A cost-effectiveness analysis of prenatal carrier screening for cystic fibrosis
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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
Prenatal carrier screening for cystic fibrosis.

Type of intervention
Screening.

Economic study type
Cost-benefit analysis.

Study population
The economic analysis was performed for four different backgrounds: whites, blacks, Asians, and Hispanics. The screening procedure was offered to American women and their partner if the woman tested positive.

Setting
Hospital. The study was carried out in New Jersey and Connecticut, USA.

Dates to which data relate
Effectiveness data were collected from studies published between 1990 and 1997. Resource use and cost data were collected from studies published in 1997. The price year was 1997.

Source of effectiveness data
Effectiveness data were derived from a review of previously published studies.

Modelling
A mathematical model was developed from the parameters used to calculate costs and benefits (actually monetary costs of screening versus monetary costs of no screening).

Outcomes assessed in the review
The review assessed the following outcomes: prevalence of cystic fibrosis, prevalence of cystic fibrosis carrier status, screening test’s sensitivity for carrier identification, and the fertility rate for each of the four subgroups.

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
Summary statistics from each study.

Number of primary studies included
Approximately 4 studies were included.

Methods of combining primary studies
Studies were combined using the narrative method.

Investigation of differences between primary studies
Not stated.

Results of the review
The prevalence of cystic fibrosis ranged from 1/32,000 (Asians) to 1/3,300 (whites). The prevalence of cystic fibrosis carrier status varied between 0.011 (Asians) and 0.034 (whites). The screening test's sensitivity for carrier identification ranged from 0.3 (Asians) to 0.9 (whites). The fertility rate varied between 1.94 (Asians) and 2.3 (blacks). Full details are provided in the paper.

Measure of benefits used in the economic analysis
Averted costs, that is the difference between the costs of screening and lifetime costs of a newborn with cystic fibrosis, were used as the measure of benefits.

Direct costs
Screening costs were not discounted. Quantities and costs were reported separately. Direct costs included the average lifetime costs of a newborn with cystic fibrosis and the costs of the amniocentesis package (ultrasound guidance, invasive procedure, genetic counselling, AF DNA testing, karyotype determination, and AF alpha-fetoprotein determination). The quantity/cost boundary adopted was that of the health service. The estimation of quantities and costs was based on actual data. Estimates of costs were derived from two previously published studies. The price year was 1997.

Statistical analysis of costs
Not reported.

Indirect Costs
Not included.

Currency
US dollars ($).
Sensitivity analysis
A sensitivity analysis was conducted on the following parameters: cost per screening test ($50-$150), patient screening acceptance rates (25%-100%), and therapeutic abortion rates (50%-100%).

Synthesis of costs and benefits
The maximum allowable cost for the screening programme to break even should be no more than $189 for whites, $135 for blacks, $22 for Asians, and $72 for Hispanics during the first year of the programme and $369 for whites, $309 for blacks, $42 for Asians, and $144 for Hispanics when the programme reaches full maturity. Assuming a therapeutic abortion rate of 100%, there was a net benefit of $382,369 per pre-natally diagnosed case among whites, and losses of $544,808, $2,410,841, and $391,231 per diagnosed case among blacks, Asians, and Hispanics, respectively. Assuming a therapeutic abortion rate of 50%, there was a net benefit of $58,369 per pre-natally diagnosed case among whites, and losses of $768,808, $2,446,841, and $521,191 per diagnosed case among blacks, Asians, and Hispanics, respectively. The net savings by routine offering of prenatal carrier screening for cystic fibrosis among whites should be approximately $251 million annually.

Authors’ conclusions
Under most assumptions and sensitivity analyses, a prenatal cystic fibrosis carrier screening programme appears to be cost-effective for the white population only.

CRD COMMENTARY - Selection of comparators
Rationale for the choice of the comparator was clear. You, as a user of this database, should verify whether this health technology is relevant to your setting.

Validity of estimate of measure of benefit
Details about the literature review could have been provided. The benefit results would have been more reliable had a more comprehensive review of the literature been undertaken to provide data for the input variables. The authors did not consider other benefit measures such as quality-adjusted life years (QALYs), or patient's willingness to pay for carrier screening.

Validity of estimate of costs
Direct costs were included. Indirect costs in terms of time lost from work and caregiver's time were not included. This was consistent, however, with the chosen perspective (health care system). It was unclear how lifetime costs of a newborn with cystic fibrosis were estimated.

Other issues
Validity of the results depends on the data derived from the literature and the assumptions made by the authors. The generalisability of the results to other settings or countries was not discussed. The authors did not attach a monetary value to invasive procedure-related foetal losses or to the emotional burden of raising a child with cystic fibrosis. The authors did not consider how information on carrier status in the current pregnancy affects future pregnancies.

Implications of the study
The authors state that, given recent developments, the National Institutes of Health consensus statement on cystic fibrosis calls for routine offering of cystic fibrosis testing to couples currently planning a pregnancy and to couples seeking prenatal testing. The panel concluded that the cost is reasonable in relation to the benefits obtained.

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