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
Two different strategies of preconceptional cystic fibrosis (CF) screening of couples were assessed: single entry two step (SETS; start by testing one partner) and double entry two step (DETS; test both partners immediately).

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
The costs of screening were based on a theoretical population of 100,000 couples a year, who were eligible for screening. The costs of the medical care of CF patients were based on the records of 81 CF patients.

Setting
The study was carried out by researchers from the State University Groningen, the Erasmus University Rotterdam and the Free University Amsterdam, The Netherlands.

Dates to which data relate
Effectiveness data related to 1997, based on studies published between 1991 and 1996. Dates relating to resource usage are 1990-1996. The price year was not stated.

Source of effectiveness data
The effectiveness of the different screening tests was based on assumptions.

Modelling
A decision tree was used to demonstrate the different costs and consequences of the two alternative screening strategies.

Methods used to derive estimates of effectiveness
Assumptions about effectiveness were based on expert opinion, published studies and published data from the Dutch Office of Statistics.

Estimates of effectiveness and key assumptions
The first test for both SETS and DETS was delta-F508-mutation analysis with a sensitivity of 77.6%. For DETS, if one partner was positive, the negative partner would be screened again with a more comprehensive test, accounting for another 12.7% of mutations. For SETS, if the first partner was positive, the other partner would be screened with the more comprehensive test with a sensitivity of 90.3%. The authors assumed that every year 100,000 couples were eligible for screening. 10% of couples would remain without children and every other pair would have 2 children. The time between the birth of both children was on average 3 years. Additional assumptions were that 15% of identified carrier couples decided not to have any (more) children, 75% of the remaining carrier couples would use prenatal diagnostic tests, 75% of affected pregnancies would be terminated (induced abortion) and prenatal diagnostic tests had a risk of iatrogen abortion of 1%.

Measure of benefits used in the economic analysis
The number of cases of preterm CF detected and the subsequent reduction in number of CF births was the benefit measures.

Direct costs
The costs of medical care for CF patients who would be born without the screening programme were collected, i.e. hospital costs and costs for home care. Hospital costs were based on the records of 81 CF patients (adults and children). Home care costs were based on diaries of 47 CF patients. Together with data from the Dutch CF registration, giving the life expectancy of CF patients in the Netherlands, average lifetime medical costs for a CF patient were calculated. Future costs were discounted at 5% per year.

The costs of the screening programme were various. Firstly, costs for information to inform the Dutch population about the availability of screening. These costs were assumed to be equal to those incurred in screening for breast cancer. The costs of organising the programme included informing couples about the test, performing the test and sending mouth swabs of both partners from the general practice to the laboratory, DNA-extraction and analysis and reporting of results. The costs of the consequences of screening included those incurred by a involving a genetic consultant in informing identified carrier couples and couples where one partner is a carrier, the use of prenatal diagnostic tests and, in some cases, abortion of the affected foetus.

Indirect Costs
Indirect costs were not incorporated.

Currency
Dutch guilders (Dfl).

Sensitivity analysis
One-way sensitivity analysis based on different levels of screening uptake (25%, 50%, 75% and 100%) was used to assess the impact of different levels on the results.

Estimated benefits used in the economic analysis
The number of identified carrier couples for different uptake levels were as follows:

For SETS:
25% uptake, 19.5;
50% uptake, 38.9;
75% uptake, 58.4;
100% uptake, 77.9.
For DETS:
25% uptake, 22.1;
50% uptake, 44.1;
75% uptake, 66.2;
100% uptake, 88.2.
This produced the following decreases in the number of children being born with CF each year:
For SETS:
25% uptake, 5.4;
50% uptake, 10.9;
75% uptake, 16.3;
100% uptake, 21.9.
For DETS:
25% uptake, 6.2;
50% uptake, 12.4;
75% uptake, 18.6;
100% uptake, 24.8.

Cost results
Average lifetime medical costs for a CF patient in the Netherlands were estimated at Dfl541,894. The costs of both screening strategies were not specified. Savings for different uptake levels were presented in the form of a graph. The costs of screening would equal the savings if approximately 8,000 couples (10%) were screened yearly in the Netherlands.

Synthesis of costs and benefits
A synthesis of costs and benefits was not reported. The sensitivity analysis showed that the savings were no longer greater than the costs if, at a participation rate of 50%, the percentage of carrier couples who used pre-natal diagnostics dropped to about 16%. If the participation rate increased to 100%, screening of pairs would be financially attractive at a take-up rate for diagnostic tests of 6% for DETS and 7% for SETS.

Authors' conclusions
There are no financial objections to pre-conception couple screening in the Netherlands, even with an uptake ratio of around 10%. Whether screening for CF carriers should be offered should be decided on the basis of non-financial arguments.

CRD COMMENTARY - Selection of comparators
The two screening strategies were compared to "no screening", which has a clear rationale.
Validity of estimate of measure of benefit
The estimates used in the derivation of the model were based on experts’ opinion, with reference to some literature sources. Whilst, there was no evidence of a systematic review of available literature, sensitivity analyses were performed to determine the thresholds at which the interventions became economically viable.

Validity of estimate of costs
The cost data were limited in that costs and quantities were not reported separately, the costs of screening were not reported and a price year was not given. However, discounting was applied appropriately to the lifetime costs of treating a CF patient.

Other issues
The generalisability of the cost results is limited by some omissions such as the lack of a price year, bearing in mind the variety of sources used to derive resource use and cost information.

Implications of the study
The authors suggest that the decision to offer screening for CF carriers should be decided on the basis of non-financial arguments.

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