

Mortality and volume of cases in paediatric cardiac surgery: retrospective study based on routinely collected data

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Abstract

Objectives To determine whether mortality between 1991 and 1995 in hospitals in England carrying out surgery for congenital heart disease in children was associated with the annual volume of cases and to estimate the extent to which an association could explain the apparent divergent mortality at Bristol Royal Infirmary.

Design Retrospective analysis of data from two sources, a register of returns by surgeons to their professional society and an administrative database.

Setting 12 hospitals in England carrying out surgery for congenital heart disease over the period April 1991 to March 1995.

Main outcome measure 30 day mortality.

Results For open heart operations in children under 1 year old, and in particular for arterial switches and repair of atrioventricular septal defect, there is strong and consistent evidence of an inverse association between mortality and volume of cases (not taking into account any data from Bristol). A hospital carrying out 120 open operations per year in 1991-5 on children aged under 1 year would be expected to have a mortality 25% lower than that in a hospital carrying out 40 operations. If the children in the hospitals had the same mix of operations, this reduction is 34%. Stratifying for types of operation or including the results from Bristol strengthens this association. It was also estimated that less than a fifth of the excess mortality at Bristol Royal Infirmary in open operations in children less than 1 year old was due to the hospital's lower volume of surgery.

Conclusions Using appropriate methods, this study showed that mortality in paediatric cardiac surgery was inversely related to the volume of surgery. Considerable caution is needed in interpreting these results, and it does not necessarily follow that concentrating resources in fewer centres would reduce mortality.

Introduction

As part of its remit to investigate the adequacy of Bristol Royal Infirmary's surgical services for children with heart disease, the Bristol Royal Infirmary Inquiry commissioned a range of statistical work to investigate out-

comes of paediatric cardiac surgery and compare Bristol with other centres.¹ This statistical analysis identified a high mortality at Bristol that is highly unlikely to be due to chance, particularly for open heart operations conducted between 1991 and March 1995.² That Bristol was one of the smaller centres performing paediatric cardiac surgery leads to two further questions: whether the outcome of such surgery is associated with the volume of cases; and, if so, to what extent the high mortality in Bristol can be explained by the hospital's lower volume of cases.

Materials and methods

Sources of data

The cardiac surgical register comprises voluntary returns made by surgeons to their professional society and uses diagnostic categories. The hospital episode statistics for 1991-5 comprise four years of administrative data entered by clinical coders. Data are available from 12 centres in England. In each source, operations are primarily treated as either "open" or "closed" and are further subdivided into 13 "procedure groups."² For this analysis children were grouped by age at time of operation (less than 1 year old and 1 year or older).

The role of Bristol Royal Infirmary

This study was generated by the high mortality in children who underwent heart operations at Bristol Royal Infirmary, a centre with a low volume of cases, and hence it is likely that Bristol would be very influential in any analysis. It is inappropriate to test hypotheses on the same data as those that generated the hypothesis. Thus the primary analysis excluded results from Bristol. This also provided an unbiased assessment of the extent to which any excess mortality in Bristol can be explained by its lower volume of cases.

Statistical analysis

Studies of volume and outcome present a number of potential statistical problems. Firstly, results should ideally be adjusted for type of cases (case mix), to avoid some centres seeming to perform poorly because they carry out more complex surgery. Each of the 13 procedure groups was individually analysed, although there are acknowledged difficulties in the coding at this level

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BMJ 2002;324:261-4

of detail—a particular difficulty in the cardiac surgical register is distinguishing switch operations for transposition of the great arteries from Mustard or Senning repairs. The primary analysis was therefore based on pooled open operations and was stratified by procedure group. This stratification estimated a common association within procedure groups and should be more robust with respect to errors in allocation to procedure groups.

Secondly, low and high volume should be defined before the analysis. Recently, authors in the United States were accused of deliberately selecting volume thresholds after the analysis of survival rates in liver transplantation to justify their institution remaining the sole provider of the operation in the state.^{3 4} Selecting thresholds to maximise significance renders the claimed level of significance uninterpretable, and information is lost by grouping institutions into categories.⁵ No threshold was chosen in the present analysis, and volume was defined as the number of patients treated in each age group. Logistic regression was used to estimate the odds ratio of a specific change in volume; this odds ratio was assumed to be constant across the volume range unless there was strong evidence of a threshold. A degree of stratification for risk was achieved by including procedure group as a factor in the logistic regression. The odds ratio can be transformed to the relative change (*r*) in odds of death (expressed as a percentage) per additional patient per year—for example, an odds ratio of 0.98 per additional patient per year corresponds to a value of *r* of $-100 \times (1 - 0.98) = -2\%$. This would mean that for each additional operation of the type carried out, the estimated risk for each patient (expressed as odds of death) is reduced by 2%.

Thirdly, general conclusions should not be made from the very good or bad performance of just one or two centres.^{6 7} Plots show whether individual centres are having undue influence. Finally, it should be recognised that the unit of analysis is the hospital, rather than the individual patient, and so estimated standard errors should be adjusted appropriately.

The relative change in risk for all open operations was estimated in each age group, with and without the inclusion of the data from Bristol and with and without stratification for case mix, for both data sources for the period 1991-5. This analysis was repeated for all closed operations in each age group, with and without Bristol. When there was an association between risk and volume, the impact on absolute mortality and the extent to which the associ-

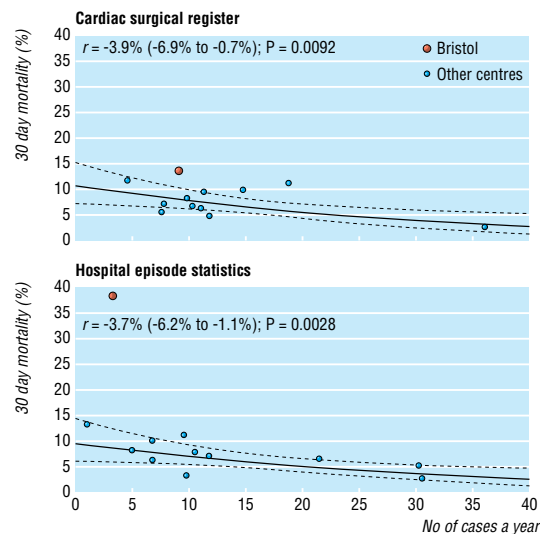


Fig 1 Relation between mortality and volume of cases in corrective surgery for transposition of the great arteries in children aged <1 year, 1991-5 (hospital episode statistics show switch operations; the cardiac surgical register includes other types). *r*—estimated change (excluding data from Bristol) in odds of death for each extra case per year; p values are for a two sided test of the hypothesis of no association. Solid line shows the relation fitted by logistic regression, dashed lines show 95% confidence intervals

ation explains the apparent excess mortality in Bristol was estimated. Reports detailing the statistical analysis carried out for the Bristol Royal Infirmary Inquiry, including a fuller version of this paper, may be found on the inquiry’s website.⁸

Results

In all open operations in children aged less than 1 year there was a significant association in both data sources between mortality and volume (table 1). Both data sources showed a consistent relation (excluding data from Bristol), despite disagreement in the data.

Stratifying the data by procedure group increased the estimated association between mortality and volume (table 1). This result might be expected if larger centres carried out a greater proportion of more complex operations. Again, inclusion of the data from Bristol strengthened this finding. The procedure groups that contributed most to the association are corrective operations for transposition of the great arteries (“switch” operations in the health episode statistics)

Table 1 Percentage change in odds of death (95% CI; P value) for each extra patient per year in 12 centres in England, including and excluding data for Bristol, according to data for 1991-5 from two data sources

Type of operation	Excluding Bristol data		Including Bristol data	
	Hospital episode statistics	Cardiac surgical register	Hospital episode statistics	Cardiac surgical register
All open operations				
Children aged <1	-0.38 (-0.71 to -0.04; 0.015)	-0.34 (-0.56 to -0.12; 0.001)	-0.58 (-1.04 to -0.09; 0.01)	-0.42 (-0.69 to -0.13; 0.002)
Children aged ≥1	-0.20 (-0.81 to 0.46; 0.27)	-0.45 (-1.22 to 0.39; 0.14)	-0.24 (-0.81 to 0.37; 0.22)	-0.47 (-1.20 to 0.31; 0.11)
Open operations (stratified by type of procedure)				
Children aged <1	-0.56 (-0.92 to -0.20; 0.0015)	-0.61 (-0.90 to -0.31; <0.0001)	-0.83 (-1.20 to -0.44; <0.0001)	-0.67 (-0.97 to -0.37; <0.0001)
Children aged ≥1	-0.22 (-0.60 to 0.18; 0.14)	-0.7 (-1.17 to -0.23; 0.002)	-0.25 (-0.61 to 0.13; 0.10)	-0.74 (-1.19 to -0.26; 0.001)
All closed operations				
Children aged <1	-0.28 (-1.60 to 1.24; 0.35)	-1.44 (-2.62 to -0.07; 0.02)	-0.28 (-1.68 to 1.36; 0.36)	-1.43 (-2.57 to -0.12; 0.02)
Children aged ≥1	-9.01 (-9.82 to -4.65; 0.004)	-3.07 (-7.08 to 6.43; 0.2)	-9.17 (-9.84 to -5.61; 0.002)	-2.99 (-6.87 to 5.70; 0.19)

Table 2 Death rate in children aged <1 year who underwent open heart operations in 1991-5 in 12 English hospitals, showing the extent to which the apparent excess mortality at Bristol Royal Infirmary can be explained by its volume of surgery.

Data source	Mortality (%) in Bristol (A)	Mortality (%) in other centres (B)	Excess mortality in Bristol, not adjusted for volume (C=A-B)	Expected mortality in Bristol, adjusted for volume* (D)	Excess mortality in Bristol, adjusted for volume (E=A-D)	% of excess mortality explained by effect of volume 100(1-E/C)
Hospital episode statistics	28.7 (41/143)	11.2	17.5	13.3	15.4	12.0
Cardiac surgical register	23.7 (43/181)	12.5	11.2	14.4	9.3	17.0

*Estimated from the fitted line in fig 1, under the assumption that the mortality at Bristol did not differ from the other centres.

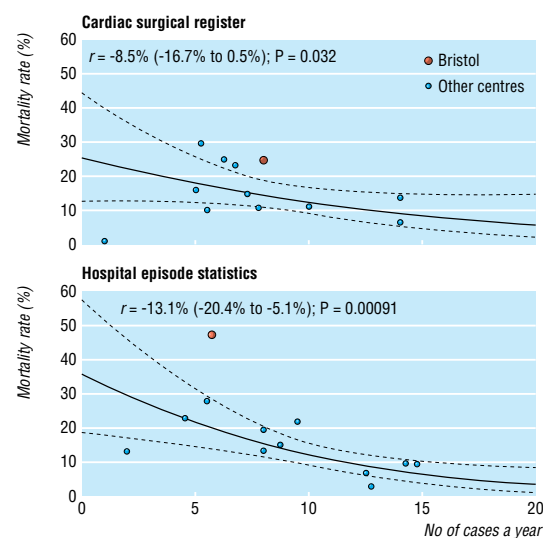


Fig 2 Relation between mortality and volume of cases in surgery for repair of atrioventricular septal defect in children aged <1 year, 1991-5. r =estimated change (excluding data from Bristol) in odds of death for each extra case per year; p values are for a two sided test of the hypothesis of no association. Solid line shows the relation fitted by logistic regression, dashed lines show 95% confidence intervals

and repair of atrioventricular septal defect (figs 1 and 2). Much of the relation shown in the data from the cardiac surgical register in figure 1 comes from one large centre.

For closed operations, no consistent pattern occurred in either data source (table 1). Including the data from Bristol had negligible influence on the relation in closed operations.

When both sets of data shown in table 1 were used, r is around -0.4% without adjustment for operation mix and around -0.6% with adjustment. A hospital carrying out 120 open operations per year on children less than 1 year old in 1991-5 would be expected to have a mortality that is 25% lower (11.3% *v* 15.0%) than that in a hospital carrying out only 40 such operations. If the hospitals had exactly the same mix of operations, this relative reduction is 34% (9.9% *v* 15.0%) (for details see full version on *BMJ's* website). Table 2 shows that only an estimated 12% (hospital episode statistics) or 17% (cardiac surgical register) of the excess mortality at Bristol can be explained by Bristol's low volume of cases.

Discussion

Mortality in children aged less than 1 year old who underwent open heart surgery in 1991-5 is significantly related to the volume of cases, even when data from Bristol are excluded. This effect was consistent

across both data sources and became more pronounced when the data were stratified according to the mix of operations. This finding is not due to the disproportionate influence of just one or two centres. The data sources were consistent in showing that only a small proportion of the excess mortality at Bristol Royal Infirmary can be attributed to its having a low volume.

Caution is needed in interpreting these findings. The data sources are not of high quality, they have different coding schemes, and they share inadequacies in reporting of data. The conclusions in terms of policy that can be drawn from the study are unclear. For example, for the data from the hospital episode statistics, it is tempting to recommend a minimum volume of around 50 operations per year, or one a week: mortality in children aged <1 year in centres with a lower volume was 14.7% (not including Bristol) or 16.7% (including Bristol), whereas the mortality in centres with a higher volume was 10%. Dudley et al take the bold step of using such data to predict the number of "potentially avoidable deaths"—based on the assumption that patients treated at "low" volume centres could have been treated at "high" volume centres, resulting in lower mortality—but this seems to be a quite unwarranted extrapolation.⁹

It is possible that concentrating certain types of operation in fewer centres will lead directly to benefits in outcome—for example, through increased opportunities for surgical learning. However, Posnett warns that such "economies of scale" cannot be guaranteed.¹⁰

What is already known on this topic

Mortality in children undergoing heart operations has been shown to be lower in hospitals with a high volume of such operations

Studies showing a relation between volume of cases and mortality have a range of methodological inadequacies, in particular the choice of a threshold defining high and low volume after the analysis to increase the significance of the results

What this study adds

Disregarding data from Bristol, there is strong and consistent evidence that in England in 1991-5 hospitals performing a higher number of open heart operations in children aged under 1 year tended to have lower mortality

This association explains only a small proportion (less than a fifth) of the excess mortality seen at the Bristol Royal Infirmary over this period

Rather than indicating causality, an association between volume and better outcome might be due to a common underlying factor, such as a hospital's longer history, better associated services (such as intensive care), its ability to attract and retain skilled staff, or its ability to attract more patients because of its reputation. None of these factors would necessarily be obtained by, say, merging the caseloads of two centres. It is also important not to extrapolate beyond the available data; further increases in the case volume in larger centres may even lead to poorer outcomes, if communication in the hospital were to start to decline. Finally, it is possible that the concordance between centres might have increased since 1995, because experience with operations such as the arterial switch has been gained.

The author is grateful to Ruth Chadwick, Paul Aylin, Gordon Murray, Stephen Evans, and Nicky Best for advice and provision of data. All views expressed in this paper are those of the author alone and do not necessarily represent the views of the Bristol Royal Infirmary Inquiry.

Funding: Bristol Royal Infirmary Inquiry.

Competing interests: None declared.

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(Accepted 29 August 2001)



The full version of this article appears on bmj.com

Relative importance of genetic effects in rheumatoid arthritis: historical cohort study of Danish nationwide twin population

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BMJ 2002;324:264-7

Abstract

Objective To determine the relative importance of environmental and genetic effects in the development of rheumatoid arthritis.

Design Historical cohort study with record linkage between a twin registry and the Danish discharge registry as well as the Danish national registry of deaths used to estimate completeness.

Setting Two population based nationwide twin birth cohorts.

Participants 37 338 twins were sent a questionnaire about rheumatic diseases. Self reported rheumatoid arthritis was verified by clinical examination and from medical records.

Main outcome measures The probandwise concordance rate of rheumatoid arthritis in monozygotic and dizygotic twins.

Results The response rate was 84.7%. Rheumatoid arthritis was verified in 13 monozygotic and 36 dizygotic twins. There were no concordant monozygotic twin pairs and two concordant dizygotic twin pairs. Based on capture-recapture methods the probability of ascertainment was 78.3%. The probandwise concordance rate was 0 (95% confidence interval 0 to 24.7) in monozygotic twins and 8.8 (1.9 to 23.7) in dizygotic twins.

Conclusion Genes are of minor importance in the development of rheumatoid arthritis.

Introduction

Rheumatoid arthritis is a systemic inflammatory autoimmune disease of unknown cause. Environmental and genetic risk factors have been identified, but no single risk factor has emerged as necessary or sufficient to cause the disease.

Twin studies represent one of the simplest ways to unravel the relative importance of genetic and environmental effects. In studies of specific diseases or traits in twins who volunteer to take part, monozygotic, concordant, and female twins tend to be over-represented.^{1,2} Hence, much of the available literature on rheumatoid arthritis in twins overestimates the contribution of genetic factors.³⁻⁵ Only two previous studies were population based, but confirmation of the diagnosis according to validated classification criteria was not performed.^{1,6}

We undertook a nationwide study among twins in Denmark to estimate the importance of genetic effects in the development of rheumatoid arthritis.

Methods

Ascertainment of twins—The study comprised two nationwide twin populations. The older birth cohort comprised 1631 same sex pairs of twins born 1921-40 in which both twins were alive in 1994.⁷ The younger birth cohort comprised 34 076 surviving twins from same and opposite sex pairs of twins born 1953-82.⁷ To