Cost-benefit analysis of a national thalassaemia prevention programme in Israel
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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
A national prenatal screening programme supplemented by educational programmes for thalassaemia major in Israel.

Type of intervention
Screening and primary prevention.

Economic study type
Cost-effectiveness analysis.

Study population
A hypothetical population of all pregnancies and live births in Israel within a calendar year.

Setting
Community. The economic study was set in Israel.

Dates to which data relate
Effectiveness and resource use data were collected from studies published between 1982 and 1991. Cost data were collected from the authors’ institutions and from a study published in 1994. The price year was 1996.

Source of effectiveness data
Effectiveness data were derived from a review of the literature.

Modelling
A decision analytic model was used to determine the cost-effectiveness of a national prevention programme of thalassaemia major.

Outcomes assessed in the review
The review assessed life expectancy, sensitivity and specificity of the initial blood test, proportion of partners accepting electrophoresis, rate of positive electrophoresis, proportion of couples receiving counselling, carrier rates, and prenatal diagnosis compliance.

Study designs and other criteria for inclusion in the review
Effectiveness estimates were based on data from the Jezreel Valley Programme, the Central Bureau of Statistics, the national diagnostic centre for thalassaemia, the national thalassaemia registry, and published studies.
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
At least 6 primary studies were included in the review.

Methods of combining primary studies
Life expectancy was the midpoint of a 25-35 year estimate of life expectancy of subjects with thalassaemia major. Some primary studies had separate inputs, while the methods of combining the remainder of the studies were not specified.

Investigation of differences between primary studies
Not stated.

Results of the review
The results of the review were as follows:

Life expectancy of subjects with thalassaemia was estimated to be 30 years.

The sensitivity of the initial blood test was 99% and the specificity 87.5%.

The proportion of partners accepting electrophoresis was 80% among Jews and 87.5% among non-Jews.

The proportion of partners having positive electrophoresis was 2.18% among Jews and 8.54% among non-Jews.

The proportion of positive couples receiving counselling was 99% among Jews and 90.5% among non-Jews.

The proportion of carrier couples having prenatal diagnosis was 100% among Jews and 57.9% among Arabs in the first years of the programme, rising to 100% in 1996-97.

A 100% prenatal diagnosis compliance was assumed among Jews and 80% among Arabs.

The natural miscarriage rate from 16 weeks to term was 1.5%.

Measure of benefits used in the economic analysis
The number of thalassaemia cases averted was used as the measure of benefits. Modelling was used to estimate benefits.

Direct costs
Direct costs were discounted at an annual rate of 5%. Quantities and costs were reported separately. Direct costs covered the costs of treatment and drugs (initial blood test, electrophoresis, genetics counselling, abortion, and miscarriage). The quantity/cost boundaries adopted were that of the health service. The estimation of quantities and
costs was based on actual data. Electrophoresis costs were obtained from the Ministry of Health laboratory test price list. Laboratory costs were taken from a hospital. Genetics counselling, abortion, and miscarriage costs were taken from a previous study. The price year was 1996.

**Statistical analysis of costs**
No statistical analysis of costs was carried out.

**Indirect Costs**
Indirect costs related to work losses of parent and patient because of thalassaemia. Mortality costs were estimated using the gross national product per head method of valuing life. The quantity/cost boundaries adopted were those of society. The price year was 1996.

**Currency**
US dollars ($).

**Sensitivity analysis**
Sensitivity analyses were conducted on the discount rate, electrophoresis costs, cost of introducing a national health education programme, and the compliance rate.

**Estimated benefits used in the economic analysis**
The number of thalassaemia major births was 26.4 in the absence of screening and 13.0 with screening.

**Cost results**
The total costs of the screening programme amounted to $900,197. The lifetime medical care costs with screening amounted to $4.6 million. In the absence of screening, lifetime medical care costs amounted to $7.5 million.

**Synthesis of costs and benefits**
The screening programme was dominant over the do-nothing alternative as it provided more benefits and was cheaper. Given that the averted lifetime costs of having 13.4 fewer thalassaemia major cases amount to $3.8 million to the health services, and $5.4 million to society, the benefit/cost ratios were 4.22/1 and 6.01/1, respectively. The results were sensitive to changes in the discount rate.

**Authors’ conclusions**
Clinical and public health authorities should act and invest in a national programme for prevention of thalassaemia based on health education and screening activities, which could change the pattern of this disease within a short number of years.

**CRD COMMENTARY - Selection of comparators**
A justification was given for the comparator used, namely the do nothing strategy. You, the user of this database, should decide if this health technology is relevant to your setting.

**Validity of estimate of measure of benefit**
The authors did not state that a systematic review of the literature had been undertaken. More details about the design, conduct of the review and the method of combining primary effectiveness data could have been provided. However, the epidemiologic data were derived from national sources and would, therefore, apply to the Israeli population. Estimation
of benefits was obtained directly from the effectiveness analysis.

Validity of estimate of costs
All categories of costs relevant to the perspective adopted were included in the analysis. As acknowledged by the authors, the costs of bone marrow transplants were not considered. Details of the methods of cost estimation were provided. Quantities and costs were reported separately. The price year and the discount rate were reported. A sensitivity analysis was conducted on costs, but not on quantities.

Other issues
The authors made relevant comparisons of their findings with those from other studies. The issue of generalisability to other settings was not addressed. The authors do not appear to have presented their results selectively. The study examined couples at risk of thalassaemia and this was reflected in the authors’ conclusions. The authors assumed that health educational activities do not have any additional effect on improving compliance with the programme.

Implications of the study
Israel should start to provide a nationwide thalassaemia screening programme, as the monetary benefits to society (and even to the health services alone) will exceed the screening programme’s costs. The use of deferiprone as a substitute for Desferal should also be examined.

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