those provided from registry data linked to national administrative educational records in four areas of England (41.6% in our study compared with 44.0% from the EUROlinkCAT report) [10]. The decline in the proportion of children with SEN support (formerly 'School Action'/'School Action Plus') following 2014 government reforms fits with patterns reported elsewhere [26]. We add to this evidence by quantifying this decrease (approximately 1 in 20 fewer children receiving SEN provision) and showing that changes affected children with and without MCAs who had additional learning needs at the lower level of provision.

As illustrated in our time series plot, the decrease in SEN support had already begun several years before formal introduction of reforms. Sweeping public sector austerity policies in the early 2010's, including cuts to school budgets, alongside criticism of the current system of SEN identification likely contributed to this decrease [27]. Prior to 2010, SEN provision had been rising [20], culminating in a report by the Office for Standards in Education, Children's Services and Skills, which stated that there was "over-identification" of children at the lower level of SEN provision in lieu of "better teaching" [21]. Decreases in recorded SEN provision over this period may therefore reflect changing criteria of the children who should receive SEN provision and at what age. This may explain the trend differences across MCA groupings, although one should not over interpret these differences given the relatively small numbers involved. The intended impact of the 2014 SEN reform on better teaching contrasts with evidence on the experiences of children, their families and teachers, which indicate that changes in SEN provision reflects rising unmet need [28–30]. The possible detrimental impact of these changes is further supported by reports published in 2019 by the House of Commons Education Committee and the National Audit Office that present a picture of a fragmented and increasingly unsustainable SEN system [28, 29].

Strengths and limitations

A strength of our study is the cohort size and comprehensive national coverage. We used a cohort of over six million children (5,189,922 in the SEN provision analysis) to describe less commonly investigated MCA subgroups and conditions. Longitudinal records over a 10-year study period meant that

Table 2: Number and proportion of children in the school cohort with SEN provision recorded at least once during years 1 to 6, by category of recorded SEN, MCA, system-specific subgroup[a] and selected conditions

	School cohort N	Any recorded SEN provision N (%)	SEN support in mainstream school N (%)	EHCP in mainstream school N (%)	Specialist provision N (%)
No MCA	5,008,598	1285572 (25.7)	1125783 (22.5)	88108 (1.8)	71681 (1.4)
Any MCA	181,324	75381 (41.6)	50133 (27.6)	11954 (6.6)	13294 (7.3)
Nervous system anomalies					
Any nervous system anomaly	8,676	6363 (73.3)	2251 (25.9)	1440 (16.6)	2672 (30.8)
Isolated nervous system anomaly	5,656	3868 (68.4)	1560 (27.6)	828 (14.6)	1480 (26.2)
Microcephaly	1,659	1441 (86.9)	274 (16.5)	215 (13.0)	952 (57.4)
Hydrocephaly	1,883	1526 (81.0)	477 (25.3)	392 (20.8)	657 (34.9)
Spina Bifida	1,022	803 (78.6)	345 (33.8)	296 (29.0)	162 (15.9)
Eye anomalies					
Any eye anomaly	3,599	2374 (66.0)	1152 (32.0)	555 (15.4)	667 (18.5)
Isolated eye anomaly	2,510	1446 (57.6)	922 (36.7)	316 (12.6)	208 (8.3)
Anophthalmos/Microphthalmos	506	432 (85.4)	158 (31.2)	111 (21.9)	163 (32.2)
Congenital cataract	1,069	745 (69.7)	408 (38.2)	151 (14.1)	186 (17.4)
Congenital Glaucoma	390	289 (74.1)	148 (37.9)	73 (18.7)	68 (17.4)
Ear, face, and neck anomalies					
Any ear, face, and neck anomaly	1,336	698 (52.2)	416 (31.1)	115 (8.6)	167 (12.5)
Isolated ear, face, and neck anomaly	829	332 (40.0)	265 (32.0)	36 (4.3)	31 (3.7)
Cardiac anomalies					
Any cardiac anomaly	41857	21995 (52.5)	12271 (29.3)	4536 (10.8)	5188 (12.4)
Isolated cardiac anomaly	29745	12853 (43.2)	9290 (31.2)	1878 (6.3)	1685 (5.7)
Severe cardiac	10337	5913 (57.2)	3074 (29.7)	1292 (12.5)	1547 (15.0)
Respiratory anomalies					
Any respiratory anomaly	2997	1594 (53.2)	884 (29.5)	341 (11.4)	369 (12.3)
Isolated respiratory anomaly	1755	689 (39.3)	514 (29.3)	94 (5.4)	81 (4.6)
Choanal atresia	647	391 (60.4)	160 (24.7)	98 (15.1)	133 (20.6)
Orofacial anomalies					
Any orofacial anomaly	7993	4452 (55.7)	3123 (39.1)	647 (8.1)	682 (8.5)
Isolated orofacial anomaly	5653	2771 (49.0)	2256 (39.9)	290 (5.1)	225 (4.0)
Cleft lip	2242	921 (41.1)	733 (32.7)	90 (4.0)	98 (4.4)
Cleft palate	4030	2474 (61.4)	1576 (39.1)	423 (10.5)	475 (11.8)
Cleft lip and palate	3051	1862 (61.0)	1401 (45.9)	235 (7.7)	226 (7.4)
Digestive anomalies					
Any digestive anomaly	12544	5950 (47.4)	3690 (29.4)	1081 (8.6)	1179 (9.4)
Isolated digestive anomaly	7657	2862 (37.4)	2207 (28.8)	349 (4.6)	306 (4.0)
Oesophageal atresia	1297	704 (54.3)	442 (34.1)	158 (12.2)	104 (8.0)
Small intestine atresia	1545	790 (51.1)	382 (24.7)	192 (12.4)	216 (14.0)
Hirschsprung's disease	1298	659 (50.8)	392 (30.2)	117 (9.0)	150 (11.6)
Biliary atresia	346	178 (51.4)	116 (33.5)	26 (7.5)	36 (10.4)
Anorectal malformation	896	376 (42.0)	258 (28.8)	56 (6.3)	62 (6.9)
Congenital diaphragmatic hernia	1363	783 (57.4)	477 (35.0)	167 (12.3)	139 (10.2)
Abdominal wall anomalies					
Any abdominal wall anomaly	3352	1374 (41.0)	1082 (32.3)	158 (4.7)	134 (4.0)
Isolated abdominal wall anomaly	2398	879 (36.7)	752 (31.4)	63 (2.6)	64 (2.7)
Omphalocele	968	434 (44.8)	302 (31.2)	76 (7.9)	56 (5.8)
Gastroschisis	2141	834 (39.0)	713 (33.3)	65 (3.0)	56 (2.6)
Urinary system anomalies					
Any urinary system anomaly	24203	8934 (36.9)	6674 (27.6)	1121 (4.6)	1139 (4.7)
Isolated urinary system anomaly	20949	6884 (32.9)	5681 (27.1)	653 (3.1)	550 (2.6)
Bladder exstrophy	596	255 (42.8)	181 (30.4)	40 (6.7)	34 (5.7)

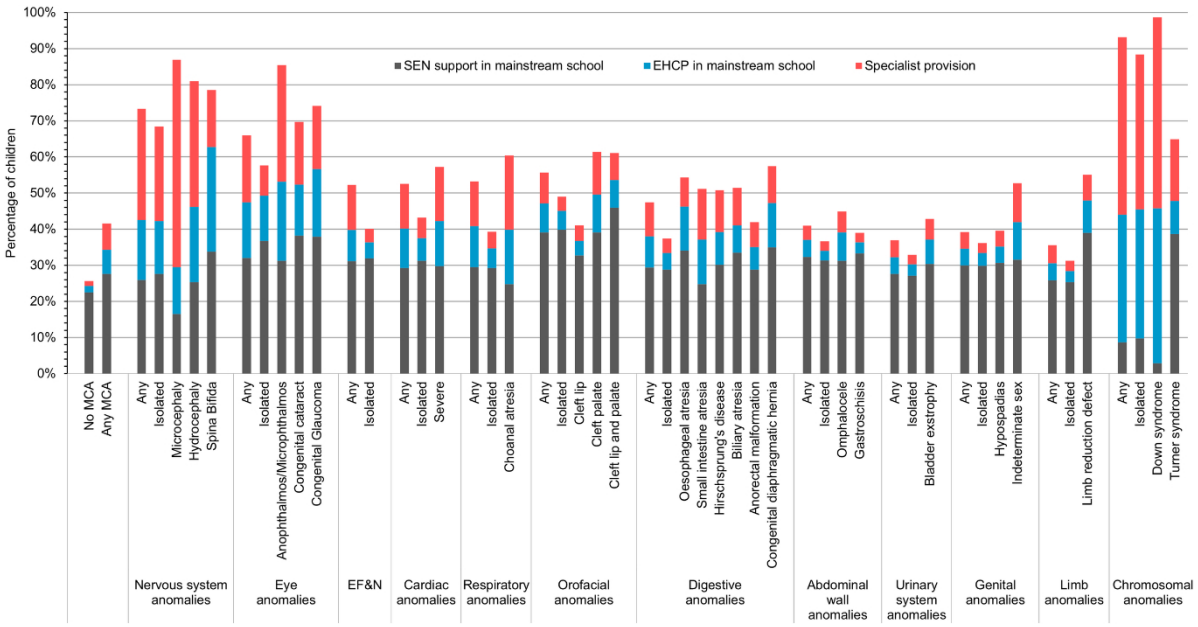
Continued

Table 2: Continued

	School cohort N	Any recorded SEN provision N (%)	SEN support in mainstream school N (%)	EHCP in mainstream school N (%)	Specialist provision N (%)
Genital anomalies					
Any genital anomaly	21923	8600 (39.2)	6575 (30.0)	1015 (4.6)	1010 (4.6)
Isolated genital anomaly	19093	6910 (36.2)	5703 (29.9)	665 (3.5)	542 (2.8)
Hypospadias	15243	6034 (39.6)	4669 (30.6)	696 (4.6)	669 (4.4)
Indeterminate sex	637	336 (52.7)	201 (31.6)	66 (10.4)	69 (10.8)
Limb anomalies					
Any limb anomaly	28951	10291 (35.5)	7468 (25.8)	1368 (4.7)	1455 (5.0)
Isolated limb anomaly	24911	7772 (31.2)	6316 (25.4)	751 (3.0)	705 (2.8)
Limb reduction defect	1783	982 (55.1)	695 (39.0)	159 (8.9)	128 (7.2)
Chromosomal anomalies					
Any chromosomal anomaly	7208	6712 (93.1)	624 (8.7)	2548 (35.3)	3540 (49.1)
Isolated chromosomal anomaly	2626	2321 (88.4)	257 (9.8)	936 (35.6)	1128 (43.0)
Down syndrome	5157	5087 (98.6)	147 (2.9)	2214 (42.9)	2726 (52.9)
Turner syndrome	305	198 (64.9)	118 (38.7)	28 (9.2)	52 (17.0)

[a]any groups contain children with and without additional anomalies in other system-specific subgroups, isolated groups do not contain children with anomalies in other system-specific subgroups. EHCP = education, health and care plan, MCA = major congenital anomaly, SEN = special educational needs.

Figure 2: Percentage of children in the school cohort with different categories of SEN provision recorded at least once during years 1 to 6, by MCA, system-specific subgroup and selected conditions



EF&N = ear, face, and neck anomalies, EHCP = education, health and care plan, MCA = major congenital anomaly, SEN = special educational needs.

we could examine SEN provision before and after a major policy change. A major limitation relates to the recording of SEN provision not being based on clear criteria for service delivery. Firstly, a record of SEN provision in the educational data does not necessarily indicate that SEN provision was received, and there is no information about the precise elements of support received (hence our label of 'recorded' SEN provision). Secondly, in practice, SEN provision varies by factors beyond a child's learning needs, as highlighted by the time-varying patterns presenting in our study. Other research has shown SEN provision to vary by school, geographical location, child's ethnicity and previous contact with social services, amongst other factors [31]. A further limitation is that diagnostic codes of MCAs are not necessarily indicate additional learning needs, as they do not capture severity or functional impairment. For example, the diagnostic codes used to define conditions in the eye anomaly group (anophthalmos/microphthalmos, congenital

Figure 3: Prevalence of SEN provision recorded in year 1, by academic year and MCA status

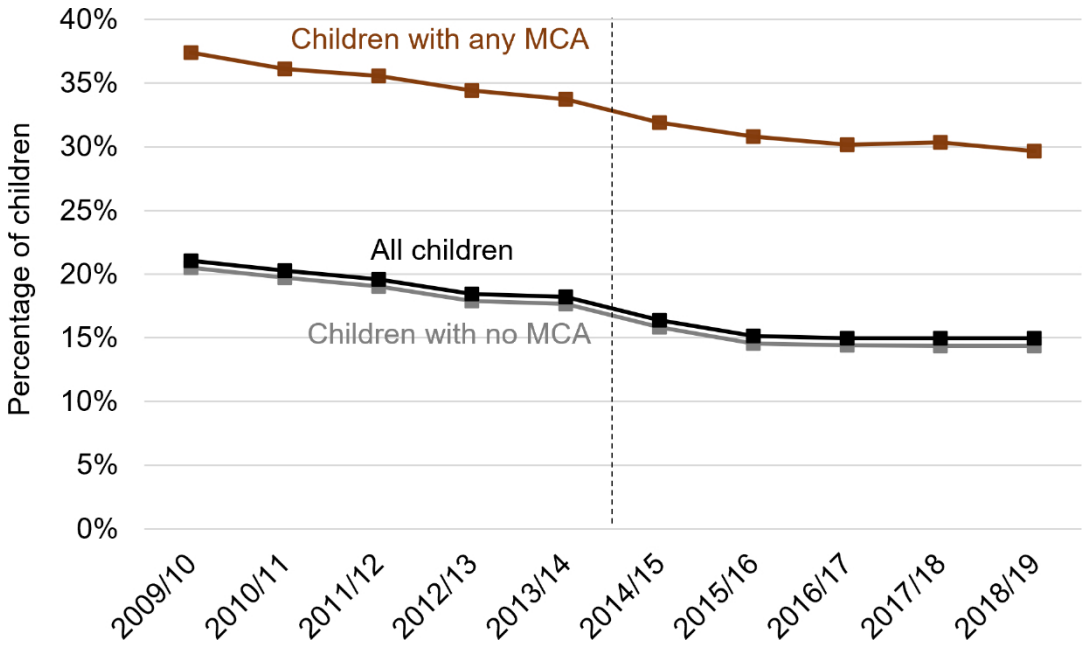
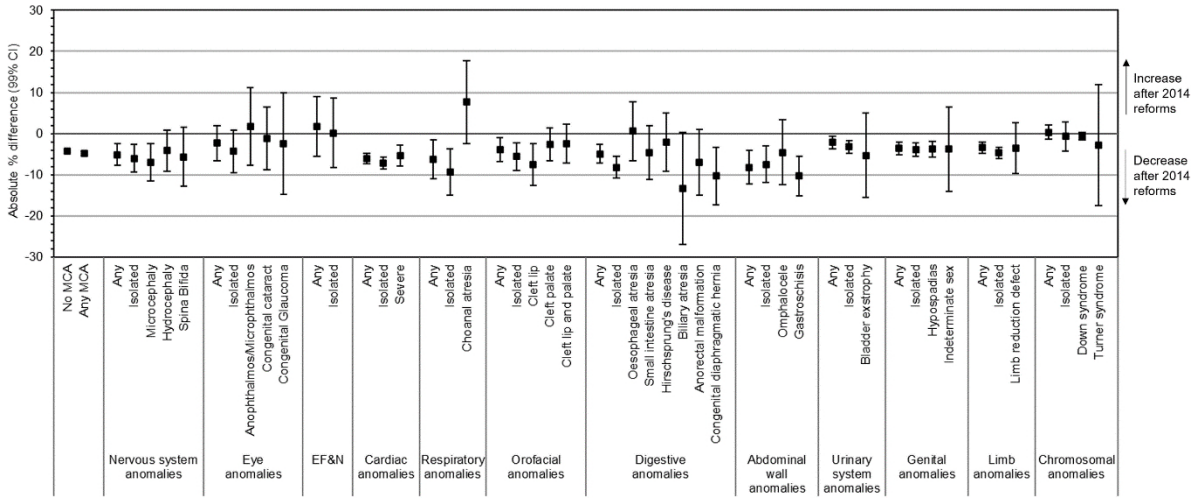


Figure 4: Absolute % difference (99% CI) between the prevalence of any recorded SEN provision amongst children in year 1 between 2014/15 and 2018/19 (after the 2014 SEN reforms) compared with children in year 1 between 2009/10 and 2013/14 (before the 2014 SEN reforms), by MCA, system-specific subgroup and selected conditions

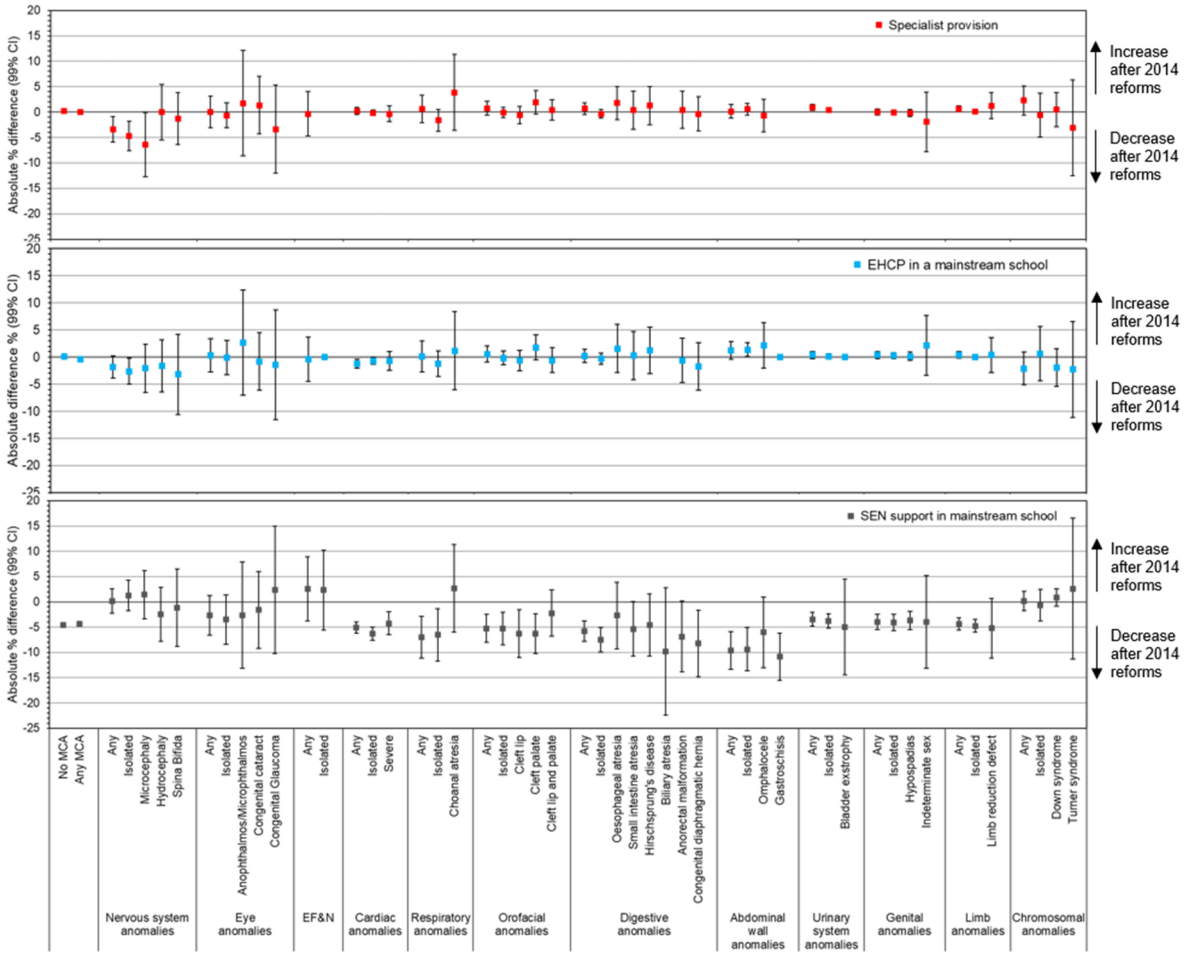


EF&N = ear, face and neck anomalies, MCA = major congenital anomaly, SEN = special educational needs.

cataract and congenital glaucoma) do not indicate whether a child has unilateral or bilateral eye disease, with the former unlikely to require SEN provision in isolation [32]. We are undertaking further work, with comprehensive clinical input, to define groups of children who are most likely to require SEN provision.

Using hospital admission and mortality records during the first year of life, we identify MCAs in 354.7 per 10,000 live singleton births, compared with 185.5 per 10,000 live and still births in registry data over the same period [3]. Differences in completeness of reporting, coverage (England-wide compared with one third of the population) and ages at data collection (infancy compared with predominately birth or newborn examinations) over the time period studied may partially explain our higher rates [4]. However, our study also misclassifies some children with minor anomalies due to the granularity of ICD-10 codes required to exclude some conditions. We also find an increase in the prevalence of MCAs in our study over time, which may indicate both increased detection and depth of coding in hospital records over time, particularly for milder congenital anomalies [7, 22]. Arguably, changes to the population of children with MCAs captured over time (potentially with less need for education support, on average), may have led to an overestimation in the difference of SEN provision after SEN reforms. However, the same pattern of results are observed for children with orofacial and abdominal anomalies—MCA subgroups with comparable rates to registry data and stable prevalence rates over the study period—, which give us confidence in the interpretation of our results. That is, that children with lower

Figure 5: Absolute % difference (99% CI) between the prevalence of recorded SEN amongst children in year 1 between 2014/15 and 2018/19 (after the 2014 SEN reforms) compared with children in year 1 between 2009/10 and 2013/14 (before the 2014 SEN reforms), by category of recorded SEN provision, MCA, system-specific subgroup and selected conditions



EF&N = ear, face and neck anomalies, EHCP = education, health and care plan, MCA = major congenital anomaly, SEN = special educational needs.

level SEN needs (with and without MCAs) were less likely to have SEN provision recorded in the latter half of the 2010's. Further studies, including validation via linkage to the national congenital anomaly and rare disease registration service (national coverage available since 2018 [33]) and primary care records, are required to improve case ascertainment and confidence in, or adjustment for, the precision of results for a wider group of congenital anomalies [34].

Overall, 84% of children in the birth cohort and alive at age 5 years were included in the school cohort; meaning the results may not be representative of all children born in England [17]. Approximately half of the children excluded would be expected to: attend non-state funded, or private, school (7% of the population) [17]; be home schooled (estimated to be 0.6% of the population) [17]; or have emigrated out of England before starting school (no reliable estimates are available but, based on emigration statistics for all ages in England, rates are unlikely to exceed 0.5%) [35]. The remaining non-links (~8%) represent missed links or potential duplicates between the health and educational datasets (leading to an inflated denominator). Nonetheless, rates of linkage across MCA groups and children without an MCA are similar, giving us confidence in our comparative results. The rate of inclusion in the school cohort was below 70% for children with indeterminate sex, suggesting that findings for this condition should be interpreted with some caution. We theorise that lower rates of follow-up in this group are the result of linkage error, since the algorithm for linkage between datasets in ECHILD uses child sex and children with indeterminate sex are more likely to have changes to sex/gender assignment over time. Continued improvements to linkage in the ECHILD database over time, will further strengthen the national application of results to children attending state-funded schools in England [17].

Implications

Clinicians and families want precise information about the prognosis of children with MCAs beyond infancy. The work presented here shows how large administrative datasets can be harnessed to provide such information, albeit through a rationed service that is subject to policy change. As the ECHILD database is accessible to government and UK researchers [36], the approach in this study could be developed to use de-identified data routinely for planning and monitoring of SEN provision. For example, together with our ongoing

work to phenotype children in need of SEN provision, the need for early specialised support could be identified in HES by the end of infancy to inform service planning by local authorities. Specialist paediatric services could use such information to prepare parents for the type of support likely to be received. Administrative data offers an important resource for understanding variation and inequities in the provision of SEN services and, if combined with qualitative evidence of areas of good practice, and randomised or quasi randomised methods, to assess SEN practices that have a beneficial effect and for whom. The application of these methods could extend to examining the impact of changes in clinical practices or support (for example, a new drug or therapy) on educational outcomes, such as attainment and school absence.

Future research comparing MCA cohorts across UK countries and internationally will provide evidence on how education and health outcomes in children with MCAs, compared with peers, vary in different jurisdictions with contrasting timing, intensity and pedagogical approaches to SEN provision. Continued work on validating coding for specific MCA groups is important to support this comparative work. SEN provision has been called 'a postcode lottery' in England [31], but the extent to which this represents underlying inequities has been difficult to capture to date. Our approach of defining similar groups of children using health records could be used to explore whether children with the same underlying needs are systematically managed differently in educational settings, depending on the school or local authority or both. Extension of this work to children without MCAs who are in need of SEN provision is also important, given that we find that children with MCAs only account for a minority (5.5%) of all children with any recorded SEN provision and less than two in ten children with EHCPs.

Conclusion

In this study, we described the proportions of children born in England who have MCAs, survive to primary school, and have recorded SEN provision before and after the 2014 SEN system reforms. We found that recorded SEN provision among children with major congenital anomalies was markedly higher than for those without these conditions, but over 50% had no recorded SEN provision. Our findings also suggest that following the government reforms to the SEN system in 2014/15, there was a continued reduction in recorded SEN provision for children with and without MCAs. The proportion of recorded SEN provision varied depending on the type of MCA and whether the MCA was isolated or linked to congenital anomalies affecting multiple body systems. Further validation studies, including linkage to high-quality congenital anomaly registry data and primary care records to account for misclassification, will improve the validity of results obtained from administrative records.

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NIHR GOSH-BRC had no role in the design, conduct, analysis or interpretation of the study and no role in drafting the manuscript. The ECHILD Database uses data from the Department for Education (DfE). The DfE does not accept responsibility for any inferences or conclusions derived by the authors. This work contains statistical data from ONS which is Crown Copyright. The use of the ONS statistical data in this work does not imply the endorsement of the ONS in relation to the interpretation or analysis of the statistical data. This work uses research datasets which may not exactly reproduce National Statistics aggregates. This research contributes to but was not commissioned by the NIHR Policy Research Programme. The views expressed are those of the author(s) and not necessarily those of the NIHR or the Department of Health and Social Care.

Statement of Conflicts of Interest

None declared.

Ethics statement

Existing research ethics approval has been granted for analyses of the ECHILD database for the purposes set out in the HOPE study (20/EE/0180). Permissions to use linked, de-identified data from HES and the NPD were granted by NHS Digital (DARS-NIC-381972-Q5F0V-v0.5) and DfE (DR200604.02B). Patient consent was not required to use the deidentified data in this study.

Data availability statement

The ECHILD database is made available for free for approved research based in the UK, via the ONS Secure Research Service. Enquiries to access the ECHILD database can be made by emailing ich.echild@ucl.ac.uk. Researchers will need to be approved and submit a successful application to the ECHILD Data Access Committee and ONS Research Accreditation Panel to access the data, with strict statistical disclosure controls of all outputs of analyses.

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Abbreviations

APC:	Admitted Patient Care
ECHILD:	Education and Child Health Insights from Linked Data
EHCP:	Education, Health and Care Plans
EUROCAT:	European Network of Population-based Registries for Congenital Malformations
GIAS:	Get Information about Schools
HES:	Hospital Episode Statistics
MCA:	major congenital anomaly
NPD:	National Pupil Database
ONS:	Office for National Statistics
SEN:	special educational needs

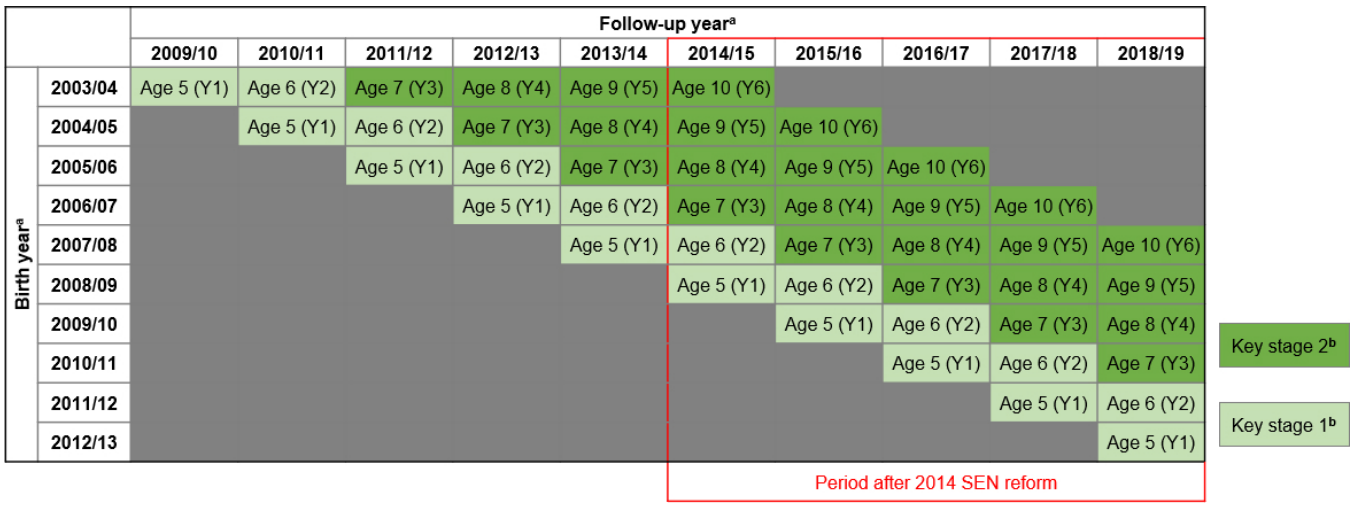
Supplementary Appendix 1

Supplementary Table 1: Pupil-level national pupil database censuses and school types included in this study

Census name (collection frequency)	School type	Examples of school type[a]	Dates of inclusion in study	Included in specialist provision[b]
School census (termly)	Mainstream schools: provide education for students with a wide range of abilities and aptitudes	Maintained, academy and free schools	2009/10–2018/19	No
	Special schools[c]: provide education specifically for students with special educational needs	Maintained and non-maintained special schools, including hospital special schools and academy special schools	2009/10–2018/19	Yes
	Pupil referral units (a type of alternative provision[d]): cater for children who aren't able to attend a mainstream school, often for reasons related to their behaviour	Maintained pupil referral units, alternative provision academies, alternative provision free schools	2013/14–2018/19	Yes
Pupil referral unit census (annual collection)	Pupil referral units (a type of alternative provision[d]): cater for children who aren't able to attend a mainstream school, often for reasons related to their behaviour	Maintained pupil referral units, alternative provision academies, alternative provision free schools	2009/10–2012/13	Yes
Alternative provision census (annual collection)	Alternative provision[d]: provision outside of school due to reasons such as exclusion from school on a permanent or fixed term basis, unable to attend school for medical reasons, or awaiting placement in a maintained school	Schools not maintained by an authority for whom the local authority is paying full tuition fees; including some independent schools, hospitals and non-maintained special schools	2009/10–2018/19	Yes

[a]School descriptors: maintained = local authority run schools (also known as community schools), academies and free schools = not-for-profit schools that are independent from the local authority; [b]Specialist provision is a category of recorded SEN provision in this study, encompassing establishments that provide an alternative educational environment for pupils who, for a variety of reasons, cannot attend mainstream school; [c]Special schools are tailored towards children with healthcare needs (including palliative care) and disabilities. Almost all children attending special school have an EHCP. Provision at special school differs from provision in a mainstream school due to availability of medical staff, teaching assistants, facilities (for example, padded classrooms, hoists and an on-site aqua therapy room), class sizes as well as alternative assessments and qualifications. In this study, 0.72% of children attended special school in Year 1 (rising to 1.34% of children in Year 6). [d]Alternative provision (including pupil referral units) provides an educational and social environment for pupils who for a variety of reasons cannot attend mainstream school, for example behavioural challenges in a mainstream setting that could disrupt other students. That vast majority of pupils at alternative provision have either a record of SEN support or an EHCP. In this study, 0.25% of children attended alternative provision in Year 6. In this study, 0.07% of children attended alternative provision in Year 1 (rising to 0.34% of children in Year 6).

Supplementary Figure 1: Expected age at entry into primary school years 1 to 6, by birth year and follow-up year



SEN = special educational needs, Y = year; [a]defined according to the academic calendar, i.e. from 1 September to 31 August; [b]key stages represent blocks of set subjects and standards (the 'national curriculum') followed by, and the basis of examinations for, most pupils in state-funded schools in England and Wales.

Supplementary Table 2: ICD-10 codes used to define major congenital anomalies[a], by system-specific subgroup and selected conditions

Subgroup Condition	Definition (ICD-10 codes)[b]	
	Include	**Exclude**
Nervous system anomalies		
Any nervous system anomaly	Q00-07	Q0461[c], Q0780[c], Q0782[c]
Microcephaly	Q02	
Hydrocephaly	Q03	
Spina Bifida	Q05	
Eye anomalies		
Any eye anomaly	Q10-15	Q101-103, Q105, Q135,
Anophthalmos/Microphthalmos	Q110-112	
Congenital cataract	Q120	
Congenital glaucoma	Q150	
Ear, face, and neck anomalies		
Any ear, face, and neck anomaly	Q16-18	Q170-175, Q179, Q180-182, Q184-187, Q1880[c], Q189
Cardiac anomalies		
Any cardiac anomaly	Q20-26	Q2111[c], Q246, Q2541[c], Q250 if gestational age <37 weeks, Q256 if gestational age <37 weeks, Q261
Severe cardiac anomaly	Q200-201, Q203-204, Q212-213, Q220, Q224-226, Q230, Q232-234, Q251-252, Q262	
Respiratory anomalies		
Any respiratory anomaly	Q300, Q32-34	Q320, Q322, Q3300[c], Q331, Q336
Choanal atresia	Q300	
Orofacial anomalies		
Any orofacial anomaly	Q35-37	Q357
Cleft lip	Q36	
Cleft palate	Q350-356, Q358-359	
Cleft lip and palate	Q37	
Digestive anomalies		
Any digestive system anomaly	Q38-Q45, Q790	Q381-382, Q3850[c], Q400-401, Q4021[c], Q430, Q4320[c], Q4381[c], Q4382[c], Q444, Q4583[c]
Oesophageal atresia	Q390, Q391	
Small intestine atresia	Q410-418	
Hirschsprung's disease	Q431	
Biliary atresia	Q442	
Anorectal malformation	As defined in Ford *et al.* (2022) [1]	
Congenital diaphragmatic hernia	As defined in Peppa *et al.* (2022) [2]	
Abdominal wall anomaly		
Any abdominal anomaly	Q792, Q793, Q795	
Omphalocele	Q792	
Gastroschisis	Q793	
Urinary system anomalies		
Any urinary system anomaly	Q60-64, Q794	Q610, Q627, Q633
Bladder exstrophy	Q640-641	
Genital anomalies		
Any genital anomaly	Q50-52, Q54-56	Q501-502, Q505, Q523, Q525, Q527, Q544, Q5520[c], Q5521[c]
Hypospadias	Q540, Q541-543, Q548, Q549	
Indeterminate sex	Q56	

Continued

Supplementary Table 2: Continued

Subgroup Condition	Definition (ICD-10 codes)[b]	
	Include	**Exclude**
Limb anomalies		
Any limb anomaly	Q65-74	Q653-656, Q658-Q659, Q661-669, Q670-678, Q680, Q6810[c], Q6821[c], Q683-685, Q7400[c]
Limb reduction defect	Q71-73	
Chromosomal anomalies[d]		
Any chromosomal anomaly	Q90-93, Q96-99	
Down syndrome	Q90	
Turner syndrome	Q96	
Other anomalies[e]	Q271-274, Q278-279, Q28, Q301-303, Q308-313, Q318-319, Q750-751, Q754-755, Q758-759, Q761-764, Q766-767, Q768-769, Q77, Q78, Q791, Q796, Q798-799, Q80, Q81, Q820-824, Q828-829, Q83, Q840-844, Q846, Q848-849, Q85, Q86, Q87, Q890-894, Q897-898, Q952-955, Q958-959	

ICD-10=International Classification of Diseases 10th Revision; [a]children without any such codes in hospital or mortality records during the first year of life were defined as not having any MCA; [b]definitions based on 'European Network of Population-based Registries for Congenital Malformations' (EUROCAT) Guide 1.5 [3], unless specified; [c]we were unable to exclude these ICD-10 codes in our dataset; [d]subset of the EUROCAT "genetic disorders sub-group"; [e]included in the definition of "any MCA" alongside the 12 system-specific anomalies.

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Supplementary Table 3: Prevalence of major congenital anomalies identified in this study compared with data from five England-based registries[a]

	This study[b]: prevalence per 10,000 live births (95% CI)	Registry data[a]: prevalence per 10,000 live and still births (95% CI; all values rounded to the nearest 0.5)
Total population of children (N)	**6,180,400 births between 1 September 2003 to 31 August 2013**	**2,000,000–2,020,000 births between 2004 and 2014**
Any MCA	354.7 (353.3–356.2)	185.5 (183.5–187.0)
Nervous system anomalies		
Any nervous system anomaly	19.4 (19.0–19.7)	11.0 (10.5–11.5)
Isolated nervous system anomaly	11.8 (11.6–12.1)	[c]
Microcephaly	3.8 (3.6–4.0)	1.5 (1.9–1.5)
Hydrocephaly	4.2 (4.0–4.3)	3.5 (3.5–4.0)
Spina Bifida	2.2 (2.1–2.3)	1.5 (1.5–2.0)
Eye anomalies		
Any eye anomaly	7.1 (6.9–7.3)	3.0 (2.5–3.0)
Isolated eye anomaly	4.6 (4.4–4.8)	[c]
Anophthalmos/Microphthalmos	1.1 (1.0–1.2)	0.5 (0.5–0.5)
Congenital cataract	2.1 (1.9–2.2)	1.0 (0.5–1.0)
Congenital Glaucoma	0.7 (0.7–0.8)	0 cases registered
Ear, face, and neck anomalies		
Any ear, face, and neck	2.7 (2.6–2.8)	1.5 (1.0–1.5)
Isolated ear, face, and neck	1.6 (1.5–1.7)	[c]
Cardiac anomalies		
Any cardiac anomaly	87.0 (86.2–87.7)	53.0 (52.0–54.0)
Isolated cardiac anomaly	60.0 (59.4–60.6)	[c]
Severe cardiac	23.1 (22.7–23.5)	21.0 (20.5–22.0)
Respiratory anomalies		
Any respiratory anomaly	6.4 (6.2–6.6)	3.5 (3.5–4.0)
Isolated respiratory anomaly	3.4 (3.3–3.6)	[c]
Choanal atresia	1.3 (1.2–1.4)	<5 cases registered
Orofacial anomalies		
Any orofacial anomaly	15.6 (15.2–15.9)	14.0 (13.5–14.5)
Isolated orofacial anomaly	10.5 (10.3–10.8)	[c]
Cleft lip	4.2 (4.0–4.4)	[c]
Cleft palate	7.9 (7.7–8.1)	6.0 (5.5–6.0)
Cleft lip and palate	5.9 (5.7–6.1)	[c]
Digestive system anomalies		
Any digestive anomaly	26.0 (25.6–26.5)	15.5 (15.0–16.5)
Isolated digestive anomaly	15.1 (14.8–15.4)	[c]
Oesophageal atresia	2.7 (2.6–2.8)	2.0 (2.0–2.5)
Small intestine atresia	3.1 (3.0–3.3)	<5 cases registered
Hirschsprung's disease	2.4 (2.3–2.6)	1.5 (1.5–1.5)
Biliary atresia	0.7 (0.6–0.8)	0 cases registered
Anorectal malformation	2.5 (2.3–2.6)	[c]
Congenital diaphragmatic hernia	2.8 (2.6–2.9)	2.5 (2.0–2.5)
Abdominal wall anomalies		
Any abdominal wall anomaly	6.7 (6.4–6.9)	6.0 (6.0–6.5)
Isolated abdominal wall anomaly	4.6 (4.5–4.8)	[c]
Omphalocele	2.1 (2.0–2.2)	1.5 (1.5–2.0)
Gastroschisis	4.1 (4.0–4.3)	4.5 (4.0–4.5)

Continued

Supplementary Table 3: Continued

	This study[b]: prevalence per 10,000 live births (95% CI)	Registry data[a]: prevalence per 10,000 live and still births (95% CI; all values rounded to the nearest 0.5)
Total population of children (N)	**6,180,400 births between 1 September 2003 to 31 August 2013**	**2,000,000–2,020,000 births between 2004 and 2014**
Urinary system anomalies		
Any urinary system anomaly	47.2 (46.6–47.7)	23.5 (22.5–24.0)
Isolated urinary system anomaly	40.0 (39.5–40.5)	[c]
Bladder exstrophy	1.1 (1.1–1.2)	0.5 (0.5–0.5)
Genital anomalies		
Any genital anomaly	41.9 (41.4–42.4)	19.0 (18.5–19.5)
Isolated genital anomaly	36.0 (35.5–36.5)	[c]
Hypospadias	28.7 (28.3–29.1)	15.0 (14.5–15.5)
Indeterminate sex	1.7 (1.6–1.8)	0.5 (0.5–1.0)
Limb anomalies		
Any limb anomaly	55.6 (55.0–56.2)	28.5 (27.5–29.0)
Isolated limb anomaly	46.8 (46.2–47.3)	[c]
Limb reduction defect	3.5 (3.4–3.7)	4.0 (3.5–4.5)
Chromosomal anomalies		
Any chromosomal anomaly	15.8 (15.4–16.1)	[c]
Isolated chromosomal anomaly	5.4 (5.2–5.6)	[c]
Down syndrome	10.1 (9.9–10.4)	11.5 (11.0–12.0)
Turner syndrome	0.6 (0.6–0.7)	1.0 (1.0–1.5)

MCA = major congenital anomaly;[a]combined average of the prevalence per 10,000 live births and still births across five England-based registries from 2004 to 2013, inclusive (including genetic anomalies): East Midlands & South Yorkshire (data gap in 2013), Northern England, South West England, Thames Valley, Wessex [1]; [b]MCAs identified in hospital and mortality records before the age of 1 year among children born in NHS-funded hospitals in England between 1 September 2003 to 31 August 2013; [c]comparator data not available.

References

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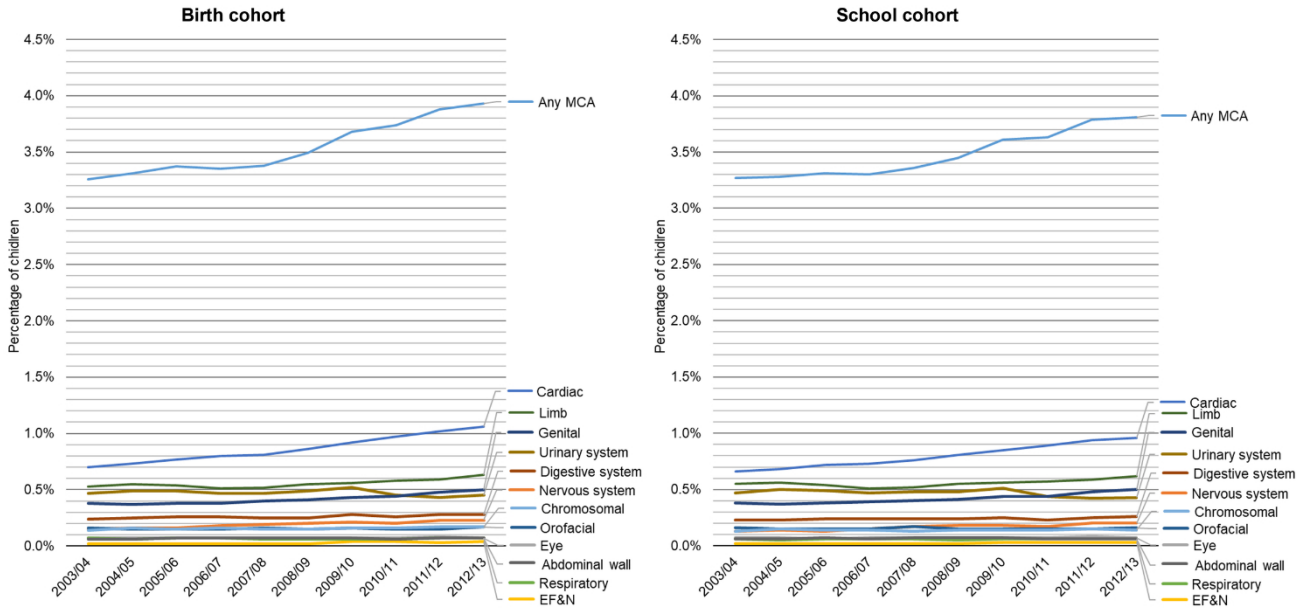
Supplementary Figure 2: Prevalence of any identified MCAs and MCA subgroups in the birth and school cohorts, by birth year[a]



EF&N = ear, face and neck anomalies, MCA = major congenital anomaly; [a]defined according to the academic calendar (i.e. 2003/04 includes 1 September 2003 to 31 August 2004, inclusive).

Supplementary Table 4: Number and proportion of children in the birth cohort (and alive at age five years) and the school cohort, by birth characteristics

		School cohort		Not in the school cohort[a]	
	Birth cohort survivors N (%)	N	% of birth cohort survivors	N	% of birth cohort survivors
Total	6,150,877	5,189,922	84.4	960,955	15.6
Birth year[b]					
2003/04	563,921	451,420	80.1	112,501	19.9
2004/05	576,193	476,102	82.6	100,091	17.4
2005/06	587,269	490,098	83.5	97,171	16.5
2006/07	597,539	499,899	83.7	97,640	16.3
2007/08	626,003	506,763	81.0	119,240	19.0
2008/09	624,394	527,775	84.5	96,619	15.5
2009/10	638,738	554,159	86.8	84,579	13.2
2010/11	652,799	567,194	86.9	85,605	13.1
2011/12	649,787	566,420	87.2	83,367	12.8
2012/13	634,234	550,092	86.7	84,142	13.3
Sex					
Female	3,146,768	2,668,336	84.8	478,432	15.2
Male	2,993,216	2,521,586	84.2	471,630	15.8
Missing	10,893	0	0.0	10,893	100.0
Region of residence					
North East	235,861	219,524	93.1	16,337	6.9
North West	650,383	572,635	88.0	77,748	12.0
Yorkshire and the Humber	476,208	438,161	92.0	38,047	8.0
East Midlands	378,006	343,111	90.8	34,895	9.2
West Midlands	463,163	417,085	90.1	46,078	9.9
East of England	464,822	412,577	88.8	52,245	11.2
London	799,122	639,527	80.0	159,595	20.0
South East	726,273	630,998	86.9	95,275	13.1
South West	438,393	396,149	90.4	42,244	9.6
Missing	1,518,646	1,120,155	73.8	398,491	26.2
Ethnic group					
White	4,193,153	3,632,233	86.6	560,920	13.4
Black	307,350	251,723	81.9	55,627	18.1
Asian	600,823	491,680	81.8	109,143	18.2
Mixed/multiple	219,785	182,998	83.3	36,787	16.7
Other	166,433	128,282	77.1	38,151	22.9
Missing	663,333	503,006	75.8	160,327	24.2
IMD groups					
1 Most deprived	1,129,032	1,026,359	90.9	102,673	9.1
2	881,819	792,960	89.9	88,859	10.1
3	730,658	652,110	89.2	78,548	10.8
4	652,141	576,170	88.4	75,971	11.6
5 Least deprived	639,392	557,453	87.2	81,939	12.8
Missing	2,117,835	1,584,870	74.8	532,965	25.2

IMD = index of multiple deprivation, MCA = major congenital anomaly; [a]these children do not appear in the school cohort for a variety of reasons, including attendance at a non-state-funded school, missed links between their hospital and school records, emigration and death before school entry; [b]defined according to the academic calendar (i.e. 2003/04 includes 1 September 2003 to 31 August 2004, inclusive).

Supplementary Table 5: Number and proportion of children from birth cohort present in school cohort, by MCA, system-specific subgroup[a] and selected conditions

	Birth cohort N	Birth cohort survivors at age 5[b] N	Present in any school year 1–6		
			N	% of birth cohort	% of birth cohort survivors
No MCA	5961151	5942586	5008598	84.0	84.3
Any MCA	219249	208558	181324	82.7	86.9
Nervous system anomalies					
Any nervous system anomaly	11971	9877	8676	72.5	87.8
Isolated nervous system anomaly	7322	6452	5656	77.2	87.7
Microcephaly	2349	1871	1659	70.6	88.7
Hydrocephaly	2574	2111	1883	73.2	89.2
Spina Bifida	1343	1169	1022	76.1	87.4
Eye anomalies					
Any eye anomaly	4404	4028	3599	81.7	89.3
Isolated eye anomaly	2849	2826	2510	88.1	88.8
Anophthalmos/ Microphthalmos	672	553	506	75.3	91.5
Congenital cataract	1271	1183	1069	84.1	90.4
Congenital Glaucoma	448	428	390	87.1	91.1
Ear, face, and neck anomalies					
Any ear, face, and neck anomaly	1663	1572	1336	80.3	85.0
Isolated ear, face, and neck anomaly	993	987	829	83.5	83.7
Cardiac anomalies					
Any cardiac anomaly	53741	47691	41857	77.9	87.8
Isolated cardiac anomaly	37080	34065	29745	80.2	87.3
Severe cardiac	14260	11676	10337	72.5	88.5
Respiratory anomalies					
Any respiratory anomaly	3941	3448	2997	76.0	86.9
Isolated respiratory anomaly	2121	2038	1755	82.7	86.1
Choanal atresia	804	722	647	80.5	89.6
Orofacial anomalies					
Any orofacial anomaly	9611	9131	7993	83.2	87.5
Isolated orofacial anomaly	6503	6474	5653	86.9	87.3
Cleft lip	2589	2538	2242	86.6	88.3
Cleft palate	4891	4591	4030	82.4	87.8
Cleft lip and palate	3656	3473	3051	83.5	87.8
Digestive anomalies					
Any digestive anomaly	16098	14367	12544	77.9	87.3
Isolated digestive anomaly	9343	8825	7657	82.0	86.8
Oesophageal atresia	1664	1464	1297	77.9	88.6
Small intestine atresia	1934	1767	1545	79.9	87.4
Hirschsprung's disease	1508	1448	1298	86.1	89.6
Biliary atresia	440	395	346	78.6	87.6
Anorectal malformation	1524	1019	896	58.8	87.9
Congenital diaphragmatic hernia	1713	1540	1363	79.6	88.5
Abdominal wall anomalies					
Any abdominal wall anomaly	4110	3818	3352	81.6	87.8
Isolated abdominal wall anomaly	2854	2757	2398	84.0	87.0
Omphalocele	1286	1131	968	75.3	85.6
Gastroschisis	2539	2400	2141	84.3	89.2
Urinary system anomalies					
Any urinary system anomaly	29158	27958	24203	83.0	86.6
Isolated urinary system anomaly	24703	24234	20949	84.8	86.4
Bladder exstrophy	704	699	596	84.7	85.1

Continued

Supplementary Table 5: Continued

	Birth cohort N	Birth cohort survivors at age 5[b] N	Present in any school year 1–6		
			N	% of birth cohort	% of birth cohort survivors
Genital anomalies					
Any genital anomaly	25914	25426	21923	84.6	86.2
Isolated genital anomaly	22252	22152	19093	85.8	86.2
Hypospadias	17727	17492	15243	86.0	87.1
Indeterminate sex	1044	925	637	61.0	68.9
Limb anomalies					
Any limb anomaly	34366	33381	28951	84.2	86.7
Isolated limb anomaly	28911	28752	24911	86.2	86.6
Limb reduction defect	2180	2038	1783	81.8	87.5
Chromosomal anomalies					
Any chromosomal anomaly	9734	8122	7208	74.0	88.7
Isolated chromosomal anomaly	3330	2983	2626	78.9	88.0
Down syndrome	6260	5768	5157	82.4	89.4
Turner syndrome	381	349	305	80.1	87.4

MCA = major congenital anomaly; [a]*any* groups contain children with and without additional anomalies in other system-specific subgroups, *isolated* groups do not contain children with anomalies in other system-specific subgroups; [b]a random number of deaths between 1 and 7 is imputed for isolated ear, face, and neck anomaly and bladder exstrophy groups due to suppressed death count (see Table 1).

Supplementary Table 6: Number and proportion of children in the school cohort with SEN provision recorded during years 1 and/or 2 (key stage 1), by category of recorded SEN provision, MCA, system-specific subgroups[a] and selected conditions

	School cohort[b]	Any recorded SEN	SEN support in mainstream school	EHCP in mainstream school	Specialist provision
	N	N (%)	N (%)	N (%)	N (%)
No MCA	4,977,468	1022149 (20.5)	919893 (18.5)	62007 (1.2)	40249 (0.8)
Any MCA	180,308	66134 (36.7)	44795 (24.8)	11429 (6.3)	9910 (5.5)
Nervous system anomalies					
Any nervous system anomaly	8,646	6072 (70.2)	2254 (26.1)	1433 (16.6)	2385 (27.6)
Isolated nervous system anomaly	5,634	3648 (64.7)	1544 (27.4)	802 (14.2)	1302 (23.1)
Microcephaly	1,652	1407 (85.2)	290 (17.6)	232 (14.0)	885 (53.6)
Hydrocephaly	1,876	1476 (78.7)	511 (27.2)	392 (20.9)	573 (30.5)
Spina Bifida	1,022	766 (75.0)	345 (33.8)	297 (29.1)	124 (12.1)
Eye anomalies					
Any eye anomaly	3,580	2240 (62.6)	1125 (31.4)	540 (15.1)	575 (16.1)
Isolated eye anomaly	2,497	1341 (53.7)	890 (35.6)	285 (11.4)	166 (6.6)
Anophthalmos/Microphthalmos	504	414 (82.1)	155 (30.8)	115 (22.8)	144 (28.6)
Congenital cataract	1,061	704 (66.4)	393 (37.0)	145 (13.7)	166 (15.6)
Congenital Glaucoma	388	272 (70.1)	147 (37.9)	70 (18.0)	55 (14.2)
Ear, face, and neck anomalies					
Any ear, face, and neck anomaly	1,328	638 (48.0)	383 (28.8)	115 (8.7)	140 (10.5)
Isolated ear, face, and neck anomaly	827	291 (35.2)	236 (28.5)	33 (4.0)	22 (2.7)
Cardiac anomalies					
Any cardiac anomaly	41,647	19964 (47.9)	11392 (27.4)	4578 (11.0)	3994 (9.6)
Isolated cardiac anomaly	29,589	11202 (37.9)	8373 (28.3)	1603 (5.4)	1226 (4.1)
Severe cardiac	10,278	5387 (52.4)	2874 (28.0)	1344 (13.1)	1169 (11.4)
Respiratory anomalies					
Any respiratory anomaly	2,983	1452 (48.7)	833 (27.9)	327 (11.0)	292 (9.8)
Isolated respiratory anomaly	1,746	594 (34.0)	459 (26.3)	74 (4.2)	61 (3.5)
Choanal atresia	647	370 (57.2)	158 (24.4)	102 (15.8)	110 (17.0)
Orofacial anomalies					
Any orofacial anomaly	7,950	4059 (51.1)	2963 (37.3)	575 (7.2)	521 (6.6)
Isolated orofacial anomaly	5,617	2474 (44.0)	2103 (37.4)	229 (4.1)	142 (2.5)
Cleft lip	2,227	788 (35.4)	649 (29.1)	74 (3.3)	65 (2.9)
Cleft palate	4,015	2290 (57.0)	1525 (38.0)	391 (9.7)	374 (9.3)
Cleft lip and palate	3,031	1708 (56.4)	1332 (43.9)	207 (6.8)	169 (5.6)
Digestive anomalies					
Any digestive anomaly	12,487	5313 (42.5)	3397 (27.2)	1022 (8.2)	894 (7.2)
Isolated digestive anomaly	7,615	2421 (31.8)	1959 (25.7)	268 (3.5)	194 (2.5)
Oesophageal atresia	1,292	647 (50.1)	421 (32.6)	151 (11.7)	75 (5.8)
Small intestine atresia	1,541	723 (46.9)	365 (23.7)	198 (12.8)	160 (10.4)
Hirschsprung's disease	1,291	591 (45.8)	358 (27.7)	124 (9.6)	109 (8.4)
Biliary atresia	342	156 (45.6)	111 (32.5)	23 (6.7)	22 (6.4)
Anorectal malformation	895	325 (36.3)	230 (25.7)	49 (5.5)	46 (5.1)
Congenital diaphragmatic hernia	1,360	714 (52.5)	466 (34.3)	155 (11.4)	93 (6.8)
Abdominal wall anomalies					
Any abdominal wall anomaly	3,339	1143 (34.2)	924 (27.7)	130 (3.9)	89 (2.7)
Isolated abdominal wall anomaly	2,389	699 (29.3)	616 (25.8)	47 (2.0)	36 (1.5)
Omphalocele	965	365 (37.8)	256 (26.5)	68 (7.0)	41 (4.2)
Gastroschisis	2,133	686 (32.2)	607 (28.5)	51 (2.4)	28 (1.3)
Urinary system anomalies					
Any urinary system anomaly	24,044	7592 (31.6)	5822 (24.2)	949 (3.9)	821 (3.4)
Isolated urinary system anomaly	20,804	5677 (27.3)	4857 (23.3)	487 (2.3)	333 (1.6)
Bladder exstrophy	592	224 (37.8)	166 (28.0)	36 (6.1)	22 (3.7)

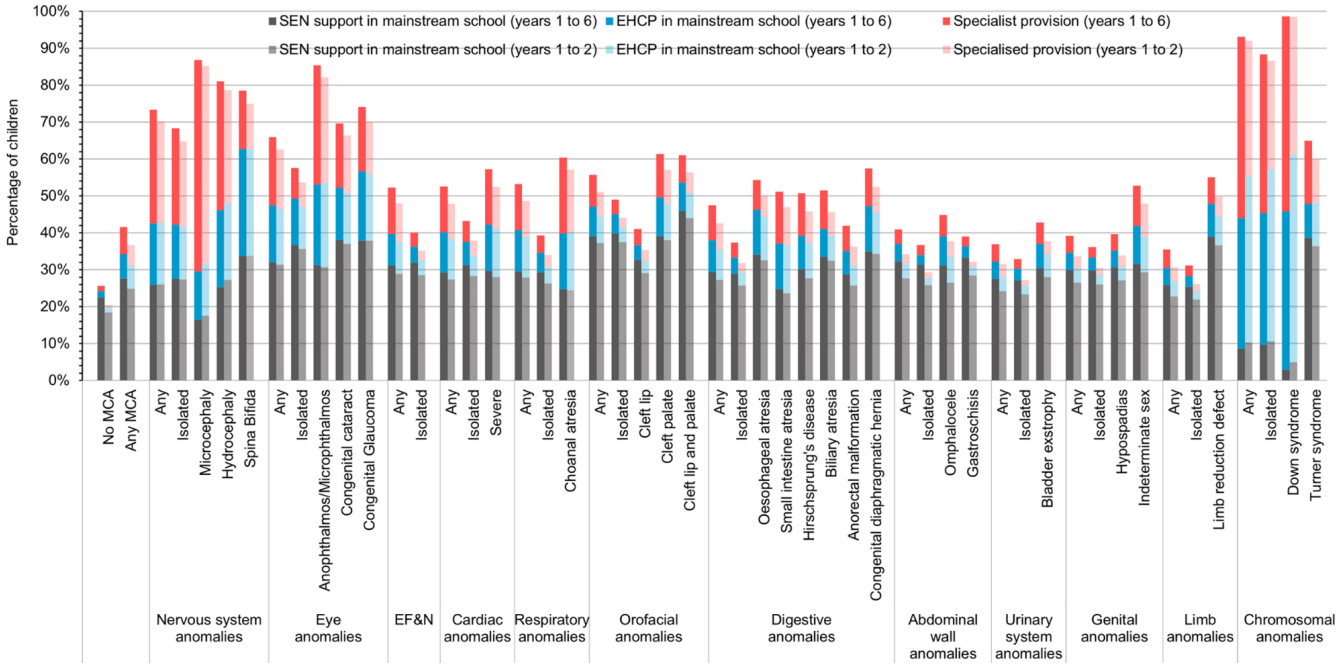
Continued

Supplementary Table 6: Continued

	School cohort[b]	Any recorded SEN	SEN support in mainstream school	EHCP in mainstream school	Specialist provision
	N	N (%)	N (%)	N (%)	N (%)
Genital anomalies					
Any genital anomaly	21,817	7351 (33.7)	5783 (26.5)	822 (3.8)	746 (3.4)
Isolated genital anomaly	18,996	5792 (30.5)	4946 (26.0)	502 (2.6)	344 (1.8)
Hypospadias	15,164	5144 (33.9)	4119 (27.2)	545 (3.6)	480 (3.2)
Indeterminate sex	635	304 (47.9)	186 (29.3)	60 (9.4)	58 (9.1)
Limb anomalies					
Any limb anomaly	28,769	8832 (30.7)	6547 (22.8)	1211 (4.2)	1074 (3.7)
Isolated limb anomaly	24,747	6464 (26.1)	5425 (21.9)	596 (2.4)	443 (1.8)
Limb reduction defect	1,770	882 (49.8)	647 (36.6)	140 (7.9)	95 (5.4)
Chromosomal anomalies					
Any chromosomal anomaly	7,173	6603 (92.1)	736 (10.3)	3242 (45.2)	2625 (36.6)
Isolated chromosomal anomaly	2,611	2260 (86.6)	277 (10.6)	1217 (46.6)	766 (29.3)
Down syndrome	5,130	5055 (98.5)	253 (4.9)	2891 (56.4)	1911 (37.3)
Turner syndrome	305	183 (60.0)	111 (36.4)	36 (11.8)	36 (11.8)

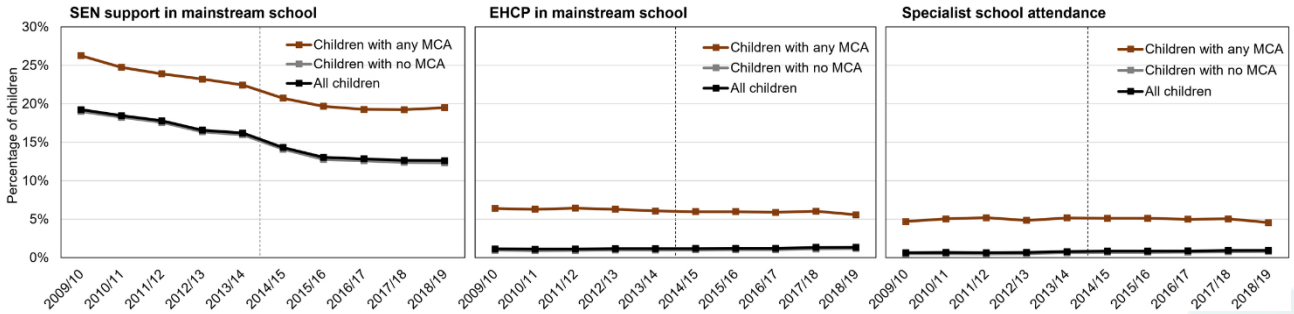
EHCP = education, health and care plan, MCA=major congenital anomaly, SEN= Special Educational Needs; [a]*any* groups contain children with and without additional anomalies in other system-specific subgroups, *isolated* groups do not contain children with anomalies in other system-specific subgroup; [b]Children in the school cohort that are present in year 1 and/or year 2 of the education data.

Supplementary Figure 3: Percentage of children in the school cohort with different categories of recorded SEN provision, by MCA, system-specific subgroup and selected conditions: comparing results for years 1 to 6 with years 1 to 2 (key stage 1 only)



EF&N = ear, face, and neck anomalies, EHCP = education, health and care plan, MCA = major congenital anomaly, SEN = special educational needs.

Supplementary Figure 4: Prevalence of recorded SEN provision in year 1, by academic year and MCA status: split by category of SEN provision (dotted line represents approximate timing of the SEN reforms)



EHCP = Education, health and care plan, MCA = major congenital anomaly, SEN = special educational needs.

Supplementary Table 7: Prevalence of (a) any recorded SEN provision, (b) SEN support, (c) EHCP and (d) specialist provision attendance in year 1 before and after 2014 SEN reforms, by MCA, system-specific subgroup[a] and selected conditions

	Before reforms[b]		After reforms[c]		
S7(a) Any recorded SEN	**Total N**	**Any recorded SEN provision N (%)**	**Total N**	**Any recorded SEN provision N (%)**	**Absolute % difference (99% CI)**
No MCA	2311628	437223 (18.9)	2643436	388885 (14.7)	−4.23 (−4.32, −4.15)
Any MCA	79242	28023 (35.4)	99965	30777 (30.8)	−4.83 (−5.40, −4.25)
Nervous system anomalies					
Any nervous system anomaly	3519	2487 (70.7)	5069	3387 (66.8)	−5.11 (−7.72, −2.50)
Isolated nervous system anomaly	2383	1558 (65.4)	3230	1945 (60.2)	−6.01 (−9.35, −2.66)
Microcephaly	759	665 (87.6)	892	736 (82.5)	−7.00 (−11.57, −2.44)
Hydrocephaly	793	625 (78.8)	1068	820 (76.8)	−4.13 (−9.17, 0.91)
Spina Bifida	426	324 (76.1)	592	424 (71.6)	−5.62 (−12.79, 1.54)
Eye anomalies					
Any eye anomaly	1660	1010 (60.8)	1893	1126 (59.5)	−2.29 (−6.52, 1.94)
Isolated eye anomaly	1192	623 (52.3)	1283	622 (48.5)	−4.23 (−9.40, 0.93)
Anophthalmos/Microphthalmos	209	164 (78.5)	290	239 (82.4)	1.73 (−7.70, 11.17)
Congenital cataract	497	318 (64.0)	557	354 (63.6)	−1.11 (−8.74, 6.53)
Congenital Glaucoma	191	129 (67.5)	191	127 (66.5)	−2.41 (−14.80, 9.98)
Ear, face, and neck anomalies					
Any ear, face, and neck anomaly	484	207 (42.8)	834	377 (45.2)	1.79 (−5.48, 9.07)
Isolated ear, face, and neck anomaly	308	92 (29.9)	513	156 (30.4)	0.19 (−8.30, 8.67)
Cardiac anomalies					
Any cardiac anomaly	17094	8156 (47.7)	24146	10218 (42.3)	−6.06 (−7.33, −4.78)
Isolated cardiac anomaly	12282	4634 (37.7)	17122	5280 (30.8)	−7.14 (−8.58, −5.70)
Severe cardiac	4518	2320 (51.4)	5668	2657 (46.9)	−5.25 (−7.81, −2.70)
Respiratory anomalies					
Any respiratory anomaly	1382	658 (47.6)	1567	658 (42.0)	−6.25 (−10.95, −1.55)
Isolated respiratory anomaly	804	271 (33.7)	921	226 (24.5)	−9.33 (−14.95, −3.70)
Choanal atresia	282	144 (51.1)	365	218 (59.7)	7.70 (−2.41, 17.80)
Orofacial anomalies					
Any orofacial anomaly	3777	1848 (48.9)	4143	1878 (45.3)	−3.92 (−6.81, −1.04)
Isolated orofacial anomaly	2718	1137 (41.8)	2872	1046 (36.4)	−5.53 (−8.88, −2.17)
Cleft lip	1043	361 (34.6)	1173	319 (27.2)	−7.49 (−12.54, −2.43)
Cleft palate	1947	1066 (54.8)	2049	1080 (52.7)	−2.60 (−6.66, 1.45)
Cleft lip and palate	1453	769 (52.9)	1577	802 (50.9)	−2.42 (−7.09, 2.25)
Digestive anomalies					
Any digestive anomaly	5655	2307 (40.8)	6737	2449 (36.4)	−4.90 (−7.15, −2.65)
Isolated digestive anomaly	3651	1136 (31.1)	3922	904 (23.0)	−8.21 (−10.83, −5.59)
Oesophageal atresia	596	274 (46.0)	677	325 (48.0)	0.66 (−6.51, 7.82)
Small intestine atresia	703	319 (45.4)	816	341 (41.8)	−4.64 (−11.16, 1.89)
Hirschsprung's disease	617	264 (42.8)	667	276 (41.4)	−2.02 (−9.09, 5.05)
Biliary atresia	153	70 (45.8)	187	61 (32.6)	−13.30 (−26.91, 0.30)
Anorectal malformation	418	147 (35.2)	475	135 (28.4)	−6.98 (−15.00, 1.03)
Congenital diaphragmatic hernia	594	322 (54.2)	756	336 (44.4)	−10.29 (−17.29, −3.28)
Abdominal wall anomalies					
Any abdominal wall anomaly	1503	492 (32.7)	1822	450 (24.7)	−8.18 (−12.24, −4.13)
Isolated abdominal wall anomaly	1112	300 (27.0)	1268	249 (19.6)	−7.40 (−11.87, −2.93)
Omphalocele	450	162 (36.0)	511	162 (31.7)	−4.54 (−12.40, 3.31)
Gastroschisis	993	305 (30.7)	1128	232 (20.6)	−10.29 (−15.16, −5.42)
Urinary system anomalies					
Any urinary system anomaly	11476	3257 (28.4)	12455	3287 (26.4)	−2.13 (−3.61, −0.64)
Isolated urinary system anomaly	10143	2479 (24.4)	10578	2251 (21.3)	−3.23 (−4.73, −1.73)

Continued

Supplementary Table 7: Continued

S7(a) Any recorded SEN	Before reforms[b] Total N	Before reforms[b] Any recorded SEN provision N (%)	After reforms[c] Total N	After reforms[c] Any recorded SEN provision N (%)	Absolute % difference (99% CI)
Bladder exstrophy	248	91 (36.7)	339	107 (31.6)	−5.22 (−15.43, 4.99)
Genital anomalies					
Any genital anomaly	9239	2862 (31.0)	12474	3436 (27.5)	−3.58 (−5.19, −1.97)
Isolated genital anomaly	8118	2250 (27.7)	10801	2590 (24.0)	−3.82 (−5.48, −2.16)
Hypospadias	6471	2022 (31.2)	8630	2387 (27.7)	−3.73 (−5.66, −1.80)
Indeterminate sex	269	120 (44.6)	359	148 (41.2)	−3.73 (−13.98, 6.53)
Limb anomalies					
Any limb anomaly	12829	3702 (28.9)	15769	4039 (25.6)	−3.37 (−4.73, −2.01)
Isolated limb anomaly	11218	2779 (24.8)	13406	2705 (20.2)	−4.65 (−6.03, −3.27)
Limb reduction defect	755	361 (47.8)	1001	448 (44.8)	−3.55 (−9.72, 2.63)
Chromosomal anomalies					
Any chromosomal anomaly	3285	2991 (91.1)	3682	3587 (97.4)	0.38 (−1.34, 2.11)
Isolated chromosomal anomaly	1344	1151 (85.6)	1201	1083 (90.2)	−0.63 (−4.20, 2.93)
Down syndrome	2389	2356 (98.6)	2543	2709 (106.5)	−0.54 (−1.45, 0.37)
Turner syndrome	145	82 (56.6)	157	86 (54.8)	−2.80 (−17.48, 11.88)

S7(b) SEN support in mainstream school	Before reforms[b] Total N	Before reforms[b] SEN support N (%)	After reforms[c] Total N	After reforms[c] SEN support N (%)	Absolute % difference (99% CI)
No MCA	2311628	402204 (17.4)	2643436	340095 (12.9)	−4.56 (−4.64, −4.48)
Any MCA	79242	19074 (24.1)	99965	19844 (19.9)	−4.38 (−4.89, −3.87)
Nervous system anomalies					
Any nervous system anomaly	3519	878 (25.0)	5069	1297 (25.6)	0.16 (−2.28, 2.59)
Isolated nervous system anomaly	2383	607 (25.5)	3230	876 (27.1)	1.27 (−1.77, 4.31)
Microcephaly	759	128 (16.9)	892	167 (18.7)	1.43 (−3.38, 6.24)
Hydrocephaly	793	227 (28.6)	1068	287 (26.9)	−2.49 (−7.85, 2.88)
Spina Bifida	426	143 (33.6)	592	195 (32.9)	−1.18 (−8.85, 6.50)
Eye anomalies					
Any eye anomaly	1660	516 (31.1)	1893	547 (28.9)	−2.64 (−6.59, 1.31)
Isolated eye anomaly	1192	420 (35.2)	1283	411 (32.0)	−3.50 (−8.38, 1.38)
Anophthalmos/Microphthalmos	209	63 (30.1)	290	82 (28.3)	−2.63 (−13.17, 7.92)
Congenital cataract	497	181 (36.4)	557	196 (35.2)	−1.61 (−9.20, 5.99)
Congenital Glaucoma	191	66 (34.6)	191	72 (37.7)	2.37 (−10.19, 14.93)
Ear, face, and neck anomalies					
Any ear, face, and neck anomaly	484	117 (24.2)	834	226 (27.1)	2.54 (−3.82, 8.90)
Isolated ear, face, and neck anomaly	308	70 (22.7)	513	130 (25.3)	2.32 (−5.54, 10.18)
Cardiac anomalies					
Any cardiac anomaly	17094	4732 (27.7)	24146	5548 (23.0)	−5.07 (−6.18, −3.95)
Isolated cardiac anomaly	12282	3520 (28.7)	17122	3859 (22.5)	−6.30 (−7.63, −4.97)
Severe cardiac	4518	1248 (27.6)	5668	1348 (23.8)	−4.24 (−6.47, −2.00)
Respiratory anomalies					
Any respiratory anomaly	1382	396 (28.7)	1567	345 (22.0)	−6.97 (−11.08, −2.86)
Isolated respiratory anomaly	804	206 (25.6)	921	177 (19.2)	−6.53 (−11.70, −1.35)
Choanal atresia	282	64 (22.7)	365	94 (25.8)	2.64 (−6.02, 11.31)
Orofacial anomalies					
Any orofacial anomaly	3777	1392 (36.9)	4143	1319 (31.8)	−5.25 (−7.99, −2.50)
Isolated orofacial anomaly	2718	979 (36.0)	2872	886 (30.8)	−5.27 (−8.51, −2.02)
Cleft lip	1043	296 (28.4)	1173	260 (22.2)	−6.27 (−11.03, −1.51)
Cleft palate	1947	757 (38.9)	2049	675 (32.9)	−6.29 (−10.18, −2.40)

Continued

Supplementary Table 7: Continued

S7(b) SEN support in mainstream school	Before reforms[b] Total N	Before reforms[b] SEN support N (%)	After reforms[c] Total N	After reforms[c] SEN support N (%)	Absolute % difference (99% CI)
Cleft lip and palate	1453	601 (41.4)	1577	621 (39.4)	−2.26 (−6.84, 2.33)
Digestive anomalies					
Any digestive anomaly	5655	1522 (26.9)	6737	1441 (21.4)	−5.79 (−7.78, −3.81)
Isolated digestive anomaly	3651	926 (25.4)	3922	704 (18.0)	−7.52 (−9.95, −5.09)
Oesophageal atresia	596	188 (31.5)	677	201 (29.7)	−2.71 (−9.31, 3.90)
Small intestine atresia	703	167 (23.8)	816	154 (18.9)	−5.36 (−10.74, 0.03)
Hirschsprung's disease	617	165 (26.7)	667	150 (22.5)	−4.59 (−10.75, 1.58)
Biliary atresia	153	50 (32.7)	187	43 (23.0)	−9.81 (−22.36, 2.75)
Anorectal malformation	418	102 (24.4)	475	84 (17.7)	−6.87 (−13.89, 0.16)
Congenital diaphragmatic hernia	594	215 (36.2)	756	214 (28.3)	−8.22 (−14.80, −1.64)
Abdominal wall anomalies					
Any abdominal wall anomaly	1503	416 (27.7)	1822	331 (18.2)	−9.62 (−13.39, −5.85)
Isolated abdominal wall anomaly	1112	280 (25.2)	1268	201 (15.9)	−9.38 (−13.64, −5.11)
Omphalocele	450	117 (26.0)	511	103 (20.2)	−6.00 (−13.00, 1.00)
Gastroschisis	993	279 (28.1)	1128	196 (17.4)	−10.84 (−15.52, −6.17)
Urinary system anomalies					
Any urinary system anomaly	11476	2568 (22.4)	12455	2371 (19.0)	−3.44 (−4.79, −2.09)
Isolated urinary system anomaly	10143	2164 (21.3)	10578	1866 (17.6)	−3.75 (−5.17, −2.33)
Bladder exstrophy	248	70 (28.2)	339	79 (23.3)	−4.99 (−14.42, 4.44)
Genital anomalies					
Any genital anomaly	9239	2300 (24.9)	12474	2623 (21.0)	−3.98 (−5.47, −2.49)
Isolated genital anomaly	8118	1967 (24.2)	10801	2184 (20.2)	−4.08 (−5.66, −2.51)
Hypospadias	6471	1636 (25.3)	8630	1875 (21.7)	−3.67 (−5.47, −1.87)
Indeterminate sex	269	76 (28.3)	359	88 (24.5)	−3.94 (−13.09, 5.21)
Limb anomalies					
Any limb anomaly	12829	2842 (22.2)	15769	2814 (17.8)	−4.40 (−5.62, −3.17)
Isolated limb anomaly	11218	2377 (21.2)	13406	2212 (16.5)	−4.73 (−6.02, −3.44)
Limb reduction defect	755	278 (36.8)	1001	320 (32.0)	−5.20 (−11.08, 0.68)
Chromosomal anomalies					
Any chromosomal anomaly	3285	362 (11.0)	3682	440 (12.0)	0.20 (−1.72, 2.11)
Isolated chromosomal anomaly	1344	149 (11.1)	1201	133 (11.1)	−0.65 (−3.77, 2.47)
Down syndrome	2389	137 (5.7)	2543	183 (7.2)	0.89 (−0.84, 2.62)
Turner syndrome	145	47 (32.4)	157	56 (35.7)	2.59 (−11.36, 16.54)

S7(c) EHCP in mainstream school	Before reforms[b] Total N	Before reforms[b] EHCP N (%)	After reforms[c] Total N	After reforms[c] EHCP N (%)	Absolute % difference (99% CI)
No MCA	2311628	22500 (1.0)	2643436	29049 (1.1)	0.12 (0.10, 0.15)
Any MCA	79242	5014 (6.3)	99965	5956 (6.0)	−0.42 (−0.71, −0.12)
Nervous system anomalies					
Any nervous system anomaly	3519	604 (17.2)	5069	792 (15.6)	−1.83 (−3.92, 0.25)
Isolated nervous system anomaly	2383	354 (14.9)	3230	401 (12.4)	−2.61 (−5.00, −0.23)
Microcephaly	759	117 (15.4)	892	122 (13.7)	−2.05 (−6.50, 2.40)
Hydrocephaly	793	164 (20.7)	1068	209 (19.6)	−1.65 (−6.45, 3.15)
Spina Bifida	426	131 (30.8)	592	166 (28.0)	−3.18 (−10.60, 4.25)
Eye anomalies					
Any eye anomaly	1660	239 (14.4)	1893	283 (14.9)	0.32 (−2.72, 3.36)
Isolated eye anomaly	1192	128 (10.7)	1283	138 (10.8)	−0.08 (−3.28, 3.11)
Anophthalmos/Microphthalmos	209	45 (21.5)	290	72 (24.8)	2.63 (−7.09, 12.35)
Congenital cataract	497	66 (13.3)	557	70 (12.6)	−0.85 (−6.16, 4.46)

Continued

Supplementary Table 7: Continued

S7(c) EHCP in mainstream school	Before reforms[b] Total N	Before reforms[b] EHCP N (%)	After reforms[c] Total N	After reforms[c] EHCP N (%)	Absolute % difference (99% CI)
Congenital Glaucoma	191	36 (18.8)	191	34 (17.8)	−1.41 (−11.52, 8.69)
Ear, face, and neck anomalies					
Any ear, face, and neck anomaly	484	42 (8.7)	834	70 (8.4)	−0.40 (−4.50, 3.70)
Isolated ear, face, and neck anomaly	308	[d]	513	[d]	[d]
Cardiac anomalies					
Any cardiac anomaly	17094	1957 (11.4)	24146	2517 (10.4)	−1.19 (−1.99, −0.39)
Isolated cardiac anomaly	12282	648 (5.3)	17122	791 (4.6)	−0.69 (−1.36, −0.03)
Severe cardiac	4518	610 (13.5)	5668	739 (13.0)	−0.68 (−2.41, 1.05)
Respiratory anomalies					
Any respiratory anomaly	1382	142 (10.3)	1567	165 (10.5)	0.10 (−2.79, 2.98)
Isolated respiratory anomaly	804	34 (4.2)	921	28 (3.0)	−1.21 (−3.54, 1.12)
Choanal atresia	282	40 (14.2)	365	57 (15.6)	1.18 (−6.02, 8.38)
Orofacial anomalies					
Any orofacial anomaly	3777	242 (6.4)	4143	291 (7.0)	0.57 (−0.88, 2.01)
Isolated orofacial anomaly	2718	101 (3.7)	2872	102 (3.6)	−0.18 (−1.46, 1.11)
Cleft lip	1043	35 (3.4)	1173	32 (2.7)	−0.63 (−2.52, 1.25)
Cleft palate	1947	157 (8.1)	2049	204 (10.0)	1.79 (−0.53, 4.10)
Cleft lip and palate	1453	98 (6.7)	1577	98 (6.2)	−0.57 (−2.87, 1.73)
Digestive anomalies					
Any digestive anomaly	5655	447 (7.9)	6737	553 (8.2)	0.20 (−1.06, 1.46)
Isolated digestive anomaly	3651	126 (3.5)	3922	124 (3.2)	−0.31 (−1.37, 0.75)
Oesophageal atresia	596	59 (9.9)	677	80 (11.8)	1.58 (−2.85, 6.01)
Small intestine atresia	703	91 (12.9)	816	111 (13.6)	0.32 (−4.13, 4.76)
Hirschsprung's disease	617	57 (9.2)	667	71 (10.6)	1.25 (−3.02, 5.52)
Biliary atresia	153	[d]	187	[d]	[d]
Anorectal malformation	418	26 (6.2)	475	27 (5.7)	−0.58 (−4.66, 3.49)
Congenital diaphragmatic hernia	594	70 (11.8)	756	77 (10.2)	−1.72 (−6.13, 2.69)
Abdominal wall anomalies					
Any abdominal wall anomaly	1503	44 (2.9)	1822	77 (4.2)	1.27 (−0.37, 2.92)
Isolated abdominal wall anomaly	1112	10 (0.9)	1268	29 (2.3)	1.38 (0.08, 2.68)
Omphalocele	450	26 (5.8)	511	41 (8.0)	2.18 (−2.00, 6.36)
Gastroschisis	993	[d]	1128	[d]	[d]
Urinary system anomalies					
Any urinary system anomaly	11476	400 (3.5)	12455	484 (3.9)	0.38 (−0.25, 1.01)
Isolated urinary system anomaly	10143	203 (2.0)	10578	221 (2.1)	0.08 (−0.42, 0.59)
Bladder exstrophy	248	[d]	339	[d]	[d]
Genital anomalies					
Any genital anomaly	9239	289 (3.1)	12474	440 (3.5)	0.38 (−0.25, 1.01)
Isolated genital anomaly	8118	161 (2.0)	10801	247 (2.3)	0.30 (−0.25, 0.84)
Hypospadias	6471	201 (3.1)	8630	282 (3.3)	0.14 (−0.60, 0.89)
Indeterminate sex	269	18 (6.7)	359	32 (8.9)	2.15 (−3.34, 7.64)
Limb anomalies					
Any limb anomaly	12829	473 (3.7)	15769	647 (4.1)	0.40 (−0.19, 0.99)
Isolated limb anomaly	11218	237 (2.1)	13406	284 (2.1)	0.00 (−0.47, 0.47)
Limb reduction defect	755	53 (7.0)	1001	75 (7.5)	0.39 (−2.81, 3.59)
Chromosomal anomalies					
Any chromosomal anomaly	3285	1616 (49.2)	3682	1848 (50.2)	−2.09 (−5.13, 0.96)
Isolated chromosomal anomaly	1344	669 (49.8)	1201	642 (53.5)	0.62 (−4.42, 5.65)
Down syndrome	2389	1473 (61.7)	2543	1650 (64.9)	−1.92 (−5.43, 1.60)
Turner syndrome	145	16 (11.0)	157	14 (8.9)	−2.28 (−11.12, 6.55)

Continued

Supplementary Table 7: Continued

S7(d) Specialist provision attendance	Before reforms[b] Total N	Before reforms[b] Specialist provision	After reforms[c] Total N	After reforms[c] Specialist provision	Absolute % difference (99% CI)
No MCA	2311628	12519 (0.5)	2643436	19741 (0.7)	0.20 (0.19, 0.22)
Any MCA	79242	3935 (5.0)	99965	4977 (5.0)	−0.03 (−0.29, 0.24)
Nervous system anomalies					
Any nervous system anomaly	3519	1005 (28.6)	5069	1298 (25.6)	−3.43 (−5.94, −0.93)
Isolated nervous system anomaly	2383	597 (25.1)	3230	668 (20.7)	−4.66 (−7.58, −1.74)
Microcephaly	759	420 (55.3)	892	447 (50.1)	−6.38 (−12.68, −0.07)
Hydrocephaly	793	234 (29.5)	1068	324 (30.3)	0.00 (−5.47, 5.47)
Spina Bifida	426	50 (11.7)	592	63 (10.6)	−1.27 (−6.42, 3.87)
Eye anomalies					
Any eye anomaly	1660	255 (15.4)	1893	296 (15.6)	0.03 (−3.08, 3.14)
Isolated eye anomaly	1192	75 (6.3)	1283	73 (5.7)	−0.65 (−3.11, 1.80)
Anophthalmos/Microphthalmos	209	56 (26.8)	290	85 (29.3)	1.73 (−8.65, 12.11)
Congenital cataract	497	71 (14.3)	557	88 (15.8)	1.34 (−4.30, 6.99)
Congenital Glaucoma	191	27 (14.1)	191	21 (11.0)	−3.37 (−12.02, 5.29)
Ear, face, and neck anomalies					
Any ear, face, and neck anomaly	484	48 (9.9)	834	81 (9.7)	−0.34 (−4.71, 4.02)
Isolated ear, face, and neck anomaly	308	[d]	513	[d]	[d]
Cardiac anomalies					
Any cardiac anomaly	17094	1467 (8.6)	24146	2153 (8.9)	0.20 (−0.53, 0.92)
Isolated cardiac anomaly	12282	466 (3.8)	17122	630 (3.7)	−0.14 (−0.72, 0.43)
Severe cardiac	4518	462 (10.2)	5668	570 (10.1)	−0.34 (−1.88, 1.20)
Respiratory anomalies					
Any respiratory anomaly	1382	120 (8.7)	1567	148 (9.4)	0.62 (−2.09, 3.33)
Isolated respiratory anomaly	804	31 (3.9)	921	21 (2.3)	−1.59 (−3.75, 0.56)
Choanal atresia	282	40 (14.2)	365	67 (18.4)	3.87 (−3.55, 11.30)
Orofacial anomalies					
Any orofacial anomaly	3777	214 (5.7)	4143	268 (6.5)	0.76 (−0.62, 2.13)
Isolated orofacial anomaly	2718	57 (2.1)	2872	58 (2.0)	−0.08 (−1.06, 0.89)
Cleft lip	1043	30 (2.9)	1173	27 (2.3)	−0.58 (−2.32, 1.16)
Cleft palate	1947	152 (7.8)	2049	201 (9.8)	1.90 (−0.39, 4.19)
Cleft lip and palate	1453	70 (4.8)	1577	83 (5.3)	0.41 (−1.63, 2.45)
Digestive anomalies					
Any digestive anomaly	5655	338 (6.0)	6737	455 (6.8)	0.69 (−0.43, 1.82)
Isolated digestive anomaly	3651	84 (2.3)	3922	76 (1.9)	−0.37 (−1.23, 0.48)
Oesophageal atresia	596	27 (4.5)	677	44 (6.5)	1.78 (−1.45, 5.01)
Small intestine atresia	703	61 (8.7)	816	76 (9.3)	0.40 (−3.34, 4.15)
Hirschsprung's disease	617	42 (6.8)	667	55 (8.2)	1.32 (−2.44, 5.08)
Biliary atresia	153	[d]	187	[d]	[d]
Anorectal malformation	418	19 (4.5)	475	24 (5.1)	0.46 (−3.21, 4.14)
Congenital diaphragmatic hernia	594	37 (6.2)	756	45 (6.0)	−0.35 (−3.71, 3.02)
Abdominal wall anomalies					
Any abdominal wall anomaly	1503	32 (2.1)	1822	42 (2.3)	0.16 (−1.15, 1.48)
Isolated abdominal wall anomaly	1112	10 (0.9)	1268	19 (1.5)	0.59 (−0.55, 1.73)
Omphalocele	450	19 (4.2)	511	18 (3.5)	−0.73 (−3.94, 2.48)
Gastroschisis	993	[d]	1128	[d]	[d]
Urinary system anomalies					
Any urinary system anomaly	11476	289 (2.5)	12455	432 (3.5)	0.93 (0.37, 1.50)
Isolated urinary system anomaly	10143	112 (1.1)	10578	164 (1.6)	0.44 (0.03, 0.85)
Bladder exstrophy	248	[d]	339	[d]	[d]

Continued

Supplementary Table 7: Continued

S7(d) Specialist provision attendance	Before reforms[b] Total N	Before reforms[b] Specialist provision	After reforms[c] Total N	After reforms[c] Specialist provision	Absolute % difference (99% CI)
Genital anomalies					
Any genital anomaly	9239	273 (3.0)	12474	373 (3.0)	0.02 (−0.58, 0.62)
Isolated genital anomaly	8118	122 (1.5)	10801	159 (1.5)	−0.04 (−0.49, 0.42)
Hypospadias	6471	185 (2.9)	8630	230 (2.7)	−0.21 (−0.90, 0.49)
Indeterminate sex	269	26 (9.7)	359	28 (7.8)	−1.93 (−7.81, 3.95)
Limb anomalies					
Any limb anomaly	12829	387 (3.0)	15769	578 (3.7)	0.63 (0.08, 1.18)
Isolated limb anomaly	11218	165 (1.5)	13406	209 (1.6)	0.08 (−0.32, 0.49)
Limb reduction defect	755	30 (4.0)	1001	53 (5.3)	1.26 (−1.31, 3.83)
Chromosomal anomalies					
Any chromosomal anomaly	3285	1013 (30.8)	3682	1299 (35.3)	2.28 (−0.56, 5.11)
Isolated chromosomal anomaly	1344	333 (24.8)	1201	308 (25.6)	−0.60 (−4.93, 3.73)
Down syndrome	2389	746 (31.2)	2543	876 (34.4)	0.49 (−2.85, 3.83)
Turner syndrome	145	19 (13.1)	157	16 (10.2)	−3.10 (−12.56, 6.35)

CI = confidence interval, MCA=major congenital anomaly, SEN= Special Educational Needs; [a]any groups contain children with and without additional anomalies in other system-specific, isolated groups do not contain children with anomalies in other system-specific subgroups; [b]before reforms group includes children born between 2003/04 and 2007/08 (entering year 1 before 2014/15); [c]after reforms group includes children born between 2008/09 and 2012/13 (entering year 1 on and after 2014/15); [d]small numbers (<8) suppressed to prevent disclosure of identities.

Supplementary Appendix 2

The RECORD statement – checklist of items, extended from the STROBE statement, that should be reported in observational studies using routinely collected health data

	Item No.	STROBE items	Location in manuscript where items are reported	RECORD items	Location in manuscript where items are reported
Title and abstract					
	1	(a) Indicate the study's design with a commonly used term in the title or the abstract (b) Provide in the abstract an informative and balanced summary of what was done and what was found		RECORD 1.1: The type of data used should be specified in the title or abstract. When possible, the name of the databases used should be included. RECORD 1.2: If applicable, the geographic region and timeframe within which the study took place should be reported in the title or abstract. RECORD 1.3: If linkage between databases was conducted for the study, this should be clearly stated in the title or abstract.	Title; Methods and findings section of the abstract Title Title
Introduction					
Background rationale	2	Explain the scientific background and rationale for the investigation being reported			Introduction paragraph 1
Objectives	3	State specific objectives, including any prespecified hypotheses			Introduction paragraph 3
Methods					
Study Design	4	Present key elements of study design early in the paper			Methods paragraph 2
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection			Methods paragraph 1, 2
Participants	6	*(a) Cohort study* - Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up *Case-control study* - Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls *Cross-sectional study* - Give the eligibility criteria, and the sources and methods of selection of participants *(b) Cohort study* - For matched studies, give matching criteria and number of exposed and unexposed *Case-control study* - For matched studies, give matching criteria and the number of controls per case		RECORD 6.1: The methods of study population selection (such as codes or algorithms used to identify subjects) should be listed in detail. If this is not possible, an explanation should be provided. RECORD 6.2: Any validation studies of the codes or algorithms used to select the population should be referenced. If validation was conducted for this study and not published elsewhere, detailed methods and results should be provided. RECORD 6.3: If the study involved linkage of databases, consider use of a flow diagram or other graphical display to demonstrate the data linkage process, including the number of individuals with linked data at each stage.	Methods paragraph 2 Methods paragraph 3 Fig 1 (flowchart)
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable.		RECORD 7.1: A complete list of codes and algorithms used to classify exposures, outcomes, confounders, and effect modifiers should be provided. If these cannot be reported, an explanation should be provided.	Code provided (GitHub link)
Data sources/ measurement	8	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group			Methods paragraphs 3-5
Bias	9	Describe any efforts to address potential sources of bias			Methods analysis section
Study size	10	Explain how the study size was arrived at			Fig 1 (flowchart)
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen, and why			Methods paragraphs 3-5
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding (b) Describe any methods used to examine subgroups and interactions (c) Explain how missing data were addressed (d) *Cohort study* - If applicable, explain how loss to follow-up was addressed			

	Item No.	STROBE items	Location in manuscript where items are reported	RECORD items	Location in manuscript where items are reported
		Case-control study - If applicable, explain how matching of cases and controls was addressed *Cross-sectional study* - If applicable, describe analytical methods taking account of sampling strategy (e) Describe any sensitivity analyses			Methods analysis section
Data access and cleaning methods		..		RECORD 12.1: Authors should describe the extent to which the investigators had access to the database population used to create the study population. RECORD 12.2: Authors should provide information on the data cleaning methods used in the study.	Method analysis section, third paragraph Code provided (GitHub link)
Linkage		..		RECORD 12.3: State whether the study included person-level, institutional-level, or other data linkage across two or more databases. The methods of linkage and methods of linkage quality evaluation should be provided.	Methods paragraph 1
Results					
Participants	13	(a) Report the numbers of individuals at each stage of the study (*e.g.*, numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed) (b) Give reasons for non-participation at each stage. (c) Consider use of a flow diagram		RECORD 13.1: Describe in detail the selection of the persons included in the study (*i.e.,* study population selection) including filtering based on data quality, data availability and linkage. The selection of included persons can be described in the text and/or by means of the study flow diagram.	Fig 1 (flowchart)
Descriptive data	14	(a) Give characteristics of study participants (*e.g.*, demographic, clinical, social) and information on exposures and potential confounders (b) Indicate the number of participants with missing data for each variable of interest (c) *Cohort study* - summarise follow-up time (*e.g.*, average and total amount)			S4 table
Outcome data	15	*Cohort study* - Report numbers of outcome events or summary measures over time *Case-control study* - Report numbers in each exposure category, or summary measures of exposure *Cross-sectional study* - Report numbers of outcome events or summary measures			S3 Fig
Main results	16	(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (e.g., 95% confidence interval). Make clear which confounders were adjusted for and why they were included (b) Report category boundaries when continuous variables were categorized (c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period			All results are unadjusted, 95% and 99% confidence intervals included
Other analyses	17	Report other analyses done—e.g., analyses of subgroups and interactions, and sensitivity analyses			Across all results
Discussion					
Key results	18	Summarise key results with reference to study objectives			Discussion paragraph 1
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias		RECORD 19.1: Discuss the implications of using data that were not created or collected to answer the specific research question(s). Include discussion of misclassification bias, unmeasured confounding, missing data, and changing eligibility over time, as they pertain to the study being reported.	Discussion paragraph 4-6
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence			Discussion paragraph 2
Generalisability	21	Discuss the generalisability (external validity) of the study results			Discussion paragraph 5

	Item No.	STROBE items	Location in manuscript where items are reported	RECORD items	Location in manuscript where items are reported
Other Information					
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based			Funding information provided in paper
Accessibility of protocol, raw data, and programming code		..		RECORD 22.1: Authors should provide information on how to access any supplemental information such as the study protocol, raw data, or programming code.	Link to code supplied in paper

*Reference: Benchimol EI, Smeeth L, Guttmann A, Harron K, Moher D, Petersen I, Sørensen HT, von Elm E, Langan SM, the RECORD Working Committee. The REporting of studies Conducted using Observational Routinely-collected health Data (RECORD) Statement. *PLoS Medicine* 2015; in press.