

A second example would be with data from the Mayo Clinic. I note that this centre of excellence is one of the centres most frequently publishing quality articles, which form part of the report. The Mayo Clinic is in a small town, i.e. Rochester, Minnesota, U.S.A. The vast majority of patients seen and treated at this clinic are referred from other national and international centres. By definition such patients will have undergone a selection process before referral and be fit enough to be transferred often large distances. It follows that such patients will not be comparable with patients arriving at a regional centre on an "all comers" non-selected basis.

I do however accept that publications from centres of excellence represent gold standards. One has to accept and believe the veracity of the data in this type of publication but equally these publications do not tell the whole truth about the results of treatment which are experienced throughout the world. Bad results are not published. Small numbers of results, which would often reflect the practice of a regional unit, would in general not be considered worthy of publication in a major journal. However and despite these reservations the gold standard data of best practice would represent a goal to which all would aspire.

The journals quoted in the report I think would be available to surgeons in this country. They are respected and have high review and editorial skills. They do however have an inherent time lapse between practice and publication. A given clinical experience would take place over at least a period of a couple of years before a sufficient experience would be obtained. The submission of such an experience followed by the peer review process and various editorial processes would take a matter of at least several months. From editorial acceptance to publication in the period in question would take probably at least another year. Relying on literature as an educational tool there would be therefore at least a three year period before knowledge would become available to the general reading surgical public. What other methods were available to the surgeon?

National meetings dealing with highly specialised topics would occur in the U.K. on at best an annual basis. International meetings are more frequent while international symposia on specific topics, for example the Management of Transposed Great Arteries would occur somewhere in the world on at least an annual or biannual basis. Attendance at such meetings would have been and still is on a very pro-active basis. Attendance demands time and can be relatively expensive and my own experience was that it only occurred with determination on the part of the surgeon and was not encouraged by management, either in terms of time away from work or financial support. Surgeons practising in centres of excellence have the frequent opportunity to meet speciality leaders during visits. These centres have the prestige and often finance to attract national and international figures of excellence and thus have the opportunity to shorten their learning curves by obtaining knowledge and experience from others more advanced in the particular area. Such visits were much less available to people working in regional centres - not defined as those of excellence. Thus the learning curves of those centres could be and were more prolonged.

The quality of facilities, equipment and staff in centres of excellence also tended to mean that the very best standards of care were available at all times and under all circumstances. This certainly was not the case in a regional centre. There is a very big difference between the first assistant at a complex infant heart operation being a fully trained experienced surgeon as opposed to a young resident in training with no specific knowledge of the complexity of the operation being performed.

I have previously referred in a previous report to the evolution of open-heart paediatric practice, which occurred throughout the 1970's and 1980's. This evolution and improvement in results is reflected in the report of the independent group. The paediatric surgeon is the central point of the complex supporting infrastructure and knowledge base at the core of this evolution and improvement. The centres of excellence producing the gold standards however surrounded the surgeon with all the conditions necessary for the surgeon to achieve best possible results. I remember from my own experience that I visited centres of excellence and most of them are named in this report as often as possible with at least one of two visits each year. Such visits often had to be made at my own expense and in my holiday time. They were hugely stimulating and educational but one's feelings were often ambivalent in that this stimulation was also accompanied by varying degrees of depression on returning to one's own place of work.

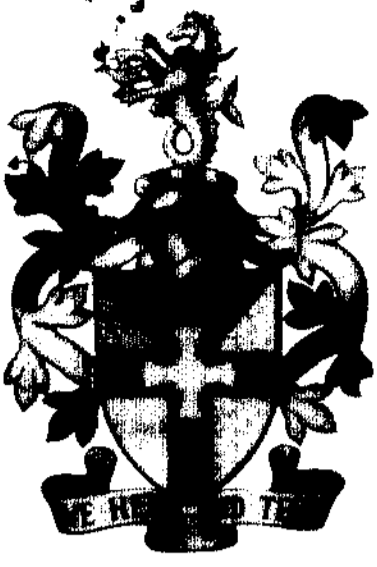
With reference to the Bristol Royal Inquiry I believe that information described in this report should have been available to the surgeons and their colleagues and using the various means of acquiring the information described in this letter. Clearly it would not be possible for individual surgeons to systematically analyse the world literature but they should have been aware of the trends and whatever reservations they may have had in regard to centres of excellence they should have been aware of the gold standards being set. I have significant doubts as to whether the busy surgeons in Bristol could have found the time and support necessary to take advantage of the means of acquiring knowledge, which I have described. I have a major doubt that even had the surgeons been able to most efficiently acquire experience and knowledge that they could apply this to their patients in an optimal way. The facilities, for example a split site, the equipment, for example echocardiography machines, and the clinical profile of the patients, for example babies presenting late in the evolutionary clinical process, would all introduce conditions making the achievement of an optimal outcome more difficult. I do agree that the surgeons should have kept a full database with as much as possible information to qualify that database. I do agree that continuing with a form of treatment in inadequate circumstances and inadequate results is unacceptable. I do agree that decisions regarding continuing or discontinuing a programme in less than ideal circumstances is a very complex question, which must involve all participants, including management at local, regional and central level.

I hope this brief response to the independent report is of value to the Inquiry.

Yours sincerely,



Philip B Deverall, MB FRCS
Consultant Cardiac Surgeon



THE NEWCASTLE UPON TYNE HOSPITALS NHS TRUST

THE FREEMAN HOSPITAL

High Heaton, Newcastle upon Tyne NE7 7DN

Tel: 0191 284 3111 Fax: 0191 213 1968

Direct Line: [REDACTED]

E-mail: [REDACTED]

Mr JRL Hamilton FRCS Eng, FRCS Ed (C/Th)
Consultant Cardiothoracic Surgeon

JRLH/KP

12 July 2000

Dr R M Chadwick
 Analysis Team Leader
 The Bristol Royal Infirmary Inquiry
 2-10 Temple Way
 Bristol
 BS2 0BY

Dear Ruth

A systematic review of the outcomes of open heart paediatric surgery
London School of Hygiene and Tropical Medicine

Thank you for your letter of 5th July and for giving me the opportunity to review this submission to the BRI Inquiry.

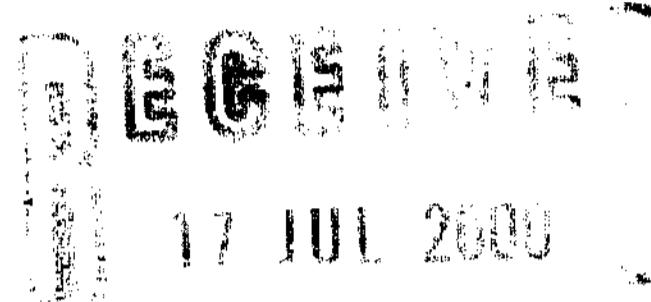
I will make some general comments, raise a few specific points, and then attempt to answer the four questions you have posed in your letter.

The team faced a difficult challenge and I think it has produced an excellent document. It is the first time that these difficult issues have been tackled in such detail and the end result will provide a framework for the future. The work on case mix is particularly valuable and the thoughts on predicting risks for individual patients are very stimulating.

I was interested in the journals surveyed – as an author one would always want to publish in the most prestigious journal and these would undoubtedly be the Journal of Thoracic and Cardiovascular Surgery and the Annals of Thoracic Surgery (in recent years the European Journal of Cardiothoracic Surgery has increased in stature). Thus, if an article appears in another journal, a practising surgeon would assume that it had been turned down by the other two first of all and therefore would not place much weight on it. Nonetheless, occasionally authors choose to publish in more cardiology orientated journals like Circulation and Heart. Interesting, too, how journals have changed over the years – Fontan and Baudet's seminal paper was published in Thorax in 1971 – I would certainly not expect to see a major surgical paper in Thorax nowadays!

The range of mortalities published in the literature for each individual lesion is interesting – surgeons would have been aware of this wide range and thus the problem was knowing which was realistic. Many of the series actually quoted 0% mortality which is obviously unrealistic for any operation.

The authors refer to the anatomical variations in case mix and also to the important of physiological factors – in complete AV septal defect the pulmonary vascular resistance is one of the key risk factors. In TAPVD, the degree of obstruction to pulmonary venous drainage is probably the most important issue of all. However, as the authors note, it is difficult to quantify these.



A few specific points:

- ◆ Table 8 (p 32): case series start date – this is given as 1953 but the earliest Fontan operation was performed in 1968.
- ◆ Paragraph 43 (p 34): pulmonary atresia – a small point, but this should be labelled as pulmonary atresia with intact ventricular septum (and thus hypoplastic right ventricle) to distinguish it from pulmonary atresia and ventricular septal defect (with a normal size right ventricle).
- ◆ Paragraph 49 (p39): was particularly striking – this suggests that the mortalities quoted would be more likely to be realistic and true confirming the scepticism of many practising surgeons of published mortality data in some series.
- ◆ Paragraph 85 (p 64): “There are well recognised methods for weighting local and national data in estimating the current risk in a local setting” – would it be possible to expand on this as I am not clear about what these methods are.

Now to answer the specific questions listed in your letter:

- i) “Common knowledge”: the report deals very successfully with the difficulties of interpreting data from the literature. The point about results improving over the time period is very well made.
- ii) Accessibility of evidence to practising clinicians: surgeons would have had a feeling of the general principles outlined in the report but would not have had the specific factual evidence on which to base their assumptions.
- iii) Risks and outcomes: surgeons would have been aware of the factors that would increase the risk for any particular procedure but again this would be based on “gut feeling” rather than evidence. This report provides that evidence. However, what was realistic mortality rate for any operation has always been a difficult question.
- iv) Journal sources: the review has been wide-ranging and I don’t think there are any significant gaps in the coverage of research evidence. Surgeons would have attempted to keep in touch by attendance at meetings but paediatric cardiac surgery is a small part of the overall specialty of cardiothoracic surgery. Thus, topics related to congenital heart disease would have formed a small part of even the largest meetings – the largest meetings would have been that of the Society of Thoracic Surgeons or the American Association for Thoracic Surgery (both American). Funding to attend these meetings has always been a problem for most surgeons in the UK.

I hope these comments are helpful.

Yours sincerely





Mr JRL Hamilton FRCS Eng FRCS Ed (C/Th)
Consultant Cardiac Surgeon

Royal Brompton & Harefield

NHS Trust

RECEIVED
24 JUL 2000

Royal Brompton Hospital
Sydney Street
London
SW3 6NP

Tel: 020 
Fax: 020 

DM/rd

20 July 2000

Dr R.M. Chadwick
Analysis Team Leader
The Bristol Royal Infirmary Enquiry
2-10 Temple Way
Bristol
BS2 0BY

Dear Dr Chadwick

Re: Systematic review of the outcomes of open heart paediatric surgery

Thank you for asking me to comment on the final version of the above document. I have now completed a detailed reading of the document, which has clearly been very carefully prepared and is presented in a relatively digestible format.

You asked me to comment specifically on a number of points from my perspective as a practising clinician with specialist expertise in congenital heart defects, with reference to the period 1994-1995.

The report reviews published literature. In the mid 1980's, paediatric cardiac surgery and in particular neonatal heart surgery was in its infancy. The tide was running rapidly and much of the knowledge acquired by individual clinicians was, I suspect, obtained at least as frequently from discussion with colleagues in particular at academic meetings, such as the series of paediatric cardiac surgical/cardiology meetings, run by hospitals such as Great Ormond Street.

I agree that the literature presented in the review does represent fairly the common published literature in the field. As the report points out, it is likely that large ("good") centres are disproportionately represented in this series of published papers. Smaller centres are probably not adequately represented, either because they accumulate too few cases to publish contemporary series or because their results are poor and therefore not deemed publishable. If the Bristol results were only compared to published outcomes, then there is a risk that they be judged unduly harshly because of this inherent publication bias towards large high-volume centres.

You asked me to comment on the extent to which the case series presented in the report would have been available to practising clinicians during the enquiry period. I think it is fair to say that the majority of the journals cited were readily available throughout that period in Medical Schools and Departmental Libraries throughout the UK.

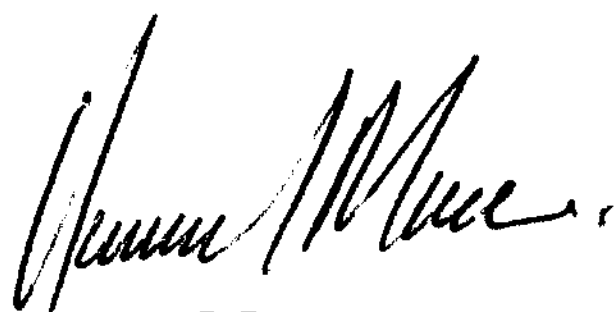
Page 2.

Further, you ask me to comment on the extent to which the evidence presented in the report matches what was commonly known at the time to practising clinicians about surgical risks and outcomes of these high risk operations. I think, if asked that question in an academic setting, the answer would have to be that the published papers do accurately reflect the known surgical risks and outcomes. In the present era, all of these risks would be discussed by a surgeon seeking consent in detail. I suspect that in the era starting in the mid '80's, risks were bundled together and perhaps a little glossed over at times. I think there may have also been a greater tendency in this period to rely on anecdotal recollections of local or regional practice, when describing results or procedures to parents, rather than quoting risks from published series.

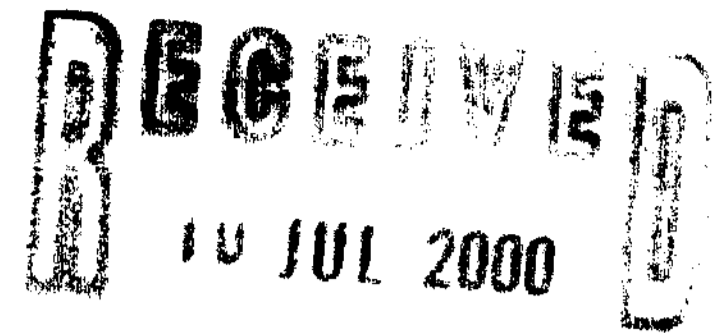
You also asked me to comment on the authority of the journals quoted. I think it is fair to say that the three journals quoted, "Annals of Thoracic Surgery", "Circulation" and the "Journal of Thoracic and Cardiovascular Surgery" were in 1985, and continue to be, the most widely read in the field. Certainly, anything published in these peer review journals is likely to disseminate widely in the paediatric cardiac/surgical and cardiology community and anyone wishing to establish current practice would feel comfortable drawing information from one of these three publications. The report also draws on a wide range of other journals, many of which are also excellent peer review publications. My own view is that the quality of the publications cited in the review is consistently good and will I think be seen as being so by a wide spectrum of the paediatric congenital heart disease community.

I hope these comments are of use to the enquiry team. I would of course be delighted to answer any queries this response may have raised, or indeed to assist you further, should you require this in the future.

Yours sincerely



Duncan Macrae
Director of Paediatric Intensive Care



Royal Brompton & Harefield NHS Trust

Sydney Street, London SW3 6NP Tel: 020 7 352 8121 Fax: 020 7 351 8473 / 8099

Direct Line: [REDACTED] – Direct Fax Line: [REDACTED]

BS/JF
6th July 2000

Dr. Ruth M. Chadwick,
Analysis Team Leader – Inquiry Secretariat,
The Bristol Royal Infirmary Inquiry,
2-10 Temple Way,
Bristol,
BS2 0BY

Dear Dr. Chadwick,

Thank you for your letter dated the 5th July 2000.

I have read the documentation on the systematic review on the outcomes of open-heart paediatric surgery. I would answer your questions as follows:

1. The report itself emphasizes that this type of meta-analysis would not have been available to individual practicing surgeons during the time period under examination. I agree with this conclusion and also I note that the number of UK publications in many of the sub-groups is relatively small. I agree with the assertion that the report essentially indicates the best levels of outcome that might have been available in the management of these five treatment groups under examination during this time frame. I would point out that the series under examination in the main examined outcomes and do not address the detailed issue of surgical risk when this does not involve mortality. The information on surgical risks (in essence the incidence of significant intermediate and long-term complications) is relatively scanty although, clearly, the mortality risks are well assessed in the compilation of this documentation.
2. I think that the case series evidence presented in this report was widely available and accessible during the time frame under consideration. I think that the attributed comments of the expert to the report who noted the problems in reviewing more than a limited number of journals on a personal basis each month is entirely apposite in the context of specialty where most surgeons work extremely intensive rotas and have limited time allocated for personal continued medical education.

The Bristol Royal Infirmary Inquiry

3. I think that the evidence presented in the report does, in the main, match what was commonly known at the time concerning surgical risks and outcomes of higher risk procedures. There is some bias in the Fontan data as it includes cases of Hypoplastic Left Heart Syndrome which were not commonly treated in many centres in the time frame under consideration. Because the number of UK contributing centres during the time under consideration are relatively free, I would hypothesize (albeit from memory) that the UK results may not have been as good as the best average outcomes indicated in the report and I suspect that the actual risks at that time for surgery in many UK centres was possibly therefore higher than the figures quoted in this report. Clearly (as noted in the report) it is impossible to evaluate this by comparison with the UK Cardiac Surgical Register as that data was not validated and, indeed, the structure of the data collection did not make it possible to analyse important sub-groups of the various conditions being assessed.
4. Most of the journals quoted in Appendix A would be perceived by the paediatric cardiac surgical community as sources of seminal or authoritative evidence on surgical risks and outcomes. During the time frame under consideration most of the best publications appeared in the American literature. An important source of evidence on surgical risks and outcomes would have been the Annual meetings of both the British and European Cardiac Surgical Societies at which a steady number of congenital presentations were made on each occasion.

In conclusion, I believe that this report is an excellent report and that the conclusions and recommendations are entirely appropriate.

With best wishes.

Yours sincerely,



B. SETHIA
Consultant Cardiac Surgeon and
Clinical Director of Paediatric Services

COMMENTS ON

A systematic review of the outcomes of open-heart paediatric surgery

The background

The report was commissioned by the Bristol Royal Infirmary Inquiry with the purposes of trying to understand certain factors related to the more complicated operations that were performed during the period 1984-1995. In particular the following specific questions were addressed:

1. The cardiac operations chosen for study and said to be relevant to infants and neonates were:
 - a. The arterial switch operation for transposition of the great arteries
 - b. Repair of complete atrioventricular septal defect
 - c. Repair of total anomalous pulmonary venous drainage
 - d. Repair of truncus arteriosus
 - e. Fontan- type operation
2. The authors set out to establish the knowledge base that might reasonably be expected to have been available to clinicians during the period 1984-1995.
3. They attempted to assess the factors that affected surgical risk and which might therefore have been expected to be taken into account when communicating risk.
4. They investigated wider research evidence on surgical outcomes in order to help inform the Inquiry's assessment of the adequacy of paediatric cardiac surgical care in Bristol.

General Comments:

The Report has been produced as a result of a great deal of thorough, meticulous work. It has certain important strengths which I will attempt to highlight. There also appear to be limitations, most of which have been identified by the authors of the report. I will attempt to indicate how those limitations may be relevant to the clinician, especially during the period covered by the Inquiry

Strengths of Report:

1. The methodology has been very thorough. It probably represents the largest literature search for relevant reports of case series within the group of operations that were considered. The selection of relevant publications appears to have been appropriate. Extraction of data has been carefully achieved and the authors have

been meticulous in scrutinising publications that might have presented duplicated data. They laid down strict criteria that had to be satisfied before any paper was included in their survey. Mortality results were stratified by dividing each of the diagnostic and operative categories into sub-types. Because there was a large number of papers to be surveyed, the task of assessing each paper was usually given to a single author but a reliability study was conducted to assess inter-observer reliability. The statistical methods appeared to be robust but I am not sufficiently expert in this regard to pass judgement.

2. They produced results for 30-day hospital mortality for the 5 groups of operations. The results were divided into periods of time. They had synthesised data from reported case series and they discuss some of the possible limitations. They acknowledge that the papers they reviewed might have had some selection bias and that they tended to emanate from a select number of centres.

Limitations of Report:

1. It is important to recognise that some of the very limitations to which the authors refer might be factors that make the data so selective that they might not be representative of the surgical results in any other than the best centres. It is noteworthy that in para 91 they list 7 centres from which about one-third of all of the case series were derived. Only one of those centres was in the United Kingdom (Great Ormond Street Hospital). From the perspective of the practising clinician, it is well-known that centres do not publish if their results are not the best or near the best. It also is self-evident that editors of the more reputable journals will not accept papers unless they have something new to offer their readers. Clearly then, the selection of the publications that were reviewed must have been significantly biased. The reported mortality rates cannot be representative of the expected results in the world as a whole, nor in any one country. We therefore need to view the mortality results as the very best that could be expected in the United States (5 centres), Australia (1 centre), and the United Kingdom (1 centre). This point is made by the authors in their Conclusions (para 110) but is not given enough emphasis in the report as a whole.
2. The authors also make the point that the knowledge base which they synthesised is not one which would realistically be expected to be available in its entirety to any clinician. No clinician would have the time to read all the papers which were extracted for the purpose of this report and clearly many more papers would have to be read in order to synthesise this selection. No clinician would have the expertise to synthesise the literature as it has been done for the purposes of the report. The authors also point to the time lag between the publication and widespread uptake of evidence in any area of health care.
3. The authors acknowledge the difficulty they would have had in trying to compare the mortality estimates that they produced with those observed at the Bristol

Royal Infirmary. They pointed to the difficulty in accepting the annual mortality results in the UK Cardiac Surgery Register, and considered that it was inappropriate for them to use that data for comparison. Unfortunately, the UKCSR data was the only pool of information readily available to all clinicians in UK who wanted to get what they might have considered to be a reasonable estimate of what the expectations were in the UK. It is unlikely that most centres would have aspired to produce results as good as the best in the United States or, for that matter at Great Ormond Street Hospital. It is also unlikely that clinicians at any centre would have taken into account the best results when communicating expected mortality rates to patients and families. It is more likely that they would have relied on the UKCSR data, however flawed it may have been.

4. There are some minor limitations.
 - a. It is only the arterial switch operation which was researched in relation to the treatment of transposition of the great arteries. Although the authors refer to the earliest operations having been performed in 1975, they were not widely practised in the UK until the early 1990's. The authors do not provide any comparative information for the "atrial switch" procedures, namely the Mustard and the Senning operations. These were widely practised in most centres in the UK during the first 60% of the period covered by the Inquiry.
 - b. The authors refer to the Fontan operation in para 34 and in that context refer to infants with tricuspid atresia who die before reaching 6 months of age if untreated. It is extremely rare for the Fontan operation to be performed in infancy. The earliest age commonly reported in the 1980's was around 2 years, and usually it was not done in children before the age of 3 or 4 years, almost always after one or more earlier palliative operations. The remainder of the discussion about the Fontan procedure is appropriate but paragraph 34 is quite misleading.
 - c. In para 78 it is stated that "surgeons should consider discussing how the specific nature of a patient's congenital anomaly affects the risk of mortality, relative to average risk". This suggestion is unrealistic. The information is not generally available to the surgeons, nor was it even available to the authors of the report.
 - d. In para 80 they suggest that ideally surgeons should collect personal data on outcomes of their operations and contribute these data to a larger national pool, e.g. UKCSR. This in fact is what surgeons thought they were doing throughout the period covered by the Inquiry. They were not to know that later, in 1999, the pool of data to which they had contributed would be so heavily criticised as flawed and inaccurate.

Summary:

I have been asked specifically to comment on the following, which I will briefly summarise:

- (i) The Report was a tour de force in trying to get to grips with the problem of analysing common knowledge about surgical risks. It represented a mountain of work and provided a valid estimate of the best results that have been achieved. However, by its very nature, the selection bias was a significant limitation because it did not provide evidence of everyday expectations in clinical practice.
- (ii) The case series evidence presented in the report would have been available to practising clinicians during the period covered by the Inquiry (1984-1995) but would not have been accessible in the analysed format of the Report. It would have been unrealistic to have expected any clinician to conduct similar analyses.
- (iii) The evidence about mortality rates in the best centres, presented in the report, probably was commonly known at the time to practising clinicians but it would not have been viewed as immediately achievable in most centres
- (iv) The journal sources listed in the Report are widely perceived by the paediatric cardiac surgical community as sources of authoritative evidence on surgical risk and outcomes, but only as found in those centres which aspire to producing the best results.

Eric D Silove
23 July, 2000