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 was to evaluate the effects of increasing the thyroid-stimulating hormone (TSH) threshold for neonatal screening to minimize false-positive results and recall rates. The authors concluded that the threshold of TSH (5mU/L) in the national congenital hypothyroidism screening programme was the most appropriate in terms of cost-effectiveness. There were a few limitations to the study but the authors’ conclusions are reasonable and their suggestions for further research are warranted.

Type of economic evaluation
Cost-effectiveness analysis

Study objective
To evaluate increasing the threshold of thyroid-stimulating hormone (TSH) in current screening for congenital hypothyroidism to minimise false-positive results and recall rates.

Interventions
Four national congenital hypothyroidism screening programmes that used four different TSH cut-off points (5mU/L current threshold, 10mU/L, 15mU/L and 20mU/L) were compared against no screening. Screening comprised a heel prick test of TSH in neonates at three to five days old. Neonates with a Guthrie TSH test equal to or more than the threshold being evaluated were recalled for more confirmatory tests.

Location/setting
Central Health Laboratory of Tehran University of Medical Sciences, Iran.

Methods
Analytical approach:
A decision tree was used to synthesise clinical and cost data to enable a comparison of the strategies. The analysis was conducted using a lifetime horizon. The authors reported that point of view was that of the caregiver.

Effectiveness data:
Screening data were obtained from 34,007 neonates in Central Health Laboratory of Tehran University of Medical Sciences in 2009 and 2,310 with heel prick tests of TSH above 5mU/L were recalled for further tests and 1,197 were tested. Tests revealed that 39 neonates had congenital hypothyroidism using the lab criteria of TSH of 10mU/L or more. Incidence of congenital hypothyroidism was considered equal in males and females. Other data included the number of permanent and transient cases of congenital hypothyroidism, programme coverage, false-negative rates, mortality rates and incidence.

Monetary benefit and utility valuations:
Not relevant

Measure of benefit:
The summary measure of benefit was disability adjusted life years (DALY). Disability weight used was similar to that of Down's Syndrome, citing a local reference study of disease burden.
Cost data:
Data were derived from a local study and Ministry of Health and State Welfare Organization data. Cost categories included were screening, confirmatory test, general and specialised physician visits, drugs, periodic laboratory tests, education and care of mentally retarded patients.

Analysis of uncertainty:
For sensitivity analysis, each input parameter changed ±1SD and its effect on ICERs (incremental cost-effectiveness ratios), incremental costs and incremental effects were calculated and shown by tornado plots. Probabilistic analysis was performed by Monte Carlo simulation using a cohort of 10,000 neonates (5,000 females and 5,000 males). Selected distributions were described.

Results
All screening strategies were less costly and more beneficial than no screening. Each screening strategy dominated no screening (hence negative ICERs). Costs were reported in thousands of US dollars ($) per gained DALY.

At a cut-off of 5mU/L (current practice) compared with no screening the cost was -4.58 (95% CI -4.95 to -4.21), at 10mU/L compared the cost was -4.57 (95% CI -4.94 to -4.20), at 15mU/L the cost was -4.50 (95% CI -4.87 to -4.13) and at 20mU/L the cost was -4.45 (95% CI -4.82 to -4.07).

Sensitivity analysis showed that the results remained robust.

Authors’ conclusions
Authors concluded that the current threshold of TSH in the national congenital hypothyroidism screening programme was the most cost-effective and appropriate.

CRD commentary
Interventions:
A justification was given for the comparator used (different TSH cut-off points for screening). Prevalence of congenital hypothyroidism may vary across settings and may impact on relevant cut-off points.

Effectiveness/benefits:
The screening data were derived from a University hospital data set that appeared to have been collected routinely and included details of neonatal heel prick tests and subsequent recall. Data sources were well presented but how they were utilised to obtain model estimates was not always reported clearly. Data sources were all relevant to the setting but it was unclear whether they would be generalisable to other countries. DALYs was an appropriate outcome measure to capture the burden of disease.

Costs:
Cost analysis was performed from the perspective of the caregiver. It appeared that all relevant categories were included. The main sources of cost data were reported along with all relevant adjustments, price year and relevant exchange rate. The estimates of private and public education and care centres may limit the transferability of the results to other settings. The costing assumptions seem reasonable and the level of reporting adequate.

Analysis and results:
The model structure was described in detail and included a graphical representation. Some sensitivity analysis, including probabilistic analysis, was conducted and demonstrated the robustness of the results. The authors discussed some of the limitations of their analysis and their simplifying assumptions. The major limitation was the lack of appropriate data to fully facilitate the modelling of the underlying condition.

Concluding remarks:
There were a few limitations to the study but the authors’ conclusions are reasonable and their suggestions for further research are warranted.

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