Screening for sickle cell and thalassaemia in primary care: a cost-effectiveness study

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.

CRD summary
The objective of the study was to assess the cost-effectiveness of sickle cell disease and thalassaemia screening. The authors concluded that sequential primary care screening was cost effective if decision makers were willing to pay £13 to achieve one additional pregnant woman screened. The quality of the methods was good and the reporting of the methods and results were satisfactory. Given the scope of the study, the authors' conclusions appear appropriate.

Type of economic evaluation
Cost-effectiveness analysis

Study objective
To assess the cost-effectiveness of antenatal sickle cell disease and thalassaemia screening in inner-city areas with high proportions of residents from ethnic minority groups.

Interventions
The authors evaluated three sickle cell disease and thalassaemia screening interventions: primary care parallel screening (test offered to the mother and father at the same time), primary care sequential screening (test offered to the mother and then subsequently to the father if the mother is a carrier) and midwife care (sequential screening at first midwife consultation by 10 weeks gestation). Midwife care was the default comparator for the two primary care interventions.

Location/setting
UK/primary care.

Methods
Analytical approach:
A Markov model was used to assess costs and outcomes associated with each of the three interventions under study. The model was based on an existing published UK model by Zeuner et al. (see Other Publications of Related Interest). The time horizon of the study covered pregnancies to their conclusion (birth, termination or pregnancy loss) but did not include the effects of screening on future pregnancies. The authors stated that the health sector perspective was adopted as the base case.

Effectiveness data:
The authors reported that judgements were made for updating parameters in the original model by Zeuner et al. using available recently published studies and UK national statistics. Parameters included ethnic composition of the population, inter-ethnic unions, failure to screen eligible women and acceptance of father to accept screening. The main measures of effectiveness were failure to screen eligible women and father's acceptance to take-up screening. These estimates were derived from a large cluster randomised trial Screening for Haemoglobinopathies In the First Trimester (SHIFT), which included all pregnant woman from 27 general practices in two London areas (see Other Publications of Related Interest).

Monetary benefit and utility valuations:
Not relevant.

Measure of benefit:
The number of women screened by 70 days (gestation).
Cost data:
Direct costs included carrier tests for mother and father, counselling for women or the couple, prenatal diagnosis, termination of pregnancy counselling and termination of pregnancy. Resource use was obtained as part of the SHIFT trial with medical and practice records of all trial participants reviewed and interviews with the personnel involved. Unit costs were derived from routine national sources and hospital finance departments. All costs were reported in UK pounds sterling (£) and updated to 2010 prices using the Hospital and Community Health Services inflation indices. As part of the sensitivity analysis, a subsample of trial patients were required to complete a patient cost questionnaire for travel costs, time off work, lost income and childcare costs to undergo screening.

Analysis of uncertainty:
A probabilistic sensitivity analysis (7,000 replications) attached probability distributions to model parameters. Base case results were presented as means with 95% confidence intervals (CIs). The authors evaluated broadening the economic perspective to include patient costs. The results were presented on a cost-effectiveness plane.

Results
For every 10,000 pregnancies, midwife care was associated with mean healthcare costs of £145,000 (95% CI 119,000 to 167,000) compared with £201,000 (95% CI 169,000 to 225,000) for primary care parallel screening and £178,000 (95% CI 149,000 to 203,000) for primary care sequential screening.

For every 10,000 pregnancies, midwife care was associated with a mean of 264 women screened (95% CI 92 to 580), compared with 2,556 (95% CI 1,276 to 4,444) for primary care parallel screening and 2,887 (95% CI 1,509 to 4,930) for primary care sequential screening.

Costs and outcomes were combined using an incremental cost-effectiveness ratio (additional cost per additional woman screened). Compared with midwife care, primary care sequential screening was associated with an additional cost of £13 per additional woman screened by 70 days. Primary care parallel screening was found to be dominated (more costly and less effective) by primary care sequential screening. Where patient costs were included in the analysis, primary care sequential screening was associated with an additional cost of £15 per additional woman screened by 70 days when compared to midwife care.

Authors' conclusions
The authors concluded that sequential primary care screening was cost effective if decision makers were willing to pay an additional £13 to achieve one additional pregnant woman screened (compared to midwife care).

CRD commentary
Interventions:
The interventions under study were reported clearly and in detail. The rationale for selection of the comparators was appropriate as the primary care screening strategies were compared against midwife care, which appeared to be usual care in authors setting.

Effectiveness/benefits:
The authors did not appear to use a systematic review to identify the published data sources used for updating parameters in the original model and this made it uncertain that all available evidence was included. The sources appeared appropriate. Adequate details of the randomised controlled SHIFT trial (undertaken specifically to inform the model) was provided with a reference for further information. The nature of the trial being performed made it likely that the effectiveness estimates were internally valid. However, the authors used a narrow outcome measure (increase in number of women screened). The authors pointed out that this involved a policy judgement that decision makers were willing to attach a certain value (in this case £13) for every additional woman screened. Use of quality-adjusted life years might have captured the impact of the intervention on quality of life as well as allowing comparisons with other programmes.

Costs:
It appeared that all relevant cost categories and costs for the stated health sector perspective were included in the analysis. Inclusion of patient costs was investigated in the sensitivity analyses. Adequate details of the cost analysis were reported. The authors adequately reported the sources from which resource use and unit costs were derived. Time
horizon, price year and currency details were reported. No discounting was necessary given the short time horizon.

Analysis and results:
The analytic modelling approach used to synthesise costs and outcomes was appropriate. Adequate details of the model structure used were provided and included a graphical depiction and key assumptions. There was an incremental analysis of the alternatives and the results were clearly presented and discussed. Uncertainty in the model was adequately tested using a probabilistic sensitivity analysis. Alternative scenarios were considered. The authors acknowledged some limitations to their analysis such as few data on additional costs associated with the screening process. The generalisability of the findings to other settings was discussed such as higher versus lower areas of prevalence of sickle cell disease and thalassaemia and the specific organisation of primary care services.

Concluding remarks:
The quality of the methods was good and the reporting of the methods and results were satisfactory. Given the scope of the study, the authors' conclusions appear appropriate.

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