Universal versus targeted screening of infants for sickle cell disease: a cost-effectiveness analysis

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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
Screening of infants for sickle cell disease (universal versus targeted to African Americans).

Type of intervention
Screening.

Economic study type
Cost-effectiveness analysis.

Study population
The study population was a hypothetical cohort of 1 million newborns with the racial and ethnic distribution of infants born in the USA in 1994. In the analysis infants were divided into 3 groups: white, African American and non-white, and non-black (the last group included Hispanics, Asians and Native Americans).

Setting
Community. The study was developed at the University of Colorado School of Medicine, Denver, USA.

Dates to which data relate
Effectiveness estimates were derived from studies published between 1981 and 1996. Resources used and cost data were obtained from actual costs incurred in 1994 and previously published average variable costs adjusted for 1994 prices.

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

Modelling
A Markov simulation model was used to evaluate costs and outcomes associated with the prevention and treatment of sepsis for newborns with sickle cell anaemia and sickle beta-thalassemia.

Outcomes assessed in the review
The review assessed the prevalence of haemoglobin SS and the prevalence of sickle beta-thalassemia in each of the three groups of newborns. Other estimated probabilities were the incidence of sepsis and the sepsis case-fatality rate for no penicillin prophylaxis and penicillin prophylaxis treatment, and presentation of the first complication of sickle cell disease.
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
19 primary studies were used to derive transition probabilities.

Methods of combining primary studies
The narrative method was used to combine primary studies.

Investigation of differences between primary studies
Not stated.

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

The prevalence of haemoglobin SS was estimated at 0.000008 for the white group, 0.0000438 for the non-white and non-black groups, and 0.0025 for the African American group.

The prevalence of sickle beta-thalassemia was estimated at 0.0000003 for the white group, 0.0000018 for the non-white and non-black groups, and 0.000006 for the African American group.

Plausible ranges were reported and were tested in the sensitivity analysis.

The incidence of sepsis was taken to be at 0.0989 with no penicillin prophylaxis (PP) and at 0.0159 with PP.

The sepsis case-fatality rate was estimated at 0.27 with no PP and 0.183 with PP.

Presentation of first complications of sickle cell disease (SCD) was estimated as 0.06 at age 6 months, 0.32 at 1 year, 0.61 at 2 years and 0.78 at 3 years.

Again plausible ranges were reported.

These data formed the principal inputs for the Markov model.

Measure of benefits used in the economic analysis
Health benefits were measured as years of life saved (based on the median life expectancy of 45 years for persons with SCD). Years of life where discounted at 3%.
Direct costs
The cost of the initial screening test was estimated by using actual costs incurred in 1994 in the Newborn Screening Laboratory. The cost of the confirmatory test was obtained from the clinical laboratory of the University Hospital in Denver. Costs of family education were obtained from the Colorado Sickle Cell Treatment and Research Center. The cost of antibiotic therapy was estimated from The Children's Hospital of Denver. Costs for treatment of sepsis and anaphylaxis were obtained from published studies. Resources and unit costs were not presented separately. The price year was 1994.

Indirect Costs
No indirect costs were included in the analysis.

Currency
US dollars ($).

Sensitivity analysis
A univariate sensitivity analysis was performed on each variable in the model.

Estimated benefits used in the economic analysis
Given the hypothetical cohort of 1 million infants, the no screening strategy identified no case of disease and 13 deaths, the targeted screening identified 316 cases of disease and 7 deaths, and the universal screening identified 404 cases of disease and 5 deaths would have occurred. The incremental years of life saved (discounted at 3%) by the targeted screening with respect to no screening was estimated at 146, while the incremental years of life saved of the universal versus the targeted screening was equal to 42.

Cost results
Total costs (discounted at 3%) were estimated at $418,812 for the no-screening strategy, $1,398,388 for targeted screening, and $2,690,307 for universal screening.

Synthesis of costs and benefits
In the base-case analysis, compared with no screening strategy, the targeted screening strategy cost $6,709 dollars per additional life saved, while, compared with the targeted screening, the universal strategy cost $30,760 per additional year of life saved. These results were highly sensitive to 3 parameters: the delivery rate for targeted screening (proportion of infants who are intended to be screened and are actually screened), the proportion of African Americans in the population, and the incidence of sepsis. At a baseline delivery rate of 80% for targeted screening, an increase of the African Americans proportion by 50% (baseline 16%) leads to a decrease of the incremental cost-effectiveness ratio of universal screening to $8,000 per additional year of life saved, while a decrease of the African American proportion to 2.5% leads to a cost-effectiveness ratio between universal and targeted screening equal to $110,000 per additional year of life saved. This cost-effectiveness ratio decreases to $17,500 per additional year of life saved when the delivery rate is decreased to 60% (given the baseline African American proportion), and increases again to $86,605 when the delivery rate is increased to 95%. Finally the incremental cost-effectiveness ratio of universal screening versus targeted screening ranges from $29,000 to $205,000 per year of life saved according to the variation in the absolute difference of sepsis incidence between penicillin-treated and untreated patients (0.12 and 0.44 respectively).

Authors' conclusions
Targeted screening compared to no screening was always cost-effective; while universal compared to targeted screening always identifies more infants with disease and prevents more deaths but incurs greater dollar costs. Given the results of the sensitivity analysis, the choice between the universal and targeted screening strategies should be related to the delivery rate of targeted screening and the proportion of African Americans in the population.
CRD COMMENTARY - Selection of comparators
The choice of comparators reflects the common practice in the different states of the USA. You, as a user of this database, should determine if they are relevant to your own setting.

Validity of estimate of measure of benefit
The internal validity of effectiveness estimation cannot be fully assessed since all the data used in the model were not derived from a systematic review of the literature. More information on the search method should be specified. However, to compensate for this limitation, comprehensive sensitivity analyses were conducted on all parameters to increase the robustness of the results.

Validity of estimate of costs
All relevant direct costs were included and calculated on actual costs in Colorado's laboratories. This leads to strong internal validity of the cost result but might lead to problems of generalisability (external validity).

Other issues
Since the data used in the model were not derived from a systematic review of the literature, the issue of comparability with other studies should be addressed. Other limitations are reported by the authors: sickle cell disease is prevalent not only in African Americans but also in other ethnic groups (Mediterranean and Arab) that were classified as white because of lack of data; the implementation of a targeted screening programme might include additional costs (e.g. costs for in-service education of hospital staff, time spent by the nursery personnel to determine the race of infants etc.) that were not considered in the study. Finally the issue of the generalisability of these findings to other settings can be addressed.

Implications of the study
The use of universal or targeted screening should be related to each particular racial context.

Source of funding
None stated.

Bibliographic details

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Other publications of related interest

Indexing Status
Subject indexing assigned by NLM

MeSH
African Continental Ancestry Group; Anemia, Sickle Cell /diagnosis /ethnology /prevention & control; Cost-Benefit Analysis; Decision Support Techniques; Humans; Infant, Newborn; Markov Chains; Neonatal Screening /economics /methods; Outcome Assessment (Health Care); Sensitivity and Specificity; United States /epidemiology
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