

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.

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