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# The Evaluation of Congenital Hypothyroidism Screening Program in Indonesia: A Literature Review

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#### ABSTRACT

Congenital hypothyroidism is one of the main causes of mental retardation which can be prevented. Reasearch in 2020 found that the prevalence of congenital hypothyroidism in Indonesia was 1:1167. During the previous 18 years of impelementation of the Congenital Hypothyroidsm Screening Program (CHSP), it was found that only 4,6% of newborns. This study aims to conduct a literature review to identify problems and obtain information on improving the implementation of congenital hypothyroidism screening programs in Indonesia. This research was carried out between November-December 2021 and consists of a review of a range of research articles and journals which have been publishes within the last decade. Several studies have shown that during the implementation of the CHSP, several onstacles were found which hindered the effectiveness of the program, namely the lack of training for health workers, sources of financing which have not been fully borne by the center, accuracy of collection and sample delivery, and less than optimal management function in regulating the implementation of the program thus a more in-depth evaluation needed, especially in terms of economic benefits and the level of effectiveness of the screening program.

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#### ABSTRAK

Hipotiroid kongenital merupakan salah satu penyebab utama keterbelakangan mental yang dapat dicegah. Penelitian pada tahun 2020 menemukan bahwa prevalensi hipotiroid kongenital di Indonesia adalah 1:1167. Selama 18 tahun terakhir pelaksanaan Congenital Hypothyroidsm Screening Program (CHSP), ditemukan hanya 4,6% bayi baru lahir. Penelitian ini bertujuan untuk melakukan studi literatur untuk mengidentifikasi masalah dan memperoleh informasi dalam meningkatkan pelaksanaan program skrining hipotiroid kongenital di Indonesia. Penelitian ini dilakukan antara November-Desember 2021 dan terdiri dari review berbagai artikel penelitian dan jurnal yang telah diterbitkan dalam dekade terakhir. Beberapa penelitian menunjukkan bahwa selama pelaksanaan CHSP ditemukan beberapa kendala yang menghambat efektifitas program, yaitu kurangnya pelatihan bagi tenaga kesehatan, sumber pembiayaan yang belum sepenuhnya ditanggung oleh pusat, ketepatan pengumpulan dan penyampaian sampel, dan kurang optimalnya fungsi manajemen dalam mengatur pelaksanaan program sehingga diperlukan evaluasi yang lebih mendalam terutama dari segi manfaat ekonomi dan tingkat efektivitas program skrining hipotiroid kongenital.

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#### INTRODUCTION

Congenital hypothyroidism is an inherited disorder that causes a deficiency of thyroid hormone in the body. Congenital hypothyroidism is one of the causes of mental retardation or mental retardation that can be prevented by early identification and treatment in the baby's life (Hatam et al., 2016). The global prevalence of congenital hypothyroidism varies widely. The prevalence of congenital hypothyroidism is estimated at 1:3000-4000 newborns and increases to 1:300-900 in areas with iodine deficiency. Quebec Canada reported prevalence of congenital hypothyroidism 1:2976 (Deladoëy et al., 2011), Saudi Arabia 1:2470 (Shaikh et al., 2021), Mongolia 1:2091 (Tsevgee et al., 2021), Sri Lanka 1:1682 (Hettiarachchi and Amarasena, 2014). Malaysia reported an incidence of congenital hypothyroidism studied from January 2009 to December 2013 of 1:1170 births (Hussain, Taib and Zainol, 2013). Between 2000-2014 of the 213,699 newborns who were screened for congenital hypothyroidism, positive results were obtained for 85 babies with the disease with a prevalence of 1:2513. When compared with the global prevalence of 1: 3000, the prevalence in Indonesia is significantly higher (Center for Data and Information of the Indonesian Ministry of Health, 2015). According to Deliana et al (2016), cases of congenital hypothyroidism in Indonesia occur sporadically with an estimated prevalence of 1:2916. However, a recent study carried out in 2020 displayed a higher prevalence of 1:1167 (Pulungan et al., 2020).

The prevalence of congenital hypothyroidism in a country can reflect the true incidence if screening can cover all births. In Quebec, Canada, since 1987, screening for congenital hypothyroidism has been performed on 98.8% of 2011). newborns (Deladoëy et al., Congenital hypothyroidism screening coverage in Iran during the 13 years of implementation was 98% of the total number of babies born (Yarahmadi et al., 2020). Evaluation of the implementation of congenital hypothyroidism screening in Malaysia from 2004 - 2013 at a hospital showed that the program's coverage was 98.8% of newborns (Hussain, Taib and Zainol, 2013). In Indonesia, congenital hypothyroidism screening was first carried out in 2000, and according to the 2018 Basic Health Research report, only 4.6% of newborns were screened for congenital hypothyroidism.

Congenital hypothyroidism screening is not only about laboratory tests. However, it is a system of combining processes/procedures with the individuals involved, namely the management of health facilities, the person in charge of the program, health workers, parents, the community, both central and local governments. The system in question is the component of Communication, Information, Education (KIE), diagnosis, management, case monitoring, organization, and program monitoring and evaluation. The description above is the basis for the researchers to write a literature review because they want to know more about the evaluation of the implementation of the congenital hypothyroidism screening program (SHK) in Indonesia so that problems can be identified that affect the congenital hypothyroid screening system so that it can be used as information for policy holders in improving the implementation of the following program.

#### METHOD

The research method used is a qualitative literature review study by collecting data from various literature sources (books, journals, theses, articles that have been proceeded) regarding national and international congenital hypothyroidism screening published on Google Scholar, PUV Med, Science Direct, Research gate and Elsevier. Due to the limited number of journals regarding congenital hypothyroidism screening, especially journals on evaluation of hypothyroid screening in Indonesia, the authors use the publication period of the last ten years, namely in 2011-2021. Moreover, the authors used references from international journals to analyse the journals reviewed even though Indonesia is mentioned in the title as the research area of this literature review. Searches were performed using the keywords "newborn screening", "congenital hypothyroidism" or "congenital hypothyroid screening evaluation". The findings of all existing journals will then be examined based on the suitability between the themes discussed by the journals studied and the themes chosen by the authors. The resulting journals will be re-sorted according to the characteristics of the research place discussed with the characteristics that exist in Indonesia. The results of this sorting are then used as material for analysis and reference in writing this literature review.

The theory of evaluation of the CHSP used by the authors in this literature review is a systems-approach theory, through which various factors are examined, including the input, process, and output of the program. The author chose a systems approach as the underlying theory in writing this literature review because, according to Azwar (1996) and Adisasmito (2007), the systems approach provides a function in overcoming health problems by increasing the coverage of health services. This is in line with the problem with the implementation of the CHSP in Indonesia, namely the low coverage of the program. The activities of collecting journal articles used as references as well as literature review analysis and the result presentation are carried out for 2 months, November - December 2021.

#### RESULTS

Based on the search results for articles/journals published on Google Scholar, PUV Med, Science Direct, Research Gate and Scopus which have been published within the last decade, namely in 2011-2021, 45 journals were found related to congenital hypothyroidism screening research. However, after the filtering was carried out, ten articles have obtained that match the research topic, consisting of 7 national and three international journals. The obtained journal search results are tabulated in Table 1 as follows :

#### DISCUSSION

The discussion regarding the evaluation of the implementation of the CHSP in Indonesia will be reviewed based on three variables, namely input variables, process variables, and output variables.

#### Table 1. Research Result Matrix

No	Researcher Name	Title	Results
1	Adelia Anggraini, Chriswardani Suryawati, Eka Yunila Fatmasari	Evaluation of the Implementation of the Congenital Hypothyroid Screening Program by the Karangrejo Health Center, Metro Lampung City	In the implementation of the congenital hypothyroid screening program, there are many obstacles starting from the input stage, namely many health workers who have not been trained in congenital hypothyroidism screening, sources of financing that adjust to regional capabilities, the facilities provided are incomplete, and the absence of procedur operational standart in the implementation of congenital hypothyroid screening at the Puskesmas. At this stage of the process there are obstacles to the in effectiveness of socialization to pregnant women due to the absence of supporting facilities, health workers who do not understand the procedure for taking blood samples, and monitoring evaluations that do not work. At the output stage, there are obstacles to the low coverage of the congenital hypothyroid screening program.
2	Resty Noflidaputri, Vittria Meilinda	Evaluation Analysis of the Implementation of Congenital Hypothyroid Screening in Newborns	The results of the evaluation analysis of the implementation of congenital hypothyroid screening in newborns showed that there were obstacles from input, namely there were still untrained health workers, lack of confidence in health workers to take SHK blood samples, incomplete facilities and infrastructure. From the process, there are obstacles that are not optimal in planning, organizing and supervising. Meanwhile, the output of the SHK examination coverage has not reached the 100% target due to the lack of confidence of health workers to take SHK blood samples, the refusal of families who refuse to do SHK and also the slow delivery of reports from the posyandu supervisor to the field supervisor.
3	Rini Anggraini, Madarina Julia, Suryono Yudha Patria	Timeliness of Congenital Hypothyroidism Screening Services in Yogyakarta	The implementation of the congenital hypothyroid screening program in Yogyakarta D.I showed that there were still health workers who had difficulty taking specimens. The distribution of program funds and facilities has been going well with the result that the percentage of accuracy in taking specimens from the independent fund group is around 11.2% - 21.2% compared to the government fund group. The percentage of timeliness in sending specimens to the laboratory from the government fund group is around 30.5% - 37.0% higher than the independent fund group.
4	Carmencita David Padilla dan Bradford L. Therrell Jr	Consolidating newborn screening efforts in the Asia Pacific region	This journal writes about the results of the consolidation of various countries under the Asia Pacific region, including Indonesia. The consolidated results that have been implemented highlight the ongoing and unresolved challenges of financing newborn screening programs including congenital hypothyroid screening in Asia Pacific countries. Various financing models applied in Asia Pacific countries ranging from grants, government loan programs to the regions and national insurance. Of the various financing models, one model that can increase the success of the long-term program as well as for program stabilization is to include screening activities in national health planning priorities. In addition, the addition of an examination laboratory is also one of the efforts that can be implemented to develop a newborn screening program.
5	Adi Wirawan, Sunartini, Bikin Suryawan, Soetjiningsih	Child Development Congenital Hypothyroidism Treated Early with Levo- thyroxine and High Initial Dose	This study demonstrated the effectiveness of congenital hypothyroid screening for early case finding in children with congenital hypothyroidism. There is a significant difference regarding the growth and development of congenital hypothyroid children who received early hormone therapy compared to those who received hormone therapy late
6	Aman Bhakti Pulungan, Myrte Everarda Oldenkamp, Adrianus Sarinus Paulus van Trotsenburg, Wiwik Windarti, Hartono Gunardi	Effect of delayed diagnosis and treatment of congenital hypothyroidism on intelligence and quality of life: an observational study	This study revealed that the delay in hormone therapy in patients with congenital hypothyroidism caused by the absence of a national congenital hypothyroid screening program had a negative impact on the intellectual abilities of children suffering from congenital hypothyroidism, thereby reducing the quality of life of children.
7	Nahid Hatam, Mehrdad Askarian, Peivand Bastani, Kimia Pourmohammadi, dan Samad Shirvani	Cost-Utility of Screening Program for Neonatal Hypothyroidism in Iran	The results showed that screening for congenital hypothyroidism is not only economically beneficial but also prevents mental retardation and complications in children with congenital hypothyroidism. This is evidenced by the additional cost-effectiveness ratio (ICER) of children with hypothyroidism, calculated by comparing the cost ( $\triangle$ C) with the difference in quality of life ( $\triangle$ QALY) showing that Iran by conducting congenital hypothyroid screening saves costs of 13413 US dollars.

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9Yusi Dwi Nurcahyani, Donny K Mulyantoro, Prihatin BrotoSensitivity and Specificity of Hypothyroid Screening Instruments for Diagnosis of Sukandar, M. Samsudin, Nur IhsanThis study revealed that blood tes hypothyroidism are more sensitiv diagnosis compared to using a hy instrument.9Yusi Dwi Nurcahyani, Prihatin Broto Sukandar, M. Samsudin, Nur IhsanSensitivity and Specificity of Hypothyroid Screening Instruments for Diagnosis of in Toddlers instrument.This study revealed that blood tes hypothyroidism are more sensitiv diagnosis compared to using a hy instrument.	ts to screen for congenital e and specific in supporting the pothyroid screening assessment
10Anggraini, CharuniaThe Role of the Health Center in the Implementation of Congenital HypothyroidThis study reveals the importance supporting the implementation o in the regions by establishing regi Screening to Ensure Child Health at the Health Center 	of the role of local governments in F congenital hypothyroid screening onal policies that regulate the othyroid screening in integrative y health centers.



Picture 1. Journal Selection Flowchart

#### **Input Evaluation**

According to research conducted by Anggraini et al (2019) and Anggraini et al (2016) the skills of health workers who play a role in congenital hypothyroid screening blood sampling are considered lacking because there are still health workers who have not attended training for congenital hypothyroid screening blood sampling. The results of this study are supported by Noflidaputri et al (2021) who revealed that the cause of the lack of confidence in health workers in taking hypothyroid screening samples was caused by health workers who had not received training on procedures for taking blood samples for congenital hypothyroid screening. Judging from the results of the input evaluation on the human resource component, it is known that there are still health workers who have not attended training causing a lack of sampling skills so that health workers are less confident in carrying out their duties as implementers of the CHSP.

In terms of facilities and infrastructure, according to research by Anggraini et al (2019), the quantity of blood sample collection facilities in the form of filter paper and lancet is still limited, so that if targets are found but facilities are not available, the CHSP cannot be implemented. Likewise, the completeness of the facilities is considered to be lacking because the supporting facilities for activities such as drying racks, informed consent, ziplock plastic, posters, leaflets and brochures are not yet available. Similar results were shown in the study of Noflidaputri et al (2021) regarding the completeness of the CHSP. In contrast to the results of research by Anggraini et al (2019) which stated that the distribution of supporting facilities for the CHSP has gone well. The need for a CHSP should be evaluated regularly so as to minimize the loss of blood sampling if a target is found. In addition, the completeness of the facilities must also be considered so that the CHSP can run optimally.

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The method used in the CHSP according to Permenkes number 78 of 2014 regarding congenital hypothyroid

screening is to take a blood sample from the heel to be dripped directly on filter paper then dried and sent to the laboratory for examination. According to Nurcahyani et al (2017) the method of examining congenital hypothyroidism using blood samples is more sensitive and specific in establishing a diagnosis than using a hypothyroid screening instrument. Reducing the threshold for examination results has a very significant impact on finding cases of congenital hypothyroidism in children (Yarahmadi et al., 2020). This is supported by Pulungan et al (2019), children who show symptoms are a sign of delay in establishing a diagnosis of congenital hypothyroidism so that it affects the growth and development of children. The most effective method used in establishing the diagnosis of congenital hypothyroidism is to perform a blood test through a laboratory.

In terms of time, the results of research by Anggraini et al (2016) show that the timeliness in sending samples of CHSP funded by the government is better than independent funds from the community. Meanwhile, the timeliness of sampling for congenital hypothyroid screening from an independent fund is better than the sample of CHSP funded by the government. Research by Anggraini et al (2019) revealed the problem of sending sample results which takes 7-14 days and for negative results, the laboratory will provide results in a long time, namely one year. The research of Noflidaputri et al (2021) did not reveal the results of the timeliness of examination, delivery and delivery of results. The results of the research by Wirawan et al (2016) found that children with congenital hypothyroidism who received hormonal therapy early had more optimal growth and development than children who received hormonal therapy late. Pulungan et al (2019) showed a negative effect due to the delay in giving hormone therapy in children with congenital hypothyroidism on the level of intelligence and also the occurrence of complications due to congenital hypothyroid disease. Another study revealed significant differences in the growth and development of children with congenital hypothyroidism who received hormone therapy on time and those who received hormone therapy late (Wirawan et al., 2016). Early management of hormonal therapy in cases of congenital hypothyroidism greatly affects the severity that results in mental retardation so that it requires timeliness in terms of examination, delivery and delivery of screening results.

#### **Process Evaluation**

The role of planning, organizing, implementing and monitoring in the implementation of the CHSP is important in providing good guidance to the implementers of activities so that they can run optimally. In the research, Anggarini et al (2019) revealed that planning needs needed collaboration between the Puskesmas as field implementers and the Health Office as the implementing coordinator for the CHSP. In the stage of organizing blood sampling at the Puskesmas, it is absolutely necessary because it reduces the loss of opportunity in taking blood samples if a target is found, but the Department of Health as the coordinator of the CHSP has never discussed the implementation of the program. At the pre-screening implementation stage, providing information to the public about the importance of the CHSP for children's growth and development greatly affects public acceptance during the screening and post-screening process, namely case tracking as a follow-up to the examination results if there are positive results. Monitoring of the implementation of the CHSP must be carried out by the Health Service because the program coordinator, however, the role of monitoring has not been carried out optimally. While the research of Anggraini et al (2017) and Noflidaputri et al (2021) did not show in detail the results of the evaluation of the CHSP process. Yarahmadi et al's research (2020) which shows the success of the program comes from the collaboration between the community and the government that supports each other in the implementation of the CHSP. The same thing was expressed by Anggraini (2017) that the government plays a role in formulating policy formulations regarding the implementation of congenital hypothyroid screening in their regions.

#### **Output Evaluation**

From research conducted by Anggraini et al (2017, 2019) and Noflidaputri et al (2021), it was revealed that the achievements of the CHSP still do not cover 100% of all newborns in Indonesia. This is echoed by the results featured in the Indonesian Ministry of Health's Basic Health Research carried out in 2018, which revealed that 4.6% of all newborns tested for congenital hypothyroid screening through blood samples in Indonesia. When compared with other countries such as Iran which has successfully screened 98% of newborns (Yarahmadi et al., 2020), and Sri Lanka which has successfully screened 96% of newborns (Karunarathna and Hettiarachchi, 2021), Indonesia's achievements are still far from being successful in this regard. As such, a comprehensive evaluation of the problem factors that affect the success rate of the program is needed in order to identify and mitigate the issues.

Apart from the scope of the program, the output evaluation can also be calculated economically by calculating value of the economic effectiveness of the the implementation of the CHSP. Congenital hypothyroidism was not screened for before the introduction of the program. A study on the economic benefits of the CHSP was conducted by Hatam et al (2016) in Iran. The study found the incremental cost-effectiveness ratio (ICER) of congenital hypothyroid children by comparing the cost ( $\triangle C$ ) and the difference in the quality of life ( $\triangle$ QALY), which demonstrated savings of 13413 US dollars. In Indonesia, research conducted by Rochmah et al (2020) revealed that congenital hypothyroid children who received early hormone therapy had a significant difference in the quality of life (QALY) when compared to those who received treatment later. . As for the research which discusses the economic evaluation of the implementation of the CHSP in Indonesia, there is still no such research in existence. The results of the economic evaluation of congenital hypothyroid screening are needed because the results can be used as a basis for making policies which include congenital hypothyroid screening as a priority within the national health system.

#### LIMITATION OF THE STUDY

The advantage of this literature review is that it analyzes more data in comparison to the results of the evaluation of CHSP in various regions of Indonesia into one journal article. One limitation of this review lies in the lack of references to aid in evaluating the financing both in terms of input and economic evaluation in terms of output. There exists a limited number of journals which discuss and debate the CHSP within Indonesia.

#### CONCLUSIONS AND SUGGESTIONS

Based on the results of the literature review, it can be concluded that various factors cause the low coverage of CHSP in Indonesia. Discussions regarding the implementation of the program in Indonesia are still very limited, especially in terms of economic benefits and the level of effectiveness of the programs on preventing mental retardation in congenital hypothyroid children. Resultantly, further research is needed so that effective conclusions and data can be drawn. The results of this evaluation contribute towards and act as a basis for a follow-up plan and improvement of the implementation of CHSP which has been implemented.

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#### ETHICAL CONSERATIONS

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#### **Conflict of Interest Statement**

There are no possible conflicts of interest with respect to the authoring and publishing of this work, according to the authors.

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