Reviewer's report

Title: The cost-effectiveness of tracking newborns with bilateral hearing impairment in Bavaria: A decision-analytic model

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Reviewer: Scott D. Grosse

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The authors make the case in this paper that centralized tracking and follow-up of infants who fail hearing screening during the newborn period is not only essential to maximize the detection of bilateral congenital hearing impairment but is also cost-effective. I am in full agreement with this conclusion. A previous article in German by this group of investigators demonstrated the importance of centralized tracking for detection of children with hearing loss in Bavaria in reducing loss to follow-up. This paper is a useful complement to the previous publication. First, it shows that such tracking is cost-effective as well. Second, it will bring this point to a wider, English-language readership. This is the first paper I am aware of that has modeled the economic impact of centralized tracking of hearing screening results in terms of cost per case detected. This paper will be an important contribution to the UNHS literature.

Major compulsory revisions

1. Some clarification is required. The paper indicates that this analysis is based on the experience of tracking of hearing screening results in Bavaria. However, there are discrepancies with published reports of hearing screening in Bavaria, specifically both the pilot project conducted in the northern part of Bavaria from 2003 to 2008 and described in reference 7 and the state-wide screening programme which was implemented in 2009 and described in reference 6. Those discrepancies need to be explicitly addressed, not necessarily in terms of changes to the model but in documenting limitations and implications for the interpretation of results.

2. The first discrepancy is that the screening protocol described does not appear to correspond to either reference. In particular, referrals in Bavaria occur if an infant fails in either ear, not just those with bilateral refers. In the pilot project, there was a 3-step process in which infants were first screened in the birthing centre or paediatric clinic using TEOAE and, if that test failed were retested using AABR. If that test failed, infants underwent a confirmatory test by a physician, followed by a full diagnostic audiologic work-up. The report of the pilot screening project makes clear that infants who failed the test in either ear were referred for further testing. In 2008, 2.2% of infants tested were referred, among which <30%, or 0.6% failed bilaterally. The state-wide screening protocol involves a single TEOAE test in both ears, with a failure in either ear leading to a second test using AABR prior to discharge. Infants who fail both tests in either ear are then
referred for further testing. In 2010, 4.5% of all infants screening in Bavaria were referred for further testing, of which one-third, or 1.5%, failed the hearing screen in both ears. That discrepancy is not critical because tracking was limited to bilateral refers. Consequently, the cost of further testing for unilateral refers would presumably be the same in both arms and therefore not affect the estimates of incremental costs or yield.

3. Rates of diagnosed bilateral congenital hearing impairment in Bavaria appear substantially higher than was assumed in the decision analytic model. The decision model projects that of 100,000 infants screened, 51 cases of bilateral hearing impairment will be found with tracking and 21 without tracking. It is useful to compare these results with the actual experience of the hearing screening programme in Bavaria. Reference 6 reports that in the first year of operation, 2009, there were 108 cases of confirmed bilateral hearing loss detected out of 58,500 infants screened, for a rate of 1.85 per 1000. That is 3 times higher than the rate of 0.51 per 1000 with tracking assumed in the decision analytic model. Moreover, reference 6 reported that 50.7% of the children with bilateral hearing impairment were followed up only because of the tracking centre. That implies that 53 cases of bilateral hearing impairment would have been diagnosed in the absence of the tracking programme, for a rate of 0.9 per 1000, which is more than 4 times higher than the rate projected in the decision analytic model. During the pilot project, both refer rates and diagnosis rates were substantially lower, and only 0.7 per 1000 children screened were diagnosed with and received therapy for bilateral hearing loss. That implies that the sensitivity of the pilot screening protocol was less than half as high as the protocol used subsequently in the state-wide screening programme based on one year of data; confirmatory test results for 2010 were not yet available for all children when the study was published. Nonetheless, the actual rate of detection of hearing loss in the pilot screening project was still almost 40% higher than that projected in the decision analysis on the basis of a hypothetical screening protocol. If actual data on cases of hearing loss detected by screening and tracking had been used, the cost-effectiveness ratio would presumably have been lower.

Minor essential revisions

1. The statement that the protocol was adapted from reference 19 should be modified to make clear that Kezirian et al. modeled the referral of infants who failed testing in either ear, as is standard practice in the United States.

Discretionary revisions

1. The authors may wish to discuss the economic implications of extending centralized tracking to include unilateral refers, some of which may result in the diagnosis of bilateral hearing impairment.