Comparing costs and benefits over a 10 year period of strategies for familial hypercholesterolaemia screening

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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
This paper considered two strategies for screening for familial hypercholesterolaemia. One strategy involved universal screening of all 16-year-olds. All 16-year-olds with a nonfasting total cholesterol above the 95th percentile are offered a fasting cholesterol test. All individuals with a total cholesterol level above 7.5 mmol/L and a low-density lipoprotein cholesterol above 4.9 mmol/L are referred for diagnostic confirmation of familial hypercholesterolaemia, by either clinical examination by a consultant, or genetic testing of blood or buccal cells. The other screening strategy involved a clinic nurse taking family histories from all patients with familial hypercholesterolaemia and inviting all first-degree relatives for screening.

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
The study populations were different for the two screening strategies. In the universal screening strategy, the study population comprised all 16-year-olds. In the familial screening strategy, the study population comprised all people with a first-degree relative with hypercholesterolaemia.

Setting
The setting was primary, secondary and tertiary care. The economic study was carried out in England and Wales.

Dates to which data relate
The clinical effectiveness of the screening strategies was derived from data published between 1995 and 2000. The authors made assumptions about resource use. No price year was reported.

Source of effectiveness data
The effectiveness data were derived from a review or synthesis of completed studies. Estimates of effectiveness were based on opinion.

Modelling
A model was used to establish the clinical effectiveness and resources used for hypothetical populations.

Outcomes assessed in the review
The following model input parameters were identified from published primary studies:

- the prevalence of familial hypercholesterolaemia;
- the probability of a high cholesterol result;
- the probability of a low second cholesterol test;
- the probability of familial hypercholesterolaemia given a high cholesterol; and
- the age-specific death rates from familial hypercholesterolaemia, with and without treatment with statins.

Study designs and other criteria for inclusion in the review
Not reported.

Sources searched to identify primary studies
Not reported.

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

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

Number of primary studies included
The model input parameters were established from three primary studies.

Methods of combining primary studies
Not reported.

Investigation of differences between primary studies
Not reported.

Results of the review
The following model input parameters were identified:

- the prevalence of familial hypercholesterolaemia was 0.002 in the universal screening population and 0.5 in the family screening population;
- the probability of a high cholesterol result was 0.05 in the universal screening population and 0.4991 in the family screening population;
- the probability of a low second cholesterol test was 0.065 in the universal screening population and 0.065 in the family screening population; and
- the probability of familial hypercholesterolaemia given a high cholesterol level was 0.038 in the universal screening population and 0.9517 in the family screening population.
The age-specific death rates from familial hypercholesterolaemia, with and without treatment with statins, were not reported.

**Methods used to derive estimates of effectiveness**
The authors made a number of assumptions about the model input parameters.

**Estimates of effectiveness and key assumptions**
The authors assumed that the probability of attending a first appointment was 0.55 for universal screening and 0.95 for family tracing. The probability of attending a second appointment was assumed to be 0.75 for universal screening and 0.90 for family tracing.

**Measure of benefits used in the economic analysis**
The health benefit used was the number of deaths averted. This was derived from the model (see Modelling).

**Direct costs**
The direct costs of the health care provider were assessed in this analysis. These costs were for the screening programme, statin treatment for all individuals diagnosed with familial hypercholesterolaemia, and cardiac events. The resource use data were derived from the model (see Modelling). It was possible to establish resource use data for each individual covered in the screening programme. The source of the unit costs was not reported. Some unit costs were reported, but others were not. No price year was reported. The paper did not indicate whether or not the future costs were discounted.

**Statistical analysis of costs**
No statistical analysis of the costs was undertaken.

**Indirect Costs**
No indirect costs were included in the economic analysis.

**Currency**
UK pounds sterling ( ).

**Sensitivity analysis**
The paper indicated that sensitivity analyses had been undertaken. However, the type of analysis and the methods used to identify appropriate ranges were not reported.

**Estimated benefits used in the economic analysis**
Over 10 years, the universal screening strategy would avert 11.7 deaths and the family tracing strategy would avert 560 deaths.

**Cost results**
The total cost was 6,176,649 for universal screening and 46,430,681 for family tracing.

The paper reported that differences in drug costs, attendance rates, discount rates, the cost of a coronary event and the life-years gained did not alter the ranking of cost-effectiveness between the two strategies.
Synthesis of costs and benefits
The cost per death averted was 527,919 for universal screening and 3,187 for family tracing.

Authors' conclusions
Over a 10-year period, screening for hypercholesterolaemia by family tracing was more cost-effective than the universal screening of all 16-year-olds.

CRD COMMENTARY - Selection of comparators
This study considered two screening strategies but neither was used explicitly as the comparator. You should consider how these two options relate to current practice in your setting prior to applying the results of this study.

Validity of estimate of measure of effectiveness
The clinical effectiveness evidence used in this study was taken from the literature. The authors also assumed a number of the model parameters. However, a clear rationale for their choice of values was included in the paper. Other model parameters were taken from primary studies. The paper contained little detail of the review used to identify potential primary studies. A systematic review of the literature does not appear to have been undertaken. Only one source for each model parameter appears to have been used. There was no information on whether other relevant studies were also identified.

Validity of estimate of measure of benefit
The measure of health benefit used in the economic analysis was taken from the model that utilised the clinical effectiveness information.

Validity of estimate of costs
The paper did not report the economic perspective adopted in the study, but it appears to have been that of a health care provider. All the relevant costs seem to have been included in the analysis, with appropriate adjustment to reflect adherence to screening programmes and treatment. A sensitivity analysis of resource use and costs appears to have been undertaken. This will increase the generalisability of the study findings, although a lack of detail in the paper means it is not possible to comment on how comprehensive this analysis was. The paper did not state whether the future costs and benefits were discounted. However, it was reported that a sensitivity analysis, which varied the discount rate for the costs and benefits, was undertaken. This lack of clarity makes it difficult to judge whether this issue has been addressed appropriately. No price year was reported. This reduces the generalisability of the study and also prevents any future reflation exercises.

Other issues
The authors’ conclusions represented their analysis, which was presented in a comprehensive manner. The authors did not compare their findings with those of similar studies, nor consider how their findings could be generalised to other settings.

Implications of the study
The authors suggested that family tracing strategies for screening for familial hypercholesterolaemia should be piloted.

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