A comparative cost analysis of newborn screening for classic congenital adrenal hyperplasia in Texas


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 two-test newborn screening programme for congenital adrenal hyperplasia (CAH).

Type of intervention
Screening and diagnosis.

Economic study type
Cost-effectiveness analysis.

Study population
Newborn infants.

Setting
Hospital. The economic study was carried out in Texas, USA.

Dates to which data relate
Effectiveness and resource use data refer to 1994. Costs were reported in 1994 prices.

Source of effectiveness data
Evidence for final outcomes was derived from a single study.

Link between effectiveness and cost data
Costing was undertaken retrospectively on the same sample as that used in the effectiveness study.

Study sample
The study sample consisted of infants of whom 319,011 were first screens, and 292,969 were second screens. Infants with detected non-classic CAH were excluded from the study. Power calculations were not used to determine the sample size.

Study design
This cohort study involved specimens submitted from 2,700 health care providers. Loss to follow up was 1.6% of infants with positive tests. The duration of follow-up was not reported.
Analysis of effectiveness
The clinical study was based on treatment completers only. Primary health outcomes were cumulative incidence of classic CAH, success rate of detection of classic CAH through clinical methods, and first and second screens. The same group of infants was used as a comparator in order to estimate the effects if CAH was clinically diagnosed.

Effectiveness results
The cumulative incidence of classic CAH was 1:21,701. In the study 2,437 of the screens were positive with 94% of these being first screens and 6% second screens. Low birthweight infants accounted for 613 (25%) of the positive screens gain and were screened and found normal. 15 infants were diagnosed as having classic CAH, of whom 7 were first detected clinically, six were detected with the first screen and two with the second screen.

Clinical conclusions
For detection of classic CAH, the best result is achieved by applying all three procedures together: clinical diagnosis, first screening and second screening tests.

Measure of benefits used in the economic analysis
The benefit measure was the number of cases detected.

Direct costs
The following costs were included in the analysis: costs of specimen collection, specimen testing, follow-up of positive screens and diagnostic evaluation. Quantities and costs were reported separately. Estimation of quantities was based on real data. Estimation of costs was based on real data. The authors stated that the economic analysis was carried out from a societal perspective. 1994 price data were used. It is not clear whether costs were incurred over a period longer than 1 year and, hence, whether discounting would have been appropriate in the economic analysis.

Statistical analysis of costs
Not performed.

Indirect Costs
Indirect costs, in the sense of opportunity costs to society, were not estimated in the study.

Currency
US dollars ($).

Sensitivity analysis
The costs were evaluated in three different alternatives: low, moderate and high. These levels were based on different evaluations of the level of personnel, overheads and number of diagnostic evaluations. In evaluating the base case, the moderate estimates were used.

Estimated benefits used in the economic analysis
15 infants were diagnosed as having classic CAH, of whom 7 were first detected clinically, 6 were detected with the first screen and 2 with the second screen.

Cost results
The costs of newborn screening was estimated to be $1,874,401, with $1,054,686 being the cost of the first screening.
test and $819,715 being the cost of the second screening test. Clinical diagnosis without screening cost $11,312 per diagnosis, clinical diagnosis plus first test cost $115,169 per diagnosis, and clinical diagnosis plus first and second tests cost $242,865 per diagnosis.

Authors' conclusions
A single screen is effective when the purpose of the hospital is to detect newborn infants with severe salt wasting from CAH. However, if the purpose of the hospital is to detect newborn infants with the simple virilizing form of disorder in order to apply early treatment, then the second test screening is necessary but not cost-effective.

CRD COMMENTARY - Selection of comparators
The selection of comparator was appropriately justified.

Validity of estimate of measure of benefit
The study examined a large sample of infants and screening tests, and the estimate of measure of benefits is therefore likely to be internally valid.

Validity of estimate of costs
Only the results of the base-case (medium cost estimates) were reported but not those for cases with low and high cost estimates, i.e. the results of the sensitivity analysis were not reported. Although the study's purpose was to estimate the societal costs of the procedures studied, the authors only considered health care costs in the analysis.

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