Cost-effectiveness of strategies that are intended to prevent kernicterus in newborn infants

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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
Three strategies for the prevention of kernicterus in newborn infants were compared with current practice. Strategy 1 was universal follow-up in the office or at home within 1 to 2 days of early newborn discharge.

Strategy 2 was routine predischarge serum bilirubin with selective follow-up and laboratory testing.

Strategy 3 was routine predischarge transcutaneous bilirubin with selective follow-up and laboratory testing.

Type of intervention
Primary prevention.

Economic study type
Cost-effectiveness analysis.

Study population
The hypothetical study population comprised healthy, term (37 weeks’ gestation or greater) infants who were being discharged from the normal newborn nursery within 48 hours of an uncomplicated vaginal birth. The hypothetical population was based on an annual cohort of 2,800,000 healthy term newborns (70% of all live births) who were eligible for early discharge. No power calculations appear to have been carried out. The sample size was obtained by subtracting 30% of the 4,000,000 total live births per year in the USA to account for births occurring by Caesarean section, preterm births, births with an obvious setting for haemolysis (e.g. Rh incompatibility), or other high-risk conditions.

Setting
The setting was unclear, but it appears to have been a combination of primary and secondary care. The economic study was carried out in the USA.

Dates to which data relate
The effectiveness data were derived from studies published between 1994 and 2004. The resource use data were obtained for 2000 to 2003. The price year was 2002.

Source of effectiveness data
The effectiveness data were derived from a synthesis of completed studies, supplemented by interviews with paediatricians and assumptions.

Modelling
A decision analytical model was constructed to evaluate three different strategies intended to prevent kernicterus,
compared with the current practice for the prevention and treatment of jaundice in newborn infants.

**Outcomes assessed in the review**
The outcomes assessed in the review were the probabilities of:

- a home nurse visit 1 to 2 days after discharge (including probabilities for referral for an office visit after the home nurse visit, laboratory testing after the office visit, hospitalisation for phototherapy after laboratory testing, and treatment with home phototherapy after laboratory testing);

- an office visit 1 to 2 days after discharge (including probabilities of laboratory testing after the office visit, hospitalisation for phototherapy after laboratory testing, and treatment with home phototherapy after laboratory testing); and

- not being examined by a health care provider in the 2 days after discharge.

**Study designs and other criteria for inclusion in the review**
Not reported.

**Sources searched to identify primary studies**
Not reported.

**Criteria used to ensure the validity of primary studies**
Not reported.

**Methods used to judge relevance and validity, and for extracting data**
Not reported.

**Number of primary studies included**
Ten studies were included in the review.

**Methods of combining primary studies**
Not reported.

**Investigation of differences between primary studies**
Not reported.

**Results of the review**
The probability of a home nurse visit 1 to 2 days after discharge was 0.2 under current practice, 0.2 under the universal follow-up strategy, 0.1 under the predischarge serum bilirubin strategy, and 0.1 under the predischarge transcutaneous bilirubin strategy.

The probability of an office visit 1 to 2 days after discharge was 0.3 under current practice, 0.7 under the universal follow-up strategy, 0.5 under the predischarge serum bilirubin strategy, and 0.6 under the predischarge transcutaneous bilirubin strategy.

The probability of not being examined by health care provider in the 2 days of discharge was 0.5 under current practice,
0.1 under the universal follow-up strategy, 0.4 under the predischarge serum bilirubin strategy, and 0.3 under the predischarge transcutaneous bilirubin strategy, respectively.

Methods used to derive estimates of effectiveness
Some estimates of effectiveness were based on authors’ assumptions and interviews with paediatricians.

Estimates of effectiveness and key assumptions
The authors adjusted the probabilities of laboratory testing and receiving phototherapy treatment in their model so that approximately 2% of the cohort infants under the "current practice" strategy received phototherapy. They obtained the remaining probabilities from interviews with paediatricians who represented four group practices and who were interviewed about their practice patterns. Some of the probabilities for predischarge serum bilirubin and predischarge transcutaneous bilirubin were estimates based on the first author's knowledge about neonatal jaundice and its treatment, and on discussions with paediatric colleagues about likely paediatric changes in response to promulgated prevention strategies. The authors assumed that in all three prevention strategies, the threshold for treatment would be lowered.

Measure of benefits used in the economic analysis
The measure of benefits used appears to have been the estimated number of kernicterus cases prevented per year. The authors assumed that the effectiveness of each strategy in preventing kernicterus was the same.

Direct costs
The direct costs were derived from charges by applying a cost-to-charge ratio of 0.58, the Medicare state-wide average operating cost-to-charge ratio for an urban hospital in Vermont, USA. The charges included provider charges for laboratory tests, the costs for supplies for blood sampling, nursing time, the costs of transcutaneous bilirubinometry, the cost of the disposables for each infant tested, the charges for an office visit, the charges for home visits by skilled nurses, hospital charges and the charges for home phototherapy. Provider charges for laboratory tests and hospital charges per day were from the Fletcher Allen Health Care Hospital, Burlington, VT. The costs of transcutaneous bilirubinometry were from the company (Respironics, Murrysville, PA). The charges for an office visit were from the charges for paediatric office visits in Burlington, VT. The charges for home visits by skilled nurses were from the Visiting Nurses Association of Vermont. Savings resulting from the prevention of kernicterus were estimated on the basis of the average lifetime direct and indirect costs per person of cerebral palsy and mental retardation, discounted at 3%. The quantities and the costs were measured separately. All costs were estimated in 2002 dollars.

Statistical analysis of costs
No statistical analysis of the costs was reported.

Indirect Costs
Indirect costs, such as work-loss costs for the parents, were not included.

Currency
US dollars ($).

Sensitivity analysis
One-way sensitivity analyses were performed to investigate the true population incidence of kernicterus, which was unknown, and the relative risk reduction (RRR) with each strategy.

Estimated benefits used in the economic analysis
At an annual incidence of 1:100,000 healthy term births and an RRR of 0.7, the universal follow-up, predischarge serum bilirubin and predischarge transcutaneous bilirubin strategies each led to 20 additional cases prevented per year for the cohort, compared with current practice.

Cost results
At an annual incidence of 1:100,000 healthy term births and an RRR of 0.7, the annual cost for the cohort was $152,392,631 with current practice.

The incremental annual cost for the cohort over current practice was $202,300,671 under the universal follow-up strategy, $112,580,535 under the predischarge serum bilirubin strategy, and $180,150,494 under the predischarge transcutaneous bilirubin.

Synthesis of costs and benefits
The cost per case prevented was highest with universal early follow-up ($10,321,463) and lowest with routine predischarge serum bilirubin screening ($5,743,905).

The results of a one-way sensitivity analysis across a range of RRR assumptions (with a fixed kernicterus incidence of 1:100,000 live births) showed that estimates of the cost per case prevented ranged from $3,750,733 to $77,650,240 when the RRR was varied from 1.0 to 0.1, respectively, across the three strategies.

In all situations, at comparative incidence and relative risk figures, the highest costs resulted from the universal follow-up strategy and the lowest from the predischarge serum bilirubin strategy.

Authors’ conclusions
Widespread implementation of these strategies is likely to increase health care costs significantly with uncertain benefits. It is premature to implement routine predischarge serum or transcutaneous bilirubin screening on a large scale. However, universal follow-up may have benefits beyond kernicterus prevention, which the authors stated were not included in their model.

CRD COMMENTARY - Selection of comparators
A justification was given for the comparator used (i.e. current practice). You should decide if this is a widely used health technology in your own setting.

Validity of estimate of measure of effectiveness
The effectiveness evidence used in this study was taken from the literature, supplemented by interviews with paediatricians and a several assumptions on the model parameters. A systematic review of the literature does not appear to have been undertaken.

Validity of estimate of measure of benefit
The measure of benefit was the cost per case prevented. The estimation of benefits was modelled. The instrument used to derive the measure of health benefit, a decision analytical model, was appropriate.

Validity of estimate of costs
Although the authors reported that a societal perspective was adopted, no indirect costs were included (except the lifetime costs of kernicterus). The authors acknowledged that some of their cost estimates might be lower than actual nationwide costs. The quantities and the costs were measured separately.
Other issues
The authors made some comparisons of their findings with those from other studies. The reliability of their study and the issue of generalisability to other settings were addressed. The authors did not present their results selectively. The authors reported a number of limitations to their study. First, the source of the effectiveness data. Second, the cost per case prevented might have been underestimated for two reasons: the cost estimates might have been lower than actual nationwide costs, and the indirect costs were excluded. Third, other benefits arising from follow-up office or home nurse visits were not identified.

Implications of the study
The authors emphasised that their results suggest that it is premature to implement large-scale routine bilirubin screening (either serum or transcutaneous) before hospital discharge because of the potential for high costs, uncertain effectiveness, and the uncertain population incidence of kernicterus. Research is required to determine the epidemiology, risk factors and causes of kernicterus. Also, to evaluate the effectiveness of strategies intended to prevent kernicterus, and to determine the cost per quality-adjusted life-year with any proposed preventive strategy.

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Other publications of related interest


Indexing Status
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MeSH
Bilirubin /blood; Blood Chemical Analysis /economics; Cost-Benefit Analysis; Decision Trees; Health Care Costs; Hematologic Tests /economics; Hospital Charges; Humans; Hyperbilirubinemia /diagnosis /economics /therapy; Infant, Newborn; Kernicterus /economics /prevention & control; Phototherapy /economics; Quality-Adjusted Life Years