Evaluation of heart murmurs in children: cost-effectiveness and practical implications

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
Six diagnostic strategies used to evaluate heart murmurs in children were compared.

Strategy 1: the paediatrician performs a clinical examination and selectively refers those with suspected pathologic murmurs to a cardiologist, (PED/selective to CARD).

Strategy 2: the paediatrician performs a clinical examination, obtains a chest radiograph (CXR) and electrocardiogram (ECG), then selectively refers those with suspected pathologic murmurs to a cardiologist (PED-CXR-EGG/selective to CARD).

Strategy 3: the paediatrician performs a clinical examination and selectively refers those with suspected pathologic murmurs for an echocardiogram (ECHO), (PED/selective to ECHO).

Strategy 4: the paediatrician performs a clinical examination, obtains a CXR and ECG, and selectively refers those with suspected pathologic murmurs for an ECHO (PED-CXR-ECG/selective to ECHO).

Strategy 5: the paediatrician refers all patients with murmurs to a cardiologist (PED/all to CARD).

Strategy 6: the paediatrician refers all patients with murmurs for an ECHO (PED/all to ECHO).

Type of intervention
Diagnosis.

Economic study type
Cost-effectiveness analysis.

Study population
The hypothetical study population comprised patients aged between 1 month and 18 years of age.

Setting
The setting was a hospital. The economic study used data from the USA.

Dates to which data relate
The effectiveness data were gathered from studies published between 1955 and 2001. No dates for resource use were reported. The price year was 2000.

Source of effectiveness data
The effectiveness evidence came from published studies, augmented by the authors’ assumptions.
Modelling
A decision tree model was used to calculate the costs and benefits of the six diagnostic strategies. Details of the decision model were not reported and the tree was not represented.

Outcomes assessed in the review
The model parameters estimated from the review of published studies were:

- the prevalence of pathologic lesions in asymptomatic murmurs; and
- the sensitivity and specificity of the paediatrician's clinical examination, the paediatrician's clinical examination plus CXR plus ECG, the cardiologist's clinical examination, and ECHO.

Study designs and other criteria for inclusion in the review
The study designs were not reported. Except for the evidence on ECHO, the authors stated that all articles included children aged between 1 month and 18 years and used ECHO as the 'gold' standard.

Sources searched to identify primary studies
Not stated.

Criteria used to ensure the validity of primary studies
Not stated.

Methods used to judge relevance and validity, and for extracting data
Not stated.

Number of primary studies included
The effectiveness data were derived from 15 primary studies.

Methods of combining primary studies
The authors used data from the available studies selectively to determine the baseline data. The ranges for the sensitivity analysis were derived from the literature.

Investigation of differences between primary studies
Not stated.

Results of the review
The prevalence of pathologic lesions in asymptomatic murmurs was 0.02 (range: 0.005 - 0.14).

The sensitivity was 0.86 (range: 0.79 - 0.96) for paediatrician's clinical examination, 0.88 (range: 0.89 - 0.93) for paediatrician's clinical examination plus CXR plus ECG, and 0.95 (range: 0.88 - 0.97) for cardiologist's clinical examination. The specificity was 0.61 (range: 0.55 - 0.68) for paediatrician's clinical examination, 0.59 (range: 0.60 - 0.64) for paediatrician's clinical examination plus CXR plus ECG, and 0.83 (range: 0.70 - 0.95) cardiologist's clinical examination. In the base-case, the sensitivity and specificity of ECHO were assumed to be 100%. However, for the sensitivity analysis, the sensitivity was 0.96 (range: 0.93 - 0.96) and the specificity was 0.99 (range: 0.98 - 0.99).
Methods used to derive estimates of effectiveness
The authors made some assumptions that were used in the decision model. Some of the assumptions were made on the basis of published studies.

Estimates of effectiveness and key assumptions
The main assumption used in the analysis was that ECHO was considered as the 'gold' standard for the correct diagnosis of heart murmurs. Other assumptions were that the cardiologist would obtain a 12-lead ECG at the time of initial consultation and an ECHO would be obtained if a pathologic murmur was suspected, and paediatric echocardiographers in experienced centres would perform ECHO evaluations. For the main analysis, it was assumed that ECHO had perfect diagnostic accuracy in determining the presence or absence of pathologic lesions in patients with asymptomatic heart murmurs.

Measure of benefits used in the economic analysis
The benefit measure used in the economic analysis was the percentage of pathologic murmurs detected with each strategy. It was obtained using the decision model. No discounting was applied since the time horizon of the study was very short.

Direct costs
Discounting was not carried out since the costs were incurred over a short time period. The unit costs and quantities of resources were not given. Only the average costs per patient and per case detected were calculated. The health service costs included in the economic evaluation were for CXR plus ECG, ECHO, and cardiologist consultation. The costs of treating heart murmurs were not included. The costs for the initial paediatrician visit were excluded since they were incurred in all strategies. The cost/resource boundary adopted in the analysis was not explicitly stated, but it seems to have been that of the hospital. Outpatient physician costs and test costs were assessed using the resource-based relative value. The Current Procedural Terminology codes were used to estimate the costs for tests and visits. No dates for resource use were reported. The price year was 2000.

Statistical analysis of costs
The costs were treated deterministically in the base-case.

Indirect Costs
The indirect costs were not included.

Currency
US dollars ($).

Sensitivity analysis
One-way sensitivity analyses were performed to assess the robustness of the study results to variations in all the effectiveness inputs and cost items used in the decision model. The ranges used in the sensitivity analyses were derived from the parameter values found in the literature (see 'Results of the Review' section). All of the model inputs were also estimated assuming imperfect ECHO sensitivity and specificity and a correction for related bias.

Estimated benefits used in the economic analysis
The percentage of pathologic murmurs detected was 82% with strategy 1, 83% with strategy 2, 86% with strategy 3, 88% with strategy 4, 95% with strategy 5, and 100% with strategy 6.
Cost results
The average costs were $72 with strategy 1, $137 with strategy 2, $133 with strategy 3, $200 with strategy 4, $174 with strategy 5, and $332 with strategy 6.

Synthesis of costs and benefits
An incremental cost-effectiveness analysis was performed to combine the costs and benefits of the six strategies. In the base-case, strategies 2, 3, and 4 were dominated. The incremental cost per pathologic murmur detected was $38,000 with strategy 5 relative to strategy 1, and $158,000 with strategy 6 relative to strategy 5. The sensitivity analyses showed that the prevalence rate and costs of both cardiologist and ECHO were the variables that most affected the study results. The report of the analyses focused mainly on the threshold where strategy 3 became viable, which was the case within many of the ranges specified.

Authors’ conclusions
Strategies 1 (paediatric clinical examination with selective referral to cardiologist; PED/selective to CARD), 5 (all patients with murmurs referred to cardiologist; PED/all to CARD) and 6 (all patients with murmurs referred for an echocardiogram; PED/all to ECHO) were not dominated by the other options. The choice of the optimal strategy depended on the resources that society was willing to pay for, for the appropriate diagnosis of heart murmurs in children. However, referring children with murmurs to an experienced paediatric cardiologist is clinically, and probably economically, prudent.

CRD COMMENTARY - Selection of comparators
The rationale for the choice of the comparators was clear. The authors included all the feasible strategies for the detection of heart murmurs in children. You should decide whether they represent widely used diagnostic approaches in your own setting.

Validity of estimate of measure of effectiveness
The analysis of the effectiveness used data derived from published studies. However, a formal review of the literature was not undertaken and the results were not combined. The authors reported minimal inclusion criteria for the primary studies, and it was unclear whether differences between the primary studies were considered when estimating the effectiveness. The effectiveness estimates were chosen selectively. The authors made some assumptions used in the decision model. All these assumptions were investigated in sensitivity analyses, as well as the model inputs derived from the literature.

Validity of estimate of measure of benefit
The percentage of pathologic murmurs detected with each strategy was used as the benefit measure. It was obtained using a decision model populated with data derived from the literature and the authors’ assumptions.

Validity of estimate of costs
The perspective adopted in the study was not explicitly reported. The authors stated that only those costs relevant to the diagnostic process were included in the analysis. Formal costing procedures were used to identify the relevant categories of costs. The unit costs and resource use were not given. The costs were treated deterministically, but several sensitivity analyses were conducted on key cost items.

Other issues
The authors did not compare their findings with those from other studies. They also did not address the issue of the generalisability of the study results to other settings, although sensitivity analyses were performed, which aids external validity. However, the costing methodology and values are American. You will need to assess whether or not they are applicable in your setting. The study included paediatric patients and this was reflected in the conclusions of the study.
In addition, the authors stated that the diagnosis of heart murmurs may be more cost-effective in more selected populations (i.e. by age). The authors highlighted some potential limitations to the validity of their analysis. First, the analysis assumed that referral was made to an experienced paediatric cardiologist. Second, ECHO has diagnostic limitations. Third, the impact of the patients’ age was not evaluated.

**Implications of the study**
The study suggested that CXR plus ECG added little information to the initial evaluation carried out by the paediatrician. The strategies of PED/selective to CARD, PED/all to CARD, and PED/all to ECHO were those which appeared more cost-effective for the detection of heart murmurs in children. Future studies should focus on the long-term analysis of the costs and clinical outcomes of the different diagnostic strategies.

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