

- Quality; Ms Lawrence and Professor Murray, "...conclude, that the quality of the UKCSR data is inappropriate for comparative purposes at the level of diagnostic categories. The systematic differences between centres in the reporting of mortality will introduce substantial bias in comparing units." (Lawrence and Murray, Page 3, (5))

2.6 MORTALITY AND VOLUME OF SURGERY

Dr Spiegelhalter has investigated the possible variation in mortality in different centres and whether this is related to the volume of surgery carried out in those centres(3). His analysis shows that there is an inverse relationship between the volume of surgery and mortality for (i) open operations in the first year of life, (ii) the switch operation and (iii) operations to correct complete AVSD.

He estimates that 12 – 17% of the apparent excess mortality in Bristol could be explained by this relationship. I ask whether this remains a secure conclusion in the light of the uncertainties associated with the reliability of the data he has used?

3. RESULTS OF OPERATIONS IN CHILDREN UNDER ONE YEAR, 1991 – 1995

3.1

In response to my question as to whether there are still excess deaths in this group of patients after the figure's for complete AVSD and the Switch operations have been excluded, Dr Spiegelhalter has confirmed that indeed there are still excess deaths. My views on this group of patients are set out below, and in subsequent sections I enlarge on some of the points where that is needed.

3.2

- I accept that there are excess deaths as a statistical finding, amongst the Switch operations in neonates and my operations for correction of complete AVSD.
- I accept the figures for Total Anomalous Pulmonary Venous Drainage (TAPVD).
- I believe that the ASD figures are wrong (see section 3.6 below).
- For data in this section (as for other sections) there are serious questions to be answered, which are summarised in the following paragraphs. Some of these questions stem from the findings summarised in Paras 2.3-2.6 above.

3.3 DATA QUALITY

- Incomplete HES data could have increased the apparent mortality in HES by up to 10% (see 4.1 below).

- Under reporting of death in HES has been shown to be **at least** 8.4% overall, and 5.2% for children under one year having open surgery (4).
- The UKCSR is deemed by Ms Lawrence and Professor Murray to be inappropriate as a comparator at the level of diagnostic categories (5).

3.4 STATISTICAL METHODS.

- *Aggregation and Pooling.*

Aggregation of estimates arising from stratified and homogenous groups of patients is a sound and elegant technique. I do not believe that pooling of non-homogenous and disparate data can be an authoritative or widely accepted basis for this statistical analysis. In the consultation with Professor SJW Evans on 3rd December 1999 in Mr Langstaff's chambers, I understood that in clarifying the distinction between pooling and aggregation, he indicated that pooling heterogeneous data would be an inappropriate or invalid method (My personal contemporaneous memo, 3/12/99).

At Page 7, of his response to my submission (6), Para 2.3.3.3., Dr Spiegelhalter explains why the aggregation technique is used and I agree with his reasons and conclusions. He goes on to say "*That is why we present data both for pooled operations and aggregated over operative groups.*" His preceding remarks are a reason to use aggregated data, but **not** a reason to use pooled data. Even if it is correct that the estimates arising from the use of pooled and aggregated data are similar, this does not constitute a justification for the use of pooled data.

At Page 8, para 2.4.1 and 2.4.2, Dr Spiegelhalter rejects my assertion that the use of pooled data makes the analysis "unreliable." Perhaps I used the wrong term. The fact that he goes on to comment on the difficulties in obtaining agreed coding categories for diagnosis and operations, which may favour the use of a pooling technique, only

underlines the problems which are inherent in the analysis of this data and does not justify the use of pooling.

I am very disappointed that in his responses to my various submissions, Dr Spiegelhalter has not engaged in a dialogue which acknowledges the problems inherent in the use of pooled data in his analysis. I believe the use of, and dependence on pooled data, is one of the chief weaknesses of the statistical analysis.

- *Number of Deaths*

In response to some points that I made concerning discrepancies in data and coding, Dr Spiegelhalter has stated that the discrepancies were not important because there was agreement about the number of deaths which occurred in Bristol (6). I do not accept that there is agreement about the total number of deaths. There are two reasons for this –

FIRST:

TABLE 1. OPERATIONS AND MORTALITY IN BRISTOL, CHILDREN UNDER ONE YEAR OF AGE: 1991 AND 1995.

Source of Data	Groups 1-11		Open Operations	
	No	D(%)	No	D(%)
HES	140	42(30%)	143	41(28.7%)
UKCSR	157	30(19.1%)	181	43(23.6%)

This Table is based on the figures in the document INQ 0015 Tables 7.3.1, 7.3.2 and 7.4.5.

The numbers presented in Tables 1 are drawn from HES and UKCSR, analysed in two different ways, and do not agree with each for either operations or deaths. The estimates of the number of operations ranges from 140 to 181 and the estimates of percentage mortality range from 19 to 32. Agreement about the total number of deaths would be meaningless, unless there was also agreement about the total number of operations .

SECOND:

I am uncomfortable with a response to a serious question about discrepancies in various figures which seems to say that it doesn't actually matter if we get the details right or wrong, as long as the total is right.

It follows that I continue to believe that resolution of the discrepancies is needed. There shall be further discussion of the discrepancies which I have raised at paras 3.5,4.2, 4.3 and 4.4 below.

- *Statistical Significance.*

What level of probability that excess deaths exceed zero, is significant, remains unclear. I accept the notion that in reality there is not a black and white difference between findings on either side of some arbitrary line. However Dr Spiegelhalter has also stated that one should not be too confident until "*Things are really up high, 98%,99% ...*", (Day 70, Page 171, lines 15-16). I take this to mean that the P value should exceed 0.97. There needs to be clarity and consistency in how this matter is handled.

3.5 THE EXPLORATORY STAGE.

Discrepancies - ASDs

- Our records indicate that between 1st January 1991 and 31st March 1995 we carried out 102 operations for closure of Secundum Atrial Septal Defects with no deaths (Bristol Royal Infirmary Cardiac Unit: Paediatric Open-Heart Procedures 1990-1995; Bristol

Paediatric Cardiac Surgery 1990-1995, United Bristol Health Care Trust, January 1996).

- I apologise that when making my previous submissions I had misunderstood what made up Group 6 – ASDs. I had taken the Tables in INQ 0013 0054 and at INQ 0015 0043 to indicate that Group 6 consisted of Secundum ASDs, including sinus venosus defects. However I have now identified the more complete guidance at INQ 0014 0043 and realise that Group 6 includes Primum Atrial Septal Defects and Common Atrium in addition to the above.
- The revised figures for Group 6 (ASDs) for 1st January 1991 to 31st March 1995 for Bristol are in Table 2 below.

TABLE 2. OPERATIONS IN GROUP 6 (ASDS) FOR 1991 – 1995

Operations for	Under 1 yr age		Over 1 year age	
	NO	D	NO	D
Secundum ASD	2	0	100	0
Primum ASD	1	1	17	0
Common Atrium	2	2	1	0
TOTAL	5	3	118	0

- The Inquiry's statistician's figures may be seen at INQ 0015, Tables 7.3.1, 7.3.2, and 7.4.5 and may be seen in Table 3 below.

TABLE 3.

<u>Table No.</u>	<u>Source</u>	<u>Age</u>	<u>No of Operations</u>	<u>Deaths</u>
7.3.1,	HES	Under 90 days	2	0
7.3.2	HES	90 days to 1 yr	8	5
7.4.5	CSR	Under 1 yr	5	2

The discrepancies in the figures from the 2 sources is clear but needs to be resolved before statements are made about ASD mortality in Bristol.

- Beyond pointing out these discrepancies I cannot comment further on Mr Dhasmana's patients but can make a further comment on one of my own patients.

In his response at Page 14, Dr Spiegelhalter (6) identifies one of my patients, "child D", who died following surgery. Sometime after major neonatal abdominal surgery this child presented in extremis, due to acute pulmonary embolism. I operated in an emergency in an unsuccessful attempt to save his life. En passant, I stitched closed a Patent Foramen Ovale which is a type of atrial septal defect, and which is virtually a normal finding at that age. I did so in order to prevent the possibility of a clot passing to the left side of his heart and hence to his body. This child should not be classified in Group 6 as an atrial-septal defect. He did not even have congenital heart disease, he had an acquired condition. This is why I classified him as miscellaneous.

- I therefore continue to believe that the figure of 5 deaths out of 8 operations within this Group in this epoch, in children under one year of age is wrong and needs to be reviewed in detail.

patients had a circumflex coronary artery arising from sinus 2 (known to be a risk factor in a multi-institutional study)".

- **Mortality and Volume of Operation**

Dr Speigelhalter (3) suggests that 12% – 17% of the apparent excess mortality in Bristol could be explained by the fact that it was a "low volume centre".

4. POINTS FOR FURTHER DISCUSSION.

4.1 INCOMPLETE HES DATA

In my submission at Para 2.3.3.2 (SUB 0009 0022-0027), I expressed concern that the exclusion of data with missing outcomes in HES may increase the apparent excess mortality, particularly in neonates and children in the first year of life, where the excess deaths are greatest. I went on to say that I accepted Dr Spiegelhalter's reassurance that it can only be a small contribution.

Dr Spiegelhalter (6) rejects the view that the missing outcomes makes the HES analysis unreliable. I accept that as a broad statement, but put forward the view that it could contribute up to 10% of the apparent excess deaths in children under one year of age for the years 1991 – 1995. In this Group HES estimates 29.2 excess deaths; Dr Spiegelhalter's table at Page 7, states that the missing outcomes could increase the apparent excess death in children under one year of age by 2.6.

4.2 AORTIC AND PULMONARY VALVE OPERATIONS (GROUP 10); ALL AGES, 1991 – 1995.

In INQ 0015, the Table at 004 and the further Tables numbered 7.3.1, 7.3.2 and 7.3.3 identify 50 patients having aortic and pulmonary valve surgery (Group 10), and 5 deaths as follows:

TABLE 4

<u>Age</u>	<u>NO of Operations</u>	<u>Deaths</u>
Less than 90 days	1	1
90 days to 1 year	3	1
Over 1 year	46	3

Table 7, at INQ 0014, 0043 –44 states that corrective surgery for pulmonary atresia and intact

ventricular septum is also included in Group 10. Our records (Bristol Royal Infirmary Cardiac Unit: Paediatric Open Heart Procedures 1990 – 1995; Bristol Paediatric Cardiac Surgery, 1990 – 1995, UBHT January 1996) indicate that the operations in Group 10 are as follows:

TABLE 5

Operations	Under 1 yr		Over 1 year		Total	
	NO	D	NO	D	NO	D
Aortic Valve	1	1	23	0	24	1
Pulmonary Valve	0	0	11	0	11	0
Pul Atresia with IVS	0	0	4	0	4	0
TOTAL					39	1

The discrepancies in these figures should be recognised and some attempt should be made to resolve them. This is underlined by the fact that in INQ 0015, Table 7.4.9, based on the UKCSR, indicates that the number of operations in Group 10 was 34 with one death.

4.3 MITRAL VALVE SURGERY (GROUP 11); ALL AGES, 1991 – 1995

Similarly, in the document INQ 0015, the Table at Page 0004 and the further Tables numbered 7.3.1, 7.3.2 and 7.3.3 identify 23 patients having mitral valve surgery in Group 11, with 3 deaths as follows –

TABLE 6

Age	NO of Operations	Deaths
90 days to 1 year	3	2
Over 1 year	20	1

Our own records show that:-

TABLE 7

<u>Age</u>	<u>NO of Operations</u>	<u>Deaths</u>
Under 1 year	1	0
Over 1 year	10	1
TOTAL	11	1

There is in addition a patient who died who had mitral valve surgery and also relief of sub-aortic obstruction, which may have been allocated to Group 11 by the Inquiry Statisticians. In the hierarchical framework of primary procedure grouping (INQ 0015, 0044) mitral valve surgery is higher in the hierarchy than aortic valve surgery. This would increase the total to 12 operations with 2 deaths. The discrepancies here are underlined by the fact that in Table 7.4.9, based on the Cardiac Surgical Register, there are said to be 9 operations with no deaths.

4.4 MY MISCELLANEOUS GROUP

At Page 14, (6) Dr Spiegelhalter states that there are 88 patients in my WL who are in a miscellaneous group, with 33% (29 patients) mortality. My data base has patients in the miscellaneous group as follows;

TABLE 8

<u>Age</u>	<u>NO of patients</u>	<u>Deaths</u>
Under one year	21	10
Over one year	41	9
TOTAL	62	19

If one adds to the miscellaneous group, "other valves" as a quasi-miscellaneous group, the total would become 67 patients and 21 deaths (31%).

Although the mortality rate is similar to that reported by Dr Spiegelhalter, the discrepancy in numbers between his statement and what I believe to be the case, is a cause for concern.

Para s5.2.3 *"Figure s5.2 shows that pattern of operations between 1st April 1994 and the 31st December 1995 when the funding arrangements based on numbers of open heart surgery had been withdrawn. Numbers are very small, but the peak at month 11 is no longer present. Elsewhere in England the peak occurs around month 2 and 3.*

Comment

Around 1990 the paediatric whole Team made a policy decision to carry out surgical correction for Complete Atrio-Ventricular Defect before the first birthday, if possible. The reason for this decision was that we believed it to be "best practice", and in the best interests of the children. This is reflected in the finding reported in paragraph s5.2.1. This represents a dramatic change from our practice in the 80s.

This goal was often achieved by operating towards the end of the first year, as reported in paragraph s5.2.2. The reasons for this include; i) relatively late referral of patients, ii) constant pressures on the waiting list. It was only towards the end of the period under review that we were managing to achieve earlier corrective surgery.

The implication of paragraph s5.2.3 is that the timing of surgery was influenced by funding considerations. It is my conviction that this consideration did not enter into the timing of operation at all.

6. A SYSTEMATIC REVIEW OF THE OUTCOMES OF OPEN-HEART PAEDIATRIC SURGERY

In this section I shall comment on the paper by Vardulaki et al (8) and on the letters commenting on it by Drs Arnold, Bull, Macrea and Silove and Messers Deverall, Hamilton and Sethia.

6.1

This paper should be welcomed as a useful source of information and references and for its instructive methodology in evaluating papers and collating their contents. I shall make comments below, but agree with many of the points made in this review and the accompanying letters, such as:

- *“The pooled 30-day mortality estimates...reported in the review are likely to represent ‘best achievable performance’ rather than the performance to be expected in everyday practice.” (Page 6.)*
- *“Inevitably, the review of case series evidence can be subject to publication bias.” (Page 67, Para 94.)*
- The debate about the 10 criteria for patient selection for the Fontan operation. (Page 31, Para 36.)
- The difficulty which existed in obtaining leave and expenses to attend meetings and to visit centres of acknowledged excellence. Also, there was a lack of access to international surgeons of excellence in the context of learning new techniques. These points are made in the letters of Mr Deverall and Mr Hamilton.

6.2 DEFINITION OF 'INFANT'

"The review was to focus on.....open cardiac procedures in infants and neonates." (Page 13, Para2). The definition of infancy used by the Inquiry, generally in the UK, in the UK Cardiac Surgical Register (UKCSR) and in the wider paediatric cardiac surgical community is under one year.

The papers used in this review described procedures in children under two years of age except for the Fontan procedure (Page 15, Para 7.) The failure to use the common definition of infancy limits the value of the review.

6.3 THE REVIEW AND UK PRACTICE

The majority of case series collected in the review were from the USA, and about one-third of the total came from seven centres, of which just one was from the UK or Europe. (Page 4 and Page 66, Para 91.) Approximately 10% of the papers listed in Appendix H are from the UK.

The UKCSR was not used by the reviewers because of concerns about misclassification and validity of its data. (Page 70, Para108.) In addition the UKCSR gives results for children under one year of age, but not for children under two years, and therefore would be expected to show higher mortality rates. The opinion of Lawrence and Murray that it is not appropriate to use the UKCSR to compare results at the level of diagnostic categories (5) confirms the authors' decision.

6.4 AGE AT CORRECTION OF AVSD

The reviewers state that, *"In the late 1970s and early 1980s most patients with a complete AVSD were operated on before two years of age* (Page 25, Para 27). I do not believe that practice in the UK is accurately reflected in that statement. The UKCSR, with all its shortcomings, reports the number of operations carried out for complete AVSD shown in

Table 9. While it is possible that many operations were carried out in the second year of life between 1977 and 1985, it seems unlikely to be enough to confirm this statement.

TABLE 9. OPERATIONS REPORTED IN THE UKCSR FOR C-AVSD

<u>YEARS</u>	<u>NUMBER OF OPERATIONS</u>	
	<u>Under One Year</u>	<u>Over One Year</u>
1977-80	43	117
1981-85	68	176
1986-90	256	248
1991-95	532	256

6.5 CASE-MIX AND RISK STRATIFICATION

The reviewers note variations in mortality for clinical sub-groups based on anatomical differences (Page 61, Para 78; Page 63, Para 63; Page 64, Para85). They accept that unidentified physiological factors may, in some cases, be even more important than the anatomical factors (Page 61, Para79).

I believe this to be correct and would like to add that the reviewers only take account of some anatomical factors. There are other anatomical factors which will have a significant influence on mortality when they are present. The presence or absence of obstruction in TAPVD, referred to by Mr Hamilton, is but one example.

I support Dr Arnold's comment at Page 3 of his letter; "*Perhaps it would be more helpful to consider wider groups and within those groups identify factors which increase risk.*" (My

underlining). In essence that is risk stratification and Dr Arnold clearly believes that such factors exist.

6.6 TOTAL ANOMALOUS PULMONARY VENOUS DRAINAGE

The value of the findings about TAPVD in the review may be limited by the fact that the most recent mid-point date for a case series is 1983 (Page 45, Fig 5.)

6.7 FONTAN OPERATION

On Page 32, Table 8, there is a series referred to which began in 1953; this seems to be anomalous as the Fontan operation was first reported 15 or more years later.

6.8 ACCESSIBILITY OF EVIDENCE

At Paras 88 and 89, Page 65, the issues of accessibility and synthesis of evidence are discussed. It is my belief that the majority, but not all, of these papers would have been available to practising surgeons. Over the period of the review I read The Journal of Thoracic and Cardiovascular Surgery, the Annals of Thoracic Surgery, the European Journal of Cardiothoracic Surgery and Circulation. While I would have been familiar with many of these individual reports, I neither had the time, expertise or facilities to process and collate the information as the reviewers have done.

6.9 CURRENT RISKS

Like Dr Bull and Mr Hamilton, I do not know what the authors are referring to when they say, "*There are well recognised methods for weighting local and national data in estimating the current risk in a local setting.*" (Page 64, Para 85.) I did my best to do this, but without the assistance of such a method.

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