Author's response to reviews

Title: Neonatal hearing screening: modelling cost and effectiveness of hospital- and community-based screening

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1. Minor essential revisions (as proposed by reviewer Abbey Berg) have all been carried out.

2. Response to reviewer’s report (Gil L’Italien)

General
We do appreciate the reviewer’s point of view and we are grateful for his thorough critique of the manuscript.
Dr L’Italien is right to question the purpose of this study. Any equivocal finding provokes this question. Nevertheless, there is overall consensus that equivocal results should be published, although a real “positive” finding might have been more interesting. In order to add value to our findings and to show the possible effects of differing assumptions we performed extensive sensitivity analyses.

Major compulsory revisions
The transition probabilities marked as “authors’ estimate” (Follow-up after screening, Healthy children under suspicion of hearing impairment) were not sufficiently covered by the literature and therefore are an educated guess by pedaudiologists (KU and JB in collaboration with the Human Communication and Deafness Group at the University of Manchester). In a separate publication [1,2] we could already show that variation in these variables did not alter the results substantially.
Admittedly, the range for sensitivity and specificity of screening for sensitivity analysis is tight, but we could also show that variation of the test parameters has only little influence on effectiveness [1]. Additionally, newborn screening devices are sufficiently established. Nevertheless we used a rather wide range (70-99) for the extremes analysis to show the potential impact of the test parameters. The transition probabilities of “natural” discovery without systematic screening were derived from a representative survey, covering all diagnosed cases and the age of diagnosis in Upper Bavaria in 1998 and 1999 [3]. The resulting rates were then smoothed to avoid artefacts. They were not entered as range but as value. Percentage of coverage and follow-up of screening both are very high, so there are only a few children left who would not undergo screening. We felt that changing this probability would not change the results.
Aside from the sensitivity analysis in our opinion the extremes analysis sufficiently showed which parameters would destabilise the model.
We decided not to change the range of the test parameters (as used by the extremes analysis) because a wider range would not be in line with literature and pedaudiologic reality.
We hope to have answered all major concerns.