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ORIGINAL ARTICLE

Cost–Benefit and Effectiveness of Newborn Screening of Congenital Hypothyroidism: Findings from a National Program in Iran

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In this study, the cost-benefit of a screening program based on the rial, the unit of currency used, was analyzed. The intelligence quotient (IQ), and height and weight were evaluated as indices for a population of children suffering from congenital hypothyroidism (CH). Materials and Methods: The total cost for the screening program, including hormone tests, diagnosis, medicine, treatments and care was identified and calculated up to the age of seventy years and this was compared to the costs related to training and caring for patients suffering from mental retardation, who had not been screened. The screening test was done using S&S filter paper and thyroid stimulating hormone (TSH) was determined by the ELISA test.The future costs and benefits with an annual rate of 3% discount of their current value was estimated. To evaluate the effectiveness of the screening program, IQs of 32 patients were identified and compared to 36 healthy children between 2004-2005. The IQs were evaluated according to "Good Enough" and "Proteus Maze" tests. Results: During 2008, 1165169 of 12489136 newborns (51% male, 49% female) underwent screening for CH, 92% coverage of all newborns for that year. Four percent of recalled infants, with TSH>5 were subjected to diagnostic tests (T4, TSH, T3RU) and finally 2745 patients were identified. Benefit to cost ratios, based on a 3% annual discount rate, were 22, 41, 32, 34, 47 and 60 times lower, respectively. No significant differences were found with regard to diferences in IQ scores between cases and controls (cases: 105±19.3, controls: 111±19.4), height (cases: 106±0.7, controls: 102±4.9 cm) and weight (cases: 15.6±4.6, control 15.3±3.2 kg). Conclusion: The national Newborn Screening (NBS) program for CH has been successful and quite effective in Iran. The method not only has economical advantages but also reduces capital expenditures and preserves normal IQ of the patients under treatment and prevents mental retardation and growth complications.

Key Words: Newborn screening, Congenital hypothyroidism, Cost-benefit, Mental retardation

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Introduction

Congenital hypothyroidism (CH) is one of the most preventable causes of mental retardation[1]. National screening provides an opportunity for early detection and timely cure of newborns suffering from various genetic, glandular, vascular, hematological and lung diseases[2]. Screening for CH is regularly performed in most developed countries and in some of the third world countries as well[3-7]. Congenital hypothyroidism is a relatively common congenital disorder occurring in about 1 of 3000 to 1 of 4000 live births. Previous studies have reported a high incidence of CH in Iran[8-10]. In 2006, US preventive Services Tash Force (USPSTF) following a brief literature review reported the benefits of screening for CH continue to be well established. This update included a search for new and substantial evidence on the benefits and disadvantages of screening. The American Academy of Pediatrics (AAP) and the American Academy of Family Physicians recommend universal newborn screening for congenital hypothyroidism, and both in conjunction with the American Thyroid Association, and the Lawson Wilkins Pediatric Endocrine Society have recently published guidelines for screening and treatment for congenital hypothyroidism[11,12]. The cost-benefit and effectiveness of health services play important roles and have implications in the design and evaluation of health policies. The high incidence and prevalence of metabolic diseases, particularly congenital hypothyr-oidism (CH) and the damage caused thereby, such as mental retardation should considered as a critical issue and health priority in related policies. The object of this study was to evaluate the cost-benefit of the national newborn screening (NBS) program for CH. The intelligent quotient (IQ), height and weight were evaluated as indices to assess effectiveness of screening programs for children suffering from CH.

Materials and Methods

Total costs for screening program consisting of hormone tests, diagnosis, med-icine, treatments and caring were identified and calculated up to the age of seventy years and this was compared to the costs related to training and caring for patients suffering from mental retardation, in whom screening had not been applied. Excel was used to analyze data. The disability-adjusted life year (DALY) index was determinate by (DALY= YLL+YLD, YLL= N (0) * L (70), YLL=0 formula, standard life expectancy, incidence rate and aggrigation of disability according to the Dutch table were 70 years, 2.2 and 0.35 respectively. IQ score was determined from one of several different standardized tests, designed to assess intelligence. To evaluate the effectiveness of the screening program, IQs of 32 patients were identified and compared to 36 healthy children during 2004-2005; both groups were aged between 4-5 years and were of the same geographical area, social and eco-nomical classes; their IQs were evaluated using the "Good Enough" and "Proteus Maze" tests. SPSS software was employed to analyze the results. T-test was used to compare the mean values of the two groups, the cases and controls. Normal distribution and equal variance of the groups were assured by the application of “Kolmogorov-Smirnov" (KS) and "Leven" tests of evaluation.

Results

During 2008, 1165169 out of 12489136 newborns (51% males, 49% females) were subjected to the screening test, indicating 92.6% coverage of the total number of newborns for that year. Of recalled infants having TSH>5, 4.1% were subjected to diagnostic confirmation tests (T4, TSH, T3RU) and finally 2745 patients were identified. Incidence rate, coverage, number of recall and percent of recall were 2.38 per 1000, 92.6%, N: 46979 and 4.1 percent respectively. The DALY index was 53.9. In 2008, the total cost of screening, patient

finding per each recalled newborn, identifying a patient in the program, medical care for each transient CH patient during 3 years, and for each permanent CH patient during 70 years and mean of total screening cost, patient finding and medical care per healthy and sick patients newborns during 70 years and are shown in table1.

Table 1. Total cost of Screening, patient finding per each recalled newborn, identification of a patient in program, medical care for each transient CH patient during 3 years and for each permanent CH patient during 70 years

Cost	(Million Rials)	$
Screening	2630	2.6
Cost of patient finding per each recalled newborn	95106	9.5
Cost of identifying a patient in program	12704554	1270
Total cost of medical care for each transient CH patient during 3 years	1425865	143
Total cost of medical care for each permanent CH patient during 70 years	3377778	338
Mean of total screening cost, patient finding and medical care per each healthy and patient newborns during 70 years	35868	3.6

Table 2 shows that benefit to cost ratio with regard to education and care of patients with mental retardation. 100% in the public sector, 100% in the private sector, 50% in the public sector and 50% in the private sector, 100% in the public sector day and night, 50% in the public sector and 50% in the private sector day and night, 100% in private sector day and night were 22, 41, 32, 34, 47 and 60 times lower respectively (Table 2).

Table 2. Proportion of benefit to cost ratios in congenital hypothyroidism screening

Education and care of patients with mental retardation	benefit / Cost (million Rials)	Times lower
100% in the public sector	923693/41649	22
100% in the private sector	1693801/41469	41
50% in the public sector and 50% in the private sector	1308747/41469	32
100% in the public sector day night	1407432/41469	34
50% in the public sector and 50% in the private sector day night	195031/41469	47
100% in the private sector day night	2493830/41469	60

Evaluation of the effectiveness of this program between the case and the control groups in respect of IQ score are presented in Table 3.

Table 3. Comparison of age, weight, height and IQ indexes in case and control group

Variables	Cases Girls=14, Boys=18	Controls Girls=14, Boys=21	P value
Age(year)	4.9±1.4	5.0±3.5	NS
IQ(score)	111±19.4	105±19.3	NS
Weight(kg)	15.3±3.2	15.6±4.6	NS
Height(cm)	102±4.9	106±0.7	NS

In 2008 with the implementation of a screening program and identifying 2745 patients, the IQ score and DALY were CH in respective order of 98820 points and 1479555 years.

Discussion

The national NBS program for CH has been successful and quite effective in Iran. The method not only has economical advantages, and helps lower expenses, it also preserves the normal IQ of the patients under treatment and, last but not least, prevents mental retardation and growth complications. Other studies from Iran on national screening programs have documented a cost to benefit ratio of around 1 to 14 [2]. In 2008, of 46,979 infants recalled 2745 patients (6 %) were identified. In a similar research from the Zhejiang Province of China, of 6750 recalled newborns, 764 (approximately 11%) were screened[13]. In Scotland a study confirmed that after screening, the prevalence rate of disease in the first period (1979-1993) was 1 in 3655 live births, and in the second period (1994-2003), it reached 1 in 4363 live births[14]. Policy makers in accordance with the standards of America, interventions cost less than 50 to 60 dollars per quality-adjusted life year (QALY) is quite efficient[15]. National Institute for Health and Clinical Excellence (NICE) in the UK a reference laboratory for clinical eval-uation of interventions found that interven-tions amounting to less than 20 to 30000 pounds per QALY are acceptable[16]; the program conducted in Iran, was defi-nitely more cost effective. A cost benefit ratio of 1:12 was documented for a screening program to identify congenital hypoth-yroidism, conducted in Layl of France[17]. Comprehensive screening programs to control congenital hypothyroidism (CH), a preventable form of mental retardation, are today seriously being considered by public health agencies[18-20]. A study form the UK documented congenital hypothyroidism screening programs to be a cost effective means for follow up of objectives[21]. A 1995 report from the USA declared the cost benefit of screening was 10 -fold, a finding which in line with previous reports.[22-24] Using the first report doucument after implementation of the national screening program, in the current study, effectiveness of such screening programs was evaluated by IQ score and maintaining growth (height, weight and age) in identified and treated patients; non significant differences in IQ score average between aged 4 to 5 years in Esfahani children was observed 3. There was only one study in Iran which assessed children's IQ score by Bender Gestalt and Ravn test in Mahdieh hospital of Tehran. Mean ± SD for case and control groups were (98±11) and (106±8)[25]. Treatment in all patients studied was begun a month earlier, the first dose of drug being adjusted according to previous studies (10-15 µg/kg) of infant's body weight[26,27]. Several studies have shown a strong relationship between intelligence and early treatment and disease severity in patients with congenital hypothyroidism[28-30]. It seems that adequate doses of the drug is one of the causes for having normal intelligence quotientsin congenital hypothyroidism patients. Average date of treatment starting was 15.5 days after diagnosis; this finding was in line with previous reports[31, 32].

In line with our findings, one study showed that there was an inverse relation between age of patient at diagnosis and IQ score. Earlier diagnosis leads to higher IQ score. In this study, the patients who were treated at birth time (12 to 30 days after birth) had 15.7 higher average IQ scores compared to those treated 30 days after birth.

In conclusion, he national NBS program for CH has been successful and quite effective in Iran. The method not only has economical implications which reduces capital costs, it also maintains the normal IQ of patients under treatment and, last but not least, prevents mental retardation and growth complications. Hence a suitably designed screening cost effective program opens new pathways for applications of such programs for other metabolic diseases.

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