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# BMJ Open

## Healthcare use for children with complex needs: using routine health data linked to an ongoing birth cohort

Journal:	<i>BMJ Open</i>
Manuscript ID	bmjopen-2017-018419
Article Type:	Research
Date Submitted by the Author:	03-Jul-2017
Complete List of Authors:	Bishop, Chrissy; University of Bradford, Faculty of Health Small, Neil; University of Bradford, School of Health Studies Parslow, Roger; University of Leeds, Leeds Institute of Genetics, Health and Therapeutics Kelly, Brian; Bradford Institute for Health Research
<b>Primary Subject Heading</b>:	Health services research
Secondary Subject Heading:	Health services research, Paediatrics
Keywords:	Community child health < PAEDIATRICS, PRIMARY CARE, HEALTH SERVICES ADMINISTRATION & MANAGEMENT

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Manuscripts

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3 **Healthcare use for children with complex needs: using routine health data linked to an**  
4 **ongoing birth cohort**  
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23 **ABSTRACT**

24 **Objectives**

25 Congenital anomalies (CA) are a leading cause of global disease, death and disability for  
26 children. The pattern and variability of healthcare they require is complex and not well  
27 understood. Our aim was to examine healthcare use of children with CA and consequential  
28 complex needs, assessing their need and demand.  
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33 **Design**

34 Observational secondary analysis of data from a large prospective birth cohort, linked to  
35 routine health data. We repeated a sub analysis for referrals to specialists using a paper  
36 medical record review for a sample of 400 children.  
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41 **Setting**

42 Primary care data, hospital episode statistics, and referrals to specialists linked to birth cohort  
43 data.  
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47 **Participants**

48 All children who were recruited to the birth cohort between March 2007 and December 2010  
49 were eligible for the study. A total of 706 and 10768 children were included in the analyses  
50 with and without CA respectively.  
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54 **Primary and secondary outcome measures**

55 Healthcare use for children with and without CA aged 0<5 years was the primary outcome  
56 measure before and after adjustment for confounders.  
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## Results

Primary care consultations, use of hospital services and referrals to specialists were higher for children with CA than those without. Children in economically deprived neighborhoods were more likely to be admitted to hospital than consult primary care, and children with CA had a higher use of hospital services ( $\beta$  1.48, 95% CI 1.36-1.59) than primary care consultations ( $\beta$  0.24, 95% CI 1.18-0.30). Children from higher educated families were less likely to consult primary care and use hospital services. Children with CA had on average 3-4 multidisciplinary (MDT) specialists simultaneously managing their care needs.

## Conclusions

Hospital services are most in demand for children with CA, especially in economically deprived and ethnically diverse neighborhoods. The complex nature of CA in children requires multidisciplinary management and strengthened coordination between primary and secondary care.

## Article summary

### Strengths and limitations of this study

- Linking routine health data to a large prospective birth cohort, provided increased socio demographic and clinical information than that available from routine health data alone
- Ninety seven per cent of children from the birth cohort were linked to primary care and hospital episode data
- Data linkage permitted a multi-service, longitudinal evaluation of healthcare use
- We did not have access to electronic referrals, thus performed a medical record review to extract this information.

## INTRODUCTION

Around 93% of children with a congenital anomaly (CA) survive,<sup>1</sup> and a 20 year survival rate is estimated at 85.5% for children born with at least one CA,<sup>2</sup> some of whom will have complex conditions requiring multi-agency continuing care.<sup>3-4</sup> The healthcare needs of children with complex conditions have not been particularly well quantified in the past.<sup>5</sup> It has been suggested this could be partly attributable to the lack of longitudinal data reflecting the diverse professional input these children require to meet the demands of their complex healthcare needs.<sup>1</sup> The primary care practice is ideally positioned for monitoring the care requirements of children with complex conditions such as CA, whose prognosis or care needs

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3 may change as they develop.<sup>1,6-8</sup> Monitoring childhood development across the life course,  
4 provides invaluable insight into the multidisciplinary care regimes children with complex  
5 needs require due to their challenging prognoses and multiple comorbidities.<sup>9-15</sup> These  
6 complexities pose challenges in terms of coordinating multiple specialists, potential late  
7 diagnosis which may cause reliance on emergency care rather than preventative solutions  
8 offered at primary care level, leading to increases in costs.<sup>16,17-20</sup> To represent the  
9 interchangeable service requirements for children with CA, it has been suggested a  
10 combination of primary care consultations, use of hospital services, diagnosis codes,  
11 prescribed medications<sup>21</sup> and referral information<sup>22</sup> produce the best estimates of healthcare  
12 use.<sup>23</sup> The literature addressing this comprehensive map of healthcare use for children with  
13 CA is limited, with the bulk of evidence being American studies investigating primarily  
14 hospital use and heart CA.<sup>16,22-25</sup> Only two studies were found which addressed the demand on  
15 primary care services for children with CA.<sup>26,27</sup> The need and demand for healthcare services  
16 is also known to be aggravated by patient complexity, levels of deprivation, and primary care  
17 practice provision.<sup>14</sup>

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27 Our aims were therefore to select children with and without CA from a large prospective birth  
28 cohort covering a deprived and ethnically diverse population, linked to their primary care  
29 records, hospital episode statistics and referral information. We compare longitudinal  
30 healthcare use for children ages 0<5 diagnosed with and without a major CA. We examine the  
31 effects of having a CA and consequential complex healthcare needs on predicted rates of  
32 primary care consultations, use of hospital services and referrals to multidisciplinary  
33 specialists, as well as addressing the influence of demographic and socioeconomic factors.

## 34 35 36 37 38 39 **METHODS**

40 We used data from the Born in Bradford (BiB) cohort study, which is an on-going prospective  
41 birth cohort, recruiting 12450 pregnant women who gave informed consent for the study  
42 between 2007 and 2011. It monitors the health of mothers, their partners and birth outcomes  
43 for 13857 children. Detailed information on socioeconomic deprivation, demographics,  
44 clinical outcomes and risk factors are recorded. The methods for the BiB study are reported in  
45 detail elsewhere.<sup>28</sup>

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52 *Insert figure 1 here*

### 53 54 55 **Case ascertainment and coding methods**

56 BiB recruits gave their consent for access to electronic primary care records and hospital  
57 episode statistics, which are split into elective, accident and emergency and other emergency  
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3 admissions, thus referred to as use of hospital services. We linked children's primary care  
4 data held on SystmOne,<sup>29</sup> the patient contact single source system which has complete  
5 coverage in Bradford, and use of hospital services to BiB questionnaire data. Linkage was  
6 performed using NHS number, surname, date of birth and gender between SystmOne,<sup>29</sup> use of  
7 hospital services and BiB. Of 13857 recruits, 97% were matched to primary care and use of  
8 hospital services data, forming the study population. The amount of children with at least one  
9 (non-birth) hospital event was 5223 (38%). The average time over which data were recorded  
10 was 5.5 years, with a maximum of 7.6 years, in all 74386 person years of data. As not all  
11 children in the cohort had reached age seven, we censored our follow-up of these cases to  
12 ages 0<5.  
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20 Primary care data is a trusted source of ascertainment of CA, including those diagnosed later  
21 in childhood.<sup>30,31</sup> We used cross mapping of SystemOne<sup>31</sup> diagnostic CTV3 Read medical  
22 codes to ICD-10 codes<sup>32</sup> to classify and extract children with CA from the primary care  
23 database. We followed the European Surveillance of Congenital Anomalies (EUROCAT)  
24 guidelines, using the British Isles National Organisation of CA Registers (BINOCAR)  
25 methodology<sup>33</sup> to differentiate between major and minor CA. Minor anomalies were  
26 excluded, and a clinical geneticist reviewed classifications.  
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32 Extracting information from paper medical records was necessary to capture multidisciplinary  
33 outpatient data and referral activity, as this is sometimes not routinely included in electronic  
34 records.<sup>34</sup> We extracted referral information from a review of paper medical records using a  
35 convenience sample of 200 children with and 200 without CA, selected at random from the  
36 BiB cohort. The small sample size was chosen based on the exploratory nature of the medical  
37 record review, and the feasibility of performing this by hand within the time scale of this  
38 study. A standardised data extraction form was designed, reviewed by a clinician, and piloted  
39 to accumulate the number and type of referrals to different multidisciplinary services.  
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### 46 **Statistical analysis**

47 We had three outcomes for this study. The number of primary care consultations, use of  
48 hospital services and referrals to multidisciplinary specialists. Both primary care consultations  
49 and use of hospital services were counted as one per day, even if multiple appointments in the  
50 same day were recorded, as many of the appointments occurring on the same day were  
51 episodes that ran over time, or were duplicates. Negative binomial poisson regression models  
52 were used to model primary consultations and use of hospital services as they account for the  
53 over dispersion in count data. These models use an exposure variable, which indicates the  
54 number of times the event could have happened. Primary care consultations and use of  
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3 hospital services were expressed per year of observed primary care registered time, which  
4 takes into account any periods the child may not have been registered with the primary care  
5 practice, withdrawals from the cohort, or deaths. We performed a sub analysis reporting  
6 regression coefficients for the outcome multidisciplinary referrals.  
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10 We used three models to compare regression coefficients for CA for each of our three  
11 outcomes. Model 1 included univariate analyses, thought of as what is actually observed.  
12 Model 2 adds other covariates we determined as confounding factors. Model 3 controls for  
13 confounders and adds measures of underlying ill health. Controlling for ill health using a  
14 measure of multi-morbidity, is a recognised method of risk adjustment for evaluations of  
15 healthcare use, as severity of illness may not solely be due to multi-morbidity and ill health, it  
16 may also be due to other patient characteristics.<sup>35</sup> We used a count of unique prescriptions and  
17 a count of the number of comorbidities per child as measures of ill health. Simple counts of  
18 distinct medications have been suggested as an accurate measure of ill health, given chronic  
19 conditions frequently require repeat prescriptions<sup>35,36</sup> as have counts of comorbidities in  
20 primary care settings.<sup>37,38</sup> We performed a test for interaction between whether the child had a  
21 CA and level of deprivation for primary care consultations and use of hospital services. We  
22 also report the predicted rates of healthcare use for children with CA and without (figure 2).  
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### 31 **Confounders**

32 We used directed acyclic graphs to determine the minimally sufficient confounding set for all  
33 of our models and checked whether the inclusion of additional covariates improved the model  
34 more than would be expected by chance using appropriate model fit statistics. These consisted  
35 of maternal age (<20, 20-34, >34 years); educational attainment (Low education [ $<5$  GCSE  
36 equivalents or other education] High education [ $5 >$  GCSE equivalents at grade A-C or two  
37 advanced level certificates or diploma, degree or higher degree]), economic deprivation  
38 (Economically deprived, not economically deprived), ethnicity (White British, Pakistani,  
39 Other), and consanguinity (Non-consanguineous, first cousin, second cousin, other blood).  
40 All covariates were entered into the model as a categorical variable to allow for possible non-  
41 linearity in the relationship between the multi-morbidity measure and relevant outcome.  
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### 50 **Ethics**

51 Ethics approval for the cohort study was provided by Bradford Local Research Ethics  
52 Committee (reference 06/Q1202/48).  
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### 55 **Role of the funding source**

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3 The sponsors of this study had no role in the study design, data collection, analysis or  
4 interpretation, or writing of the report. CB, and BK had full access to all the data in the study  
5 and all authors had final responsibility for the decision to submit for publication.  
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## 8 9 **RESULTS**

10 Table 1 shows the characteristics of mothers who gave birth to children with a CA in the BiB  
11 cohort (CA = 706, no CA= 10768), and average healthcare use up to the child's fifth birthday.  
12 The BiB cohort is multiethnic, 40% white British, 45% Pakistani and 15% 'other' ethnicities.  
13 Children of Pakistani heritage were born with the highest proportion of CA (53%), compared  
14 to white British children (35%) and 'other' ethnicities (13%) (table 1). Forty nine per cent of  
15 children with CA were from Pakistani first cousin unions compared to <1% of children with  
16 CA from white British first cousin unions. Children of Pakistani heritage with CA had on  
17 average 1.61 more primary care consultations, and 0.39 more hospital admissions per year  
18 than children without CA. Children with CA of Pakistani heritage had the highest number of  
19 primary care appointments over the five-year period, with on average 2.4 more primary care  
20 appointments than children of white British heritage. Children with CA of Pakistani heritage  
21 and 'other ethnicities', had the same use of hospital services on average, but this was almost  
22 double for children with CA of white British heritage (Table 1). Although the most common  
23 reason for use of hospital services for children with and without CA was respiratory  
24 conditions, when stratified by admission type, children with CA had the most 'other  
25 emergency' admissions overall (40%), followed by elective admissions (34%) whereas  
26 children without CA had an increase of 'Accident & Emergency' (45%) admissions (Table 3).  
27 Diagnoses on admission were also different between groups, with neoplasms and clinical lab  
28 findings recorded as the most common reason for admission for children with CA, not  
29 recorded in children without CA (table 3). Table 2 reports the regression coefficients for the  
30 univariate and multivariable analysis of primary care and use of hospital services. Sixty three  
31 per cent of children with CA were born into economically deprived neighborhoods (table 1).  
32 Both the adjusted and unadjusted rates suggest children from economically deprived  
33 neighborhoods have an increased use of hospital services ( $\beta$  0.35, 95% CI 0.27-0.42), but do  
34 not use more primary care consultations.  
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49 Children from both Pakistani heritage and other ethnicities were predicted to require an  
50 increase in primary care consultations in both the univariate and multivariable analyses.  
51 Children who had older mothers (>34) were predicted to use hospital services less ( $\beta$  -0.17,  
52 95% CI -0.28 -0.06), but not primary care consultations ( $\beta$  -0.03, 95% CI -0.08 -0.01) after  
53 adjustment for confounders. Children born into consanguineous families were predicted to  
54 have an increased use of hospital services, but not primary care consultations. Children with a  
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CA had the largest increase in use of primary care consultations ( $\beta$  0.24, 95% CI 1.18-0.30), and use of hospital services ( $\beta$  1.48, 95% CI 1.36-1.59) after adjustment for confounders, for which usage was almost three times higher. Higher maternal education reduced primary care consultations and use of hospital services (Table 2). The sub analysis of multidisciplinary referrals predicts an increased use of specialist referrals for children with CA after adjustment for confounders ( $\beta$  3.59, 95% CI 3.11-4.08). The only other significant factor was children born into economically deprived neighborhoods ( $\beta$  0.55, 95% CI 0.01-1.09). We adjusted for ill health for services with the highest predicted usage, those being hospital services and multidisciplinary referrals for children with and without CA (figure 2). After controlling for ill health, the predicted increased use of hospital services for children with CA reduces by almost half but still remains ( $\beta$  0.80, 95% CI 0.63-0.97), as does the predicted increased use of multidisciplinary referrals ( $\beta$  2.27, 95% CI 1.59-2.94) (Figure 2).

*Insert figure 2 here*

From a possible 41 different specialists, the most common referral was to consultant pediatricians (14% 59/400), followed by neonatology (13% 57/400), pediatric surgery (8% 34/400) and local cardiology services (7% 30/400). On average children had between three and four specialists involved in their care simultaneously.

	All		White British		Pakistani		Other	
	No anomaly	Anomaly	No anomaly	Anomaly	No anomaly	Anomaly	No anomaly	Anomaly
<b>Ethnic origin</b>	10768 (93.85%)	706(6.15 %)	4288(94.60%)	245(5.40%)	4804(92.83 %)	371(7.17 %)	1653(94.84%)	90(5.16%)
<b>Economic deprivation</b>								
Economically deprived	6147(57.09%)	444(62.8 9%)	2072(48.32%)	116(47.35%)	3315(69.00 %)	280(75.4 7%)	758(45.86%)	48(53.33%)
Not economically deprived	4166(38.69%)	247(34.9 9%)	2034(47.43%)	122(49.80%)	1340(27.89 %)	85(22.91 %)	792(47.91%)	40(44.44%)
Missing	455(4.23%)	15(2.12%)	182(4.24%)	7(2.86%)	149(3.10%)	6(1.62%)	103(6.23%)	2(2.22%)
<b>Age of mother</b>								
20-34	8716(80.94%)	554(78.4 7%)	3231(75.35%)	180(73.47%)	4099(85.32 %)	312(84.1 0%)	1366(82.64%)	62(68.89%)
<20	776(7.21%)	51(7.22%)	536(12.50%)	29(11.84%)	148(3.08%)	15(4.04%)	92(5.57%)	7(7.78%)
>34	1276(11.85%)	101(14.3 1%)	521(12.15%)	36(14.69%)	557(11.59%)	44(11.86 %)	195(11.80%)	21(23.33%)
<b>Consanguinity</b>								
Non-consanguineous	7850(72.90%)	424(60.0 6%)	4284(99.81%)	224(99.59%)	2008(41.80 %)	102(27.4 9%)	1538(93.04%)	78(86.67%)
First Cousin	1834(17.03%)	192(27.2 0%)	1(<1%)	1(<1%)	1753(36.49 %)	183(49.3 3%)	79(4.78%)	8(8.89%)
Second Cousin	637(5.92%)	55(7.79%)	0	0	611(12.72%)	51(13.75 %)	25(1.51%)	4(4.44%)
Other blood	447(4.15%)	35(4.96%)	3(<1%)	0	432(8.99%)	35(9.43%)	11(0.67%)	0
<b>Maternal Education</b>								

Lower education	2894(26.88%)	217(30.74%)	1228(28.64%)	74(30.20%)	1374(28.60%)	123(33.15%)	286(17.30%)	20(22.22%)
Higher education	7617(70.74%)	470(66.57%)	3014(70.29%)	167(68.16%)	3357(69.88%)	243(65.50%)	1223(74.59%)	60(66.67%)
<b>Healthcare use</b>								
Average number of GP appointments per year	5.21	6.82	4.21	5.49	6.20	7.89	4.91	6.01
Average number of admissions per year	0.11	0.50	0.11	0.32	0.12	0.60	0.08	0.60

Table 1: Characteristics of the cohort

	Outcome 1: primary care consultation rates				Outcome 2: Admission rates				Outcome 3: Referrals			
	Model 1: Univariate		Model 2: Multivariate		Model 1: Univariate		Model 2: Multivariate		Model 1: Univariate		Model 2: Multivariate	
	Coefficient	p	Coefficient	p	Coefficient	P	Coefficient	p	Coefficient	p	Coefficient	p
	(95% CI)		(95% CI)		(95% CI)		(95% CI)		(95% CI)		(95% CI)	
<b>Economic deprivation</b>												
Economically deprived	0.11(0.08-0.14)	<0.001	0.01(-0.02-0.04)	0.48	0.35(0.27-0.42)	<0.001	0.21(0.13-0.29)	<0.001	0.95(0.35-1.56)	0.002	0.55(0.01-1.09)	0.045
Not Economically deprived	-	-	-	-	-	-	-	-	-	-	-	-
<b>Ethnicity</b>												
White British	-	-	-	-	-	-	-	-	-	-	-	-
Pakistani	0.43(0.40-0.46)	<0.001	0.40(0.36-0.44)	<0.001	0.27(0.19-0.35)	<0.001	-0.02(-0.12-0.07)	0.64	0.72(0.09-1.36)	0.003	-0.28(-0.95-0.40)	0.42
Other	0.24(0.20-0.29)	<0.001	0.26(0.21-0.30)	<0.001	-0.08(-0.19-0.03)	0.15	-0.14(-0.25-0.03)	0.02	-0.03(-0.92-0.85)	0.94	-0.36(-1.11-0.39)	0.35
<b>Mothers age</b>												
20-34	-	-	-	-	-	-	-	-	-	-	-	-
<20	-0.13(-0.19-0.07)	<0.001	-0.01(-0.07-0.04)	0.68	0.11(-0.03-0.24)	0.13	0.07(-0.07-0.21)	0.32	-0.31(-1.39-0.77)	0.58	-0.42(-1.32-0.48)	0.36
>34	-0.06(-0.10-0.01)	<0.002	-0.03(-0.08-0.01)	0.13	-0.15(-0.27--0.04)	0.01	-0.17(-0.28-0.06)	0.002	0.83(-0.08-1.74)	0.007	0.64(-0.10-1.38)	0.09
<b>Consanguinity</b>												
Non-consanguineous	-	-	-	-	-	-	-	-	-	-	-	-
First cousin	0.03(-0.01-0.08)	<0.17	0.03(-0.01-0.08)	0.17	0.48(0.39-0.58)	<0.001	0.28(0.17-0.39)	<0.001	1.19(0.48-1.91)	0.001	0.24(-0.50-0.97)	0.53
Second cousin	0.24(0.18-0.30)	<0.001	-0.01(-0.07-0.05)	0.78	0.24(0.09-0.39)	0.002	-0.01(-0.17-0.15)	0.94	1.17(0.11-2.23)	0.031	0.26(-0.70-1.21)	0.59
Other blood	0.25(0.18-0.32)	<0.001	0.01(-0.07-0.05)	0.86	0.31(0.14-0.48)	<0.001	0.23(0.05--0.41)	0.012	-0.03(-1.54-1.48)	0.96	-0.32(-1.60-0.97)	0.63
<b>Congenital Anomaly</b>												
Yes	0.29(0.23-0.35)	<0.001	0.24(0.18-0.30)	<0.001	1.46(1.35-1.57)	<0.001	1.48(1.36-1.59)	<0.001	3.71(3.28-4.15)	<0.001	3.59(3.11-4.08)	<0.001
No	-	-	-	-	-	-	-	-	-	-	-	-

Maternal Education												
Lower education	-	-	-	-	-	-	-	-	-	-	-	-
Higher education	-0.04(-0.08-0.01)	<0.007	-0.04(-0.07-0.003)	0.031	-0.21(-0.29--0.13)	<0.001	-0.09(-0.17-0.01)	0.021	-0.70(-1.34-0.06)	0.032	0.06(-0.49-0.61)	0.83

Table 2: Univariate and multivariable coefficients for primary care consultations, use of hospital services and referrals to specialists adjusted for demographic and lifestyle factors in the BiB cohort.

Admittance type	Total number of admissions over 5 years		Most common reason for admission	
	No anomaly	Anomaly	No anomaly	Anomaly
A&E	3985(49%)	609(26%)	1. Respiratory 2. Injury/poison	1. Respiratory 2. Infectious parasitic
Other	2522(31%)	932(40%)	1. Respiratory 2. Infectious parasitic	1. Respiratory 2. Clinical lab findings
Emergency				
Elective	1632(20%)	801(34%)	1. Eye/ear 2. Respiratory	1. Neoplasms/blood/immune 2. Congenital abnormalities

Table 3: Proportion of admissions by type, and most common reason for admission.

### Interactions

Interaction effects between whether the child had a CA and economic deprivation were not significant in multivariable models.

### DISCUSSION

Our data suggest children with CA have higher numbers of primary care consultations, admissions to hospital, and referrals to multidisciplinary specialists on average per year than children without CA. Children of Pakistani heritage have almost double the number of hospital admissions per year than children of white British heritage. When accounting for confounders, children with CA were predicted to require an increase in primary care consultations ( $\beta$  0.24, 95% CI 1.18-0.30) and hospital services compared to children without a CA, although the increase in use of hospital services was predicted to be ( $\beta$  1.48, 95% CI 1.36-1.59) much larger. We found only one study reporting an increase in primary care consultations for children with CA, but heart CA specifically.<sup>26</sup> Although we find children from Pakistani heritage are predicted to use more primary care consultations ( $\beta$  0.40, 95% CI 0.36-0.44) than children without CA, this might be explained by more than half (53%) of children with CA in the BiB cohort being of Pakistani heritage (53%). We also find children with CA from economically deprived neighborhoods have an increased risk of using hospital services, but not primary care consultations. This might be explained by previous findings from the BiB cohort suggesting mothers from poorer backgrounds are less likely to use primary care services due to variation in primary care practice provision.<sup>39</sup>

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3 An increased use of hospital services can be expected for children with CA<sup>12,22,24-26</sup> we find  
4 the type of hospital admission differs between children with and without CA. This is most  
5 likely explained by CA requiring more severe treatment. For example although respiratory  
6 conditions were the most common reason for using hospital services overall (table 3), a  
7 finding similar to other studies,<sup>25</sup> ‘other emergency’ admissions were the most frequently  
8 used hospital service for children with CA. Other emergency refers to procedures of urgency  
9 requiring corrective and sometimes surgical interventions that are initiated by health  
10 professionals, rather than parents presenting with their child at A & E. This increase in other  
11 emergency and elective procedures is a finding similar to that of other CA studies.<sup>12,27</sup>  
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18 Children with a CA were predicted to require more referrals to MDT specialists ( $\beta$  37.97,  
19 95% CI 18.19-79.26) than children without CA, and have more than one specialist involved  
20 in their care simultaneously compared to children without CA. Although patient complexity  
21 increases the need for healthcare,<sup>14</sup> coordination of appointments for the multiple specialists  
22 required is also susceptible to variation, and is some times exacerbated by the divide between  
23 primary care, community and hospital services,<sup>3</sup> which is also associated with patient  
24 complications, late diagnosis, and an increased reliance on emergency care.<sup>16</sup> This suggests  
25 that although the predicted increase in use of hospital services for children with CA may be  
26 primarily due to their complex needs and ill health, there may be scope for this to be reduced  
27 through increasing the efficiency of care coordination.<sup>40</sup> Our findings therefore support the  
28 suggestion of key workers as a potential solution for efficient coordination, with the aim of  
29 streamlining navigation through services.<sup>1</sup> Also, when adjusting for ill health, the predicted  
30 increase in use of hospital services and multidisciplinary referrals for children with CA  
31 reduces (figure 2), suggesting higher usage may not be completely attributable to ill health,  
32 but affected by other factors such as deprivation and ethnicity.  
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43 There are limitations. Using a subset of diseases (CA), does not cover all children with  
44 conditions that may also be complex, however in order to extract a population we could be  
45 sure were both representative of complexity, and prevalent enough to create sample size  
46 groups large enough for comparison, CA were chosen based on the knowledge that they are  
47 high in numbers in Bradford and known to require complex care.<sup>41</sup> These results are based on  
48 the Bradford population, which might be interpreted as a limitation in terms of generalisability.  
49 However we feel these results are applicable to other populations or NHS trusts serving  
50 highly deprived and ethnically diverse groups of patients, determinants which are also known  
51 to be associated with CA.<sup>41</sup> Despite the successful linkage of primary care to cohort data in  
52 this study (97%), attributable to the complete SystemOne coverage of primary care practices in  
53 Bradford, the use of paper medical records for capturing referral activity is susceptible to  
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3 missed information due fluctuations in consultant record keeping and missing records. This  
4 research therefore illustrates the advantages of implementing a completely 'paperless' record  
5 keeping ethos, and the future emphasis for ensuring the exchange of data between IT systems  
6 in all clinical and care settings. This will only further strengthen the interpretability of key  
7 information at the point of care for patients with complex healthcare needs.<sup>42</sup>  
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### 11 **What is already known on this topic?**

12 Children with CA do require increased healthcare use from both primary and secondary  
13 services, and the long-term management of their care requires careful coordination and  
14 multidisciplinary input. Longitudinal data is lacking to support this claim and evidence on  
15 healthcare use is mainly in relation to congenital heart defects and hospital care based in the  
16 USA.  
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### 21 **What does this study add?**

22 We predict healthcare use combining primary care consultations, use of hospital services and  
23 referrals to specialists for children with CA linked to detailed socio demographic data from a  
24 longitudinal birth cohort. We find an increase in primary care consultations for children with  
25 CA than children without, however they have a greater burden on use of hospital services due  
26 to the severity of their conditions, and specialist management required.  
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### 33 **Contributors**

34 CB and BK had full access to all the data, conducted the analysis and statistical interpretation.  
35 RCP and NS contributed to the conception and design of the work, drafting and critical  
36 revision for intellectual content. All authors had final responsibility for the decision to submit  
37 for publication.  
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### 43 **No competing interests**

44 All authors have completed the ICMJE uniform disclosure form  
45 at [www.icmje.org/coi\\_disclosure.pdf](http://www.icmje.org/coi_disclosure.pdf) and declare: no financial support or relationships from  
46 any organisation for the submitted work that might have an interest in the submitted work in  
47 the previous three years; no other relationships or activities that could appear to have  
48 influenced the submitted work.  
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### 53 **Funding**

54 This paper presents independent research by a PhD candidate supported by a Bradford  
55 University studentship, in conjunction with the White Rose Consortium, and the National  
56 Institute for Health Research (NIHR), Collaboration for Leadership in Applied Health  
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3 Research and Care (CLAHRC) Yorkshire and Humber programme “Healthy Children  
4 Healthy Families Theme”, IS-CLA-0113-10020.  
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6

### 7 8 **Acknowledgements**

9 The views expressed in this paper are those of the authors, and not necessarily those of the  
10 National Health Service, the NIHR, or the Department of Health. We thank the families who  
11 took part in the Born in Bradford study, the midwives for their help in recruitment, the  
12 paediatricians and health visitors, the Born in Bradford team, which included interviewers,  
13 data managers, laboratory staff, clerical workers, research scientists, volunteers, and  
14 managers,  
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### 20 **Data sharing statement**

21 No additional data are available  
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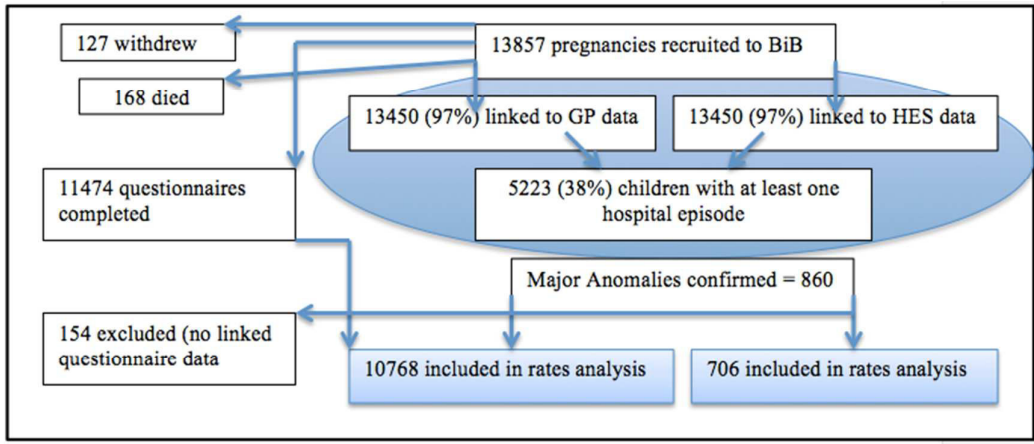
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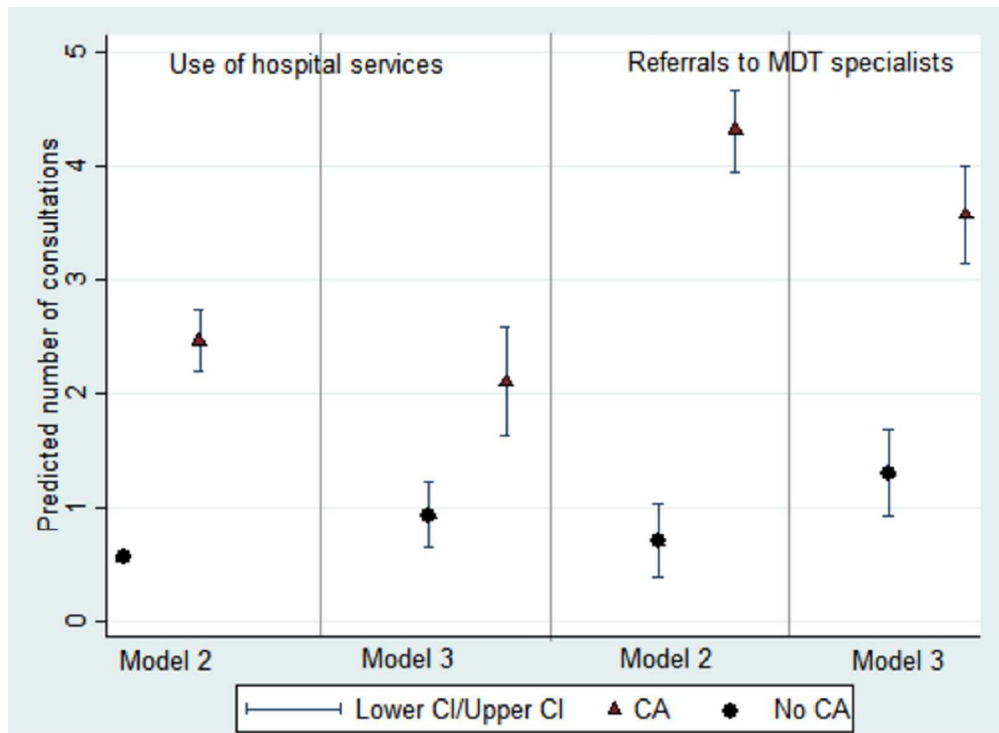
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or peer review only



Predicted use of hospital services and referrals to MDT specialists for children with and without CA, before and after controlling for ill health (Model 2 adjusted for confounders, model 3 adjusted for confounders and ill health).

view only

# BMJ Open

## Healthcare use for children with complex needs: using routine health data linked to a multiethnic, ongoing birth cohort

Journal:	<i>BMJ Open</i>
Manuscript ID	bmjopen-2017-018419.R1
Article Type:	Research
Date Submitted by the Author:	28-Nov-2017
Complete List of Authors:	Bishop, Chrissy; University of Bradford, Faculty of Health Small, Neil; University of Bradford, School of Health Studies Parslow, Roger; University of Leeds, Leeds Institute of Genetics, Health and Therapeutics Kelly, Brian; Bradford Institute for Health Research
<b>Primary Subject Heading</b>:	Health services research
Secondary Subject Heading:	Health services research, Paediatrics
Keywords:	Community child health < PAEDIATRICS, PRIMARY CARE, HEALTH SERVICES ADMINISTRATION & MANAGEMENT

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3 **Healthcare use for children with complex needs: using routine health data linked to a**  
4 **multiethnic, ongoing birth cohort**  
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7 Chrissy Bishop<sup>a</sup>, Neil Small<sup>a</sup>, Roger Parslow<sup>b</sup>, Brian Kelly<sup>c</sup>

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21

22 **ABSTRACT**

23 **Objectives**

24 Congenital anomalies (CA) are a leading cause of disease, death and disability for children  
25 throughout the world. Many have complex and varying healthcare needs which are not well  
26 understood. Our aim was to analyse the healthcare needs of children with CA and examine  
27 how that healthcare is delivered.  
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32 **Design**

33 Secondary analysis of observational data from the Born in Bradford study, a large prospective  
34 birth cohort, linked to primary care data and hospital episode statistics. Negative binomial  
35 regression with 95% confidence intervals was performed to predict healthcare use. The  
36 authors conducted a sub analysis on referrals to specialists using paper medical records for a  
37 sample of 400 children.  
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43 **Setting**

44 Primary, secondary and tertiary healthcare services in a large city in the north of England.  
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47 **Participants**

48 All children recruited to the birth cohort between March 2007 and December 2010. A total of  
49 706 children with CA and 10768 without were included in the analyses.  
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53 **Primary and secondary outcome measures**

54 Healthcare use for children with and without CA aged 0-<5 years was the primary outcome  
55 measure after adjustment for confounders.  
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## Results

Primary care consultations, use of hospital services and referrals to specialists were higher for children with CA than those without. Children in economically deprived neighborhoods were more likely to be admitted to hospital than consult primary care. Children with CA had a higher use of hospital services ( $\beta$  1.48, 95% CI 1.36-1.59) than primary care consultations ( $\beta$  0.24, 95% CI 1.18-0.30). Children with higher educated mothers were less likely to consult primary care and hospital services.

## Conclusions

Hospital services are most in demand for children with CA, but also for children who were economically deprived whether they had a CA or not. The complex nature of CA in children requires multidisciplinary management and strengthened coordination between primary and secondary care.

## Article summary

### Strengths and limitations of this study

- Linking birth cohort data to routine health data produces an enhanced dataset of socio demographic and clinical information.
- Ninety seven per cent of children from the birth cohort were linked to primary care and hospital episode data.
- Data linkage permitted a multi-service, longitudinal evaluation of healthcare use.
- We did not have access to electronic referrals, thus we performed a medical record review to extract this information for a subsample of 400 children.

## INTRODUCTION

Congenital anomalies (CA) are an abnormality of structure, function or metabolism, present at birth, which may result in mental, physical disability, or fatality (Misra et al 2005). The incidence of CA in Bradford is high; previously reported at 306 per 10,000 live births, compared to a national average of 227 per 10,000 live births (Sheridan et al 2013; BINOCAR 2012). Around 93% of children with a congenital anomaly (CA) survive to adulthood,<sup>1</sup> and a 20 year survival rate is estimated at 85.5% for children born with at least one CA,<sup>2</sup> some of whom will have complex conditions requiring multi-agency continuing care.<sup>3-4</sup> The healthcare needs of children with complex conditions have not been particularly well quantified in the past.<sup>5</sup> This may be due in part to a lack of longitudinal data related to the nature of multi-disciplinary involvement required by children with complex needs.<sup>1</sup> In the UK, primary care

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3 practice is ideally positioned for monitoring the care requirements of children with complex  
4 conditions such as CA, whose prognosis or care needs may change as they develop.<sup>1,6-8</sup>  
5 Monitoring childhood development across the life course, provides invaluable insight into the  
6 multidisciplinary care regimes that children with complex needs require.<sup>9-15</sup> Multidisciplinary  
7 care requires the coordination of multiple specialists which may result in late diagnosis  
8 leading to reliance on emergency care rather than preventative solutions offered at primary  
9 care level, with contaminant increases in costs to the healthcare services.<sup>16,17-20</sup> To represent  
10 the multidisciplinary care needs of children with CA, it has been suggested a combination of  
11 primary care consultations, use of hospital services, diagnosis codes, prescribed medications<sup>21</sup>  
12 and referral information<sup>22</sup> produce the best estimates of healthcare use.<sup>23</sup> The literature  
13 addressing this comprehensive map of healthcare use for children with CA is limited, with the  
14 bulk of evidence coming from American studies investigating primarily hospital use required  
15 for the treatment of heart CA.<sup>16,22-25</sup> Only two studies were found which addressed the demand  
16 on primary care services for children with CA.<sup>26,27</sup> The need and demand for primary care  
17 services in particular are intensified by patient complexity, levels of deprivation, and primary  
18 care practice provision.<sup>14</sup>

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28 Our aims were therefore to explore the longitudinal healthcare use for children from birth up  
29 to their fifth birthday (0-5) with CA and without. We do this by linking demographic and  
30 socioeconomic data from a large prospective birth cohort covering a deprived and ethnically  
31 diverse population, to children's primary care records, hospital episodes statistics and referral  
32 information. In doing so this study examines the effects of having a CA, and consequential ill  
33 health, on primary care use, use of hospital services and referrals to multidisciplinary  
34 specialists. We also investigate the influence of demographic and socioeconomic factors on  
35 health care use.

## 36 37 38 39 40 41 **METHODS**

42 We used data from the Born in Bradford (BiB) cohort study, an on-going prospective birth  
43 cohort, which recruited 12450 pregnant women who gave informed consent for the study  
44 between 2007 and 2011. It monitors the health of mothers, their partners and birth outcomes  
45 for 13857 children. Detailed information on socioeconomic deprivation, demographics,  
46 clinical outcomes and risk factors are recorded. The methods for the BiB study are reported in  
47 detail elsewhere.<sup>28</sup>

### 48 49 50 51 52 53 54 55 **Case ascertainment and coding methods**



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3 BiB recruits gave their consent for access to electronic primary care records and hospital  
4 episode statistics, which are split into elective, accident and emergency and other emergency  
5 admissions, here referred to as use of hospital services. We linked children's primary care  
6 data held on SystmOne,<sup>29</sup> the patient contact single source system which has complete  
7 coverage in Bradford, and use of hospital services to BiB questionnaire data. Linkage was  
8 performed using NHS number, surname, date of birth and gender between SystmOne<sup>29</sup> use of  
9 hospital services data and BiB. Of 13857 recruits, 97% were matched to primary care and use  
10 of hospital services data, forming the study population. The number of children with at least  
11 one (non-birth) hospital event was 5223 (38%). Hospital events included admissions for  
12 elective procedures, other emergencies, and A and E presentations. The average time over  
13 which data were recorded was 5.5 years, with a maximum of 7.6 years, in all 74386 person  
14 years of data. As not all children in the cohort had reached age seven, we censored our  
15 follow-up of these cases to aged 0-<5.

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23 Primary care data is a trusted source of ascertainment of CA, including for those diagnosed  
24 later in childhood.<sup>30,31</sup> We used cross mapping of SystemOne<sup>31</sup> diagnostic Clinical Terms  
25 Version 3 (CTV-3) Read medical codes to International Classification of Diseases version10  
26 codes (ICD-10)<sup>32</sup> to classify and extract children with CA from the primary care database. We  
27 followed the European Surveillance of Congenital Anomalies (EUROCAT) guidelines, using  
28 the British Isles National Organisation of CA Registers (BINOCAR) methodology,<sup>33</sup> which  
29 advise selection of major CA and removal of minor CA. A clinical geneticist reviewed  
30 classifications. A total of 860 children with CA were identified, and 154 were excluded, as  
31 they had no linked BiB questionnaire data.

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Extracting information from paper medical records was necessary to capture multidisciplinary  
outpatient data and referral activity, as this is sometimes not routinely included in electronic  
records.<sup>34</sup> We extracted referral information from a review of paper medical records using a  
sample of 200 children with and 200 without CA, selected at random from the BiB cohort.  
The small sample size was chosen based on the exploratory nature of the medical record  
review, and the feasibility of performing this by hand within the time scale of this study. A  
standardised data extraction form was designed, reviewed by a clinician, and piloted to  
accumulate the number and type of referrals to different multidisciplinary services.

### Statistical analysis

We had three outcomes for this study. The number of primary care consultations, use of  
hospital services and referrals to multidisciplinary specialists. Both primary care consultations  
and use of hospital services were counted as one per day, even if multiple appointments in the

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3 same day were recorded, as many of the appointments occurring on the same day were  
4 episodes that ran over time, or were duplicates. Negative binomial regression models were  
5 used to model primary consultations and use of hospital services as they account for the over  
6 dispersion in count data. These models use an exposure variable, which indicates the number  
7 of times the event could have happened. Primary care consultations and use of hospital  
8 services were expressed per year of observed primary care registered time, which takes into  
9 account any periods the child may not have been registered with the primary care practice,  
10 withdrawals from the cohort, or deaths. We performed a sub analysis reporting regression  
11 coefficients for the outcome multidisciplinary referrals.  
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17 We used three models to compare regression coefficients for each of our three outcomes.  
18 Model 1 included univariate analyses, thought of as what is actually observed. Model 2  
19 adjusts for other covariates we determined as confounding factors. Model 3 adjusts for  
20 confounders and measures of underlying ill health. Adjusting for ill health using a measure of  
21 multi-morbidity, is a recognised method of risk adjustment for evaluations of healthcare use,  
22 as severity of illness may not solely be due to multi-morbidity and ill health, it may also be  
23 due to other patient characteristics.<sup>35</sup> We used a count of unique prescriptions and a count of  
24 the number of comorbidities per child as measures of ill health. Simple counts of distinct  
25 medications have been suggested as an accurate measure of ill health, given chronic  
26 conditions frequently require repeat prescriptions<sup>35,36</sup> as have counts of comorbidities in  
27 primary care settings.<sup>37,38</sup> We performed a test for interaction between whether the child had a  
28 CA and level of deprivation for primary care consultations and use of hospital services. We  
29 also report the predicted rates of healthcare use for children with CA and without (figure 1).  
30 We also stratified the analyses for all three outcomes by CA (results not shown).  
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### 40 **Confounders**

41 We used directed acyclic graphs to determine the minimally sufficient confounding set for all  
42 of our models and checked whether the inclusion of additional covariates improved the model  
43 more than would be expected by chance using appropriate model fit statistics. These consisted  
44 of maternal age (<20, 20-34, >34 years); educational attainment (Low education [<5 GCSE  
45 equivalents or other education] High education [5> GCSE equivalents at grade A–C or two  
46 advanced level certificates or diploma, degree or higher degree])<sup>39</sup>, economic deprivation  
47 (Economically deprived, not economically deprived), which was measured using means-  
48 tested benefit status. In the UK, being in receipt of means-tested benefits is recognised as  
49 measure of income poverty, as these benefits are frequently the only source of income and are  
50 paid at rates that put individuals below standard poverty lines,<sup>40</sup> ethnicity (White British,  
51 Pakistani, Other), and consanguinity (Non-consanguineous, first cousin, second cousin, other  
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3 blood[any relation]). All covariates were entered into the model as a categorical variable to  
4 allow for possible non-linearity in the relationship between the multi-morbidity measure and  
5 relevant outcome.  
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### 8 9 **Ethics**

10 Ethics approval for the cohort study was provided by Bradford Local Research Ethics  
11 Committee (reference 06/Q1202/48).  
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### 14 15 **Role of the funding source**

16 The sponsors of this study had no role in the study design, data collection, analysis or  
17 interpretation, or writing of the report. CB, and BK had full access to all the data in the study  
18 and all authors had final responsibility for the decision to submit for publication.  
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### 21 22 **RESULTS**

23 Table 1 shows the characteristics of mothers who gave birth to children with a CA in the BiB  
24 cohort (CA = 706, no CA= 10768), and average healthcare use up to the child's fifth birthday.  
25 The BiB cohort is multiethnic, 40% white British, 45% Pakistani and 15% 'other' ethnicities.  
26 Of all children with a CA, 53% were of Pakistani heritage, compared to 35% white British  
27 and 13% 'other' ethnicities (13%). Forty nine per cent of Pakistani children with CA were  
28 from first cousin unions compared to <1% of white British children with CA from first cousin  
29 unions. Children of Pakistani heritage with CA had on average 1.61 more primary care  
30 consultations, and 0.39 more hospital admissions per year than children without CA. Children  
31 with CA of Pakistani heritage had the highest number of primary care appointments over the  
32 five-year period, with on average 2.4 more primary care appointments than children of white  
33 British heritage.  
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40 Children with CA of Pakistani heritage and 'other ethnicities', had the same use of hospital  
41 services on average, but this was almost double for children with CA of white British heritage  
42 (Table 1). Table 2 reports the regression coefficients for the univariate and multivariable  
43 analysis of primary care and use of hospital services. Sixty three per cent of children with CA  
44 were born into economically deprived neighborhoods (table 1). Both the adjusted and  
45 unadjusted rates suggest children from economically deprived neighborhoods have an  
46 increased use of hospital services ( $\beta$  0.35, 95% CI 0.27-0.42), but do not use more primary  
47 care consultations. Although the most common reason for use of hospital services for children  
48 with and without CA was respiratory conditions, when stratified by admission type, children  
49 with CA had the most 'other emergency' admissions overall (40%), followed by elective  
50 admissions (34%) whereas children without CA had an increase of 'Accident & Emergency'  
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(45%) admissions (Table 3). Diagnoses on admission were also different between groups, with neoplasms and clinical lab findings recorded as the most common reason for admission for children with CA, not recorded in children without CA (table 3).

Children from both Pakistani heritage and other ethnicities were predicted to require an increase in primary care consultations in both the univariate and multivariable analyses. Children who had older mothers (>34) were predicted to use hospital services less ( $\beta$  -0.17, 95% CI -0.28 -0.06), but not primary care consultations ( $\beta$  -0.03, 95% CI -0.08 -0.01) after adjustment for confounders. Children born into consanguineous families were predicted to have an increased use of hospital services, but not primary care consultations. Children with a CA had the largest increase in use of primary care consultations ( $\beta$  0.24, 95% CI 1.18-0.30), and use of hospital services ( $\beta$  1.48, 95% CI 1.36-1.59) after adjustment for confounders, for which usage was almost three times higher. Higher maternal education reduced primary care consultations and use of hospital services (Table 2). The sub analysis of multidisciplinary referrals predicts an increased use of specialist referrals for children with CA after adjustment for confounders ( $\beta$  3.59, 95% CI 3.11-4.08). The only other significant factor was children born into economically deprived neighborhoods ( $\beta$  0.55, 95% CI 0.01-1.09). We adjusted for ill health for services with the highest predicted usage, those being hospital services and multidisciplinary referrals for children with and without CA (figure 1). After controlling for ill health, the predicted increased use of hospital services for children with CA reduces by almost half but still remains ( $\beta$  0.80, 95% CI 0.63-0.97), as does the predicted increased use of multidisciplinary referrals ( $\beta$  2.27, 95% CI 1.59-2.94) (Figure 1).

*Insert figure 1 here*

From a possible 41 different specialists, the most common referral was to consultant pediatricians (14% 59/400), followed by neonatology (13% 57/400), pediatric surgery (8% 34/400) and local cardiology services (7% 30/400). On average children had between three and four specialists involved in their care simultaneously.

	All		White British		Pakistani		Other	
	No anomaly	Anomaly	No anomaly	Anomaly	No anomaly	Anomaly	No anomaly	Anomaly
<b>Ethnic origin</b>	10768 (93.9%)	706(6.2%)	4288(94.6%)	245(5.4%)	4804(92.8%)	371(7.2%)	1653(94.8%)	90(5.2%)
<b>Economic deprivation</b>								
Economically deprived	6147(57.1%)	444(62.9%)	2072(48.3%)	116(47.4%)	3315(69.0%)	280(75.5%)	758(45.9%)	48(53.3%)
Not economically deprived	4166(38.7%)	247(35.0%)	2034(47.4%)	122(49.8%)	1340(27.9%)	85(22.9%)	792(47.9%)	40(44.4%)
Missing	455(4.2%)	15(2.1%)	182(4.2%)	7(2.9%)	149(3.1%)	6(1.6%)	103(6.2%)	2(2.2%)

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Age of mother								
20-34	8716(80.9%)	554(78.5%)	3231(75.4%)	180(73.5%)	4099(85.3%)	312(84.1%)	1366(82.6%)	62(68.9%)
<20	776(7.2%)	51(7.2%)	536(12.5%)	29(11.8%)	148(3.1%)	15(4.0%)	92(5.6%)	7(7.8%)
>34	1276(11.9%)	101(14.3%)	521(12.2%)	36(14.7%)	557(11.6)	44(11.9%)	195(11.8%)	21(23.3%)
Consanguinity								
Non-consanguineous	7850(72.9%)	424(60.1%)	4284(99.8%)	224(99.6%)	2008(41.8%)	102(27.5%)	1538(93.0%)	78(86.7%)
First Cousin	1834(17.0%)	192(27.2%)	1(<1%)	1(<1%)	1753(36.5%)	183(49.3%)	79(4.8%)	8(8.9%)
Second Cousin	637(5.9%)	55(7.8%)	0	0	611(12.7%)	51(13.8%)	25(1.5%)	4(4.4%)
Other blood	447(4.2%)	35(5.0%)	3(<1%)	0	432(9.0%)	35(9.4%)	11(0.7%)	0
Maternal Education								
Lower education	2894(26.9%)	217(30.7%)	1228(28.6%)	74(30.2%)	1374(28.6%)	123(33.2%)	286(17.3%)	20(22.2%)
Higher education	7617(70.7%)	470(66.6%)	3014(70.3%)	167(68.2%)	3357(69.9%)	243(65.5%)	1223(74.6%)	60(66.7%)
Healthcare use								
Average number of primary care consultations per year	5.21	6.82	4.21	5.49	6.20	7.89	4.91	6.01
Average number of admissions per year	0.11	0.50	0.11	0.32	0.12	0.60	0.08	0.60

Table 1: Characteristics of the cohort

	Outcome 1: primary care consultation rates				Outcome 2: Admission rates				Outcome 3: Referrals			
	Model 1: Univariate		Model 2: Multivariate		Model 1: Univariate		Model 2: Multivariate		Model 1: Univariate		Model 2: Multivariate	
	Coefficient	p	Coefficient	p	Coefficient	p	Coefficient	p	Coefficient	p	Coefficient	p
	(95% CI)		(95% CI)		(95% CI)		(95% CI)		(95% CI)		(95% CI)	
<b>Economic deprivation</b>												
Not Economically deprived	-	-	-	-	-	-	-	-	-	-	-	-
Economically deprived	0.11(0.08-0.14)	<0.001	0.01(-0.02-0.04)	0.48	0.35(0.27-0.42)	<0.001	0.21(0.13-0.29)	<0.001	0.95(0.35-1.56)	0.02	0.55(0.01-1.09)	0.045
<b>Ethnicity</b>												
White British	-	-	-	-	-	-	-	-	-	-	-	-
Pakistani	0.43(0.40-0.46)	<0.001	0.40(0.36-0.44)	<0.001	0.27(0.19-0.35)	<0.001	-0.02(-0.12-0.07)	0.64	0.72(0.09-1.36)	0.03	-0.28(-0.95-0.40)	0.42
Other	0.24(0.20-0.29)	<0.001	0.26(0.21-0.30)	<0.001	-0.08(-0.19-0.03)	0.15	-0.14(-0.25-0.03)	0.02	-0.03(-0.92-0.85)	0.94	-0.36(-1.11-0.39)	0.35
<b>Mothers age</b>												
20-34	-	-	-	-	-	-	-	-	-	-	-	-
<20	-0.13(-0.19-0.07)	<0.001	-0.01(-0.07-0.04)	0.68	0.11(-0.03-0.24)	0.13	0.07(-0.07-0.21)	0.32	-0.31(-1.39-0.77)	0.58	-0.42(-1.32-0.48)	0.36
>34	-0.06(-0.10-0.01)	<0.002	-0.03(-0.08-0.01)	0.13	-0.15(-0.27--0.04)	0.01	-0.17(-0.28-0.06)	0.002	0.83(-0.08-1.74)	0.07	0.64(-0.10-1.38)	0.09
<b>Consanguinity</b>												
Non-consanguineous	-	-	-	-	-	-	-	-	-	-	-	-

<b>First cousin</b>	0.03(-0.01-0.08)	<0.17	0.03(-0.01-0.08)	0.17	0.48(0.39-0.58)	<0.001	0.28(0.17-0.39)	<0.0001	1.19(0.48-1.91)	0.001	0.24(-0.50-0.97)	0.53
<b>Second cousin</b>	0.24(0.18-0.30)	<0.001	-0.01(-0.07-0.05)	0.78	0.24(0.09-0.39)	0.002	-0.01(-0.17-0.15)	0.94	1.17(0.11-2.23)	0.031	0.26(-0.70-1.21)	0.59
<b>Other blood</b>	0.25(0.18-0.32)	<0.001	0.01(-0.07-0.05)	0.86	0.31(0.14-0.48)	<0.001	0.23(0.05--0.41)	0.012	-0.03(-1.54-1.48)	0.96	-0.32(-1.60-0.97)	0.63
<b>Congenital Anomaly</b>												
<b>Yes</b>	0.29(0.23-0.35)	<0.001	0.24(1.18-0.30)	<0.001	1.46(1.35-1.57)	<0.001	1.48(1.36-1.59)	<0.0001	3.71(3.28-4.15)	<0.001	3.59(3.11-4.08)	<0.001
<b>No</b>	-	-	-	-	-	-	-	-	-	-	-	-
<b>Maternal Education</b>												
<b>Lower education</b>	-	-	-	-	-	-	-	-	-	-	-	-
<b>Higher education</b>	-0.04(-0.08-0.01)	<0.007	-0.04(-0.07-0.003)	0.031	-0.21(-0.29--0.13)	<0.001	-0.09(-0.17-0.01)	0.021	-0.70(-1.34-0.06)	0.032	0.06(-0.49-0.61)	0.83

Table 2: Univariate and multivariable coefficients for primary care consultations, use of hospital services and referrals to specialists adjusted for demographic and lifestyle factors in the BiB cohort.

Admittance type	Total number of admissions over a period of 5 years		Most common reason for admission	
	No anomaly	Anomaly	No anomaly	Anomaly
<b>Accident &amp; Emergency</b>	3985(49%)	609(26%)	1. Respiratory 2. Injury/poison	1. Respiratory 2. Infectious parasitic
<b>Other Emergency</b>	2522(31%)	932(40%)	1. Respiratory 2. Infectious parasitic	1. Respiratory 2. Clinical lab findings
<b>Elective</b>	1632(20%)	801(34%)	1. Eye/ear 2. Respiratory	1. Neoplasms/blood/immune 2. Congenital abnormalities

Table 3: Proportion of admissions by type, and most common reason for admission. Reasons for admission derived from ICD-10 codes at patient discharge, using ICD-10 groupings to categorise.<sup>33</sup>

## Interactions

Interaction effects between whether the child had a CA and economic deprivation were not significant in multivariable models.

## DISCUSSION

Our data suggest that children with CA have higher numbers of primary care consultations, admissions to hospital, and referrals to multidisciplinary specialists on average per year than children without CA. This finding is perhaps not surprising, but is now quantified. Children of Pakistani heritage have almost double the number of hospital admissions per year than children of white British heritage. Children with CA were predicted to require an increase in primary care consultations and hospital services compared to children without a CA. We found only one study reporting an increase in primary care consultations for children with

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3 CA, but heart CA specifically.<sup>26</sup> Although we find children from Pakistani heritage are  
4 predicted to use more primary care consultations than children without CA, this might be  
5 explained by more than half (53%) of children with CA in the BiB cohort being of Pakistani  
6 heritage (53%). When stratifying the analysis by CA, we also find children with CA from  
7 economically deprived neighborhoods have an increased risk of using hospital services, but  
8 not primary care consultations. This might be explained by previous findings from the BiB  
9 cohort suggesting mothers from poorer backgrounds are less likely to use primary care  
10 services due to variation in primary care practice provision.<sup>41</sup>  
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16 Our data suggests the use of hospital services are in higher demand than primary care  
17 consultations. Although an increased use of hospital services can be expected for children  
18 with CA<sup>12,22,24-26</sup> we find the type of hospital admission differs between children with and  
19 without CA. This is most likely explained by CA requiring more intensive treatment. For  
20 example although respiratory conditions were the most common reason for using hospital  
21 services overall (table 3), a finding similar to other studies,<sup>25</sup> 'other emergency' admissions  
22 were the most frequently used hospital service for children with CA. Other emergency refers  
23 to urgent referrals requiring corrective and sometimes surgical interventions that are initiated  
24 by health professionals, rather than parents presenting with their child at A & E. This increase  
25 in other emergency and elective procedures is a finding similar to that of other CA studies.<sup>12,27</sup>  
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32 Children with a CA were predicted to require more referrals to MDT specialists than children  
33 without CA, and have more than one specialist involved in their care simultaneously  
34 compared to children without CA. Although patient complexity increases the need for  
35 healthcare,<sup>14</sup> coordination of appointments for the multiple specialists required is also  
36 susceptible to variation, and is some times exacerbated by the divide between primary care,  
37 community and hospital services,<sup>3</sup> which is also associated with patient complications, late  
38 diagnosis, and an increased reliance on emergency care.<sup>16</sup> This suggests that although the  
39 predicted increase in use of hospital services for children with CA may be primarily due to  
40 their complex needs and ill health, there may be scope for this to be reduced through  
41 increasing the efficiency of care coordination.<sup>42</sup> In terms of clinical implications, our findings  
42 provide the quantified, longitudinal evidence requested by the Chief Medical Officer,  
43 supporting the suggestion of key workers as a catalyst for efficient patient navigation through  
44 services.<sup>1</sup> Also, when adjusting for ill health, the predicted increase in use of hospital services  
45 and multidisciplinary referrals for children with CA reduces (figure 1), suggesting higher  
46 usage may not be completely attributable to ill health, but affected by other factors such as  
47 deprivation and ethnicity.  
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3 There are limitations. Using a subset of diseases (CA), does not cover all children with  
4 conditions that may also be complex, however in order to extract a population we could be  
5 sure were both representative of complexity, and prevalent enough to create sample size  
6 groups large enough for comparison, CA were chosen based on the knowledge that they are  
7 high in numbers in Bradford and known to require complex care.<sup>43</sup> These results are based on  
8 the Bradford population, which might be interpreted as a limitation in terms of  
9 generalisability. However we feel these results are applicable to other populations or NHS  
10 trusts serving highly deprived and ethnically diverse groups of patients, characteristics which  
11 are known to be associated with CA.<sup>43</sup> Despite the successful linkage of primary care to  
12 cohort data in this study (97%), attributable to the complete SystemOne coverage of primary  
13 care practices in Bradford, the use of paper medical records for capturing referral activity is  
14 susceptible to missed information due to fluctuations in consultant record keeping and  
15 missing records. This research therefore illustrates the potential advantages of implementing a  
16 completely 'paperless' record keeping ethos, and the future emphasis for ensuring the  
17 exchange of data between IT systems in all clinical and care settings. This will only further  
18 strengthen the interpretability of key information at the point of care for patients with  
19 complex healthcare needs.<sup>44</sup>  
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### 29 **What is already known on this topic?**

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31 Anecdotally, it is felt that children with CA require increased healthcare use from both  
32 primary and secondary services, and the long-term management of their care requires careful  
33 coordination and multidisciplinary input. Longitudinal data is lacking to support this claim,  
34 however, and evidence on healthcare use is mainly in relation to congenital heart defects and  
35 hospital care based in the USA.  
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### 40 **What does this study add?**

41 We predict healthcare use combining primary care consultations, use of hospital services and  
42 referrals to specialists for children with CA linked to detailed socio demographic data from a  
43 longitudinal birth cohort. There are more primary care consultations for children with CA  
44 than children without, and also greater use of hospital services due to the severity of their  
45 conditions, and the specialist management these require.  
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### 50 **Contributors**

51 CB and BK had full access to all the data, conducted the analysis and statistical interpretation.  
52 RCP and NS contributed to the conception and design of the work, drafting and critical  
53 revision for intellectual content. All authors had final responsibility for the decision to submit  
54 for publication.  
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### No competing interests

All authors have completed the ICMJE uniform disclosure form at [www.icmje.org/coi\\_disclosure.pdf](http://www.icmje.org/coi_disclosure.pdf) and declare: no financial support or relationships from any organisation for the submitted work that might have an interest in the submitted work in the previous three years; no other relationships or activities that could appear to have influenced the submitted work.

### Funding

This paper presents independent research by a PhD candidate supported by a Bradford University studentship, in conjunction with the White Rose Consortium, and the National Institute for Health Research (NIHR), Collaboration for Leadership in Applied Health Research and Care (CLAHRC) Yorkshire and Humber programme “Healthy Children Healthy Families Theme”, IS-CLA-0113-10020.

### Acknowledgements

The views expressed in this paper are those of the authors, and not necessarily those of the National Health Service, the NIHR, or the Department of Health. We thank the families who took part in the Born in Bradford study, the midwives for their help in recruitment, the paediatricians and health visitors, the Born in Bradford team, which included interviewers, data managers, laboratory staff, clerical workers, research scientists, volunteers, and managers.

### Data sharing statement

No additional data are available

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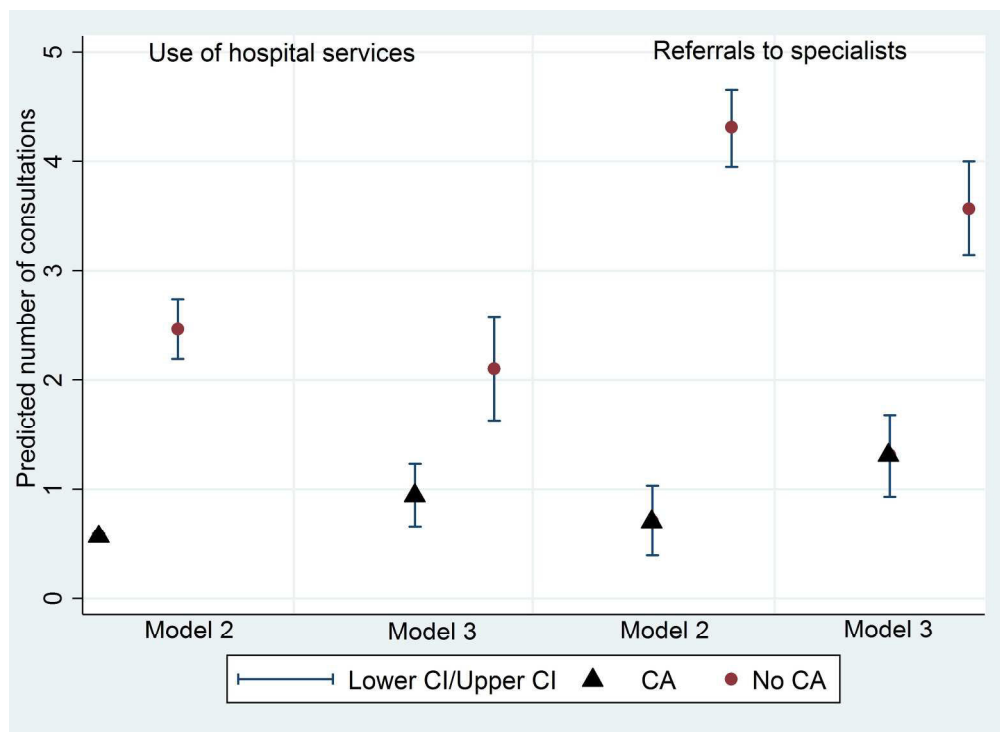
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Predicted use of hospital services and referrals to MDT specialists for children with and without CA, before and after controlling for ill health (Model 2 adjusted for confounders, model 3 adjusted for confounders and ill health)† † †

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# BMJ Open

## Healthcare use for children with complex needs: using routine health data linked to a multiethnic, ongoing birth cohort

Journal:	<i>BMJ Open</i>
Manuscript ID	bmjopen-2017-018419.R2
Article Type:	Research
Date Submitted by the Author:	15-Jan-2018
Complete List of Authors:	Bishop, Chrissy; University of Bradford, Faculty of Health Small, Neil; University of Bradford, School of Health Studies Parslow, Roger; University of Leeds, Leeds Institute of Genetics, Health and Therapeutics Kelly, Brian; Bradford Institute for Health Research
<b>Primary Subject Heading</b>:	Health services research
Secondary Subject Heading:	Health services research, Paediatrics
Keywords:	Community child health < PAEDIATRICS, PRIMARY CARE, HEALTH SERVICES ADMINISTRATION & MANAGEMENT

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3 **Healthcare use for children with complex needs: using routine health data linked to a**  
4 **multiethnic, ongoing birth cohort**  
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22 **ABSTRACT**

23 **Objectives**

24 Congenital anomalies (CA) are a leading cause of disease, death and disability for children  
25 throughout the world. Many have complex and varying healthcare needs which are not well  
26 understood. Our aim was to analyse the healthcare needs of children with CA and examine  
27 how that healthcare is delivered.  
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32 **Design**

33 Secondary analysis of observational data from the Born in Bradford study, a large prospective  
34 birth cohort, linked to primary care data and hospital episode statistics. Negative binomial  
35 regression with 95% confidence intervals was performed to predict healthcare use. The  
36 authors conducted a sub analysis on referrals to specialists using paper medical records for a  
37 sample of 400 children.  
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43 **Setting**

44 Primary, secondary and tertiary healthcare services in a large city in the north of England.  
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47 **Participants**

48 All children recruited to the birth cohort between March 2007 and December 2010. A total of  
49 706 children with CA and 10768 without were included in the analyses.  
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53 **Primary and secondary outcome measures**

54 Healthcare use for children with and without CA aged 0-<5 years was the primary outcome  
55 measure after adjustment for confounders.  
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## Results

Primary care consultations, use of hospital services and referrals to specialists were higher for children with CA than those without. Children in economically deprived neighborhoods were more likely to be admitted to hospital than consult primary care. Children with CA had a higher use of hospital services ( $\beta$  1.48, 95% CI 1.36-1.59) than primary care consultations ( $\beta$  0.24, 95% CI 1.18-0.30). Children with higher educated mothers were less likely to consult primary care and hospital services.

## Conclusions

Hospital services are most in demand for children with CA, but also for children who were economically deprived whether they had a CA or not. The complex nature of CA in children requires multidisciplinary management and strengthened coordination between primary and secondary care.

## Article summary

### Strengths and limitations of this study

- Linking birth cohort data to routine health data produces an enhanced dataset of socio demographic and clinical information.
- Ninety seven per cent of children from the birth cohort were linked to primary care and hospital episode data.
- Data linkage permitted a multi-service, longitudinal evaluation of healthcare use.
- We did not have access to electronic referrals, thus we performed a medical record review to extract this information for a subsample of 400 children.

## INTRODUCTION

Congenital anomalies (CA) are an abnormality of structure, function or metabolism, present at birth, which may result in mental, physical disability, or fatality.<sup>1</sup> The incidence of CA in Bradford is high; previously reported at 306 per 10,000 live births, compared to a national average of 227 per 10,000 live births.<sup>2-3</sup> Around 93% of children with a congenital anomaly (CA) survive to adulthood,<sup>2</sup> and a 20 year survival rate is estimated at 85.5% for children born with at least one CA,<sup>4</sup> some of whom will have complex conditions requiring multi-agency continuing care.<sup>5-6</sup> The healthcare needs of children with complex conditions have not been particularly well quantified in the past.<sup>7</sup> This may be due in part to a lack of longitudinal data capturing the multi-disciplinary care required by children with complex needs.<sup>2</sup> In the UK, primary care practice is ideally positioned for monitoring the care requirements of

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3 children with complex conditions such as CA, whose prognosis or care needs may change as  
4 they develop.<sup>2,8-10</sup> Monitoring childhood development across the life course, provides  
5 invaluable insight into the multidisciplinary care regimes children with complex needs  
6 require.<sup>11-17</sup> Multidisciplinary care requires the coordination of multiple specialists which may  
7 result in late diagnosis leading to reliance on emergency care rather than preventative  
8 solutions offered at primary care level, with significant increases in costs.<sup>18,19-22</sup> To represent  
9 the multidisciplinary care needs children with CA require, it has been suggested a  
10 combination of primary care consultations, use of hospital services, diagnosis codes,  
11 prescribed medications<sup>23</sup> and referral information<sup>24</sup> produce the best estimates of healthcare  
12 use.<sup>25</sup> The literature addressing such a comprehensive map of healthcare use for children with  
13 CA is limited, with the bulk of evidence coming from American studies investigating hospital  
14 use for the treatment of heart CA.<sup>18,24-27</sup> Only two studies were found which addressed the  
15 demand on primary care services for children with CA.<sup>28,29</sup> The need and demand for primary  
16 care services in particular are intensified by patient complexity, levels of deprivation, and  
17 primary care practice provision.<sup>16</sup>

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26 Our aims were therefore to explore healthcare use longitudinally for children with CA and  
27 without from birth up to their fifth birthday (0-5). We do this by linking demographic and  
28 socioeconomic data from a large prospective birth cohort covering a deprived and ethnically  
29 diverse population, to children's primary care records, hospital episodes statistics and referral  
30 information. In doing so this study examines the effects of having a CA, and consequential ill  
31 health, on primary care use, use of hospital services and referrals to multidisciplinary  
32 specialists. We also investigate the influence of demographic and socioeconomic factors on  
33 health care use.

## 34 35 36 37 38 39 40 **METHODS**

41 We used data from the Born in Bradford (BiB) cohort study, an on-going prospective birth  
42 cohort, which recruited 12450 pregnant women who gave informed consent for the study  
43 between 2007 and 2011. It monitors the health of mothers, their partners and birth outcomes  
44 for 13857 children. Detailed information on socioeconomic deprivation, demographics,  
45 clinical outcomes and risk factors are recorded. The methods for the BiB study are reported in  
46 detail elsewhere.<sup>30</sup>

### 47 48 49 50 51 **Case ascertainment and coding methods**

52 BiB recruits gave their consent for access to electronic primary care records and hospital  
53 episode statistics, which are split into elective, accident and emergency and other emergency  
54 admissions, here referred to as use of hospital services. We linked children's primary care  
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3 data held on SystmOne,<sup>31</sup> the patient contact single source system which has complete  
4 coverage in Bradford, and use of hospital services to BiB questionnaire data.<sup>30</sup> Linkage was  
5 performed using NHS number, surname, date of birth and gender between SystmOne<sup>31</sup> use of  
6 hospital services data and BiB. Of 13857 recruits, 97% were matched to primary care and use  
7 of hospital services data, forming the study population. The number of children with at least  
8 one (non-birth) hospital event was 5223 (38%). Hospital events included admissions for  
9 elective procedures, other emergencies, and A and E presentations. The average time over  
10 which data were recorded was 5.5 years, with a maximum of 7.6 years, in all 74386 person  
11 years of data. As not all children in the cohort had reached age seven, we censored our  
12 follow-up of these cases to aged 0–<5.

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19 Primary care data is a trusted source of CA ascertainment, including those diagnosed later in  
20 childhood.<sup>32,33</sup> We used cross mapping of SystemOne<sup>31</sup> diagnostic Clinical Terms Version 3  
21 (CTV-3) Read medical codes to International Classification of Diseases version 10 codes  
22 (ICD-10)<sup>34</sup> to classify and extract children with CA from the primary care database. We  
23 followed the European Surveillance of Congenital Anomalies (EUROCAT) guidelines, using  
24 the British Isles National Organisation of CA Registers (BINOCAR) methodology,<sup>35</sup> which  
25 advise selection of major CA and removal of minor CA. A clinical geneticist reviewed  
26 classifications. A total of 860 children with CA were identified, and 154 were excluded, as  
27 they had no linked BiB questionnaire data.

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34 Extracting information from paper medical records was necessary to capture multidisciplinary  
35 outpatient data and referral activity, as this is sometimes not routinely included in electronic  
36 records.<sup>36</sup> We extracted referral information from a review of paper medical records using a  
37 sample of 200 children with and 200 without CA, selected at random from the BiB cohort.  
38 The small sample size was chosen based on the exploratory nature of the medical record  
39 review, and the feasibility of performing this by hand within the time scale of this study. A  
40 standardised data extraction form was designed, reviewed by a clinician, and piloted to  
41 accumulate the number and type of referrals to different multidisciplinary services.

### 42 43 44 45 46 47 **Statistical analysis**

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49 We had three outcomes for this study. The number of primary care consultations, use of  
50 hospital services and referrals to multidisciplinary specialists. Both primary care consultations  
51 and use of hospital services were counted as one per day, even if multiple appointments in the  
52 same day were recorded, as many of the appointments occurring on the same day were  
53 episodes that ran over time, or were duplicates. Negative binomial regression models were  
54 used to model primary consultations and use of hospital services as they account for the over  
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3 dispersion in count data. These models use an exposure variable, which indicates the number  
4 of times the event could have happened. Primary care consultations and use of hospital  
5 services were expressed per year of observed primary care registered time, which takes into  
6 account any periods the child may not have been registered with the primary care practice,  
7 withdrawals from the cohort, or deaths. We performed a sub analysis reporting regression  
8 coefficients for the outcome multidisciplinary referrals.  
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13 We used three models to compare regression coefficients for each of our three outcomes.  
14 Model 1 included univariate analyses, thought of as what is actually observed. Model 2  
15 adjusts for other covariates we determined as confounding factors. Model 3 adjusts for  
16 confounders and measures of underlying ill health. Adjusting for ill health using a measure of  
17 multi-morbidity, is a recognised method of risk adjustment for evaluations of healthcare use,  
18 as severity of illness may not solely be due to multi-morbidity and ill health, it may also be  
19 due to other patient characteristics.<sup>37</sup> We used a count of unique prescriptions and a count of  
20 the number of comorbidities per child as measures of ill health. Simple counts of distinct  
21 medications have been suggested as an accurate measure of ill health, given chronic  
22 conditions frequently require repeat prescriptions<sup>37,38</sup> as have counts of comorbidities in  
23 primary care settings.<sup>39,40</sup> We performed a test for interaction between whether the child had a  
24 CA and level of deprivation for primary care consultations and use of hospital services. We  
25 also report the predicted rates of healthcare use for children with CA and without (figure 1).  
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### 34 **Confounders**

35 We used directed acyclic graphs to determine the minimally sufficient confounding set for all  
36 of our models and checked whether the inclusion of additional covariates improved the model  
37 more than would be expected by chance using appropriate model fit statistics. These consisted  
38 of maternal age (<20, 20-34, >34 years); educational attainment (Low education [ $<5$  GCSE  
39 equivalents or other education] High education [ $5 >$  GCSE equivalents at grade A–C or two  
40 advanced level certificates or diploma, degree or higher degree]),<sup>41</sup> economic deprivation  
41 (Economically deprived, not economically deprived [measured using a means-tested benefit  
42 status. In the UK, being in receipt of means-tested benefits is recognised as measure of  
43 income poverty, as these benefits are frequently the only source of income and are paid at  
44 rates that put individuals below standard poverty lines]),<sup>42</sup> ethnicity (White British, Pakistani,  
45 Other), and consanguinity (Non-consanguineous, first cousin, second cousin, other blood[any  
46 relation]). All covariates were entered into the model as a categorical variable to allow for  
47 possible non-linearity in the relationship between the multi-morbidity measure and relevant  
48 outcome.  
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### **Ethics**

Ethics approval for the cohort study was provided by Bradford Local Research Ethics Committee (reference 06/Q1202/48).

### **Role of the funding source**

The sponsors of this study had no role in the study design, data collection, analysis or interpretation, or writing of the report. CB, and BK had full access to all the data in the study and all authors had final responsibility for the decision to submit for publication.

### **RESULTS**

Table 1 shows the characteristics of mothers who gave birth to children with a CA in the BiB cohort (CA = 706, no CA= 10768), and average healthcare use up to the child's fifth birthday. The BiB cohort is multiethnic, 40% white British, 45% Pakistani and 15% 'other' ethnicities. Of all children with a CA, 53% were of Pakistani heritage, compared to 35% white British and 13% 'other' ethnicities. Forty nine per cent of Pakistani children with CA were from first cousin unions compared to <1% of white British children with CA from first cousin unions. Children of Pakistani heritage with CA had on average 1.61 more primary care consultations, and 0.39 more hospital admissions per year than children without CA. Children with CA of Pakistani heritage had the highest number of primary care appointments over the five-year period, with on average 2.4 more primary care appointments than children of white British heritage.

Children with CA of Pakistani heritage and 'other ethnicities', had the same use of hospital services on average, but this was almost double for children with CA of white British heritage (Table 1). Table 2 reports the regression coefficients for the univariate and multivariable analysis of primary care and use of hospital services. Sixty three per cent of children with CA were born into economically deprived neighborhoods (table 1). Both the adjusted and unadjusted rates suggest children from economically deprived neighborhoods have an increased use of hospital services ( $\beta$  0.35, 95% CI 0.27-0.42), but do not use more primary care consultations. Although the most common reason for use of hospital services for children with and without CA was respiratory conditions, when stratified by admission type, children with CA had the most 'other emergency' admissions overall (40%), followed by elective admissions (34%) whereas children without CA had an increase of 'Accident & Emergency' (45%) admissions (Table 3). Diagnoses on admission were also different between groups, with neoplasms and clinical lab findings recorded as the most common reason for admission for children with CA, not recorded in children without CA (table 3).

Children from both Pakistani heritage and other ethnicities were predicted to require an increase in primary care consultations in both the univariate and multivariable analyses. Children who had older mothers (>34) were predicted to use hospital services less ( $\beta$  -0.17, 95% CI -0.28 -0.06), but not primary care consultations ( $\beta$  -0.03, 95% CI -0.08 -0.01) after adjustment for confounders. Children born into consanguineous families were predicted to have an increased use of hospital services, but not primary care consultations. Children with a CA had the largest increase in use of primary care consultations ( $\beta$  0.24, 95% CI 1.18-0.30), and use of hospital services ( $\beta$  1.48, 95% CI 1.36-1.59) after adjustment for confounders, for which usage was almost three times higher. Higher maternal education reduced primary care consultations and use of hospital services (Table 2). The sub analysis of multidisciplinary referrals predicts an increased use of specialist referrals for children with CA after adjustment for confounders ( $\beta$  3.59, 95% CI 3.11-4.08). The only other significant factor was children born into economically deprived neighborhoods ( $\beta$  0.55, 95% CI 0.01-1.09). We adjusted for ill health for services with the highest predicted usage, those being hospital services and multidisciplinary referrals for children with and without CA (figure 1). After controlling for ill health, the predicted increased use of hospital services for children with CA reduces by almost half but still remains ( $\beta$  0.80, 95% CI 0.63-0.97), as does the predicted increased use of multidisciplinary referrals ( $\beta$  2.27, 95% CI 1.59-2.94) (Figure 1).

*Insert figure 1 here*

From a possible 41 different specialists, the most common referral was to consultant pediatricians (14% 59/400), followed by neonatology (13% 57/400), pediatric surgery (8% 34/400) and local cardiology services (7% 30/400). On average children had between three and four specialists involved in their care simultaneously.

	All		White British		Pakistani		Other	
	No anomaly	Anomaly	No anomaly	Anomaly	No anomaly	Anomaly	No anomaly	Anomaly
<b>Ethnic origin</b>	10768 (93.9%)	706(6.2%)	4288(94.6%)	245(5.4%)	4804(92.8%)	371(7.2%)	1653(94.8%)	90(5.2%)
<b>Economic deprivation</b>								
Economically deprived	6147(57.1%)	444(62.9%)	2072(48.3%)	116(47.4%)	3315(69.0%)	280(75.5%)	758(45.9%)	48(53.3%)
Not economically deprived	4166(38.7%)	247(35.0%)	2034(47.4%)	122(49.8%)	1340(27.9%)	85(22.9%)	792(47.9%)	40(44.4%)
Missing	455(4.2%)	15(2.1%)	182(4.2%)	7(2.9%)	149(3.1%)	6(1.6%)	103(6.2%)	2(2.2%)
<b>Age of mother</b>								
20-34	8716(80.9%)	554(78.5%)	3231(75.4%)	180(73.5%)	4099(85.3%)	312(84.1%)	1366(82.6%)	62(68.9%)
<20	776(7.2%)	51(7.2%)	536(12.5%)	29(11.8%)	148(3.1%)	15(4.0%)	92(5.6%)	7(7.8%)
>34	1276(11.9%)	101(14.3%)	521(12.2%)	36(14.7%)	557(11.6%)	44(11.9%)	195(11.8%)	21(23.3%)
<b>Consanguinity</b>								

Non-consanguineous	7850(72.9%)	424(60.1%)	4284(99.8%)	224(99.6%)	2008(41.8%)	102(27.5%)	1538(93.0%)	78(86.7%)
First Cousin	1834(17.0%)	192(27.2%)	1(<1%)	1(<1%)	1753(36.5%)	183(49.3%)	79(4.8%)	8(8.9%)
Second Cousin	637(5.9%)	55(7.8%)	0	0	611(12.7%)	51(13.8%)	25(1.5%)	4(4.4%)
Other blood	447(4.2%)	35(5.0%)	3(<1%)	0	432(9.0%)	35(9.4%)	11(0.7%)	0
<b>Maternal Education</b>								
Lower education	2894(26.9%)	217(30.7%)	1228(28.6%)	74(30.2%)	1374(28.6%)	123(33.2%)	286(17.3%)	20(22.2%)
Higher education	7617(70.7%)	470(66.6%)	3014(70.3%)	167(68.2%)	3357(69.9%)	243(65.5%)	1223(74.6%)	60(66.7%)
<b>Healthcare use</b>								
Average number of primary care consultations per year	5.21	6.82	4.21	5.49	6.20	7.89	4.91	6.01
Average number of admissions per year	0.11	0.50	0.11	0.32	0.12	0.60	0.08	0.60

Table 1: Characteristics of the cohort

	Outcome 1: primary care consultation rates				Outcome 2: Admission rates				Outcome 3: Referrals			
	Model 1: Univariate		Model 2: Multivariate		Model 1: Univariate		Model 2: Multivariate		Model 1: Univariate		Model 2: Multivariate	
	Coefficient	p	Coefficient	p	Coefficient	P	Coefficient	p	Coefficient	p	Coefficient	p
	(95% CI)		(95% CI)		(95% CI)		(95% CI)		(95% CI)		(95% CI)	
<b>Economic deprivation</b>												
Not Economically deprived	-	-	-	-	-	-	-	-	-	-	-	-
Economically deprived	0.11(0.08-0.14)	<0.001	0.01(-0.02-0.04)	0.48	0.35(0.27-0.42)	<0.001	0.21(0.13-0.29)	<0.001	0.95(0.35-1.56)	0.02	0.55(0.01-1.09)	0.045
<b>Ethnicity</b>												
White British	-	-	-	-	-	-	-	-	-	-	-	-
Pakistani	0.43(0.40-0.46)	<0.001	0.40(0.36-0.44)	<0.001	0.27(0.19-0.35)	<0.001	-0.02(-0.12-0.07)	0.64	0.72(0.09-1.36)	0.03	-0.28(-0.95-0.40)	0.42
Other	0.24(0.20-0.29)	<0.001	0.26(0.21-0.30)	<0.001	-0.08(-0.19-0.03)	0.15	-0.14(-0.25-0.03)	0.02	-0.03(-0.92-0.85)	0.94	-0.36(-1.11-0.39)	0.35
<b>Mothers age</b>												
20-34	-	-	-	-	-	-	-	-	-	-	-	-
<20	-0.13(-0.19-0.07)	<0.001	-0.01(-0.07-0.04)	0.68	0.11(-0.03-0.24)	0.13	0.07(-0.07-0.21)	0.32	-0.31(-1.39-0.77)	0.58	-0.42(-1.32-0.48)	0.36
>34	-0.06(-0.10-0.01)	<0.002	-0.03(-0.08-0.01)	0.13	-0.15(-0.27-0.04)	0.01	-0.17(-0.28-0.06)	0.002	0.83(-0.08-1.74)	0.07	0.64(-0.10-1.38)	0.09
<b>Consanguinity</b>												
Non-consanguineous	-	-	-	-	-	-	-	-	-	-	-	-
First cousin	0.03(-0.01-0.08)	<0.17	0.03(-0.01-0.08)	0.17	0.48(0.39-0.58)	<0.001	0.28(0.17-0.39)	<0.001	1.19(0.48-1.91)	0.01	0.24(-0.50-0.97)	0.53
Second cousin	0.24(0.18-0.30)	<0.001	-0.01(-0.07-0.05)	0.78	0.24(0.09-0.39)	0.002	-0.01(-0.17-0.15)	0.94	1.17(0.11-2.23)	0.031	0.26(-0.70-1.21)	0.59
Other blood	0.25(0.18-	<0.001	0.01(-0.07-	0.86	0.31(0.14-0.48)	<0.001	0.23(0.05--0.41)	0.012	-0.03(-1.54-	0.96	-0.32(-1.60-	0.63



	0.32)	0.05)			1.48)	0.97)						
<b>Congenital Anomaly</b>												
<b>Yes</b>	0.29(0.23-0.35)	<0.001	0.24(1.18-0.30)	<0.001	1.46(1.35-1.57)	<0.001	1.48(1.36-1.59)	<0.001	3.71(3.28-4.15)	<0.001	3.59(3.11-4.08)	<0.001
<b>No</b>	-	-	-	-	-	-	-	-	-	-	-	-
<b>Maternal Education</b>												
<b>Lower education</b>	-	-	-	-	-	-	-	-	-	-	-	-
<b>Higher education</b>	-0.04(-0.08-0.01)	<0.007	-0.04(-0.07-0.003)	0.031	-0.21(-0.29--0.13)	<0.001	-0.09(-0.17-0.01)	0.021	-0.70(-1.34-0.06)	0.032	0.06(-0.49-0.61)	0.83

Table 2: Univariate and multivariable coefficients for primary care consultations, use of hospital services and referrals to specialists adjusted for demographic and lifestyle factors in the BiB cohort.

Admittance type	Total number of admissions over a period of 5 years		Most common reason for admission	
	No anomaly	Anomaly	No anomaly	Anomaly
<b>Accident &amp; Emergency</b>	3985(49%)	609(26%)	1. Respiratory 2. Injury/poison	1. Respiratory 2. Infectious parasitic
<b>Other Emergency</b>	2522(31%)	932(40%)	1. Respiratory 2. Infectious parasitic	1. Respiratory 2. Clinical lab findings
<b>Elective</b>	1632(20%)	801(34%)	1. Eye/ear 2. Respiratory	1. Neoplasms/blood/immune 2. Congenital abnormalities

Table 3: Proportion of admissions by type, and most common reason for admission. Reasons for admission derived from ICD-10 codes at patient discharge, using ICD-10 groupings to categorise.<sup>35</sup>

### Interactions

Interaction effects between whether the child had a CA and economic deprivation were not significant in multivariable models.

### DISCUSSION

Our data suggest that children with CA have higher numbers of primary care consultations, admissions to hospital, and referrals to multidisciplinary specialists on average per year than children without CA. This finding is perhaps not surprising, but is now quantified. Children of Pakistani heritage have almost double the number of hospital admissions per year than children of white British heritage. Children with CA were predicted to require an increase in primary care consultations and hospital services compared to children without a CA. We found only one study reporting an increase in primary care consultations for children with CA, but heart CA specifically.<sup>28</sup> Although we find children from Pakistani heritage are predicted to use more primary care consultations than children without CA, this might be explained by more than half (53%) of children with CA in the BiB cohort being of Pakistani heritage (53%). When stratifying the analysis by CA, we also find children with CA from

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3 economically deprived neighborhoods have an increased risk of using hospital services, but  
4 not primary care consultations. This might be explained by previous findings from the BiB  
5 cohort suggesting mothers from poorer backgrounds are less likely to use primary care  
6 services due to variation in primary care practice provision.<sup>43</sup>  
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10 Our data suggests the use of hospital services are in higher demand than primary care  
11 consultations. Although an increased use of hospital services can be expected for children  
12 with CA<sup>14,24,26-28</sup> we find the type of hospital admission differs between children with and  
13 without CA. This is most likely explained by CA requiring more intensive treatment. For  
14 example although respiratory conditions were the most common reason for using hospital  
15 services overall (table 3), a finding similar to other studies,<sup>27</sup> ‘other emergency’ admissions  
16 were the most frequently used hospital service for children with CA. Other emergency refers  
17 to urgent referrals requiring corrective and sometimes surgical interventions that are initiated  
18 by health professionals, rather than parents presenting with their child at A & E. This increase  
19 in other emergency and elective procedures is a finding similar to that of other CA studies.<sup>14,29</sup>  
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26 Children with a CA were predicted to require more referrals to MDT specialists than children  
27 without CA, and have more than one specialist involved in their care simultaneously  
28 compared to children without CA. Although patient complexity increases the need for  
29 healthcare,<sup>16</sup> coordination of appointments for the multiple specialists required is also  
30 susceptible to variation, and is some times exacerbated by the divide between primary care,  
31 community and hospital services,<sup>2</sup> which is also associated with patient complications, late  
32 diagnosis, and an increased reliance on emergency care.<sup>18</sup> This suggests that although the  
33 predicted increase in use of hospital services for children with CA may be primarily due to  
34 their complex needs and ill health, there may be scope for this to be reduced through  
35 increasing the efficiency of care coordination.<sup>44</sup> In terms of clinical implications, our findings  
36 provide the quantified, longitudinal evidence requested by the Chief Medical Officer,  
37 supporting the suggestion of key workers as a catalyst for efficient patient navigation through  
38 services.<sup>3</sup> Also, when adjusting for ill health, the predicted increase in use of hospital services  
39 and multidisciplinary referrals for children with CA reduces (figure 1), suggesting higher  
40 usage may not be completely attributable to ill health, but affected by other factors such as  
41 deprivation and ethnicity.  
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51 There are limitations. Using a subset of diseases (CA), does not cover all children with  
52 conditions that may also be complex, however in order to extract a population we could be  
53 sure were both representative of complexity, and prevalent enough to create sample size  
54 groups large enough for comparison, CA were chosen based on the knowledge that they are  
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3 high in numbers in Bradford and known to require complex care.<sup>45</sup> These results are based on  
4 the Bradford population, which might be interpreted as a limitation in terms of  
5 generalisability. However we feel these results are applicable to other populations or NHS  
6 trusts serving highly deprived and ethnically diverse groups of patients, characteristics which  
7 are known to be associated with CA.<sup>45</sup> Despite the successful linkage of primary care to  
8 cohort data in this study (97%), attributable to the complete SystemOne coverage of primary  
9 care practices in Bradford, the use of paper medical records for capturing referral activity is  
10 susceptible to missed information due to fluctuations in consultant record keeping,  
11 interpreting handwritten entries and missing records. This research therefore illustrates the  
12 potential advantages of implementing a completely 'paperless' record keeping ethos, and the  
13 future emphasis for ensuring the exchange of data between IT systems in all clinical and care  
14 settings. This will only further strengthen the interpretability of key information at the point  
15 of care for patients with complex healthcare needs.<sup>46</sup>  
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#### 24 **What is already known on this topic?**

25 Anecdotally, it is felt that children with CA require increased healthcare use from both  
26 primary and secondary services, and the long-term management of their care requires careful  
27 coordination and multidisciplinary input. Longitudinal data is lacking to support this claim,  
28 however, and evidence on healthcare use is mainly in relation to congenital heart defects and  
29 hospital care based in the USA.  
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#### 34 **What does this study add?**

35 We predict healthcare use combining primary care consultations, use of hospital services and  
36 referrals to specialists for children with CA linked to detailed socio demographic data from a  
37 longitudinal birth cohort. There are more primary care consultations for children with CA  
38 than children without, and also greater use of hospital services due to the severity of their  
39 conditions, and the specialist management these require.  
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#### 44 **Contributors**

45 CB and BK had full access to all the data, conducted the analysis and statistical interpretation.  
46 RCP and NS contributed to the conception and design of the work, drafting and critical  
47 revision for intellectual content. All authors had final responsibility for the decision to submit  
48 for publication.  
49  
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#### 53 **No competing interests**

54 All authors have completed the ICMJE uniform disclosure form  
55 at [www.icmje.org/coi\\_disclosure.pdf](http://www.icmje.org/coi_disclosure.pdf) and declare: no financial support or relationships from  
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any organisation for the submitted work that might have an interest in the submitted work in the previous three years; no other relationships or activities that could appear to have influenced the submitted work.

### Funding

This paper presents independent research by a PhD candidate supported by a Bradford University studentship, in conjunction with the White Rose Consortium, and the National Institute for Health Research (NIHR), Collaboration for Leadership in Applied Health Research and Care (CLAHRC) Yorkshire and Humber programme “Healthy Children Healthy Families Theme”, IS-CLA-0113-10020.

### Acknowledgements

The views expressed in this paper are those of the authors, and not necessarily those of the National Health Service, the NIHR, or the Department of Health. We thank the families who took part in the Born in Bradford study, the midwives for their help in recruitment, the paediatricians and health visitors, the Born in Bradford team, which included interviewers, data managers, laboratory staff, clerical workers, research scientists, volunteers, and managers.

### Data sharing statement

No additional data are available

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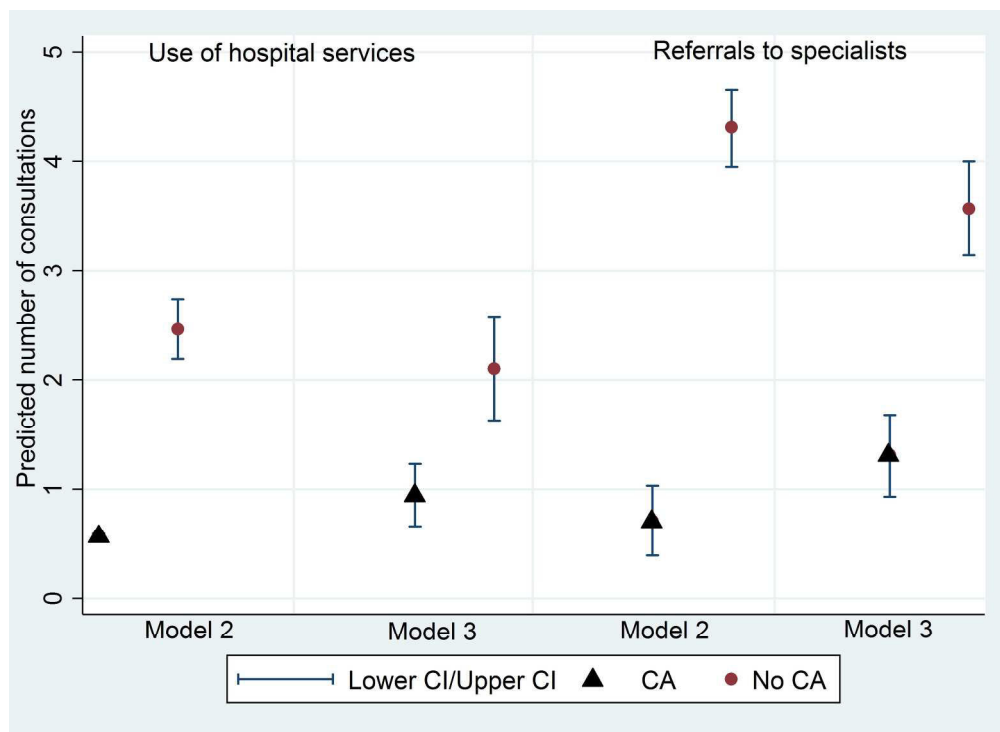
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10 Figure legend  
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13 Figure 1: Predicted use of hospital services and referrals to MDT specialists for children with  
14 and without CA, before and after controlling for ill health (Model 2 adjusted for confounders,  
15 model 3 adjusted for confounders and ill health)  
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Predicted use of hospital services and referrals to MDT specialists for children with and without CA, before and after controlling for ill health (Model 2 adjusted for confounders, model 3 adjusted for confounders and ill health)† † †

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