Universal or targeted screening for fetal alcohol exposure: a cost-effectiveness analysis

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
This study examined the cost-effectiveness of testing meconium to detect foetal alcohol spectrum disorder in newborns. The authors concluded that, from a societal perspective, screening and treatment was economically attractive if it was implemented universally and was a dominant strategy if it was targeted to those at high risk. The study was based on valid methodology and, despite the limited reporting of the data sources, the extensive use of sensitivity analyses enhanced the validity of the authors' conclusions.

Type of economic evaluation
Cost-utility analysis

Study objective
The objective was to examine the cost-effectiveness of testing meconium for alcohol exposure in all newborns or only those who have an older sibling diagnosed with foetal alcohol spectrum disorder (FASD).

Interventions
The universal and the targeted screening strategies were compared with no screening. The laboratory test identified alcohol consumption during the second or third trimester of the mother's pregnancy, but was unable to detect consumption only during the first trimester. The test was performed on the newborn's first stool (meconium).

Location/setting
Canada/hospital.

Methods
Analytical approach:
The analysis was based on a decision-analytic model for the short-term and a Markov model with a lifetime horizon. The authors stated that a societal perspective was adopted.

Effectiveness data:
The clinical evidence came from a selection of known, relevant studies. The prevalence and incidence of FASD were based on a review that included observational studies carried out in the USA and Canada. The benefits associated with special educational programmes for infants (intervention) were derived from a clinical trial supplemented with data from other published studies, the details of which were not given. The key clinical input was the accuracy of the meconium test, which was derived from published studies, the full details of which were not provided.

Monetary benefit and utility valuations:
The utility values were based on published estimates and the details were not given.

Measure of benefit:
Quality-adjusted life-years (QALYs) were the summary benefit measure. The use of discounting was not clearly reported.

Cost data:
The economic analysis considered the costs of screening, treatment, which was a pre-school programme that tested for educational needs and fostered self-learning, and the lifetime burden of disease, including the financial benefit of literacy to society. The current charges at Toronto’s Hospital for Sick Children were used to estimate the cost of
meconium test. The other two main cost categories were based on published studies. All costs were in Canadian dollars (CAD) and the price year was 2004. Long-term costs were discounted at an annual rate of 5%.

**Analysis of uncertainty:**

The issue of uncertainty was investigated by one-way sensitivity analyses on the key model inputs using plausible minimum and maximum values. A probabilistic sensitivity analysis was also carried out, using a Monte Carlo simulation, and generated cost-effectiveness acceptability curves.

**Results**

In comparison with no screening, universal screening increased the costs by CAD 232 and gained 0.0035 QALYs, resulting in an incremental cost per QALY gained of CAD 65,875.

The sensitivity analysis showed that these results were sensitive to variations in the discount rates, probability of disease, the probability of detecting mild cases with or without the meconium test, the cost of special education, and the improvement in quality of life with treatment. The probability that universal screening was cost-effective was 0.153 at a societal willingness-to-pay (WTP) of CAD 50,000 per QALY and 0.586 at a WTP of CAD 100,000. A reduction in the cost of the test to CAD 93 (it was CAD 150 in the base case) reduced the incremental cost per QALY gained to CAD 50,000.

In comparison with no screening, targeted screening reduced the costs by CAD 3,000 and gained 0.0898 QALYs, resulting in it being a dominant strategy, which means it was more effective and less expensive. This result was sensitive only to the cost of education. The probability that targeted screening was cost-effective over no screening was 99.9%.

**Authors' conclusions**

The authors concluded that, from a societal perspective, meconium testing for newborns was economically attractive if implemented universally, and was a dominant strategy if targeted at high-risk newborns.

**CRD commentary**

**Interventions:**
The selection of no screening as the comparator was appropriate as it was the current pattern of care in the authors’ setting. The consideration of the two screening strategies was useful in providing options relevant for decision makers.

**Effectiveness/benefits:**
The authors used a selective approach to identify the relevant sources of clinical evidence. Little information on the design and other characteristics of the primary sources was provided, which limits the possibility of judging the internal validity of the estimates. Issues regarding the use of data from different sources were not investigated. Limited details were given of the methods used in the sources for utility values. In general, QALYs are a valid benefit measure, especially for a disease that has a major impact on quality of life. The discounting of QALYs was not clearly reported, but it is likely that, in accordance with Canadian guidelines, a 5% discount rate was used.

**Costs:**
The economic analysis was consistent with the perspective and considered a broad range of costs. The sources of costs were only partially described and more in-depth reporting would have been useful to improve the transparency of the study and to allow replication of the analysis in other settings. Most of the costs were presented as macro-categories and data on resource consumption were not reported. The use of discounting and the price year were reported.

**Analysis and results:**
The analytic approach used to identify the optimal strategy was appropriate. Both total and incremental results (costs and benefits) were reported for all strategies, but universal and targeted screening were only compared with no screening. A direct comparison of universal versus targeted screening would have been useful. The issue of uncertainty was satisfactorily addressed by means of appropriate sensitivity analyses, the results of which were clearly presented and discussed. The authors stated that, although the study should be considered specific for Canada, it is likely that the results could be transferred to other locations, given their robustness to changes in the parameters. It was stated that an issue of equity might exist in the implementation of universal screening, since alcohol abuse was higher in
disadvantaged families.

Concluding remarks:
The study was based on valid methodology. Despite the limited reporting of the data sources, the extensive use of sensitivity analyses enhanced the validity of the authors’ conclusions.

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Other publications of related interest

Chan D, Klein J, Karashov T, Koren G. Fetal exposure to alcohol as evidenced by fatty acid ethyl esters in meconium in the absence of maternal drinking history in pregnancy. Therapeutic Drug Monitoring 2004; 26: 474-481.

Indexing Status
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