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

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## ABSTRACT

### Objectives

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

### Design

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

### Setting

Primary care data, hospital episode statistics, and referrals to specialists linked to birth cohort data.

### Participants

All children who were recruited to the birth cohort between March 2007 and December 2010 were eligible for the study. A total of 706 and 10768 children were included in the analyses with and without CA respectively.

### Primary and secondary outcome measures

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

**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 (β 1.48, 95% CI 1.36-1.59) than primary care consultations (β 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,[1] and a 20 year survival rate is estimated at 85.5% for children born with at least one CA,[2] some of whom will have complex conditions requiring multi-agency continuing care.[3-4] The healthcare needs of children with complex conditions have not been particularly well quantified in the past.[5] 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.[1] 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

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

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

## METHODS

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

*Insert figure 1 here*

### Case ascertainment and coding methods

BiB recruits gave their consent for access to electronic primary care records and hospital episode statistics, which are split into elective, accident and emergency and other emergency

admissions, thus referred to as use of hospital services. We linked children's primary care data held on SystmOne,[29] the patient contact single source system which has complete coverage in Bradford, and use of hospital services to BiB questionnaire data. Linkage was performed using NHS number, surname, date of birth and gender between SystmOne,[29] use of hospital services and BiB. Of 13857 recruits, 97% were matched to primary care and use of hospital services data, forming the study population. The amount of children with at least one (non-birth) hospital event was 5223 (38%). The average time over which data were recorded was 5.5 years, with a maximum of 7.6 years, in all 74386 person years of data. As not all children in the cohort had reached age seven, we censored our follow-up of these cases to ages 0<5.

Primary care data is a trusted source of ascertainment of CA, including those diagnosed later in childhood.[30,31] We used cross mapping of SystemOne[31] diagnostic CTV3 Read medical codes to ICD-10 codes[32] to classify and extract children with CA from the primary care database. We followed the European Surveillance of Congenital Anomalies (EUROCAT) guidelines, using the British Isles National Organisation of CA Registers (BINOCAR) methodology[33] to differentiate between major and minor CA. Minor anomalies were excluded, and a clinical geneticist reviewed classifications.

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.[34] We extracted referral information from a review of paper medical records using a convenience 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 same day were recorded, as many of the appointments occurring on the same day were episodes that ran over time, or were duplicates. Negative binomial poisson regression models were used to model primary consultations and use of hospital services as they account for the over dispersion in count data. These models use an exposure variable, which indicates the number of times the event could have happened. Primary care consultations and use of

hospital services were expressed per year of observed primary care registered time, which takes into account any periods the child may not have been registered with the primary care practice, withdrawals from the cohort, or deaths. We performed a sub analysis reporting regression coefficients for the outcome multidisciplinary referrals.

We used three models to compare regression coefficients for CA for each of our three outcomes. Model 1 included univariate analyses, thought of as what is actually observed. Model 2 adds other covariates we determined as confounding factors. Model 3 controls for confounders and adds measures of underlying ill health. Controlling for ill health using a measure if multi-morbidity, is a recognised method of risk adjustment for evaluations of healthcare use, as severity of illness may not solely be due to multi-morbidity and ill health, it may also be due to other patient characteristics.[35] We used a count of unique prescriptions and a count of the number of comorbidities per child as measures of ill health. Simple counts of distinct medications have been suggested as an accurate measure of ill health, given chronic conditions frequently require repeat prescriptions[35,36] as have counts of comorbidities in primary care settings.[37,38] We performed a test for interaction between whether the child had a CA and level of deprivation for primary care consultations and use of hospital services. We also report the predicted rates of healthcare use for children with CA and without (figure 2).

**Confounders**

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

**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. Children of Pakistani heritage were born with the highest proportion of CA (53%), compared to white British children (35%) and 'other' ethnicities (13%) (table 1). Forty nine per cent of children with CA were from Pakistani first cousin unions compared to <1% of children with CA from white British 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). 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). 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 (β 0.35, 95% CI 0.27-0.42), but do not use more primary care consultations.

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 (β -0.17, 95% CI -0.28 -0.06), but not primary care consultations (β -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 (β 0.24, 95% CI 1.18-0.30), and use of hospital services (β 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 (β 3.59, 95% CI 3.11-4.08). The only other significant factor was children born into economically deprived neighborhoods (β 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 (β 0.80, 95% CI 0.63-0.97), as does the predicted increased use of multidisciplinary referrals (β 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 |
| **Ethnic origin** | 10768 (93.85%) | 706(6.15 %) | 4288(94.60%) | 245(5.40%) | 4804(92.83 %) | 371(7.17 %) | 1653(94.84%) | 90(5.16%) |
| **Economic deprivation** | | | | | | | | |
| 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%) |
| **Age of mother** | | | | | | | | |
| 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%) |
| **Consanguinity** | | | | | | | | |
| 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 |
| **Maternal Education** | | | | | | | | |

| | | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| 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%) |
| **Healthcare use** | | | | | | | | |
| 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 (95% CI)* | *p* | *Coefficient (95% CI)* | *p* | *Coefficient (95% CI)* | *P* | *Coefficient (95% CI)* | *p* | *Coefficient (95% CI)* | *p* | *Coefficient (95% CI)* | *p* |
| **Economic deprivation** | | | | | | | | | | | | |
| **Economically deprived** | 0.11(0.08-0.14) | <0.0001 | 0.01(-0.02-0.04) | 0.48 | 0.35(0.27-0.42) | <0.0001 | 0.21(0.13-0.29) | <0.0001 | 0.95(0.35 1.56) | 0.002 | 0.55(0.01-1.09) | 0.045 |
| Not Economically deprived | - | - | - | - | - | - | | | - | | - | - |
| **Ethnicity** | | | | | | | | | | | | |
| **White British** | - | - | - | - | - | - | - | - | - | - | - | - |
| **Pakistani** | 0.43(0.40-0.46) | <0.0001 | 0.40(0.36-0.44) | <0.0001 | 0.27(0.19-0.35) | <0.0001 | -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) | <0.0001 | 0.26(0.21-0.30 | <0.0001 | -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 |
| **Mothers age** | | | | | | | | | | | | |
| **20-34** | - | - | - | - | | | - | - | - | - | - | - |
| **<20** | -0.13(-0.19- -0.07) | <0.0001 | -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) | <0.02 | -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 |
| **Consanguinity** | | | | | | | | | | | | |
| **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) | <0.0001 | 0.28(0.17-0.39) | <0.0001 | 1.19(0.48-1.91) | 0.001 | 0.24(-0.50-0.97) | 0.53 |
| **Second cousin** | 0.24(0.18-0.30) | <0.0001 | -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) | <0.0001 | 0.01(-0.07-0.05) | 0.86 | 0.31(0.14-0.48) | <0.0001 | 0.23(0.05 - -0.41) | 0.012 | -0.03(-1.54-1.48) | 0.96 | -0.32(-1.60-0.97) | 0.63 |
| **Congenital Anomaly** | | | | | | | | | | | | |
| **Yes** | 0.29(0.23-0.35) | <0.0001 | 0.24(1.18-0.30) | <0.0001 | 1.46(1.35-1.57) | <0.0001 | 1.48(1.36-1.59) | <0.0001 | 3.71(3.28-4.15) | <0.00001 | 3.59(3.11-4.08) | <0.0001 |
| **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<br>2. Injury/poison | 1. Respiratory<br>2. Infectious parasitic |
| **Other Emergency** | 2522(31%) | 932(40%) | 1. Respiratory<br>2. Infectious parasitic | 1. Respiratory<br>2. Clinical lab findings |
| **Elective** | 1632(20%) | 801(34%) | 1. Eye/ear<br>2. Respiratory | 1. Neoplasms/blood/immune<br>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 (β 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 (β 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.[26] Although we find children from Pakistani heritage are predicted to use more primary care consultations (β 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.[39]

An increased use of hospital services can be expected for children with CA[12,22,24-26] we find the type of hospital admission differs between children with and without CA. This is most likely explained by CA requiring more severe treatment. For example although respiratory conditions were the most common reason for using hospital services overall (table 3), a finding similar to other studies,[25] 'other emergency' admissions were the most frequently used hospital service for children with CA. Other emergency refers to procedures of urgency requiring corrective and sometimes surgical interventions that are initiated by health professionals, rather than parents presenting with their child at A & E. This increase in other emergency and elective procedures is a finding similar to that of other CA studies.[12,27]

Children with a CA were predicted to require more referrals to MDT specialists (β 37.97, 95% CI 18.19-79.26) than children without CA, and have more than one specialist involved in their care simultaneously compared to children without CA. Although patient complexity increases the need for healthcare,[14] coordination of appointments for the multiple specialists required is also susceptible to variation, and is some times exacerbated by the divide between primary care, community and hospital services,[3] which is also associated with patient complications, late diagnosis, and an increased reliance on emergency care.[16] This suggests that although the predicted increase in use of hospital services for children with CA may be primarily due to their complex needs and ill health, there may be scope for this to be reduced through increasing the efficiency of care coordination.[40] Our findings therefore support the suggestion of key workers as a potential solution for efficient coordination, with the aim of streamlining navigation through services.[1] Also, when adjusting for ill health, the predicted increase in use of hospital services and multidisciplinary referrals for children with CA reduces (figure 2), suggesting higher usage may not be completely attributable to ill health, but affected by other factors such as deprivation and ethnicity.

There are limitations. Using a subset of diseases (CA), does not cover all children with conditions that may also be complex, however in order to extract a population we could be sure were both representative of complexity, and prevalent enough to create sample size groups large enough for comparison, CA were chosen based on the knowledge that they are high in numbers in Bradford and known to require complex care.[41] These results are based on the Bradford population, which might be interpreted as a limitation in terms of generisability. However we feel these results are applicable to other populations or NHS trusts serving highly deprived and ethnically diverse groups of patients, determinants which are also known to be associated with CA.[41] Despite the successful linkage of primary care to cohort data in this study (97%), attributable to the complete SystmOne coverage of primary care practices in Bradford, the use of paper medical records for capturing referral activity is susceptible to

missed information due fluctuations in consultant record keeping and missing records. This research therefore illustrates the advantages of implementing a completely 'paperless' record keeping ethos, and the future emphasis for ensuring the exchange of data between IT systems in all clinical and care settings. This will only further strengthen the interpretability of key information at the point of care for patients with complex healthcare needs.[42]

**What is already known on this topic?**

Children with CA do require increased healthcare use from both primary and secondary services, and the long-term management of their care requires careful coordination and multidisciplinary input. Longitudinal data is lacking to support this claim and evidence on healthcare use is mainly in relation to congenital heart defects and hospital care based in the USA.

**What does this study add?**

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

**Contributors**

CB and BK had full access to all the data, conducted the analysis and statistical interpretation. RCP and NS contributed to the conception and design of the work, drafting and critical revision for intellectual content. All authors had final responsibility for the decision to submit for publication.

**No competing interests**

All authors have completed the ICMJE uniform disclosure form at 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.

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**Data sharing statement**

No additional data are available

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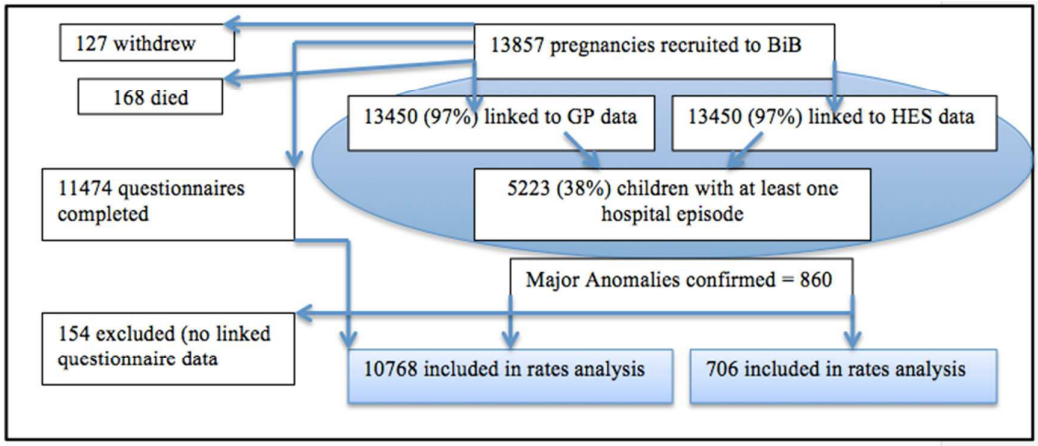

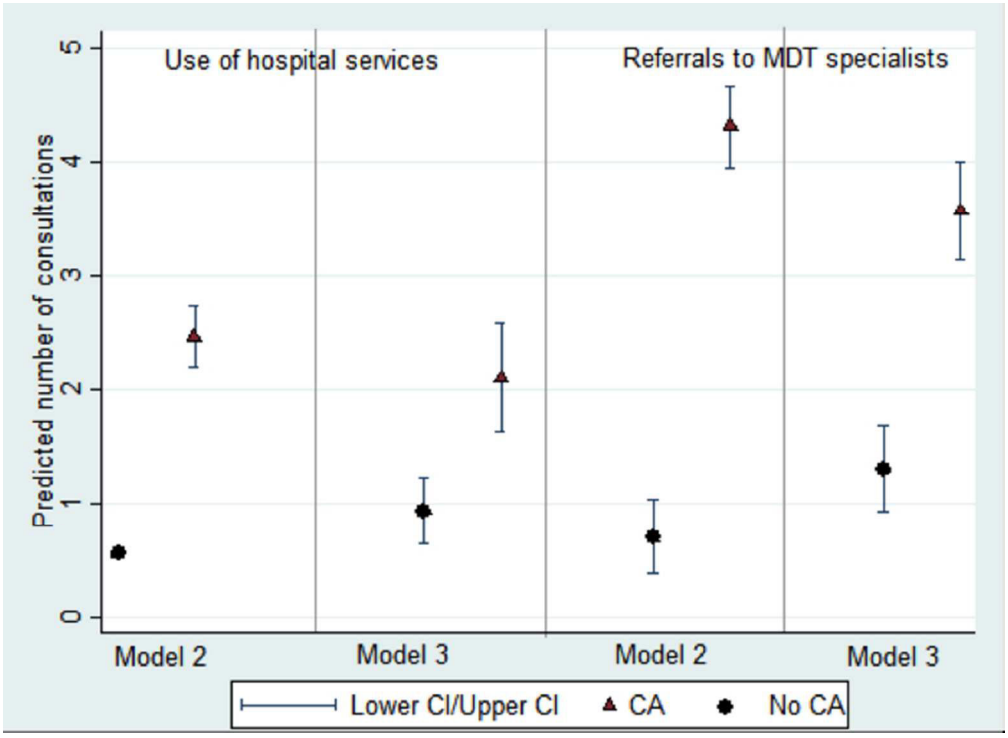


Predicted use of hospital services and referrals to MDT specialists for children with and without CA, before and after controlling for ill health (Model 2 adjusted for confounders, model 3 adjusted for confounders and ill health).

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



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

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## ABSTRACT

### Objectives

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

### Design

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

### Setting

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

### Participants

All children recruited to the birth cohort between March 2007 and December 2010. A total of 706 children with CA and 10768 without were included in the analyses.

### Primary and secondary outcome measures

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

**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 (β 1.48, 95% CI 1.36-1.59) than primary care consultations (β 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,[1] and a 20 year survival rate is estimated at 85.5% for children born with at least one CA,[2] some of whom will have complex conditions requiring multi-agency continuing care.[3-4] The healthcare needs of children with complex conditions have not been particularly well quantified in the past.[5] 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.[1] In the UK, primary care

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

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

## METHODS

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

### Case ascertainment and coding methods

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

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

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.[34] 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

same day were recorded, as many of the appointments occurring on the same day were episodes that ran over time, or were duplicates. Negative binomial regression models were used to model primary consultations and use of hospital services as they account for the over dispersion in count data. These models use an exposure variable, which indicates the number of times the event could have happened. Primary care consultations and use of hospital services were expressed per year of observed primary care registered time, which takes into account any periods the child may not have been registered with the primary care practice, withdrawals from the cohort, or deaths. We performed a sub analysis reporting regression coefficients for the outcome multidisciplinary referrals.

We used three models to compare regression coefficients for each of our three outcomes. Model 1 included univariate analyses, thought of as what is actually observed. Model 2 adjusts for other covariates we determined as confounding factors. Model 3 adjusts for confounders and measures of underlying ill health. Adjusting for ill health using a measure of multi-morbidity, is a recognised method of risk adjustment for evaluations of healthcare use, as severity of illness may not solely be due to multi-morbidity and ill health, it may also be due to other patient characteristics.[35] We used a count of unique prescriptions and a count of the number of comorbidities per child as measures of ill health. Simple counts of distinct medications have been suggested as an accurate measure of ill health, given chronic conditions frequently require repeat prescriptions[35,36] as have counts of comorbidities in primary care settings.[37,38] We performed a test for interaction between whether the child had a CA and level of deprivation for primary care consultations and use of hospital services. We also report the predicted rates of healthcare use for children with CA and without (figure 1). We also stratified the analyses for all three outcomes by CA (results not shown).

**Confounders**

We used directed acyclic graphs to determine the minimally sufficient confounding set for all of our models and checked whether the inclusion of additional covariates improved the model more than would be expected by chance using appropriate model fit statistics. These consisted of maternal age (<20, 20-34, >34 years); educational attainment (Low education [<5 GCSE equivalents or other education] High education [5> GCSE equivalents at grade A–C or two advanced level certificates or diploma, degree or higher degree])[39], economic deprivation (Economically deprived, not economically deprived), which was measured using means-tested benefit status. In the UK, being in receipt of means-tested benefits is recognised as measure of income poverty, as these benefits are frequently the only source of income and are paid at rates that put individuals below standard poverty lines,[40] ethnicity (White British, Pakistani, Other), and consanguinity (Non-consanguineous, first cousin, second cousin, other

blood[any relation]). All covariates were entered into the model as a categorical variable to allow for possible non-linearity in the relationship between the multi-morbidity measure and relevant outcome.

**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 (13%). 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 (β 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 (β -0.17, 95% CI -0.28 -0.06), but not primary care consultations (β -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 (β 0.24, 95% CI 1.18-0.30), and use of hospital services (β 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 (β 3.59, 95% CI 3.11-4.08). The only other significant factor was children born into economically deprived neighborhoods (β 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 (β 0.80, 95% CI 0.63-0.97), as does the predicted increased use of multidisciplinary referrals (β 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 |
| **Ethnic origin** | 10768 (93.9%) | 706(6.2%) | 4288(94.6%) | 245(5.4%) | 4804(92.8%) | 371(7.2%) | 1653(94.8%) | 90(5.2%) |
| **Economic deprivation** | | | | | | | | |
| 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%) |

| | | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| **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 (95% CI)* | *p* | *Coefficient (95% CI)* | *p* | *Coefficient (95% CI)* | *P* | *Coefficient (95% CI)* | *p* | *Coefficient (95% CI)* | *p* | *Coefficient (95% CI)* | *p* |
| **Economic deprivation** | | | | | | | | | | | | |
| Not Economically deprived | - | - | - | - | - | - | | | | - | - | - |
| **Economically deprived** | 0.11(0.08-0.14) | <0.0001 | 0.01(-0.02-0.04) | 0.48 | 0.35(0.27-0.42) | <0.0001 | 0.21(0.13-0.29) | <0.0001 | 0.95(0.35 1.56) | 0.002 | 0.55(0.01-1.09) | 0.045 |
| **Ethnicity** | | | | | | | | | | | | |
| **White British** | - | - | - | - | - | - | - | - | - | - | - | - |
| **Pakistani** | 0.43(0.40-0.46) | <0.0001 | 0.40(0.36-0.44) | <0.0001 | 0.27(0.19-0.35) | <0.0001 | -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) | <0.0001 | 0.26(0.21-0.30 | <0.0001 | -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 |
| **Mothers age** | | | | | | | | | | | | |
| **20-34** | - | - | - | - | | | - | - | - | - | - | - |
| **<20** | -0.13(-0.19- -0.07) | <0.0001 | -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) | <0.02 | -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 |
| **Consanguinity** | | | | | | | | | | | | |
| **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) | <0.0001 | 0.28(0.17-0.39) | <0.0001 | 1.19(0.48-1.91) | 0.001 | 0.24(-0.50-0.97) | 0.53 |
| **Second cousin** | 0.24(0.18-0.30) | <0.0001 | -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) | <0.0001 | 0.01(-0.07-0.05) | 0.86 | 0.31(0.14-0.48) | <0.0001 | 0.23(0.05--0.41) | 0.012 | -0.03(-1.54-1.48) | 0.96 | -0.32(-1.60-0.97) | 0.63 |
| **Congenital Anomaly** | | | | | | | | | | | | |
| **Yes** | 0.29(0.23-0.35) | <0.0001 | 0.24(1.18-0.30) | <0.0001 | 1.46(1.35-1.57) | <0.0001 | 1.48(1.36-1.59) | <0.0001 | 3.71(3.28-4.15) | <0.00001 | 3.59(3.11-4.08) | <0.0001 |
| **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) | <0.0001 | -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 |
| **Accident & Emergency** | 3985(49%) | 609(26%) | 1. Respiratory<br>2. Injury/poison | 1. Respiratory<br>2. Infectious parasitic |
| **Other Emergency** | 2522(31%) | 932(40%) | 1. Respiratory<br>2. Infectious parasitic | 1. Respiratory<br>2. Clinical lab findings |
| **Elective** | 1632(20%) | 801(34%) | 1. Eye/ear<br>2. Respiratory | 1. Neoplasms/blood/immune<br>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.*[33]

### 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.[26] 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 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.[41]

Our data suggests the use of hospital services are in higher demand than primary care consultations. Although an increased use of hospital services can be expected for children with CA[12,22,24-26] we find the type of hospital admission differs between children with and without CA. This is most likely explained by CA requiring more intensive treatment. For example although respiratory conditions were the most common reason for using hospital services overall (table 3), a finding similar to other studies,[25] 'other emergency' admissions were the most frequently used hospital service for children with CA. Other emergency refers to urgent referrals requiring corrective and sometimes surgical interventions that are initiated by health professionals, rather than parents presenting with their child at A & E. This increase in other emergency and elective procedures is a finding similar to that of other CA studies.[12,27]

Children with a CA were predicted to require more referrals to MDT specialists than children without CA, and have more than one specialist involved in their care simultaneously compared to children without CA. Although patient complexity increases the need for healthcare,[14] coordination of appointments for the multiple specialists required is also susceptible to variation, and is some times exacerbated by the divide between primary care, community and hospital services,[3] which is also associated with patient complications, late diagnosis, and an increased reliance on emergency care.[16] This suggests that although the predicted increase in use of hospital services for children with CA may be primarily due to their complex needs and ill health, there may be scope for this to be reduced through increasing the efficiency of care coordination.[42] In terms of clinical implications, our findings provide the quantified, longitudinal evidence requested by the Chief Medical Officer, supporting the suggestion of key workers as a catalyst for efficient patient navigation through services.[1] Also, when adjusting for ill health, the predicted increase in use of hospital services and multidisciplinary referrals for children with CA reduces (figure 1), suggesting higher usage may not be completely attributable to ill health, but affected by other factors such as deprivation and ethnicity.

There are limitations. Using a subset of diseases (CA), does not cover all children with conditions that may also be complex, however in order to extract a population we could be sure were both representative of complexity, and prevalent enough to create sample size groups large enough for comparison, CA were chosen based on the knowledge that they are high in numbers in Bradford and known to require complex care.[43] These results are based on the Bradford population, which might be interpreted as a limitation in terms of generalisability. However we feel these results are applicable to other populations or NHS trusts serving highly deprived and ethnically diverse groups of patients, characteristics which are known to be associated with CA.[43] Despite the successful linkage of primary care to cohort data in this study (97%), attributable to the complete SystmOne coverage of primary care practices in Bradford, the use of paper medical records for capturing referral activity is susceptible to missed information due to fluctuations in consultant record keeping and missing records. This research therefore illustrates the potential advantages of implementing a completely 'paperless' record keeping ethos, and the future emphasis for ensuring the exchange of data between IT systems in all clinical and care settings. This will only further strengthen the interpretability of key information at the point of care for patients with complex healthcare needs.[44]

**What is already known on this topic?**

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

**What does this study add?**

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

**Contributors**

CB and BK had full access to all the data, conducted the analysis and statistical interpretation. RCP and NS contributed to the conception and design of the work, drafting and critical revision for intellectual content. All authors had final responsibility for the decision to submit for publication.

**No competing interests**

All authors have completed the ICMJE uniform disclosure form at 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.

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**Data sharing statement**

No additional data are available

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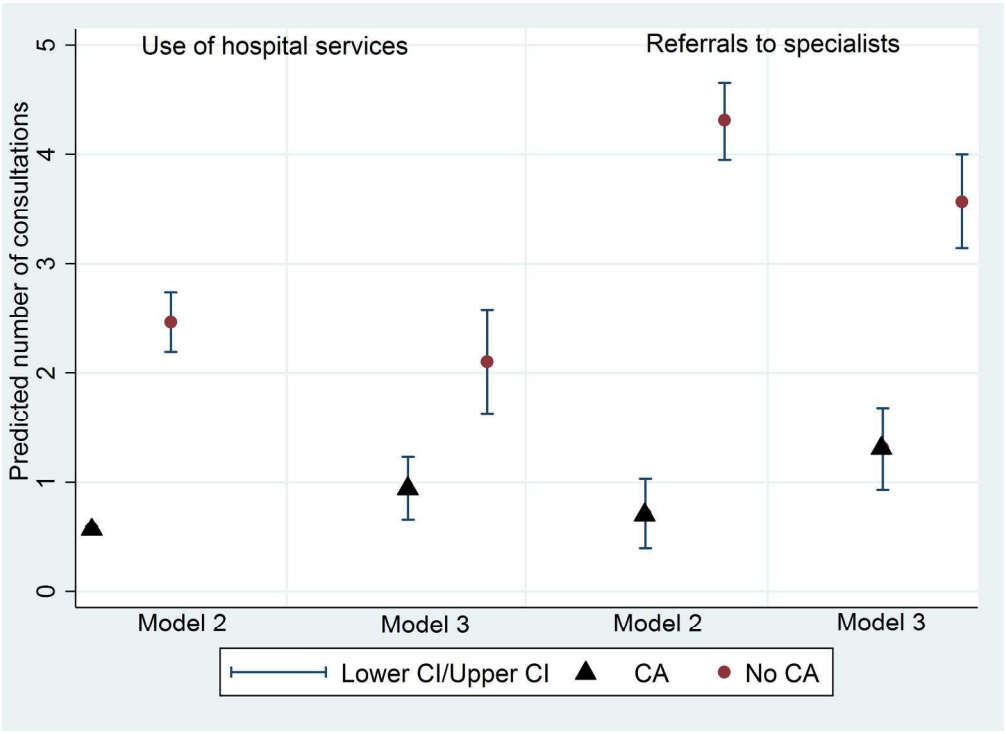


Predicted use of hospital services and referrals to MDT specialists for children with and without CA, before and after controlling for ill health (Model 2 adjusted for confounders, model 3 adjusted for confounders and ill health)

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



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

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## ABSTRACT

### Objectives

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

### Design

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

### Setting

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

### Participants

All children recruited to the birth cohort between March 2007 and December 2010. A total of 706 children with CA and 10768 without were included in the analyses.

### Primary and secondary outcome measures

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

**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 (β 1.48, 95% CI 1.36-1.59) than primary care consultations (β 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.[1] 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.[2-3] Around 93% of children with a congenital anomaly (CA) survive to adulthood,[2] and a 20 year survival rate is estimated at 85.5% for children born with at least one CA,[4] some of whom will have complex conditions requiring multi-agency continuing care.[5-6] The healthcare needs of children with complex conditions have not been particularly well quantified in the past.[7] This may be due in part to a lack of longitudinal data capturing the multi-disciplinary care required by children with complex needs.[2] In the UK, primary care practice is ideally positioned for monitoring the care requirements of

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

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

## METHODS

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

### Case ascertainment and coding methods

BiB recruits gave their consent for access to electronic primary care records and hospital episode statistics, which are split into elective, accident and emergency and other emergency admissions, here referred to as use of hospital services. We linked children's primary care

data held on SystmOne,[31] the patient contact single source system which has complete coverage in Bradford, and use of hospital services to BiB questionnaire data.[30] Linkage was performed using NHS number, surname, date of birth and gender between SystmOne[31] use of hospital services data and BiB. Of 13857 recruits, 97% were matched to primary care and use of hospital services data, forming the study population. The number of children with at least one (non-birth) hospital event was 5223 (38%). Hospital events included admissions for elective procedures, other emergencies, and A and E presentations. The average time over which data were recorded was 5.5 years, with a maximum of 7.6 years, in all 74386 person years of data. As not all children in the cohort had reached age seven, we censored our follow-up of these cases to aged 0–<5.

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

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.[36] 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 same day were recorded, as many of the appointments occurring on the same day were episodes that ran over time, or were duplicates. Negative binomial regression models were used to model primary consultations and use of hospital services as they account for the over

dispersion in count data. These models use an exposure variable, which indicates the number of times the event could have happened. Primary care consultations and use of hospital services were expressed per year of observed primary care registered time, which takes into account any periods the child may not have been registered with the primary care practice, withdrawals from the cohort, or deaths. We performed a sub analysis reporting regression coefficients for the outcome multidisciplinary referrals.

We used three models to compare regression coefficients for each of our three outcomes. Model 1 included univariate analyses, thought of as what is actually observed. Model 2 adjusts for other covariates we determined as confounding factors. Model 3 adjusts for confounders and measures of underlying ill health. Adjusting for ill health using a measure of multi-morbidity, is a recognised method of risk adjustment for evaluations of healthcare use, as severity of illness may not solely be due to multi-morbidity and ill health, it may also be due to other patient characteristics.[37] We used a count of unique prescriptions and a count of the number of comorbidities per child as measures of ill health. Simple counts of distinct medications have been suggested as an accurate measure of ill health, given chronic conditions frequently require repeat prescriptions[37,38] as have counts of comorbidities in primary care settings.[39,40] We performed a test for interaction between whether the child had a CA and level of deprivation for primary care consultations and use of hospital services. We also report the predicted rates of healthcare use for children with CA and without (figure 1).

**Confounders**

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

**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 (β 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 (β -0.17, 95% CI -0.28 -0.06), but not primary care consultations (β -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 (β 0.24, 95% CI 1.18-0.30), and use of hospital services (β 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 (β 3.59, 95% CI 3.11-4.08). The only other significant factor was children born into economically deprived neighborhoods (β 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 (β 0.80, 95% CI 0.63-0.97), as does the predicted increased use of multidisciplinary referrals (β 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 |
| **Ethnic origin** | 10768 (93.9%) | 706(6.2%) | 4288(94.6%) | 245(5.4%) | 4804(92.8%) | 371(7.2%) | 1653(94.8%) | 90(5.2%) |
| **Economic deprivation** | | | | | | | | |
| 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%) |
| **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 (95% CI)* | *p* | *Coefficient (95% CI)* | *p* | *Coefficient (95% CI)* | *P* | *Coefficient (95% CI)* | *p* | *Coefficient (95% CI)* | *p* | *Coefficient (95% CI)* | *p* |
| **Economic deprivation** | | | | | | | | | | | | |
| Not Economically deprived | - | - | - | - | - | - | | | | - | - | - |
| **Economically deprived** | 0.11(0.08-0.14) | <0.0001 | 0.01(-0.02-0.04) | 0.48 | 0.35(0.27-0.42) | <0.0001 | 0.21(0.13-0.29) | <0.0001 | 0.95(0.35 1.56) | 0.002 | 0.55(0.01-1.09) | 0.045 |
| **Ethnicity** | | | | | | | | | | | | |
| **White British** | - | - | - | - | - | - | - | - | - | - | - | - |
| **Pakistani** | 0.43(0.40-0.46) | <0.0001 | 0.40(0.36-0.44) | <0.0001 | 0.27(0.19-0.35) | <0.0001 | -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) | <0.0001 | 0.26(0.21-0.30 | <0.0001 | -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 |
| **Mothers age** | | | | | | | | | | | | |
| **20-34** | - | - | - | - | | | - | - | - | - | - | - |
| **<20** | -0.13(-0.19- -0.07) | <0.0001 | -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) | <0.02 | -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 |
| **Consanguinity** | | | | | | | | | | | | |
| **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) | <0.0001 | 0.28(0.17-0.39) | <0.0001 | 1.19(0.48-1.91) | 0.001 | 0.24(-0.50-0.97) | 0.53 |
| **Second cousin** | 0.24(0.18-0.30) | <0.0001 | -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- | <0.0001 | 0.01(-0.07- | 0.86 | 0.31(0.14-0.48) | <0.0001 | 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) | |
| **Congenital Anomaly** | | | | | | | | | | | | |
| **Yes** | 0.29(0.23-0.35) | <0.0001 | 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) | <0.00001 | 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 a period of 5 years** | | **Most common reason for admission** | |
|---|---|---|---|---|
| | No anomaly | Anomaly | No anomaly | Anomaly |
| **Accident & Emergency** | 3985(49%) | 609(26%) | 1. Respiratory<br>2. Injury/poison | 1. Respiratory<br>2. Infectious parasitic |
| **Other Emergency** | 2522(31%) | 932(40%) | 1. Respiratory<br>2. Infectious parasitic | 1. Respiratory<br>2. Clinical lab findings |
| **Elective** | 1632(20%) | 801(34%) | 1. Eye/ear<br>2. Respiratory | 1. Neoplasms/blood/immune<br>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.*[35]

### 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.[28] 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

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.[43]

Our data suggests the use of hospital services are in higher demand than primary care consultations. Although an increased use of hospital services can be expected for children with CA[14,24,26-28] we find the type of hospital admission differs between children with and without CA. This is most likely explained by CA requiring more intensive treatment. For example although respiratory conditions were the most common reason for using hospital services overall (table 3), a finding similar to other studies,[27] 'other emergency' admissions were the most frequently used hospital service for children with CA. Other emergency refers to urgent referrals requiring corrective and sometimes surgical interventions that are initiated by health professionals, rather than parents presenting with their child at A & E. This increase in other emergency and elective procedures is a finding similar to that of other CA studies.[14,29]

Children with a CA were predicted to require more referrals to MDT specialists than children without CA, and have more than one specialist involved in their care simultaneously compared to children without CA. Although patient complexity increases the need for healthcare,[16] coordination of appointments for the multiple specialists required is also susceptible to variation, and is some times exacerbated by the divide between primary care, community and hospital services,[2] which is also associated with patient complications, late diagnosis, and an increased reliance on emergency care.[18] This suggests that although the predicted increase in use of hospital services for children with CA may be primarily due to their complex needs and ill health, there may be scope for this to be reduced through increasing the efficiency of care coordination.[44] In terms of clinical implications, our findings provide the quantified, longitudinal evidence requested by the Chief Medical Officer, supporting the suggestion of key workers as a catalyst for efficient patient navigation through services.[3] Also, when adjusting for ill health, the predicted increase in use of hospital services and multidisciplinary referrals for children with CA reduces (figure 1), suggesting higher usage may not be completely attributable to ill health, but affected by other factors such as deprivation and ethnicity.

There are limitations. Using a subset of diseases (CA), does not cover all children with conditions that may also be complex, however in order to extract a population we could be sure were both representative of complexity, and prevalent enough to create sample size groups large enough for comparison, CA were chosen based on the knowledge that they are

high in numbers in Bradford and known to require complex care.[45] These results are based on the Bradford population, which might be interpreted as a limitation in terms of generalisability. However we feel these results are applicable to other populations or NHS trusts serving highly deprived and ethnically diverse groups of patients, characteristics which are known to be associated with CA.[45] Despite the successful linkage of primary care to cohort data in this study (97%), attributable to the complete SystmOne coverage of primary care practices in Bradford, the use of paper medical records for capturing referral activity is susceptible to missed information due to fluctuations in consultant record keeping, interpreting handwritten entries and missing records. This research therefore illustrates the potential advantages of implementing a completely 'paperless' record keeping ethos, and the future emphasis for ensuring the exchange of data between IT systems in all clinical and care settings. This will only further strengthen the interpretability of key information at the point of care for patients with complex healthcare needs.[46]

**What is already known on this topic?**

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

**What does this study add?**

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

**Contributors**

CB and BK had full access to all the data, conducted the analysis and statistical interpretation. RCP and NS contributed to the conception and design of the work, drafting and critical revision for intellectual content. All authors had final responsibility for the decision to submit for publication.

**No competing interests**

All authors have completed the ICMJE uniform disclosure form at 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.

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**Data sharing statement**

No additional data are available

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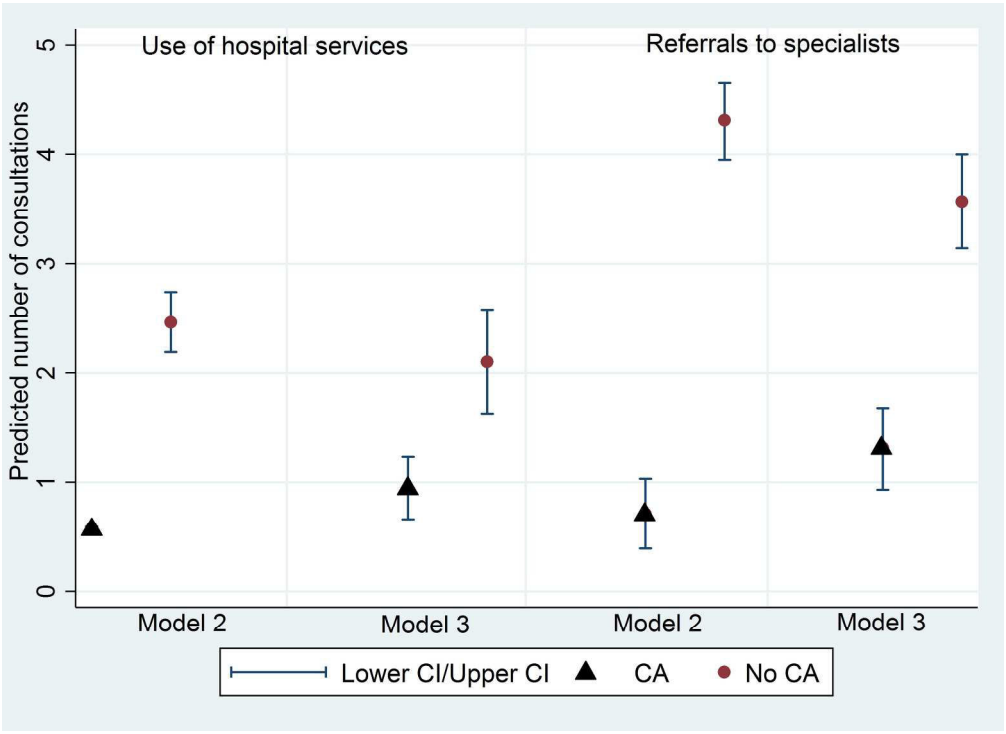
Figure legend

Figure 1: 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)




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)

120x87mm (600 x 600 DPI)