Cost-effectiveness of screening for hereditary hemochromatosis
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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
Hereditary hemochromatosis screening.

Type of intervention
Screening, primary prevention.

Economic study type
Cost-effectiveness analysis.

Study population
The model assumes a base case of asymptomatic 30-year-old white man.

Setting
The practice setting was hospital/primary care. The study was carried out in the USA.

Dates to which data relate
Prices related to 1990. Effectiveness data were mainly derived from studies published between 1988-1989.

Source of effectiveness data
Literature and opinions.

Modelling
Decision analysis methods were used to construct a decision tree in order to estimate costs and outcomes.

Outcomes assessed in the review
Probability of effectiveness and side-effects used in the decision model, such as:
bleeding following liver biopsy;
survival following a liver transplant due to cirrhosis;
sensitivity and specificity for: a) initial transferrin saturation test; b) repeated transferrin saturation test and serum ferritin determination.

Study designs and other criteria for inclusion in the review
Not stated.
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
3 studies were used.

Methods of combining primary studies
Combination of studies was not applicable since each was used to get data for a given outcome.

Investigation of differences between primary studies
Not applicable.

Results of the review
Probability of bleeding following liver biopsy was 0.007; probability of survival following liver transplant was 0.55. Sensitivity and specificity for initial transferrin saturation test were 0.96 and 0.94 respectively. For repeated transferrin test and serum ferritin determination, the sensitivity rate was 0.83 and the specificity rate was 0.98.

Methods used to derive estimates of effectiveness
The authors' assumptions were used to estimate a few baseline effectiveness and side-effects probabilities to be included in the decision model.

Estimates of effectiveness and key assumptions
Probabilities for: death following liver biopsy was 0.0001;
diabetes mellitus being insulin dependent was 0.6;
 improvement in insulin-dependent diabetes with phlebotomy was 0.45;
 improvement in non-insulin-dependent diabetes with phlebotomy was 0.50;
 improvement in heart failure with phlebotomy was 0.90.

Sensitivity and specificity of liver biopsy were 1 in both the cases, while for magnetic resonance imaging these were 0.8 in both the cases.

Measure of benefits used in the economic analysis
The measure of benefits used was 'Life-Years Gained' for the different clinical outcomes. Life-years gained were based on the literature or authors' assumptions.
Direct costs
Costs and quantities were not reported separately. The discount rate was 3\% for the base case. Direct costs were based on the authors' assumptions that the cost of testing would fall to a fraction of current costs due to economies of scale. Health service costs were considered, including costs for: serum transferrin saturation test, serum ferritin determination, liver biopsy, treatment of bleeding due to liver biopsy, phlebotomy treatment, magnetic resonance imaging, treatment of hepatocellular carcinoma, treatment of insulin-dependent diabetes mellitus (per year), treatment of non-insulin-dependent diabetes mellitus (per year), hospitalization for congestive heart failure (last year of life) and liver transplantation. 1990 prices were used.

Costs data were derived from the published literature and from actual data (costs at local hospitals). Final costs were calculated using a decision analysis model.

Currency
US dollars ($).

Sensitivity analysis
One-way simple sensitivity analysis was used to test variations in: prevalence rates; proportion of homozygous subjects developing the disease; cost of serum transferrin saturation test; discount rate; homozygotes disease development.

Estimated benefits used in the economic analysis
Early diagnosis and therapy by phlebotomies prevent disease manifestations. Therefore, for a 30 year old individual, the life expectancy in a normal condition is 47 years. However, under different disease scenarios, the life expectancies would be: 38 years in case of hereditary hemochromatosis with cirrhosis; 1 year in case of hereditary hemochromatosis with hepatocellular carcinoma; 18 years after liver transplantation; 34 years in case of hereditary hemochromatosis with diabetes; 30 years in case of hemochromatosis with heart failure.

Cost results
Total and incremental costs were not reported separately but were included in the cost-effectiveness ratio (see following field).

Synthesis of costs and benefits
The total cost per life years saved with a test cost of $20 was $1658. Screening is a dominant strategy at a baseline test cost of $12 although the authors claimed that testing the general population resulted in a test cost of $5. A base case discount rate of 3\% or lower, results in a net benefit of screening. At a discount rate of 8\% the cost per life saved is $25,174 whilst at a rate of 10\% the cost rises to $59,585.

Authors' conclusions
Screening for hereditary hemochromatosis (in young white male patients) has a favourable cost-effectiveness ratio over a wide range of assumptions. Large prospective screening studies are called for to test this hypothesis.

CRD Commentary
1) The clinical evidence was based on previous prevalence studies, with no mention of potential bias therein.

2) There was poor reporting and justification of study synthesis basis for clinical analysis.

3) Base case is an asymptomatic 30 year old white man, thus multi-racial application of the results is difficult.

4) Total and incremental costs could have been reported separately and not directly incorporated in the cost-
Implications of the study
More research may be necessary to incorporate a wider (ethnic) population at risk.

Source of funding
Supported in part by a grant from the Mary M. Gooley Hemophilia Center Inc, Rochester, NY.

Bibliographic details

PubMedID
8147681

Indexing Status
Subject indexing assigned by NLM

MeSH
Adult; Cost-Benefit Analysis; Decision Making; Hemochromatosis /complications /diagnosis /economics /genetics; Humans; Life Expectancy; Male; Mass Screening /economics; Sensitivity and Specificity

AccessionNumber
21995005505

Date bibliographic record published
25/03/1997

Date abstract record published
25/03/1997