Echocardiographic screening for congenital heart disease: a randomized study

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Record Status
This is a critical abstract of an economic evaluation that meets the criteria for inclusion on NHS EED. Each abstract contains a brief summary of the methods, the results and conclusions followed by a detailed critical assessment on the reliability of the study and the conclusions drawn.

Health technology
The addition of echocardiographic screening (ECS) of newborns to the use of scheduled surveillance, in order to detect congenital heart disease (CHD), was studied. Scheduled surveillance consisted of clinical examinations conducted after birth, before discharge from hospital, at 6 to 8 weeks, and at the pre-school assessment.

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
The study population comprised newborns. Infants were excluded if:

- their mothers had insulin dependent diabetes mellitus, a strong family history of CHD, and/or a detected antenatal abnormality of the cardiovascular system; or because of
- Down's syndrome, stillbirth, perinatal death, admission to a special care baby unit, and/or a detected postnatal abnormality relating to the cardiovascular system.

Setting
The setting was a hospital. The economic study was performed in Belfast, UK.

Dates to which data relate
The effectiveness and resource use data were collected between November 1994 and February 2001 (approximately). The price year might have been 1995, although this was not explicitly stated.

Source of effectiveness data
The effectiveness data were derived from a single study.

Link between effectiveness and cost data
The costing appears to have been conducted on the same patient population as that used in the effectiveness analysis. It was not stated whether the cost data were collected prospectively or retrospectively.

Study sample
In the planning phase of the study, it was estimated that a sample size of at least 8,142 eligible cases was required to
assure the study had 80% power, at the 5% significance level, to find a significant difference between the intervention and the control groups in terms of the pick-up rate of CHD. Among 9,697 deliveries occurring during the study period, 4,875 mothers were allocated to ECS and 4,822 were allocated to the control group. The infants were considered for the effectiveness analysis if their mothers were not discharged early and accepted the scan, and the other inclusion criteria were also met (see Study Population). A total of 3,965 newborns were scanned in the ECS group, while 4,022 newborns were examined in the control group. In total, 910 infants in the intervention group and 800 in the control group did not meet the inclusion criteria. The authors did not report any evidence that the study sample was representative of the study population.

Study design
This was a randomised controlled trial that was performed at a single centre. The method of randomisation used was not reported. Randomisation was carried out at the level of the pregnant women instead of newborns. The newborns were followed up for a minimum of 3 years. No losses to follow-up were reported to have occurred. The outcome assessment does not appear to have been conducted in a blinded manner.

Analysis of effectiveness
The authors reported that the basis of the effectiveness analysis was "intention to scan". The results of the effectiveness analysis considered all the infants, independently of whether they met the inclusion criteria. However, the manner in which the paper was written permitted included and excluded infants to be explicitly identified. The primary health outcomes assessed in the clinical study for both groups were:

- the number of cases of significant CHD found before discharge,
- the number of late diagnoses of CHD, and
- the main defect associated with each diagnosis.

A survey of parental satisfaction was also carried out to assess the approval rating for screening in parents whose child had just been scanned. Although the authors stated that there were no significant differences between the study groups in terms of maternal age, birth weight, Apgar scores and social class, they did not provide any evidence to support this statement.

Effectiveness results
When considering all the infants, 124 cases of significant CHD were found before discharge in the ECS group (7 did not meet the inclusion criteria) versus 50 in the control group (23 did not meet the inclusion criteria), (p<0.001).

The relative difference in the detection rate was 2.49 (95% confidence interval: 1.79 - 3.47).

There were 27 late diagnoses in the control group versus 1 in the ECS group.

The diagnoses most frequently found before discharge were ventricular septal defect, patent ductus arteriosus, and aortic valve disease.

The main lesions diagnosed after discharge were ventricular septal defect and pulmonary artery stenosis.

The approval rating for screening in parents whose child had just been scanned was 98%. Eleven per cent of the parents felt the screening intervention caused some unnecessary anxiety, while 99% thought ECS should be used for all newborns.

Clinical conclusions
Adding echocardiography to the usual clinical examination makes screening for CHD among infants 2.5 times more effective, while keeping the parents' satisfaction considerably high.
Measure of benefits used in the economic analysis
No summary measure of benefit was used in the economic analysis. The study was therefore categorised as a cost-consequences analysis.

Direct costs
The direct costs considered in the economic analysis appear to have been those of the hospital. They included the costs of the ultrasound machinery, sonographer's salary, and the use of hospital space and utilities. Moreover, the additional costs incurred by those children with an abnormal echocardiogram, and those incurred in the follow-up of insignificant findings, were also included and reported separately. It was not possible to assess whether the costs were estimated on the basis of actual data or on a guess, as the sources of the direct costs were not reported. Incremental rather than total costs were estimated since the health technology under analysis (i.e. echocardiography) was added to the traditional clinical practice (i.e. usual clinical screening). The resource quantities and the costs were not reported separately. Discounting was not performed, but it might not have been necessary as only costs for the first year of the screening programme were estimated. The price year was not stated clearly, but it might have been 1995.

Statistical analysis of costs
No statistical analyses of the costs were reported.

Indirect Costs
No indirect costs were reported.

Currency
UK pounds sterling (). 

Sensitivity analysis
No sensitivity analyses were performed.

Estimated benefits used in the economic analysis
See the 'Effectiveness Results' section.

Cost results
The initial start-up cost of the screening programme was 109,996 for the first year (i.e. 22 per infant scanned, considering that all infants were included). An additional further expense of 12,408 per year would be incurred to follow-up abnormal findings found with echocardiography, while 6,500 would be required to follow-up insignificant findings.

Synthesis of costs and benefits
The estimated benefits and costs were not combined.

Authors' conclusions
Echocardiography in the first few days of life is very sensitive in detecting cases of significant congenital heart disease (CHD). It can provide vital reassurance for many parents at significant, but not prohibitive, costs.

CRD COMMENTARY - Selection of comparators
Scheduled surveillance was chosen as the comparator since it represented the current practice in the authors' setting. The authors reported that this practice may differ slightly between centres in the UK, therefore, there may be differences between settings. An alternative method of screening for CHD that was not considered in the analysis was the use of foetal echocardiography, although the authors reported that this had limitations in comparison with ECS. You should identify which health technology is currently used in your own setting to detect CHD.

Validity of estimate of measure of effectiveness
This was a randomised controlled trial that was performed at a single centre. It cannot be assessed whether the method of randomisation was appropriate or not, as no details were reported. In addition, although the authors reported that the patient groups were comparable at analysis, they did not provide any evidence of this. Moreover, some risk factors during pregnancy (e.g. drug or alcohol intake, use of over-the-counter drugs) were not considered and might have acted as confounding factors, although they might, to some extent, have been picked up by the Apgar scores. Infants that did not meet the inclusion criteria were finally considered for the assessment of the clinical results. The effectiveness results were analysed on an intention to treat basis. The extent to which this might have biased the results cannot be assessed. Since the inclusion criteria considered initially were not actually followed in the final assessment of the clinical results, the external validity of the results would not apply exclusively to low-risk infants, but to infants in general.

Validity of estimate of measure of benefit
No summary measure of benefit was used in the economic analysis. In effect, the study was a cost-consequences analysis. The use of a summary measure of benefit such as the life-years gained or quality-adjusted life-years gained would have been useful for making comparisons with the benefits of different interventions.

Validity of estimate of costs
All the costs appropriate to the perspective adopted in the economic analysis appear to have been considered. However, as the authors reported, ECS may be time-consuming and the costs associated with the time lost by parents in attending the screening programme were not reported. There was uncertainty about whether this time lost should have been considered as a productivity loss since ECS was performed before discharge. Thus, only the time lost by parents on account of follow-up procedures should be included if a broader perspective (i.e. societal) were to be adopted. In addition, the authors stated that the costs were likely to have been underestimated. No statistical analyses of the costs were reported, which introduces uncertainty into the results of the cost estimation. Since the resource quantities were not reported separately from the costs, and the sources and dates used for the cost estimation were not reported, it would be difficult to carry out reflation exercises in other settings.

Other issues
Comparisons with the results from other studies showed that clinical examination alone may lead to a high proportion of babies with CHD being missed. Although the study population included only infants at low risk of CHD, the final clinical analysis was about infants in general, including high-risk newborns.

Implications of the study
The authors recommended that ECS be extended to all newborns, not only those at a high risk. Moreover, they reported that further research involving a very large number of patients is required, in order to detect differences in morbidity and mortality between scanned and control infants.

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Other publications of related interest


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