

論文の内容の要旨

論文題目 Cost-Effectiveness Analysis of Neonatal Hearing Screening Program in China

和訳 中国における新生児聴覚スクリーニングの費用対効果分析

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Introduction

Globally, hearing impairment is the third leading type of disability. As adequate auditory stimulation in early childhood is fundamental for optimal speech and language development as well as for the acquisition of literacy skills, a failure to undertake early hearing detection and intervention (EHDI) within the first year of life for permanent congenital and early-onset hearing impairment (PCEHI) can lead to significantly and irreversibly impaired language acquisition. The neonatal hearing screening (NHS) program is the only way to realize EHDI, contributing to improved language development of children with PCEHI and benefiting linguistic, cognitive and psychosocial function.

The NHS program has two strategies: 1) universal screening, covering all live births; and 2) targeted screening, which targets infants with one or more acoustic risk factors. Universal screening can detect infants with the disorder who have no known risk factors; such infants account for approximately 50% of cases. Obviously, a major limitation attributes to large amount of false positive cases due to low prevalence, causing unnecessary follow-up costs and much more parental anxiety.

In China, in 2009 the Ministry of Health decided to scale-up a hospital-based NHS program to all 31 provinces of China. Recently, based on the national agenda, provincial health agencies respond to the implementation of health programs and resource allocation at the regional level. Several questions consequently may arise from the current policy: which strategy should be prioritized; and which protocol should be

adopted? What the conditions to scale-up the NHS program from a targeted strategy to a universal strategy? The key considerations for the decision making in a region would appear to be “optimum coverage” and “cost-effectiveness”.

Therefore, aiming to provide evidence for the decision making of national and provincial policy makers on the implementation of nationwide NHS programs, the objectives of this study are to assess the cost-effectiveness of different NHS strategies, to explore the impact of the accessibility of screening, diagnosis and interventions on cost-effectiveness, and to determine the conditions that are necessary to scale-up NHS from a targeted strategy to a universal strategy.

Methods

The decision model is developed based on the natural history and incorporated the prevalence and the current practices in China. Costs and health effects for the simulated neonates were compared among five strategies: 1) universal screening using OAE and AABR, 2) universal screening using OAE, 3) targeted screening using OAE and AABR, 4) targeted screening using OAE, and 5) no screening. This model determines the proportion of infants with PCEHI in the simulated cohort who would potentially benefited from the NHS program, where detection before 6 months of age and intervention before 12 months of age. The baseline and the ranges of the transition probability parameters for sensitivity analysis derived from empirical and literature data.

I followed the guideline of World Health Organization (WHO) to estimate costs for the program implementation. All costs were discounted to their net present values at an annual rate of 3% and adjusted to international dollars based on the GDP deflator and Purchasing Power Parities.

Apart from the costs of program implementation, I preliminarily attempted to estimate the long-term costs saving resulting from the NHS program. It was estimated based on data from hospitals, disabled people’s federations the provincial health and education agency.

For data collection, a field survey to collect data was carried out in six provinces: Beijing (capital), Shandong (eastern developed region), Hebei (eastern developed region), Henan (moderately developed region), Jiangxi (moderately developed region), and Guangxi (less developed region), considering geographical and socioeconomic representativeness in China.

Population health is expressed as the number of disability-adjusted life years

(DALYs) averted as a result of the screening program. Disability weights are 0.216 and 0.168 for untreated and treated disorders, respectively. Average cost-effectiveness ratio (ACER) is calculated for each screening strategy by summing total costs and total health effects in terms of DALYs averted. WHO defined interventions with ACER less than three times of the gross domestic product (GDP) per capita as the cost-effectiveness. Incremental cost-effectiveness ratios (ICER) in different screening strategies are calculated by dividing the incremental costs by the incremental health effects, in order to determine the priority to purchasing those services in different budget levels.

Results

Benefit population of the current neonatal hearing screening program

The coverage rate, the diagnosis rate and the intervention rate determined the total number of deaf infants finally receiving early interventions and benefiting from the screening program. There was a huge disparity in this figure between different provinces: the figures in the developed eastern provinces were much higher.

Cost-effectiveness of different screening strategies

Based on the WHO's reference for cost-effectiveness ratios of three times GDP per capita (Tan-Torres et al, 2003), the two targeted strategies remained cost-effective with an ACER of 13,100 (95% CI: 8,400-17,200) and 19,100 (95% CI: 13,300-27,500) international dollars per DALY averted. On the other hand, the universal screening strategies proved to be less cost effective with ACER figures of 28,400 (95% CI: 14,500-41,900) and 35,600 (95% CI: 20,000-63,000) international dollars per DALY averted.

Among the four strategies, targeted OAE is most cost-effective. The incremental cost-effectiveness analysis suggests that shifting from targeted OAE to targeted OAE plus AABR, to universal OAE and to universal OAE plus AABR costs 127,700 (95% CI: 98,000-180,000), 43,000 (95% CI: 25,800-62,400), and 55,000 (95% CI: 32,000-87,000), respectively, for averting per DALY. Depending on the budget level, the optimal path for expansion would be to start with targeted OAE and then expand to universal OAE and universal OAE plus AABR.

Sensitivity analyses

Multivariate sensitivity analyses were performed for transition probability

parameters to determine the robustness of the model. The results suggested the variables whose range of uncertainty had a great impact on the cost-effectiveness of the screening strategies were the program coverage, diagnosis rate, and intervention rate.

The proportion of the benefit population, estimated by multiplying the three variables, reduced ACER considerably in the four strategies and achieved high cost-effectiveness as it increased. Targeted OAE, targeted OAE plus AABR, universal OAE, and universal OAE plus AABR trended to be cost-effective from the level of 7%, 10%, 20% and 30%, respectively.

Economic effects on long-term costs saving

If long-term costs saving are counted, all four strategies tend to be cost-effective. When the proportion of the benefit population expands, the effect of these screening strategies on the long-term costs saving will be more and more significant, exceeding the total costs of the screening program implementation, suggesting a good economic effect in the long term.

Discussion and conclusion

This is the first study to estimate the costs and health effects of a screening program for neonatal hearing impairment in the developing world. The goal of NHS program in China is to establish a nationwide hospital-based universal program and to continuously expand diagnosis and intervention services. The rationale for implementing a universal strategy is that it has much better health and economic effects and that if taking long-term costs saving account, it is cost-effective. Conversely, with the current level of the program coverage and the accessibility of diagnosis and intervention services, the targeted strategies were more cost-effective than the universal strategies. The decision making should depend on the conditions in different regions, as the program's implementation varies by different socioeconomic development status. The benefit population needs to be expanded to more than 20% to achieve a good level of cost-effectiveness for universal OAE and universal OAE plus AABR. In the case of the surveyed six provinces, a sufficiently high coverage rate, diagnosis rate and intervention rate ensure that the universal strategy achieves good cost-effectiveness and the relevant health and economic effects in Beijing, Shandong and Hebei; but in the other three provinces Henan, Jiangxi and Guangxi, where these three indicators were low, the targeted strategies tend to be more feasible.

To maximize the effect of the NHS program, it is thus crucial to expand the

proportion of the benefit population by improving the accessibility and utility of related services. Obstacles to the scale-up of the NHS program are huge disparity in the program's implementation and regional budgetary scale; out-of-pocket payment; and lack of audiologists and specialists for the rehabilitation.

Our study was restricted by a limited availability of data and evidence. Globally, there was no strong evidence based on randomized control trial for the effectiveness of early intervention on language development, and consequently long-term outcomes. In China, there is lack of population-based study to epidemiological survey for PCEHI. Nevertheless, this study provides evidence for the implementation of the NHS program in different Chinese regions.

In conclusion, with the expanded accessibility of screening, diagnosis and intervention services, the universal strategy will gradually become more cost-effective and bring more health and economic benefits. For the decision making on the implementation, the provincial policy makers should consider their financial capacity, conditions of the scale-up, and the potential benefits. The reference data from this study are thus expected to be of particular benefit in terms of the 'rolling out' of the national plan.