Doctoral Dissertation

Prevalence of Developmental Disorders and Screening Effects of the National Health Screening Program for Infants and Children in Korea

한국 영유아의 발달장애 유병률 및
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Prevalence of Developmental Disorders and Screening Effects of the National Health Screening Program for Infants and Children in Korea

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Abstract

As the prevalence of developmental disorders (DDs) has increased, many OECD countries now provide a national screening examination for early detection and intervention. Since 2008, the Korean government has also implemented the National Health Screening Program for Infants and Children (NHSP) to reach the target population, for early identification of children at risk of DDs. However, neither an epidemiological study for the whole set of DDs nor an evaluation of the effects of the NHSP has been completed in Korea. Therefore, studies about the prevalence of DDs and evaluation of the effects of the NHSP are needed, to provide evidence for the development and evaluation of DD-related programs and policies.

This descriptive study, using nationwide population–based data, addressed the goals of illustrating trends in prevalence and incidence of DDs among children ages six years and younger, and analyzing the screening effects of the NHSP in Korea.

The subjects of the study were divided into two groups. Children younger than seven years from 2003 to 2017 were observed for epidemiological analysis; among those, children with DDs between 2008 and 2017 were analyzed for evaluation of the screening effects of the NHSP. The study

calculated and compared prevalence and incidence, as well as the screening effects, by year, along with subconditions of DDs, demo-geographic factors, and economic status, using $\chi 2$. Simple linear regression yielded comparison of the change in linear trends of prevalence and incidence of DDs and changes in the Success-in-Screening rates (the Success-in-SCR rates).

The prevalence of DDs steadily increased by more than four times (from 0.6 to 2.5) from 2003 to 2017. Boys had higher incidence than girls throughout the period, and during this period the gap widened from 19.1% to 31.4%. The ratio of autism spectrum disorder, developmental delay, and language disorders among the total incident cases of DDs increased by 13.7%, 817.6%, and 30.7%, respectively, indicating their contribution to the trend of increasing prevalence. For the screening effects of the NHSP, 65,334 children (39%) were DD-screened, of whom 35,466 children (21%) received a false negative among the 167,050 children with DDs between 2008 and 2017. The DD-screened rate increased from 3,208 (27.2%) in 2008 to 8,471 (47.3%) in 2012, and then decreased to 5,544 (29.8%) in 2017. Change in the false negative rates—an increase from 2.7% in 2008 to 23.8% in 2017—was one of the most influential factors for these fluctuations. Both the incidence rate and the DD-screened rate were influenced by demo-geographic and economic factors, along with age-related characteristics of the subconditions. Children

living in a bigger city and with higher economic status were more likely to be identified for DDs, which indicates the higher vulnerability of children in the other group.

In conclusion, prevalence and incidence rates have steadily increased over the past fifteen years, but the NHSP has not shown many of the effects of screening for DDs. In order to suppress rapid increase in prevalence, further efforts should be applied to developing a more effective screening system for DDs and establishing related policies to support those who are more vulnerable to DDs.

Keywords: Child, Developmental disabilities, Diagnostic screening programs, Epidemiologic studies, Infant, Nationwide population–based data

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This doctoral dissertation includes the contents of the two published articles listed below.

Rah, S. S., Hong, S. B., & Yoon, J. Y. (2020). Prevalence and incidence of developmental disorders in Korea: A nationwide population-based study. *Journal of Autism and Developmental Disorders*, *50*(12), 4504–4511.

Rah, S. S., Hong, S. B., & Yoon, J. Y. (2020). Screening effects of the National Health Screening Program on developmental disorders. *Journal of Autism and Developmental Disorders*, 1–14.

This dissertation offers an expansion of the literature review, and the research framework was newly added, following the doctoral dissertation review format of Seoul National University's College of Nursing.

In addition, the study uses data collected by the National Health Insurance Service and provided through the National Health Insurance Sharing Service. It is clearly stated that neither of the organizations has been involved in the process of the study, nor did they intervene with the study's results.

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I. Introduction

1. Importance of the study

Developmental disorders (DDs) are defined as limitations on language, social, motor and cognitive abilities that occur during the developmental period (Odom et al. 2009). The conditions can vary by the domains in which the developmental problem occurs; Autism Spectrum Disorders (ASD), Attention Deficit Hyperactivity Disorder (ADHD), blindness, cerebral palsy, developmental delay, epilepsy, hearing loss, intellectual disorder, language disorders, learning disorders, and special sensory disorders are the subconditions of DDs (Boyle et al. 1994; C. A. Boyle et al. 2011; Zablotsky et al. 2017; Olusanya et al. 2018). Because people with DDs may not only have a single condition but rather show a set of the symptoms, DDs need to be considered as a whole not individually (Thapar et al. 2017). Core profiles of ADHD, for example, comprise features of language, behavioral, and emotional problems as well as cognitive impairments in combination (Thapar et al. 2017).

Because of these characteristics, the increasing prevalence of DDs has become a critical issue in communities and countries worldwide. According

to a previous study, 4.8% of school-aged children in state-funded schools or nonprofit special schools in the UK (97.2% of the total English children of that age) were identified as having developmental disorders (DDs) (Emerson 2012). In another study of Swedish children ages 0 to 17, prevalence of ASD increased by about 3.5 times, from 0.42% in 2001 to 1.44% in 2011 (Idring et al., 2015). The case of the US is more severe: prevalence of DDs in the US children from 3 to 17 years old increased by 1.23%, from 5.76% in 2014 to 6.99% in 2016 (Zablotsky et al., 2017). This phenomenon is not limited to European or North American countries but is also found in Asian countries. Prevalence of developmental delay—one of the subconditions of DDs—in Taiwanese children under six years old increased from 0.16% to 3.25%, making up 20% of the increasing ratio of prevalence over a twelve-year period, which is even higher than that of the US (Kuo et al., 2015).

In recent decades, a number of researchers have attempted to explain the change. Nonetiologic factors, such as changes in diagnostic criteria or reporting practices, use of different research methodology, or increase in diagnostic awareness, are suggested as contributors to the increasing prevalence, along with genetic factors (Matson and Kozlowski, 2011; Hansen et al., 2015; Fombonne, 2018). According to Hansen et al. (2015), a total of 60% of the rise among all reported cases of ASD in Denmark in 1995,

compared to the prevalence of ASD in 1994, was caused by the combination of the changes in diagnostic criteria from the eighth edition of International Classification of Diseases (ICD-8) to the ICD-10, and the expansion of the reported data from the inpatients to the outpatients. Environmental factors also exert influence on the increasing prevalence of DDs. Premature or low-birth-weight babies, for example, have a higher tendency toward developmental problems. Maternal age is advancing, which is a major influential factor in preterm birth and low birth weight, and this may also explain the change in the prevalence of DDs (Aras, 2013; Synnes et al., 2017; Neggers, 2014). Although researchers have suggested various factors as the causes of this phenomenon, as mentioned above, many agree that the prevalence of DDs is increasing (Boyle et al., 2011; Emerson, 2012; Zablotsky et al., 2017).

The increasing trend of DDs has aroused the attention of society, and that has led to an increasing number of epidemiological studies for DDs (Boyle et al., 2011; Boyle et al., 1994; Zablotsky et al., 2017; Emerson, 2012). Few of the epidemiological studies related to DDs in Asian countries, however, can accurately present the prevalence and characteristics of DDs in those countries. Most previous studies of this kind have small sample sizes that cannot represent the target population or analyze DDs by subcondition (Kuo

et al., 2015; Kim et al., 2017; Sachdeva et al., 2010; Kim et al., 2011). As in other Asian countries, in Korea only a limited number of studies have examined the prevalence of the subconditions of DDs. Moreover, the prevalence of a whole group of DDs has never even been analyzed, which increases the necessity of conducting an epidemiological study. Without evidence from epidemiological studies, it is difficult to grasp the trends of prevalence or the characteristics of DDs, and it is not possible to evaluate the effectiveness of DD-related national polices or programs.

Screening is another critical issue of DDs worldwide. With early detection and intervention suggested as one of the best solutions to help children vulnerable to DDs improve developmental behaviors and skills (Barger et al., 2018; Guralnick, 1998), many countries have implemented a health screening program for the younger populations. One report surveyed 21 OECD countries (and Taiwan) that provide a health examination program to younger populations (Shin et al., 2017). According to this report, most countries include developmental evaluation as part of the health examination program for as few as two times and as many as fifteen; thirteen countries run the program as a national project (Shin et al., 2017). The Korean government also launched a noninvasive systematic screening program, the National Health Screening Program for Infants and Children (NHSP) (Moon, 2010). It was

first provided to medical insurance recipients in 2007 and then expanded to the whole population of the target age who are registered at the National Health Insurance Service (NHIS) from 2008 (Moon et al., 2010). In 2018, the NHIS published a report that evaluated the effects of the NHSP throughout the previous decade (Baek et al., 2018). In this report, incident cases were calculated by using nationwide population-based data, the results showing that incident cases during the period after the implementation of the NHSP significantly decreased compared to cases before the implementation (Appendix 1) (Baek et al., 2018). The study includes several limitations, however. First, the list of the diagnosis codes did not cover all the subconditions of DDs. Diagnosis coded for special sensory disorders (F82 and F83) and intellectual disorders (F81), for example, were excluded. In addition, only the parts of the diagnosis coded for ASD and cerebral palsy appear in the list, which excludes F88 and F89 for ASD and P91.2 and P91.6 for cerebral palsy. Second, although the results cannot be verified—there has been no epidemiological study for DDs in Korea for comparison—they contrasted with the global trends of prevalence for DDs. For these reasons, the screening effects of the NHSP on DDs remain unknown.

Recent studies have also raised the problem of lower accuracy in detecting DDs in the general population. According to a previous study using data from

the Norwegian Mother and Child Cohort and the Autism Birth Cohort study, only 28.8% of children with ASD were screened by a criterion of six critical discriminative items from the Modified Checklist for Autism in Toddlers (M-CHAT), a parent-performed developmental questionnaire (Stenberg et al., 2020). Similar results were obtained in another study conducted in the US using the Modified Checklist for Autism in Toddlers with Follow-Up (M-CHAT/F) (Guthrie et al., 2019). Among the total of 454 children who were diagnosed with ASD, 176 children were screened by the M-CHAT/F, indicating 38.8% of the sensitivity for ASD.

Thus, without mentioning that only two Asian countries—Japan and Taiwan—were included among the twenty-two countries mentioned above, and that only half of the countries provided the program at a national level (Shin et al., 2017), evaluation of this national-level health screening program operating in Korea alone is meaningful.

2. Purpose of the study

The purpose of this study is to illustrate trends of the prevalence of DDs among children from 0 to 6 years old in Korea and analyze screening effects of the NHSP in children diagnosed with DDs using nationwide population-based data. The specific research questions are:

1. What are the trends of the prevalence of DDs among Korean children from 0 to 6 years old?
2. What are the characteristics of children from 0 to 6 years old who have been diagnosed with DDs?
3. What are the screening effects of the NHSP on DDs among Korean children from 0 to 6 years old?

3. Definition of terms

1) Developmental disorders

In line with previous DD-related studies (Boyle et al., 2011; Woo, 2006; Jung and Go, 2003), Autism Spectrum Disorder (ASD), Attention Deficit Hyperactivity Disorder (ADHD), cerebral palsy, developmental delay, intellectual disorder, language disorders, learning disorders, and special sensory disorders are included as subconditions of DDs. The specific diagnosis codes for DDs are F70~79 (intellectual disorder), F80 (language disorders), F81 (learning disorder), F82~83 (special sensory disorders), F84 and F88~89 (ASD), F90 (ADHD), G80, P91.2 and P91.6 (cerebral palsy), and R62.0 (developmental delay) in the Korean Standard Classification of Diseases (KCD) 7th edition (Korean Classification of Diseases 7th edition, 2016), established based on the International Classification of Diseases (ICD) 10th edition, and confirmed by experts in pediatric neurology, pediatric rehabilitation, and pediatric psychiatry. To avoid unconfirmed cases of DDs, we limit the DD-diagnosed population to those people who had two or more outpatient visits, or one or more hospitalizations, between 2003 and 2017, applying the DD diagnosis codes as above.

2) Incidence

The Centers for Disease Control and Prevention (CDC) defines incidence as new cases of disease during a specific time period (Dicker et al., 2006).

In this study, incidence refers to the new cases of DDs, calculated annually between 2003 and 2017.

3) Prevalence

The CDC defines prevalence as the sum of preexisting and new cases of disease during a specific time period (Dicker et al., 2006). Depending on whether it is calculated at a specific time cross sectionally or calculated for a specific period of time, prevalence is classified as point prevalence or period prevalence, respectively.

In this study, period prevalence was calculated annually between 2003 and 2017, which indicates the proportion of people who have been diagnosed and who were newly diagnosed with DDs.

4) The National Health Screening Program for Infants and Children

The NHSP is a non-invasive systematic screening program that comprises

body measurement and developmental evaluation (Moon, 2010).

This study analyzed the effect of the NHSP as a screening tool for DDs, however, the NHSP refers to only the developmental evaluation part throughout the study.

5) Screening effects

The primary purposes of screening are early detection and intervention. In the case of DDs, the screening effect implies prevention of the diseases for children at risk as one result, in addition to minimization of the developmental problems and ultimately for improvement of the later outcome and the quality of life (Kim et al. 2016; Barger et al. 2018; Guralnick, 1998). Developmental tasks that children need to achieve continuously change as they grow; but their parents, usually the first to identify the signs in their children, may easily miss the diagnostics at the early stage, which results in a delayed diagnosis of children with DDs. The first symptoms of pervasive developmental disorders (PDDs), for example, usually appear during infancy in about 60% of cases, but most children with PDDs are diagnosed around age 4 or even as late as 10 years old (Manea et al., 2015). In one study, the median perceived delay in diagnosis of parents who have children with PDDs was observed to be nine months (Harrington et al., 2006). Recognizing the diagnostics of children as

early as possible and referring them to a physician for further examination could be the ultimate goals for a screening program.

Therefore, this study measured the screening effect of the NHSP by the number of children who were diagnosed with DDs after they had received a positive result from the NHSP. Positive results were defined as all the results in the developmental assessment, except Good, from 2008 through 2014, and except Good, Peer level, and Fast level from 2015 through 2017.

II. Literature Review

1. Social impacts caused by developmental disorders

Developmental disorders (DDs) are chronic conditions that lead to physical, psychological, and economic burdens for families, as well as increasing the medical expenses and decreasing the labor productivity of a nation (Boyle et al., 1994; Lee & Kim, 2014). Although the mechanism of DDs has not yet been clearly discovered, they are known to occur from various causes, including high causality from genetic factors (Vorstman & Ophoff, 2013; Fitzgerald et al., 2015). DDs cannot be cured, and in many cases they persist into adulthood, resulting in a broad range of negative outcomes (Sayal et al., 2018; Reichow et al., 2018).

The association of DDs with a higher prevalence of other mental health problems has been investigated by many previous studies. One population-based study in Australia that analyzed the co-occurrence of intellectual disorder (ID) and other psychiatric illnesses found that 4,221 people had a psychiatric disease among a total of 13,295 people with ID; lifetime prevalence of psychosis among those with ID was the highest, at 8.4%, following by schizophrenia, at 3.6% (Morgan et al., 2008). Another study

compared the prevalence of psychiatric illness of two groups of school-aged children with and without ID; the results showed 7.3 times higher vulnerability among those with ID to other psychiatric illness (Emerson, 2003). Anxiety is another commonly co-occurring mental problem for people with DDs. Fuhrmann et al. (2012) studied 653 preschool children in Germany and found that depressive symptoms were associated with developmental problems, especially in motor and linguistic areas. Prevalence of developmental motor problems in the children with depressive symptoms was the highest (67.6%) among other risk factors, such as parental separation or language barrier caused by migration. These mental problems not only manifested during the childhood period but also persisted through adulthood. Shooshtari et al. (2011) compared the prevalence of depression and dementia between the DD cohort group and the matched comparison group among the population of Manitoba, Canada, from 2000 to 2004. The risk ratios of depression and dementia were both higher in the DD cohort group across all ages; the risk ratio of depression in younger adults with DDs was 2.96, and in older group with DDs it was 2.65. The risk ratios of dementia were even higher: the prevalence of dementia in the DD cohort group surpassed the comparison group by more than four times. These results clearly show that people with DDs have a high likelihood of other additional psychiatric illness, regardless of age or subcondition.

These lifelong chronic characteristics of DDs aggravate the physical, mental and financial burdens on individuals and families. According to one study, 47% of parents who had children with DDs or seizure disorder reported that their children suffered discrimination; more than half of the parents experienced a decrease in economic activity; and 15% of the parents even divorced because of issues related to their children (Spindler et al., 2017). Another study observed similar results: mothers of children with DDs showed lower family functioning, higher caregiver burden, and lower coping abilities than mothers of children without DDs (Manor-Binyamini, 2010). When accompanied by other mental problems, the family burdens increase still more; problematic behaviors, presence of psychotic disorder, and disability in social participation and self-care were identified as the main influential factors of the family burden (Irazábal et al., 2012). The family burden is also affected by the type of DD. Mugno et al. (2007) investigated quality of life among 212 parents of children with DDs and compared the score by the subconditions (ASD, cerebral palsy, and ID) to a control group of 77 parents of children without DDs. Parents of children with ASD showed a lower score in physical, mental, and social relationship areas than other subconditions and a control group; small differences were observed between the groups of other subconditions and a control group (Mugno et al., 2007).

Economic burden is another critical issue for the family and society. A previous study compared the reported income of families of children with ASD, other DDs, and without DDs with the expected income considering age, educational level, living areas, and types of family. The families of children with ASD lost 14% of their total income, which was more than 77 times greater than that of families of children without DDs, and more than five times greater than families of children with other DDs (Montes & Halterman, 2008). More recent studies match these results. Reviewing 33 articles, Liao and Li (2020) concluded that parents of children with ASD experienced adverse changes in employment and increased economic burdens, especially for mothers. In another review of the economic burden among adults with ADHD, annual medical costs were higher in the group of people with ADHD than among those without ADHD—and the gap was as high as $4,178 (US dollar value in 2004) (Matza, Paramore, & Prasad, 2005). The economic burden of ASD, calculated as a combination of annual costs of medical and nonmedical expenses and productivity, is predicted to rise from the range of 0.889–2.009% in 2015 to the range of 0.982–3.600% of the GDP in 2025 in the US (Leigh & Du, 2015).

As can be seen from these studies, children with DDs and their families have very real concerns about mental, physical, and social well-being. More

effort should be made to improve awareness of the burden of children with DDs and their families, to reduce stigmatization and establish a systemic social system that enhances the well-being of the affected people and communities.

2. The National Health Screening Program for Infants and Children

Many OECD countries have implemented a national developmental screening program targeting infants and school-aged children for the purpose of early detection of and intervention in DDs. Korea uses the National Health Screening Program for Infants and Children (NHSP). The NHSP is a noninvasive screening test comprising body measurement and developmental evaluations. The Korean government initiated it in 2007 for health insurance recipients and then expanded it to medical care recipients in 2008 for the early detection of and intervention in medical problems that occur during the early stage of the developmental process (Moon et al., 2010). For the past decade, the NHSP has undergone several changes for quality improvement: checkup frequency increased from five to seven in 2012; educational sessions and dental checkups were added in 2010 (Fig. 1) (Eun et al., 2010; Shin et al., 2017). In addition to the changes, the developmental screening tool has changed from the Korean Ages and Stages Questionnaires (K-ASQ), a Korean version of ASQ, to the Korean Developmental Screening Test for infants and children (K-DST), developed by the Korea Centers for Disease Control and Prevention (KCDC) in 2014, which reflects the differences of

Year	1st 4~6m	2nd 9~12m	3rd 18~24m	4th 30~36m	5th 42~48m	6th 54~60m	7th 66~71m	Remarks
2008	O	O	O	X	O	O	X	
2009	O	O	O	X	O	O	X	
2010	O	O	O	O	O	O	X	4th checkup, dental checkup, and educational sessions added
2011	O	O	O	O	O	O	X	
2012	O	O	O	O	O	O	O	7th checkup added
2013	O	O	O	O	O	O	O	
2014	O	O	O	O	O	O	O	K-DST developed and implemented
2015	O	O	O	O	O	O	O	
2016	O	O	O	O	O	O	O	
2017	O	O	O	O	O	O	O	

Figure 1. Changes in composition of the NHSP, 2008–2017

Note: Developmental evaluation was not performed at the first checkup

Korean cultures (Eun et al., 2014). With these extensive efforts by the government, the NHSP was equipped with systematic components and an efficient system that closely connects related institutions.

One of the strengths of the NHSP is its provision of a cumulative developmental evaluation. Except for the first screening, for infants ages 4 to 6 months, all screenings include a developmental evaluation done by parents using the K-DST, a parent-performed screening instrument. The K-DST contains 335 questions in six domains (gross motor, fine motor, cognition, language, socializing, self-help), with another category for additional questions (Eun, 2017). These questions are grouped by the target ages, so on average, 55 questions are given at each screening. Although different questions are given in each developmental evaluation, the NHSP can be considered cumulative in terms of applying the same instrument repeatedly. When the results of the screening are automatically transferred to the NHIS, they contact the children who fall into the request-for-close-examination category individually and provide financial support for closer examination, in order to facilitate early diagnosis and intervention (Eun et al., 2007). As a result, checkup rates for the NHSP have risen from 35.5% in 2008 to 76.4% in 2017 (Baek et al., 2018), along with an increase in the number of hospitals and clinics providing the NHSP—from 2,790 in 2008 to 4,165 in 2019 (Moon,

2010; “Finding clinics or hospitals,” 2017).

The next step where more effort is needed is filling the gaps between screening and referral. A previous study found that only 61% of children who screened positive for DDs were referred for early intervention in the US; pediatricians mentioned lack of time and inadequate reimbursement as barriers (King et al., 2010). In many communities, the number of facilities and professionals for early intervention are inadequate to provide the services to children at risk for DDs (Elansary & Silversterin, 2020). Shin et al. (2017) also pointed out these barriers in a government report, suggesting a web-based process to save time and increase the reimbursement. More importantly, however, few studies have evaluated the NHSP, so the screening effects of the NHSP on DDs remain unknown. Therefore, it is crucial to conduct research that evaluates the outcomes of the NHSP.

III. Methods

1. Study design

This study was conducted to describe trends in the prevalence of DDs and analyze the effect of the NHSP, as a developmental screening program, on incidence of DDs among Korean children ages 0 to 6 years. The design of the study is a descriptive study using nationwide population-based data.

2. Study data

Since 1989, all citizens in Korea have been registered with the National Health Insurance Service (NHIS) as recipients of either health insurance or medical care. To manage the insurance system, the NHIS collects necessary information from all registries, including demographic and geographic, death, inpatient and outpatient, and prescription registries. The NHIS also provides data through the National Health Insurance Sharing Service (NHISS) after transforming them into unidentifiable data through unique identification numbers.

This study used customized data provided by the NHISS.

3. Study subjects

The target population for the prevalence of DDs is children ages 0 to 6 years in Korea from 2003 to 2017. To avoid over-counting prevalent cases, however, 2002 is excluded as a "run-in" period. Among the total study subjects, children ages 0 to 6 years from 2008 to 2017 who were diagnosed with DDs were analyzed for screening effects of the NHSP (Fig. 2).

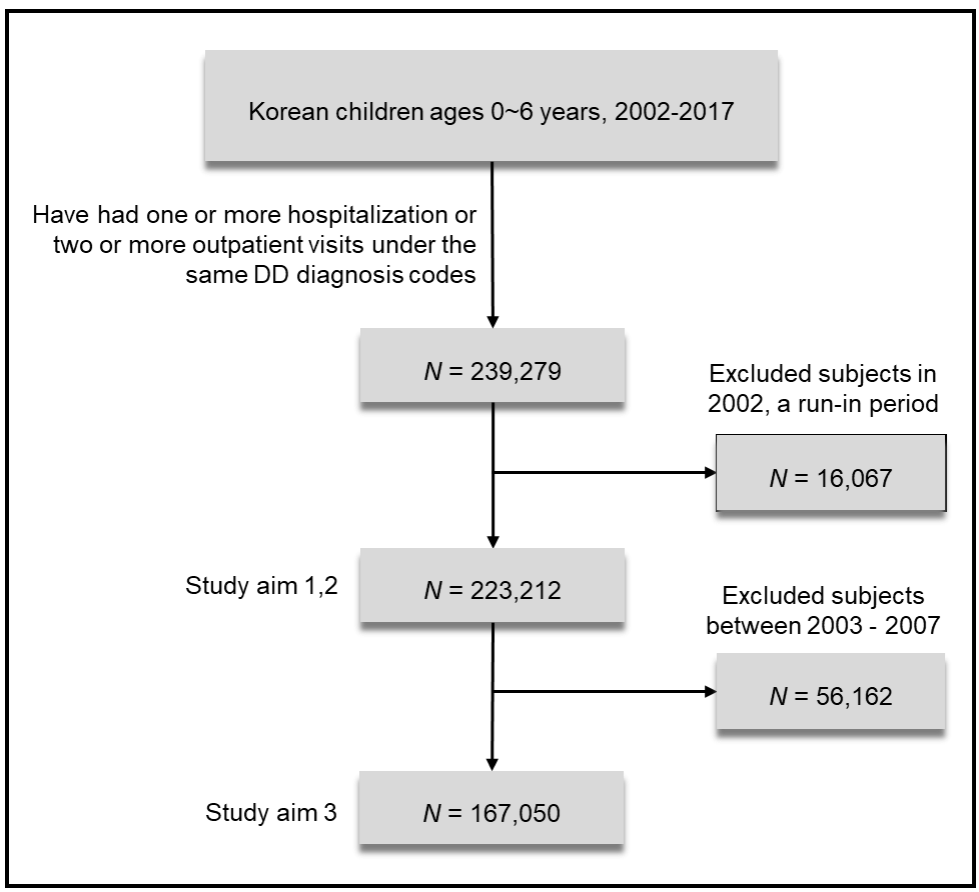


Figure 2. Flow chart of the study subjects, 2003-2017

4. Study variables

1) Prevalence and incidence of developmental disorders

To estimate the crude incidence rates (per 10,000) and prevalence (per 100) of DDs, the annual number of the target population, obtained from Statistics Korea, was used as the denominator (Status of the Targets and the Examinees for the National Health Screening Program for Infant and Children by Gender, City, and Country, 2017). With the exclusion of the incident cases in the run-in period (the year 2002), all newly diagnosed subjects with any one of the DD diagnosis codes were defined as an incident case. Prevalence was calculated by dividing the number of prevalent cases (the cumulative incident cases - the cumulative death numbers - the cumulative numbers of six-year-old children in the previous years) by the total annual number of the target population. Incidence rates and prevalence were analyzed by employing simple linear regression, categorized in five-year intervals. This made three five-year time periods, reducing the annual random bias (Boyle et al., 2011; Westerinen et al., 2017). The annual incidence rate was also calculated by gender.

In analyzing the characteristics of the incidence of DDs, the study calculated the mean ages of the subjects in each subcondition, to compare the

usual point at which the diagnosis had been made. The ratio for incident cases of each subcondition was divided into three five-year groups and then compared. The three subconditions (ASD, developmental delay, language disorders) whose ratio of incident cases showed an increase when comparing the third group (2013–2017) with the first group (2003–2007) were further stratified by demo-geographic factors.

2) Checkup rates for the National Health Screening Program for Infants and Children

The checkup rates for the NHSP were calculated by dividing the number of children who had been seen by the NHSP by the total number of children who were subject to the NHSP which excludes the first checkup from the analysis, as it does not provide developmental evaluation.

3) Screening effects of the National Health Screening Program for Infants and Children

There are two ways to analyze screening effects: prospective and retrospective. The prospective approach studies the children who were checked by the NHSP and follows the results of the screening and the

diagnosis of DDs prospectively; the retrospective approach surveys the children with DDs for their previous experience of checking the NHSP and the results retrospectively. The first method has a major limitation: the undiagnosed children among the screened subjects cannot be clearly distinguished according to whether they were not diagnosed with DDs because of the absence of the diseases or because of the absence of the opportunity to be diagnosed. Without that clarification, children with false negative results cannot be calculated, nor can children with DDs among those with positive results accurately represent the screening effect.

The second method redeems these limitations by studying only the DD-diagnosed children. Because the NHSP subjects represent the total target population, the previous experience of the NHSP and the results of these children are clear. Therefore, this study used the retrospective method.

The DD-diagnosed children ages 0 to 6 years old from 2008 through 2017 were categorized into two groups by the results of the NHSP and then compared. The Success-in-Screening (Success-in-SCR) group included children who had received a positive result before the diagnosis. The Failure-in-Screening (Failure-in-SCR) group was divided into three groups: the never-checkers, the false negatives, and the late-checkers. The never-checkers included children who had never checked the NHSP before the diagnosis, and

the false negatives were those who had received a negative result from the NHSP before the diagnosis. Children who received the positive results from the NHSP after the diagnosis were named late-checkers (Fig. 3).

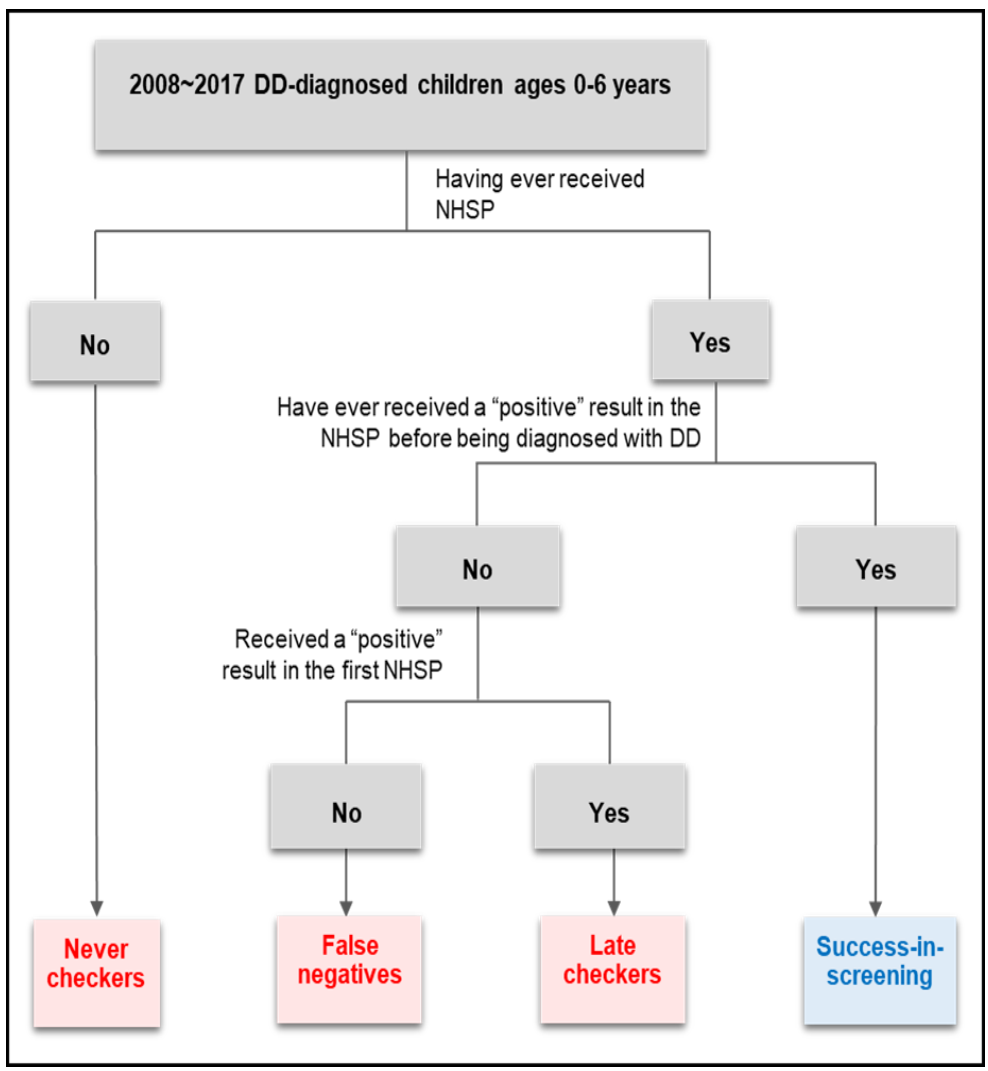


Figure 3. Research framework of screening effects analysis, 2008-2017

In this study, therefore, screening effects were defined as the case of the Success-in-SCR. The correlation between the performance of the NHSP and the incidence of DDs was analysed by comparing annual checkup rates with annual incidence rates (per 10,000). Additionally, the annual cases of the Success-in-SCR and Failure-in-SCR groups among the total study subjects were compared, to illustrate the changes in NHSP outcomes. The incident cases of DDs were analysed by age (year) and subconditions, to describe the characteristics of DDs from different perspectives. Mean age (year) at diagnosis was calculated by the case groups and the subconditions, to examine the time when the diseases were recognised. The screening effect of the NHSP was measured by analysing the incident cases in the four case groups (Success-in-SCR, never-checkers, false negatives, late-checkers) and stratifying by demo-geographic factors, as well as by subconditions. The percentages of the Success-in-SCR and Failure-in-SCR groups were also compared by these variables to determine the screening competency of the NHSP in each variable.

4) Demo-geographic variables

In this study, age, gender, city size (capital city, metropolitan city, province, and special self-governing city), and medical insurance quartile (0, 1, 2, 3, 4)

were used as demo-geographic variables. For the medical insurance quartile, subjects in the 0 medical insurance group were excluded from the analysis, as this group includes subjects whose parents have specific job categories (e.g., commissioned officer) and those with missing data.

5. Statistical analysis

(1) Descriptive statistics were used to analyze the characteristics of the children with DDs. In addition, $\chi 2$ was used to stratify the total incident cases of each subcondition and to examine DDs as a whole throughout the 15-year period by demo-geographic variables.

(2) Simple linear regression was used to analyze linear trends of the prevalence and the annual incidence rate, as well as changes in the Success-in-SCR rates.

(3) Probabilities of the incident cases in both the Success-in-SCR group and the Failure-in-SCR groups were analyzed by using a $\chi 2$ homogeneity test.

The statistical analysis mentioned above was performed using R Studio.

6. Study ethics

This study was proceeded conducted in compliance with the Helsinki Declaration and related laws such as the Bioethics and Safety Act.

For the study data, anonymized data were used after approval of the Institutional Review Board of Seoul National University (IRB No. E1811/002-001) and the review committee of the NHISS (NHISS No: NHIS-2020-1-544).

IV. Results

1. Prevalence and incidence of developmental disorders

A total of 223,212 subjects from the target population were diagnosed with DDs between from 2003 and to 2017. Like other countries around the world, Korea clearly showed trends of increase in both the incidence rate and prevalence of DDs (Fig. 4a). The incidence rate in boys surpassed that in girls, and the gap widened throughout the period, peaking at 32.7% in 2014, and then remained steady (Fig. 4b). Between 2003 and 2017, the incidence rate (per 10,000) more than doubled (from 26.30 to 60.08), and the prevalence (per 100) increased by more than four times (from 0.567 to 2.545) (Table 1). For the linear trends of the incidence rate and prevalence categorized by the three five-year groups, the increase of the prevalence in the groups remained the same, while the increase of the incidence rate rose in the second group (2008–2012) before returning to the original level of increase in the third group (2013–2017).

In 2003, the total incidence rate in boys was 35.4 (per 10,000), and in girls it was 16.3 (per 10,000); the gap of the incidence rates between boys and girls widened to 31.4 (per 10,000) in 2017 (Fig. 4b). Table 2 presents indicates a

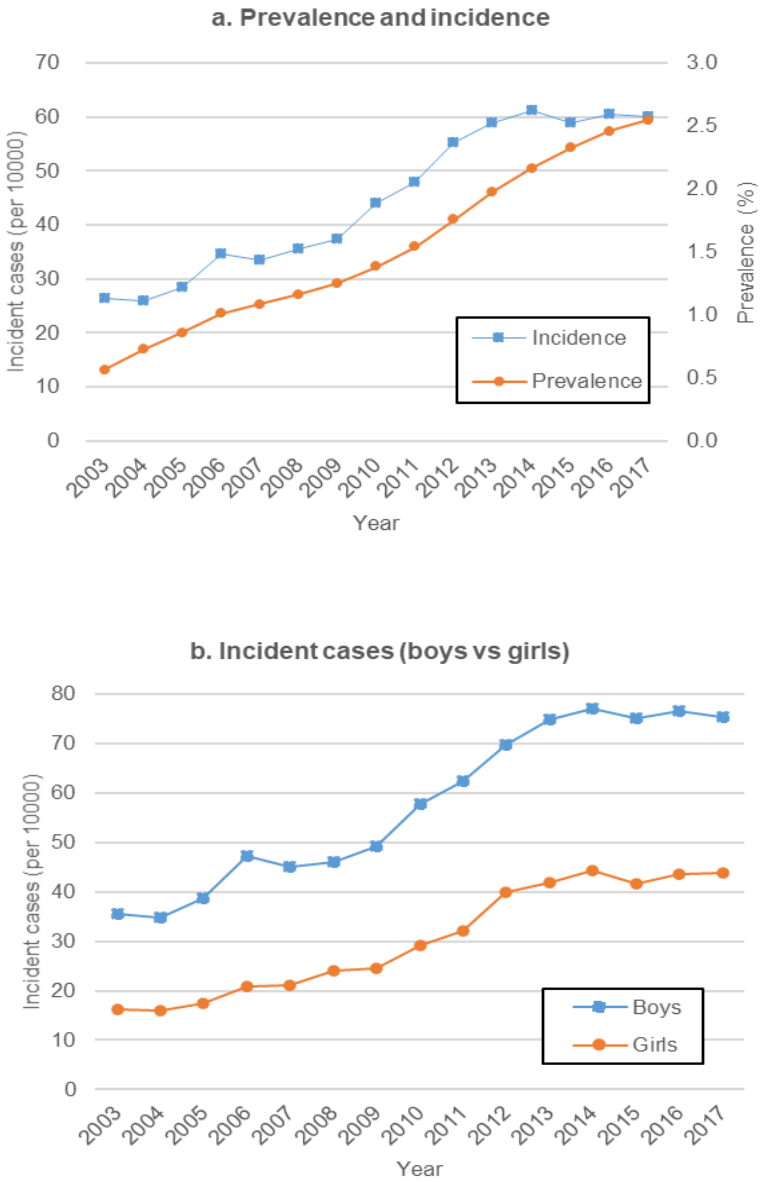


Figure 4. Trends in incidence rate and prevalence of developmental disorders among children 0 to 6 years old, 2003–2017 a. Total incidence rate and prevalence of developmental disorders b. Incident cases of developmental disorders by gender. The linear trends for boys and girls are statistically significant ($p < 0.001$)

Table 1. Linear trends in incidence rate and prevalence of developmental disorders by 3 year groups, 2003-2017

	Incidence rate				Prevalence			
	total	2003 -2007	2008 -2012	2013 -2017	total	2003 -2007	2008 -2012	2013 -2017
n	223212	56162	71363	95687	754972	159079	230314	365579
per 10,000 / per 100	43.6	29.5	43.9	59.9	1.5	0.8	1.4	2.3
ß		2.33	5.00	0.16		0.13	0.15	0.14
SE		0.64	0.52	0.36		0.01	0.01	0.01
p-value		0.04	0.002	0.7		< 0.001	0.002	< 0.001
Z		Ref.	Increased	Not different		Ref.	Not different	Not different

higher incidence rate among boys than girls in all subconditions of DDs ($p < 0.001$). Especially for ADHD, ASD, and language disorders, the incidence rates among boys were 4.2 times, 2.8 times, and 3.1 times higher, respectively, than those of girls. Unlike the incidence between boys and girls, the incidence rate in the age groups differed by subcondition that resulted to difference of the mean age in each subcondition. The mean age of the children with ADHD and learning disorder were 5.2 (±2.5) and 5.1 (±2.6) years old, while that of cerebral palsy was 1.1 (±2.4). Although the mean age of the children in each subcondition differed, the mean age of the total subjects located in the middle (2.9 years old, ±2.0). By type of city, a linear trend of the incidence rate raised as the size of city increased in all subconditions ($p < 0.001$). In addition, the

Table 2. Ratio among the incident cases of developmental disorders by characteristics of the subjects, 2003~2017

	Total incident cases N (%)	Mean Age (SD) (years)	Age (years) N (%)[a]		χ^2 (*p*)	Sex N (%)[b]		χ^2 (*p*)	Type of city N (per 10000)[c]				χ^2 (*p*)	Medical insurance quartile N (%)[d]				χ^2 (*p*)
			0-2	3-6		Boys	Girls		Capital city[e]	Metro -politan city	Prov -ince	Special Self - governing city[f]		1	2	3	4	
ADHD	32492 (14.6)	5.2 (2.5)	609 (0.3)	31882 (14.3)	19950 (< 0.001)	26249 (11.8)	6242 (2.8)	10975 (< 0.001)	16389 (7.2)	8616 (6.7)	7106 (4.8)	121 (1.6)	112942 (<0.001)	3447 (1.5)	5848 (2.6)	10574 (4.7)	1136 (5.1)	269.22 (< 0.001)
ASD	19606 (8.8)	2.9 (1.7)	7912 (3.5)	11691 (5.2)	0.03 (0.87)	14394 (6.4)	5209 (2.3)	3688 (< 0.001)	9140 (4.0)	5017 (3.9)	5127 (3.4)	86 (1.1)	91993 (<0.001)	1834 (0.8)	3764 (1.7)	6788 (3.0)	6098 (2.7)	7.92 (< 0.05)
CP	23686 (10.6)	1.1 (2.4)	20156 (9.0)	3518 (1.6)	19793 (<0.001)	13499 (6.0)	10182 (4.6)	261.18 (< 0.001)	10268 (4.5)	6001 (4.7)	6919 (4.6)	176 (2.3)	135735 (< 0.001)	2345 (1.1)	5062 (2.3)	8455 (3.8)	5886 (2.6)	428.14 (< 0.001)
DD	77696 (34.8)	2.0 (2.0)	51104 (22.9)	26582 (11.9)	21001 (< 0.001)	44231 (19.8)	33456 (15.0)	835.91 (< 0.001)	40803 (18.0)	17320 (13.5)	18316 (12.3)	277 (3.6)	311794 (< 0.001)	7263 (3.3)	13434 (6.0)	27501 (12.3)	24750 (11.1)	175.37 (< 0.001)
LD	56251 (25.2)	3.5 (1.4)	11474 (5.1)	44765 (20.1)	9263.30 (< 0.001)	42,517 (19.0)	13722 (6.1)	12820 (< 0.001)	28206 (12.4)	13252 (10.4)	13595 (9.1)	242 (3.2)	232238 (< 0.001)	5428 (2.4)	10518 (4.7)	19863 (8.9)	17514 (7.8)	4.1066 (0.25)
LRD	1092 (0.5)	5.1 (2.6)	93 (0.0)	999 (0.4)	457.17 (< 0.001)	736 (0.3)	356 (0.2)	106.41 (< 0.001)	589 (0.3)	244 (0.2)	254 (0.2)	2 (0.0)	4323.7 (< 0.001)	125 (0.1)	191 (0.1)	332 (0.1)	392 (0.2)	18.344 (< 0.001)
ID	8587 (3.8)	4.2 (2.0)	1189 (0.5)	7396 (3.3)	2495.60 (< 0.001)	5804 (2.6)	2781 (1.2)	863.43 (< 0.001)	4087 (1.8)	1843 (1.4)	2551 (1.7)	47 (0.6)	51545 (< 0.001)	961 (0.4)	1951 (0.9)	2889 (1.3)	2161 (1.0)	200.80 (< 0.001)

SSD	3802 (1.7)	1.5 (2.1)	3062 (1.4)	740 (0.3)	2557.10 (< 0.001)	2227 (1.0)	1575 (0.7)	70.55 (< 0.001)	2092 (0.9)	730 (0.6)	944 (0.6)	17 (0.2)	17049 (< 0.001)	405 (0.2)	812 (0.4)	1350 (0.6)	988 (0.4)	50.36 (< 0.001)
All	223212 (100.0)	2.9 (2.0)	95599 (42.8)	127573 (57.2)	599.75 (< 0.001)	149657 (67.0)	73523 (32.9)	21019 (< 0.001)	111574 (49.1)	53023 (41.4)	54812 (36.7)	968 (12.6)	5093.6 (< 0.001)	21808 (9.8)	41580 (18.6)	77752 (34.8)	69153 (31.0)	–

ADHD attention deficit hyperactivity disorder *ASD* autism spectrum disorders *CP* cerebral palsy *DD* developmental delay *LD* language disorders *LRD* learning disorder *ID* intellectual disorder *SSD* special sensory disorders [a] ages 0-2 versus 3-6 [b] boys versus girls [c] a linear trend of type of city [d] a linear trend of medical insurance quartile [e] It includes the capital city of Korea, Seoul, and Gyeunggi province that located around Seoul [f] It includes Seajong and Jeju.

top 50% of the ratio of medical insurance quartile for all DDs among the total incident cases outweighed that in the lower 50% by 37.4% (83,517 cases) ($p < 0.001$), and this trend was the same when analyzed by each subcondition.

When comparing the ratios for the total incident cases of subconditions in the third group (2013–2017) to those in first group (2003–2007), the ratio of ASD, developmental delay, and language disorders increased by 13.68%, 817.59%, and 30.75%, respectively, while all others decreased (Table 3). Table 4 shows further analysis of these three subconditions, stratified by age, sex, city size, and medical insurance quartile. The effects of these variables on the incidence of the three subconditions over the three time groups appeared similar to those of the total incidence rates shown in Table 2. For ASD and language disorders, the incidence rate in the older age group (3–6 years old) was higher than that in the younger age group (0–2 years old), whereas the incidence of developmental delay was higher in the younger age group (0–2 years old). The incidence for the three subconditions increased by the size of city and the medical insurance quartile, and this trend did not change over the three time periods.

Table 3. Linear trends in ratio among the total incident cases of developmental disorders, 2003-2017

Disorders	Total Incident Cases	2003 -2007	2008 -2012	2013 -2017	2003–2007 vs. 2013–2017	R [a] (*p*)
	N (%)	N (%)	N (%)	N (%)	%	
ADHD	32492 (14.6)	14077 (6.3)	11061 (5.0)	7354 (3.3)	-47.8 *	-1.51 (0.04)
ASD	19606 (8.8)	6058 (2.7)	6661 (3.0)	6887 (3.1)	13.7	0.19 (0.16)
CP	23686 (10.6)	8916 (4.0)	7701 (3.5)	7069 (3.2)	-20.7	-0.41 (0.11)
DD	77696 (34.8)	5599 (2.5)	20721 (9.3)	51376 (23.0)	817.6	10.25 (0.12)
LD	56251 (25.2)	15488 (6.9)	20513 (9.2)	20250 (9.1)	30.7	1.07 (0.36)
LRD	1092 (0.5)	523 (0.2)	360 (0.2)	209 (0.1)	-60.0 *	-0.07 (0.02)
ID	8587 (3.8)	3902 (1.7)	2630 (1.2)	2055 (0.9)	-47.3	-0.41 (0.14)
SSD	3802 (1.7)	1599 (0.7)	1716 (0.8)	487 (0.2)	-69.5	-0.25 (0.39)

ADHD attention deficit hyperactivity disorder, *ASD* autism spectrum disorders, *CP* cerebral palsy, *DD* developmental delay, *LD* language disorders, *LRD* learning disorder, *ID* intellectual disorder, *SSD* special sensory disorders; [a] Test of linear trend between 3 year groups.

Table 4. Incident rates of autism spectrum disorder, developmental delay, and language disorders among the total population by year groups, demo-geographic factors, and economic factors, 2003-2017

	ASD, per 10000			2003–2007 vs. 2013–2017 (%)	R^d	DD, per 10000			2003–2007 vs. 2013–2017 (%)	R^d	LD, per 10000			2003–2007 vs. 2013–2017 (%)	R^d
	2003 - 2007	2008 - 2012	2013 - 2017			2003 – 2007	2008 – 2012	2013 – 2017			2003 – 2007	2008 – 2012	2013 – 2017		
Age, year															
0~2	3.0	4.7	3.9	31.2	0.005	5.9	22.2	47.8	710.9	0.21	4.1	6.7	5.9	43.1	0.009
3~6	3.3	3.7	4.6	39.0	0.007	1.2	5.8	21.1	1728.7	0.10	10.6	16.9	17.5	65.3	0.03
Sex															
Boys	4.6	5.7	6.2	33.8	0.008	2.9	13.4	36.7	1164.6	0.17	12.0	18.4	18.5	54.2	0.03
Girls	1.6	2.4	2.4	44.2	0.004	3.0	12.0	27.4	817.4	0.12	4.0	6.4	6.6	65.6	0.01
Type of city															
Capital [a]	3.2	4.5	4.5	41.9	0.007	3.6	14.2	38.6	959.5	0.18	9.8	14.2	13.6	37.8	0.02
Metropolitan	3.4	4.3	4.2	21.7	0.004	2.3	11.7	28.9	1171.4	0.13	7.5	12.0	12.1	61.2	0.02
Province	3.0	3.4	4.0	35.1	0.005	2.5	11.3	25.3	933.1	0.11	6.1	10.5	11.4	86.4	0.03
Special Self -governing [b]	2.3	<0.1	0.9	-59.6	-0.007	1.1	<0.1	8.3	644.4	0.03	7.3	0.1	1.9	-73.6	-0.03
Medical insurance quartile [c]															
1	10.9	8.5	7.2	-34.2	-1.86	10.5	26.5	49.5	369.3	19.46	25.0	28.1	22.6	-9.8	-1.22
2	11.3	9.0	7.5	-33.9	-1.92	10.8	28.2	51.5	377.5	20.36	25.8	28.7	22.1	-14.5	-1.87
3	11.1	9.2	6.9	-37.9	-2.11*	10.0	30.3	54.7	446.7*	22.34*	28.5	29.0	21.2	-25.8	-3.67
4	10.2	9.9	7.3	-27.7	-1.41	9.0	28.7	55.5	516.1	23.24	29.4	29.2	20.3	-31.1	-4.57

ASD autism spectrum disorders, *DD* developmental delay, *LD* language disorders; [a] It includes the capital city of Korea, Seoul, and Gyeunggi Province located around Seoul; [b] It includes Sejong and Jeju; [c] Incidence rate among the total incident cases of developmental disorders, (%); [d] Test of linear trend between 3 year groups *$p < 0.05$.

2. Screening effects of the National Health Screening Program for Infants and Children on developmental disorders

Among the total of 7,374,284 children who were subjects of the NHSP from 2008 to 2017, 5,079,364 children (69%) checked the NHSP and 167,050 (2%) were diagnosed with DDs (Fig. 5).

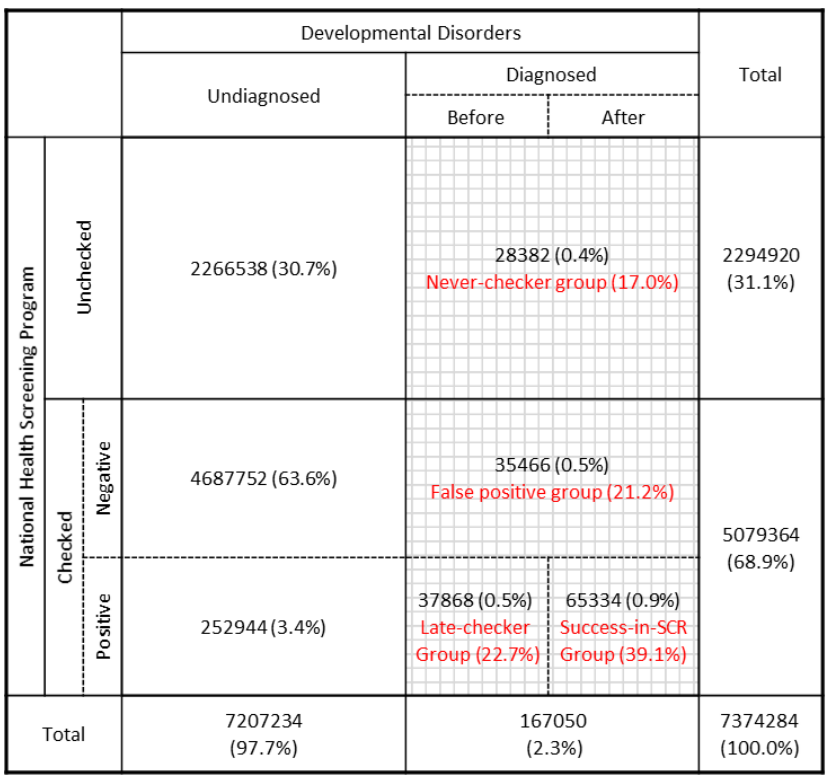
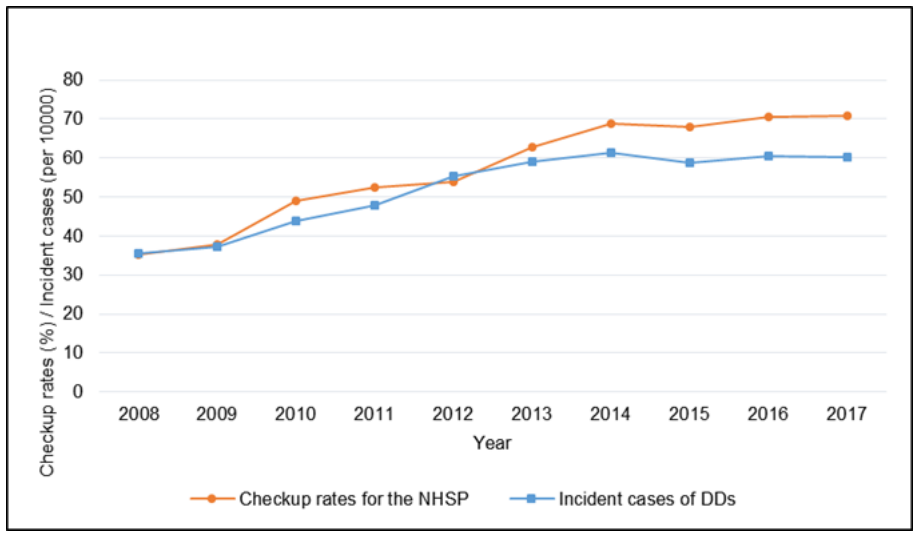


Figure 5. Cross-analysis between the NHSP checkups and the incidence of DDs, 2008–2017. *Note:* Plaid pattern describes the study subjects, and () in red color within the plaid area indicates the percentage among the study subjects.

Among the study subjects (167,050), 65,334 children (39%) comprised the Success-in-SCR group, and 35,466 (21%) children comprised the false negatives. Figure 6 and Table 5 illustrate the annual trends of checkup rates and incidence, along with the total cases of the Success-in-SCR and Failure-in-SCR groups. Both the checkup and incidence rates showed an increasing trend for the past decade. But checkup rates began increasing more rapidly than incidence rates (increases by about 35% and about 25%, respectively) since 2013, and the gap between the two linear trends was statistically significant on the Z test. When comparing the annual cases of DDs between the case groups, the percentage of never-checkers rapidly decreased, from 51.1% in 2008 to 6.6% in 2014, and then turned to an increasing trend since 2015, soaring to 24.0% in 2017. The gap between the Success-in-SCR and Failure-in-SCR rates narrowed between 2008 and 2013, following the trend of the never-checker rate; but then it widened since 2014, where the increase in the false negative rate outweighed the decrease of the never-checker rate; the false negative rate steadily increased, from 2.7% in 2008 by more than 10 times throughout the period. In 2017, however, the never-checker rate again grew higher than the false negative rate, and as a result, the Failure-in-SCR rate peaked at 13,068 (70.2%). Apart from the fluctuation of the never-checker and the false negative rates, the late-checker rate remained steady over the whole period.

a.



b.

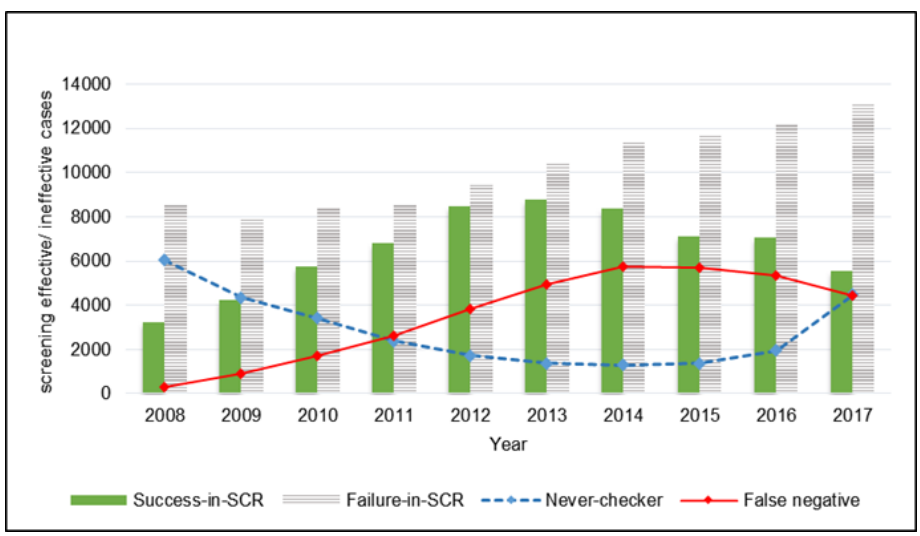


Figure 6. Linear trends of the key variables compared: the NHSP checkup rates and the incidence rates for DDs (a), the Success-in-SCR rate versus the Failure-in-SCR (b), 2008-2017

Table 5. The annual number of children of the Success-in-SCR group and the Failure-in-SCR groups, 2008–2017

YEAR	2008 n (%)	2009 n (%)	2010 n (%)	2011 n (%)	2012 n (%)	2013 n (%)	2014 n (%)	2015 n (%)	2016 n (%)	2017 n (%)
Checkup cases of the NHSP	708972 (35.1)	685009 (37.9)	1090675 (48.9)	1188166 (52.3)	1477147 (54.0)	1743673 (62.7)	1881683 (68.8)	1838084 (68.1)	1909577 (70.6)	1875921 (70.9)
Incident cases of DDs	11807 (35.5)	12153 (37.4)	14154 (44.0)	15348 (47.8)	17901 (55.3)	19237 (58.9)	19812 (61.2)	18833 (58.9)	19193 (60.6)	18612 (60.1)
Success-in-SCR group	3208 (27.2)	4248 (35.0)	5754 (40.7)	6818 (44.4)	8471 (47.3)	8767 (45.6)	8358 (42.2)	7128 (37.8)	7038 (36.7)	5544 (29.8)
Failure-in-SCR group	8599 (72.8)	7905 (65.0)	8400 (59.3)	8530 (55.6)	9430 (52.7)	10470 (54.4)	11454 (57.8)	11705 (62.2)	12155 (63.3)	13068 (70.2)
Never-checker group	6039 (51.1)	4339 (35.7)	3428 (24.2)	2373 (15.5)	1735 (9.7)	1368 (7.1)	1307 (6.6)	1371 (7.3)	1947 (10.1)	4475 (24.0)
False negative group	324 (2.7)	881 (7.2)	1703 (12.0)	2599 (16.9)	3814 (21.3)	4921 (25.6)	5751 (29.0)	5692 (30.2)	5359 (27.9)	4422 (23.8)
Late-checker group	2236 (18.9)	2685 (22.1)	3269 (23.1)	3558 (23.2)	3881 (21.7)	4181 (21.7)	4396 (22.2)	4642 (24.6)	4849 (25.3)	4171 (22.4)

Note: () of incident cases of DDs indicates the number of cases per 10,000.

The Success-in-SCR rate differed by general characteristics: the Success-in-SCR rate among girls was higher than among boys by 8.6%, and it increased as the size of the city was bigger and economic status higher (Table 6). When comparing the percentages among the case groups under the Failure-in-SCR group, the late-checkers had the highest percentage, regardless of city size (with the exception of the special self-governing city), while it differed by economic status—children in a higher medical insurance quartile showed the highest rate of false negatives, whereas those in the lower quartile had the highest rate of late-checkers. Probabilities of the incident cases of DDs in the case groups stratified by general characteristics were all statistically significant on the $\chi 2$ homogeneity test, except that the never-checkers were stratified by sex. These results were similar when analyzed by subconditions (Appendix 2).

Table 6. The incident cases of DDs in the case groups, stratified by demo-geographic and economic factors, 2008-2017

Case Group	Total (N, %)	Mean Age, SD (year)	Sex (N, %) [a]		χ^2 (*p*)	Type of City (N, per 10000) [b]				χ^2 (*p*)	Medical Insurance Quartile (N, %)[c]				χ^2 (*p*)
			M	F		Special Self-Governing City [d]	Prov-ince	Metro-politan City	Capital City [e]		1	2	3	4	
Success-in-SCR	65334 (39.1)	2.9 (2.0)	39791 (36.2)	25543 (44.8)	1184.20 (<0.001)	922 (33.8)	15071 (36.6)	15031 (38.2)	34234 (41.1)	291.73 (<0.001)	6123 (31.1)	10583 (35.0)	22444 (42.1)	22647 (44.1)	1435.80 (<0.001)
Failure-in-SCR	101716 (60.9)	2.7 (1.9)	70251 (63.8)	31436 (55.2)	1184.20 (<0.001)	1808 (66.2)	26146 (63.4)	24341 (61.8)	49147 (58.9)	291.73 (<0.001)	13573 (68.9)	19669 (65.0)	30856 (57.9)	28651 (55.9)	1435.80 (<0.001)
Never-checker group [f]	28382 (27.9)	3.1 (2.4)	18747 (26.7)	9635 (30.6)	0.42 (0.52)	355 (19.6)	6619 (25.3)	6356 (26.1)	14899 (30.3)	121.69 (<0.001)	2967 (21.9)	4993 (25.4)	9179 (29.7)	9626 (33.6)	159.20 (<0.001)
False negative group [f]	35466 (34.9)	2.9 (1.5)	26207 (37.3)	9247 (29.4)	1291.80 (<0.001)	787 (43.5)	9146 (35.0)	8709 (35.8)	16790 (34.2)	194.98 (<0.001)	3779 (27.8)	6614 (33.6)	12639 (41.0)	10005 (34.9)	340.63 (<0.001)
Late-checker group [f]	37868 (37.2)	2.2 (1.6)	25297 (36.0)	12554 (39.9)	19.52 (<0.001)	666 (36.8)	10381 (39.7)	9276 (38.1)	17458 (35.5)	314.08 (<0.001)	6827 (50.3)	8062 (41.0)	9038 (29.3)	9020 (31.5)	3634.30 (<0.001)
Total	167050 (100.0)	2.8 (1.9)	110042 (65.9)	56979 (34.1)	–	2730 (1.6)	41217 (24.7)	39372 (23.6)	83381 (50.0)	–	19696 (11.8)	30252 (18.1)	53300 (31.9)	51298 (30.7)	–

SCR, screening. *Note:* All data have P 0.001, except the incidence cases in the never-checker group by sex (p>0.05) [a] boys versus girls [b] linear trend of type of city [c] linear trend of medical insurance quartile [d] Seajong and Jeju [e] Seoul, the capital city of Korea, and Gyeunggi Province, located around Seoul [f] () is the percentage among the NSCR total.

Table 7 presents the percentages in the Success-in-SCR and Failure-in-SCR groups by subconditions. ADHD and developmental delay had the highest rates of the Success-in-SCR group (49.0% and 45.4%, respectively), while the Failure-in-SCR rates for ASD, cerebral palsy, and intellectual disorder were the highest (74.6%, 68.4%, and 82.3%, respectively). For ASD, language disorders, and intellectual disorder, the number of children with false negatives exceeded even that of the Success-in-SCR group by 13.4%, 12.6%, and 11.7%, respectively.

Table 7. The incident cases of DDs in the subconditions, stratified by case groups, 2008-2017

Disorders	Total (N, %)	Mean Age at diagnosis, SD (year)	Success-in-SCR (N, %)	Failure-in-SCR (N, %)	Never-checker group[a] (N, %)	False negative group[a] (N, %)	Late-checker group[a] (N, %)
ADHD	18415 (11.0)	5.3 (1.0)	9025 (49.0)	9390 (51.0)	6116 (65.1)	1956 (20.8)	1318 (14.0)
ASD	13548 (8.1)	2.8 (1.7)	3441 (25.4)	10107 (74.6)	2128 (21.1)	3917 (38.8)	4062 (40.2)
CP	14770 (8.8)	0.8 (1.3)	4667 (31.6)	10103 (68.4)	4810 (47.6)	1361 (13.5)	3932 (38.9)
DD	72097 (43.2)	2.0 (1.8)	32758 (45.4)	39339 (54.6)	8072 (20.5)	14205 (36.1)	17062 (43.4)
LD	40763 (24.4)	3.5 (1.2)	13591 (33.3)	27172 (66.7)	5524 (20.3)	12471 (45.9)	9177 (33.8)
LRD	569 (0.3)	5.0 (1.5)	229 (40.2)	340 (59.8)	201 (59.1)	77 (22.6)	62 (18.2)
ID	4685 (2.8)	4.2 (1.5)	828 (17.7)	3857 (82.3)	1207 (31.3)	1134 (29.4)	1516 (39.3)
SSD	2203 (1.3)	1.4 (1.4)	795 (36.1)	1408 (63.9)	324 (23.0)	345 (24.5)	739 (52.5)
All	167050 (100.0)	2.8 (1.9)	65334 (39.1)	101716 (60.9)	28382 (27.9)	35466 (34.9)	37868 (37.2)

ADHD attention deficit hyperactivity disorder *ASD* autism spectrum disorders *CP* cerebral palsy *DD* developmental delay *LD* language disorders *LRD* learning disorder *ID* intellectual disorder *SSD* special sensory disorders *SCR* screening. *Note:* () is percentage among the Failure-in-SCR group.

V. Discussion

1. Prevalence and incidence of developmental disorders

The study results show an evident increase over the 15-year period in both the incidence rate and prevalence of DDs among the younger population in Korea. When analyzing the trends by the three time-period groups, a more rapid increase of the incidence rate in the second group (2008–2012) was observed. Although the increase of the prevalence of each group remained the same, this may be due to the small portion of the incidence rates among the prevalence. The implementation of the National Health Screening Program for Infants and Children (NHSP) is one of the possible causes for this change. The checkup rate for the NHSP increased from about 35% in 2008 to more than 70% in 2014, and it has since maintained a similar rate (Status of the Targets and the Examinees for the National Health Screening Program for Infant and Children by Gender, City, and Country, 2017). The trend of the incidence rates for DDs followed this change, which indicates possible influence of the NHSP on the incidence of DDs.

The study results suggest clear differences between the mean ages in each subcondition. Cerebral palsy or special sensory disorders have younger mean

ages (1.1 (±1.4) and 1.5 (±2.1) years old, respectively) than ADHD, learning disorder, and intellectual disorder (5.2 (±2.5), 5.1 (±2.6), and 4.2 (±2.0) years old, respectively). Considering the nature of developmental disorders, which occur according to the developmental process, the mean age implies the onset of the subconditions.

The incidence rates differed by demo-geographic factors and by economic status of families. In all subconditions, boys had at least 1.3 to 4.2 times higher incidence rates than girls did, indicating a higher tendency for boys to have DDs. This phenomenon appeared more prominently in ADHD, ASD, and language disorders—4.2 times, 2.8 times, and 3.1 times higher in boys, respectively—which matches many previous findings (Boyle et al., 2011; Sayal et al., 2018; Canals et al., 2018; Longo et al., 2017; Supekar et al., 2017). Sayal et al. (2018), however, suggest under-identification of ADHD in girls as the reason for the gender differences in the incidence of ADHD. According to their study, the prescribing prevalence of ADHD among boys increased in earlier years, while a similar trend among girls was also found in later years (Sayal et al., 2018).

Economic status also plays an important role in detecting DDs. The prevalence of all DDs in the upper 50% of economic status was more than double that of the lower 50%, which we can interpret as a result of higher

awareness of the diseases and greater ability to pay for medical expenses to undergo close examinations for DDs. Considering these findings, along with similar results from a previous study (Roelfsema et al., 2012), financial support needs to be made available in order to achieve early detection of DDs and alleviate health inequality caused by economic status.

As a consequence of geographic factors affecting access to hospitals that provide close examination for DDs, the incidence rate increased by the size of the living area when analyzed by each subcondition and by DDs as a whole. In fact, among 168 clinics or hospitals that provide close developmental examination for infants and children in Korea, 97 (57.7%) are concentrated in the capital city (Seoul and Gyeunggi Province) and 46 in metropolitan cities (27.4%), whereas only 23 (13.7%) and 2 (1.2%) clinics and hospitals are located in provinces and special self-governing cities (Finding Clinics or Hospitals, 2017). Looking at the relationship between the incidence rates of DDs and economic status, it is clear that the incidence rates in the top 50% of the medical insurance median were more than double those of the lower 50% (65.8% and 28.4%, respectively), which may indicate an impact of financial ability on identification of DDs. Considering that the average cost to undergo a close developmental examination (Kim, 2013) was higher than 10% of the median monthly income for Korean employees (Average Income, Median

Income, Income Distribution, 2017), examinations may be burdensome for low-income families.

Compared to those of the first group (2003–2007), the incident rates for ASD, developmental delay, and language disorders in the third group (2013–2017) increased by 13.7%, 817.6%, and 30.8%, respectively, and the rest of the subconditions decreased. In addition, the total incident cases for ASD (19,606, 8.8%), developmental delay (77,696, 34.8%), and language disorders (56,251, 25.2%) throughout the 15-year period rank 5, 1, and 2, respectively. These statistical results are different from those of an epidemiological study conducted in the US. In that study, the incident cases of learning disorder (8154, 51.10%), ADHD (7652, 47.96%), and developmental delay (3,978, 24.93%) ranked 1, 2, and 3, respectively, among other subconditions (Boyle et al., 2011).

This result, however, is due to the age difference of the subjects. Our study subjects are between 0 and 6 years old; those of the other study are between 3 and 17 years old. In fact, the prevalence of ADHD and learning disorder in the older group (11–17 years old, 8.93% and 9.27%, respectively) was higher than that in the younger group (3–10 years old, 4.72% and 5.07%, respectively) (Boyle et al., 2011). Methodological difference between the studies may also contribute to the difference of the results: in the American

study (Boyle et al., 2011), all subconditions that the subjects were diagnosed with were counted as individual cases, which allowed double counting of the total subjects, whereas this study only considered an initially diagnosed subcondition as an incident case of each subject, which may lead to the underestimation of the incidence rates for subconditions with later onset, such as ADHD or learning disorder, when they coexist with other subconditions with earlier onset.

2. Screening effects of the National Health Screening Program for Infants and Children on developmental disorders

The results of the NHSP throughout the 10-year period were evaluated since 2008, when it was first implemented nationally. Of all the children who were subject to the NHSP, 69% (5,079,364) have been to the NHSP at least once, and about 2% (167,050) of the children were diagnosed with DDs. This high rate of unchecked children was due to the lower checkup rates for the first five years, where the average checkup rate was 46%. But considering that checkup rates of Japan's mandatory health screening program for children between 2009 and 2013 were consistently higher than 90%, and even the elective examination showed higher checkup rates than 80% across all ages in 2013 (Shin et al., 2017), approximately 70% of checkup rate in recent years is relatively low. The lower checkup rate caused the higher rate of never-checkers, and it resulted in the lower rate of the Success-in-SCR group. Both the increasing trend of the Success-in-SCR rate between 2008 and 2013 and the sudden drop in 2017 were consequences of the changes of the never-checker rate.

The false negative rate was another main contributor to the increasing trend of the Failure-in-SCR rate. It was higher than the never-checker rate

between 2011 and 2016—as much as four times higher in 2015. Although the screening tool changed from the K-ASQ to the K-DST in 2014, this seems to have had little effect on the phenomenon. The increasing trend of the Failure-in-SCR rate did not occur at a specific point, but rather, it happened gradually. Moreover, both the K-ASQ and the K-DST showed 65–96% and 88% of sensitivity, respectively (Eun, 2017; Chung et al., 2014), whereas the sensitivity of ASQ, one of the most widely used parent-performed screening tools for DDs, is 75% (Heo et al., 2008). Based on the survey results in the NHSP reevaluation report (Eun, 2017), we can hypothesize that the higher false negative rate may result from inaccurate parent performance on the K-DST, caused by misunderstanding of the purpose of the questions, as they lack understanding of the phenomenology of the behaviors, or not being able to recognize the behaviors in their children because of the absence of opportunities or the absence of tools to observe their children performing the tasks. This hypothesis agrees with a previous study; Øien et al. (2018) investigated the characteristics of children with false-negative results for ASD in comparison with true-negative children in the M-CHAT and found significant delays in social, communication, fine motor, and gross motor development in the false-negative group. They suggested several possible causes for these results: parents having difficulties in recognizing the behavioral markers in their children's behaviors, and understanding DD-

related behaviors, lack of opportunities to grade the behaviors, and differences in levels of the children's expression skills affecting the symptoms of ASD (Øien et al., 2018). The K-DST was revised in 2017; a short paragraph added as a parental guideline, pictures were attached or sentences rephrased to improve understanding for frequently misunderstood questions (Eun, 2017). Considering the number of frequently misunderstood questions and the causes, however, these changes may not be sufficient to improve sensitivity of the K-DST. Further efforts are needed to develop a manual to guide parents performing the K-DST, along with an evaluation tool to measure the accuracy of their performance.

Children with DDs were screened differently by demo-geographic factors and economic status, as well as the subconditions. In line with previous studies, gender differences were marked. In spite of the larger number of incident cases of DDs among boys than among girls (110,042 and 56,979, respectively), the Success-in-SCR rate in boys was less than three-quarters of the girls'. The lower Success-in-SCR rate prevents at-risk boys from early detection and intervention, which, secondarily, aggravates boys' vulnerability for DDs. A number of previous studies have pointed out the gender differences in screening for DDs, and reflecting the differences on the screening program was suggested as a solution to fill the gap (Øien et al.,

2018). Health inequality was also noted, despite the fact that the NHSP is a free program. Children with higher economic status showed a higher tendency to fall into the Success-in-SCR group. In Shin et al. (2017)'s report, limitation of time because of work or for other reasons was pointed out as the main constraint on the parents for missing their children's checkups, which contributes most to the higher Failure-in-SCR among the lower half of the medical insurance quartile (37.8%). In other words, the government's financial support should not be limited to providing the free screening program but needs to be expanded to supporting affordability of time so that health inequality caused by economic status can be ameliorated. Furthermore, the Success-in-SCR rate differed by the subconditions, and the gap was as big as 31.3%, between ADHD (9025, 49.0%) and intellectual disorder (828, 17.7%). Systemic and instrumental problems of the NHSP are possible causes. The higher rate of the Failure-in-SCR group in children with cerebral palsy (68.4%), for example, was due to the higher rate of the never-checkers, which indicates the necessity of additional checkups during the first six months. For France, the Netherlands, Estonia, and Finland, monthly developmental evaluations are provided for the first six months, but the NHSP does not include developmental evaluation for the same period (Shin et al., 2017). The high rates of the false negatives lead to a delay in intervention, and the consequences are critical, especially for children with ASD or language

disorders. For the most frequently seen communication problems, more than 65% can be improved when intervention happens before age 3 years (Mulrine & Kollia, 2015). In the case of ASD, a significant gap exists between the average diagnosis age and the age at which the diagnosis is highly stable (4 years and 18 months, respectively) (Christensen et al., 2016; Landa, 2018). Some researchers suggest widening access to the early intervention services to children at risk of ASD—such as those who have an older sibling with ASD—in order to overcome the gap and facilitate early intervention (Landa, 2018).

3. Implications and future research

Analyzing influential factors in the prevalence of DDs and the screening effects of the NHSP, we observed that economic status and geography played crucial roles in the early detection of DDs. The inability to pay for medical expenses or to take time off from the workplace, and insufficient medical services for screening or for closed examination, were barriers to the early detection of DDs. This may aggravate preexisting health inequalities, as the delay in identification of DDs will eventually lead to increased burden for the children with DDs and their families. In this study, therefore, we have suggested policy proposals.

First, a web-based developmental screening test is needed. According to a report published in 2016, 67% of adults worldwide used the internet, and 43% used smartphones (Poushter, 2016). In Korea, the percentage of internet and smartphone users among adults was 94% and 88%, respectively—first place in the world in both cases (Poushter, 2016). A web-based developmental screening test is best suited to implementation, given these high penetration rates of internet and smartphones. It will not only help overcome the geographic barrier but it will also reduce the medical expenses because of the non-face-to-face process.

Second, the Korean government needs to apply more effort to making the existing programs more feasible so that people can benefit from them. Since the government first implemented the NHSP in 2008, for example, the NHSP has also launched a financial support program to subsidize medical expenses for close examination for the vulnerable. According to data provided by the Ministry of Health and Welfare, however, the average application rate for this program over the past five years was only 13.7% (1,881) among the total number of people who were eligible to apply (13,501) in 2019 (Kim, 2020). Complex procedure and the low checkup rate for the NHSP are the major causes. Therefore, simplifying the application and refund process, as well as advertising the program more actively, are potential solutions (Kim, 2020).

Lastly, social movements for improving awareness of people with DDs must gain momentum. In Korean society, disabled people, especially those with DDs, have been stigmatized, which results in people being undiagnosed and untreated (Kim et al., 2011). This discriminating atmosphere aggravates the psychological burden and stress on parents and causes them to withdraw from actively seeking treatment for their children (Kang-Yi et al., 2013). In fact, social support can increase coping behavior and resiliency in the family (Tak & Lee, 1996). Therefore, a movement to improve awareness should accompany increased professional servicies for people with DDs and their

families.

4. Limitations

This study includes several limitations. First, although selection bias can be minimized because of the nature of the data, there is a high chance of underestimation of the real number of incident cases for DDs, as we counted only diagnosed cases of DDs. Fear of stigmatization for parents of children with DDs is one of the barriers for the children to be diagnosed. As the parents face the dilemma of whether to retain the title of "normal" for their children or to have the children diagnosed and receive subsequent remediation to be "normalized" (Russell & Norwich, 2012), many children with DDs still remain undiagnosed and untreated (Rowland et al., 2015; Kang-Yi et al., 2013). This phenomenon may be even more prevalent in Korea: from a previous study, two-thirds of ASD cases among 7–12-year-old children in a total population sample in Korea were neither diagnosed nor treated, because of parental fears about the stigmatization of ASD as a hereditary disorder (Kim et al., 2011). To the best of our knowledge, however, this study is the first epidemiological study for DDs as a whole, using nationwide population–based data in Korea.

Second, this study may not reflect the influences of the related politics or political changes, such as changes in the qualification and benefits of the

developmental rehabilitation service provided by the Ministry of Health and Welfare in Korea, that could bias either the incidence of DDs or the Success-in-SCR rate, as it was conducted at an individual level.

Third, it may not reflect the real screening effect for some subconditions, like ADHD and language disorders, due to the later onset of diagnostics.

The findings of this study should be interpreted within these limitations.

VI. Conclusion

This study shows the increasing trend of the prevalence of DDs over the past 15 years, with the incidence rates differing by demo-geographic factors and economic status. Poor access to clinics or hospitals for close examination and low affordability of the examination for low-income families are the barriers to the detection of DDs. Incidences of developmental delay and language disorders are also found as the most influential subconditions of this change.

In light of the worldwide increase in the prevalence of DDs, early detection and intervention play crucial roles. Despite many countries having implemented a national health screening program for children, many children remained undiagnosed and untreated for DDs (Rowland et al., 2015; Scherzer et al., 2012).

Continued efforts are needed to expand the study to school-aged children and to identify further influential factors. In addition, establishing a more effective screening system for DDs demands consistent effort in evaluating existing programs and solving health inequality.

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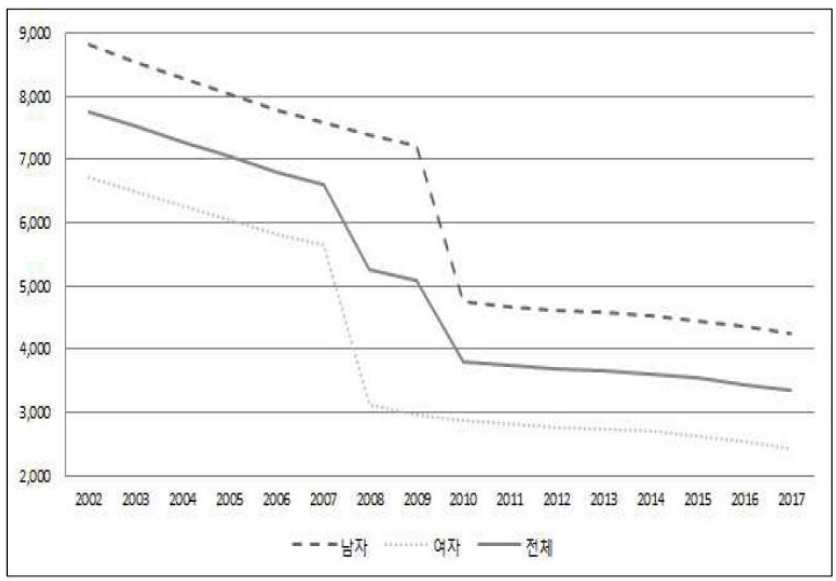
Appendix

Appendix 1. An analysis of the effects of the NHSP implementation, 2002–2017

a



b

	β	S.E	T	P
NHSP implementation (referenced by the period before the NHSP) After the NHSP	-2356.558	418.3	-5.63	0.000
The year of the first finding of DDs	-46.907	50.9	-0.92	0.358
Gender (referenced by boy) Girl	-1651.043	253.8	-7.00	0.000
Age	63.886	60.0	1.06	0.288
Population of the middle of the year by year	0.104	0.004	2.84	0.0005

a. predictive value of the incident cases for DDs; b. the use of medical service for DDs comparing the two period, before and after the NHSP. *Note:* This figure was adapted from Baek et al. (2018).

Appendix 2. The incident cases of DDs in subconditions, categorized by case groups and stratified by demo-geographic and economic factors

Disorders	Mean Age, (SD) (year)	Total (N, %)	Sex (N, %)[a]		Type of City (N, per 10,000)[b]				Medical Insurance Quartile (N, %)[c]			
			M	F	Special Self-Governing City [d]	Province	Metro -politan City	Capital City [e]	1	2	3	4
ADHD												
Success [f]	5.3 (1.0)	9025 (49.0)	7213 (48.5)	1812 (51.0)	25 (83.3)	2045 (47.9)	2502 (51.3)	4339 (48.2)	1059 (47.9)	1523 (47.1)	2855 (50.1)	3148 (49.6)
Failure [g]		9390 (51.0)	7645 (51.5)	1744 (49.0)	5 (16.7)	2225 (52.1)	2375 (48.7)	4664 (51.8)	1151 (52.1)	1709 (52.9)	2842 (49.9)	3195 (50.4)
NC [h]		6116 (65.1)	4891 (64.0)	1225 (70.2)	0 (0.0)	1292 (58.1)	1500 (63.2)	3255 (64.8)	718 (58.1)	1082 (58.3)	1771 (62.3)	2272 (71.1)
FN [h]		1956 (20.8)	1658 (21.7)	297 (17.0)	4 (80.0)	561 (25.2)	535 (22.5)	818 (21.5)	265 (25.2)	358 (25.9)	648 (22.9)	542 (17.0)
LC [h]		1318 (14.0)	1096 (14.3)	222 (12.7)	1 (20.0)	372 (16.7)	340 (14.3)	591 (14.7)	168 (16.7)	269 (16.7)	423 (14.7)	381 (11.9)
Total		18415 (100.0)	14858 (100.0)	3556 (100.0)	30 (100.0)	4270 (100.0)	4877 (100.0)	9003 (100.0)	2210 (100.0)	3232 (100.0)	5697 (100.0)	6343 (100.0)

ASD

Success [f]		3441 (25.4)	2324 (23.7)	1117 (30.0)**	9 (32.1)	843 (24.4)	928 (27.5)	1631 (23.2)	291 (24.4)	554 (24.8)	1158 (25.8)	1249 (28.3)
Failure [g]		10107 (74.6)	7499 (76.3)	2605 (70.0)	19 (67.9)	2607 (75.6)	2452 (72.5)	4843 (76.8)	1004 (75.6)	1871 (75.2)	3365 (74.2)	3167 (71.7)*
NC [h]	2.8 (1.7)	2128 (21.1)	1561 (20.8)	567 (21.8)	0 (0.0)	536 (20.6)	458 (18.7)	1115 (20.0)	208 (20.6)	363 (20.4)	629 (18.4)	774 (24.4)
FN [h]		3917 (38.8)	3042 (40.6)	873 (33.5)	13 (68.4)	958 (36.7)	942 (38.4)	1901 (40.3)	382 (36.7)	683 (36.5)	1388 (41.5)	1225 (38.7)*
LC [h]		4061 (40.2)	2896 (38.6)	1165 (44.7)	6 (31.6)	1113 (42.7)	1052 (42.9)	1827 (38.7)	414 (42.7)	825 (42.1)	1348 (40.1)	1168 (36.9)
Total		13548 (100.0)	9823 (100.0)	3722 (100.0)	28 (100.0)	3450 (100.0)	3380 (100.0)	6474 (100.0)	1295 (100.0)	2425 (100.0)	4523 (100.0)	4416 (100.0)

Cerebral Palsy

Success [f]	0.8 (1.3)	4667 (31.6)	2600 (30.6)	2067 (32.9)	15 (23.8)	1423 (33.1)	1132 (29.6)	2035 (30.1)	413 (33.1)	944 (33.9)	1726 (33.9)	1307 (33.4)
Failure [g]		10103 (68.4)	5885 (69.4)	4214 (67.1)	48 (76.2)	2881 (66.9)	2686 (70.4)	4312 (69.9)	1120 (66.9)	2018 (66.1)	3500 (67.1)	2602 (66.6)
NC [h]		4810 (47.6)	2754 (46.8)	2056 (48.8)	39 (81.3)	1265 (43.9)	1289 (48.0)	2157 (46.0)	533 (43.9)	899 (43.5)	1678 (47.5)	1344 (51.7)
FN [h]		1361 (13.5)	826 (14.0)	535 (12.7)	2 (4.2)	413 (14.3)	353 (13.1)	570 (14.2)	158 (14.3)	288 (14.3)	479 (13.3)	315 (12.1)
LC [h]		3932 (38.9)	2305 (39.2)	1623 (38.5)	7 (14.6)	1203 (41.8)	1044 (38.9)	1585 (39.8)	429 (41.8)	831 (41.2)	1343 (38.2)	943 (36.2)
Total		14770 (100.0)	8485 (100.0)	6281 (100.0)	63 (100.0)	4304 (100.0)	3818 (100.0)	6347 (100.0)	1533 (100.0)	2962 (100.0)	5226 (100.0)	3909 (100.0)

Developmental Delay

Success [f]		32758 (45.4)	16659 (40.3)	16099 (52.4)	129 (51.8)	7163 (42.3)	6860 (42.3)	18299 (40.5)	2794 (42.3)	4972 (42.9)	11497 (45.9)	11730 (50.4)
Failure [g]		39339 (54.6)	24697 (59.7)	14634 (47.6)	120 (48.2)	9771 (57.7)	9368 (57.7)	19466 (59.5)*	3946 (57.7)	7183 (57.1)	13966 (54.1)	11528 (49.6)
NC [h]	2.0 (1.8)	8072 (20.5)	4370 (17.7)	3702 (25.3)	33 (27.5)	1778 (18.2)	1598 (17.1)	4556 (17.4)	792 (18.2)	1261 (18.6)	2864 (20.6)	2741 (23.8)
FN [h]		14205 (36.1)	9892 (40.1)	4309 (29.4)	44 (36.7)	3476 (35.6)	3599 (38.4)	6870 (40.3)	1465 (35.6)	2684 (35.4)	5029 (36.4)	4051 (35.1)
LC [h]		17062 (43.4)	10435 (42.3)	6623 (45.3)	43 (35.8)	4517 (46.2)	4171 (44.5)	8040 (42.3)	1689 (46.2)	3238 (46.1)	6073 (43.1)	4736 (41.1)
Total		72097 (100.0)	41356 (100.0)	30733 (100.0)	249 (100.0)	16934 (100.0)	16228 (100.0)	37765 (100.0)	6740 (100.0)	12155 (100.0)	25463 (100.0)	23258 (100.0)

Language Disorders

Success [f]		13591 (33.3)	9850 (32.2)	3741 (37.0)	26 (44.8)	3112 (30.6)	3276 (34.0)	6912 (32.5)	1358 (30.6)	2260 (30.3)	4618 (32.3)	4609 (36.5)
Failure [g]		27172 (66.7)	20787 (67.8)	6374 (63.0)	32 (55.2)	7047 (69.4)	6367 (66.0)	13099 (67.5)	2829 (69.4)	5198 (69.7)	9434 (67.7)	8028 (63.5)
NC [h]	3.5 (1.2)	5524 (20.3)	4043 (19.4)	1481 (23.2)	1 (3.1)	1290 (18.3)	1246 (19.6)	2906 (19.2)	523 (18.3)	1004 (18.3)	1728 (18.3)	1990 (24.8)
FN [h]		12471 (45.9)	9691 (46.6)	2776 (43.6)	22 (68.8)	3262 (46.3)	2943 (46.2)	5904 (46.1)	1309 (46.3)	2294 (46.1)	4561 (48.1)	3501 (43.6)
LC [h]		9177 (33.8)	7053 (33.9)	2117 (33.2)	9 (28.1)	2495 (35.4)	2178 (34.2)	4289 (33.7)	997 (35.4)	1900 (35.6)	3145 (33.6)	2537 (31.6)
Total		40763 (100.0)	30637 (100.0)	10115 (100.0)	58 (100.0)	10159 (100.0)	9643 (100.0)	20011 (100.0)	4187 (100.0)	7458 (100.0)	14052 (100.0)	12637 (100.0)

Learning Disorder

Success [f]		229 (40.2)	153 (40.6)	76 (39.6)	1 (100.0)	52 (37.1)	39 (32.8)	135 (40.0)**	35 (37.1)	27 (37.9)	58 (34.9)	95 (44.6)
Failure [g]		340 (59.8)	224 (59.4)	116 (60.4)	0 (0.0)	88 (62.9)	80 (67.2)	172 (59.0)*	34 (62.9)	55 (62.1)	109 (65.1)	118 (55.4)*
NC [h]	5.0 (1.5)	201 (59.1)	123 (54.9)	78 (67.2)	0 (0.0)	42 (47.7)	45 (56.3)	114 (54.3)*	20 (47.7)	32 (47.2)	59 (54.2)	81 (68.6)*
FN [h]		77 (22.6)	56 (25.0)	21 (18.1)	0 (0.0)	26 (29.5)	23 (28.8)	28 (25.3)**	10 (29.5)	11 (29.0)	29 (26.0)	18 (15.3)*
LC [h]		62 (18.2)	45 (20.1)	17 (14.7)	0 (0.0)	20 (22.7)	12 (15.0)	30 (20.4)*	4 (22.7)	12 (22.8)	21 (19.8)	19 (16.1)
Total		569 (100.0)	377 (100.0)	192 (100.0)	1 (100.0)	140 (100.0)	119 (100.0)	307 (100.0)	69 (100.0)	82 (100.0)	167 (100.0)	213 (100.0)

Intellectual Disorder

Success [f]		828 (17.7)	562 (17.5)	266 (18.2)	2 (25.0)	286 (19.0)	157 (17.1)	375 (17.0)	97 (19.0)	165 (19.5)	240 (16.5)	262 (22.1)**
Failure [g]		3857 (82.3)	2658 (82.5)	1197 (81.8)	6 (75.0)	1219 (81.0)	762 (82.9)	1834 (82.0)	485 (81.0)	834 (81.5)	1240 (83.5)	926 (77.9)
NC [h]	4.2 (1.5)	1207 (31.3)	804 (30.2)	403 (33.7)	0 (0.0)	351 (28.8)	250 (32.8)	592 (30.3)	147 (28.8)	288 (28.5)	349 (28.5)	313 (33.8)
FN [h]		1134 (29.4)	815 (30.7)	318 (26.6)	4 (66.7)	386 (31.7)	212 (27.8)	523 (30.5)	152 (31.7)	223 (31.7)	384 (31.7)	270 (29.2)
LC [h]		1516 (39.3)	1039 (39.1)	476 (39.8)	2 (33.3)	482 (39.5)	300 (39.4)	719 (39.2)	186 (39.5)	323 (39.7)	507 (40.7)	343 (37.0)
Total		4685 (100.0)	3220 (100.0)	1463 (100.0)	8 (100.0)	1505 (100.0)	919 (100.0)	2209 (100.0)	582 (100.0)	999 (100.0)	1480 (100.0)	1188 (100.0)

Special Sensory Disorders

Success [f]		795 (36.1)	430 (33.4)	365 (39.8)	0 (0.0)	147 (32.3)	137 (29.1)	508 (33.2)	76 (32.3)	138 (32.7)	292 (37.7)	247 (39.7)*
Failure [g]		1408 (63.9)	856 (66.6)	552 (60.2)	0 (0.0)	308 (67.7)	334 (70.9)	757 (66.8)	154 (67.7)	272 (67.3)	480 (62.3)	375 (60.3)**
NC [h]	1.4 (1.4)	324 (23.0)	201 (23.5)	123 (22.3)	0 (0.0)	65 (21.1)	53 (15.9)	204 (23.9)	26 (21.1)	64 (21.5)	101 (21.5)	111 (29.6)*
FN [h]		345 (24.5)	227 (26.5)	118 (21.4)	0 (0.0)	64 (20.8)	102 (30.5)	176 (26.2)	38 (20.8)	73 (20.8)	121 (25.8)	83 (22.1)**
LC [h]		739 (52.5)	428 (50.0)	311 (56.3)	0 (0.0)	179 (58.1)	179 (53.6)	377 (50.8)	90 (58.1)	135 (58.6)	258 (53.6)	181 (48.3)**
Total		2203 (100.0)	1286 (100.0)	917 (100.0)	0 (0.0)	455 (100.0)	471 (100.0)	1265 (100.0)	230 (100.0)	410 (100.0)	772 (100.0)	622 (100.0)

All Subconditions

Success [f]		65334 (39.1)	39791 (36.2)	25543 (44.8)	207 (47.4)	15071 (36.6)	15031 (38.1)	34234 (36.1)	6123 (36.6)	10583 (36.6)	22444 (39.6)	22647 (43.1)
Failure [g]		101716 (60.9)	70251 (63.8)	31436 (55.2)	230 (52.6)	26146 (63.4)	24424 (61.9)	49147 (63.9)	10723 (63.4)	19140 (63.4)	34936 (60.4)	29939 (56.9)
NC [h]	2.8 (1.9)	28382 (27.9)	18747 (26.7)	9635 (30.6)	73 (31.7)	6619 (25.3)	6439 (26.4)	14899 (26.3)	2967 (25.3)	4993 (25.1)	9179 (26.1)	9626 (32.2)
FN [h]		35466 (34.9)	26207 (37.3)	9247 (29.4)	89 (38.7)	9146 (35.0)	8709 (35.7)	16790 (37.2)	3779 (35.0)	6614 (35.6)	12639 (36.6)	10005 (33.4)
LC [h]		37868 (37.2)	25297 (36.0)	12554 (39.9)	68 (29.6)	10381 (39.7)	9276 (38.0)	17458 (36.5)	3977 (39.7)	7533 (39.4)	13118 (37.4)	10308 (34.4)
Total		167050 (100.0)	110042 (100.0)	56979 (100.0)	437 (100.0)	41217 (100.0)	39455 (100.0)	83381 (100.0)	16846 (100.0)	29723 (100.0)	57380 (100.0)	52586 (100.0)

Abbreviations: ADHD, attention deficit hyperactivity disorder; ASD, autism spectrum disorders; SCR, screening; FN, False negative group; LC, Late-checker group; NC, Never-checker group.

Note: All the results for the linear trends are significant, P 0.001, except where marked.

[a] boys versus girls

[b] a linear trend of type of city

[c] a linear trend of medical insurance quartile

[d] Seajong and Jeju

[e] Seoul, the capital city of Korea, and Gyeunggi Province located around Seoul

[f] Success-in-SCR group

[g] Failure-in-SCR group

[h] () is the percentage among the Failure-in-SCR group

*p>0.05

**0.001<p<0.05

Appendix 3. IRB approval letter

심의면제 통보서

수신

책임연구자	**이름**: 나성실	**소속**: 간호대학 지역사회간호학	**직위**: 박사과정
지원기관	해당없음		

과제정보

승인번호	IRB No. E1811/002-001
연구과제명	영유아건강검진의 발달장애 선별효과 분석
연구종류	학술 연구, 학위 논문 연구, 공개된 정보를 이용하는 연구
면제일자	2018-10-24
검토의견	1. 본 연구는 연구참여자에 대한 기존의 자료인 국민건강보험공단의 '맞춤형 DB' 자료를 이용하는 연구로「생명윤리 및 안전에 관한 법률」시행규칙 제13조 제1항 제3호에 근거하여 심의를 면제합니다. 2. 분석한 연구결과는 본교 연구윤리지침에 근거하여 5년 이상 가능한 영구보관하여 주시기 바랍니다.
심의결과	면제승인

상기 연구과제에 대하여 본 위원회에서는 심의면제대상임을 확인합니다.

모든 연구자들은 아래의 사항을 준수하여야 합니다.

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서울대학교 생명윤리위원회 위원장

국문 초록

한국 영유아의 발달장애 유병률 및 영유아건강검진의 선별효과

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발달장애 유병률이 증가함에 따라 많은 OECD 국가들은 발달장애의 조기발견과 조기중재를 위해 국가 차원의 선별검사를 제공하고 있다. 한국정부 또한 2008년 이래 0~6세 영유아 전체를 대상으로 발달장애 고위험아동을 조기에 선별하고자 영유아건강검진을 실시하였다. 하지만 발달장애 역학연구나 영유아건강검진의 효과를 평가한 연구가 매우 부족한 상황이다. 따라서 이러한 연구를 통해 발달장애 관련 사업이나 정책을 개발하고 평가하기 위한 근거를 마련하는 것이 매우 시급이다.

본 기술연구의 목적은 전국민 인구기반 자료를 사용하여 0~6세 영유아의 발달장애 유병률과 발생률의 추세를 확인하고 발달장애에 대한 한국 영유아건강검진 사업의 선별효과를 분석하는 데 있다.

본 연구의 대상자는 위의 두 가지 목적에 따라 분류되었다. 2003년도

에서 2017년도 사이 0~6세 영유아를 대상으로 역학연구를 진행하였으며, 그 중 2008년도에서 2017년도 사이 발달장애를 진단받은 영유아를 대상으로 영유아건강검진의 발달선별효과를 분석하였다. 발달장애 유병률과 발생률, 그리고 영유아건강검진의 선별효과는 연도, 발달장애 하위질병, 인구지리학적 요소, 그리고 경제적 수준에 따라 χ^2을 이용하여 비교, 분석되었으며, 단순선형회귀분석 방법을 사용하여 이들의 추세 변화를 비교하였다.

발달장애 유병률은 2003년도부터 2017년도까지 4배 이상 꾸준히 증가한 것으로 나타났다. 남아 발생률은 전 기간에 걸쳐 여아에 비해 높은 수치를 보였으며, 그 차이는 19.1%에서 31.4%로 더욱 커졌다. 전체 발달장애 발생건수 중 자폐 스펙트럼 장애, 발달지연, 그리고 언어장애의 비율은 각각 13.7%, 817.6%, 30.7% 증가하였으며, 이는 발달장애 유병률의 증가에 큰 영향을 미친 것으로 나타났다.

한국 영유아건강검진 사업의 선별효과를 살펴보면, 2008년도에서 2017년도 사이 발달장애를 진단받은 167,050명의 영유아 중 65,334(39%)명의 영유아가 발달장애로 선별되었으며, 35,466(21%)명의 영유아가 위음성 판정을 받은 것으로 나타났다. 발달장애 선별률을 살펴보면, 2008년 3,208(27.2%)명에서 2012년 8,471(47.3%)명으로 증가하였으나 이후 2017년 5,544(29.8%)명으로 감소하였다. 위음성률의 경우 2008년 2.7%에서 2017년 23.8%로 증가하였으며, 이 변화는 발달장애 발생률의 파동에 가장 큰 영향요인으로 작용하였다.

발달장애 발생률과 선별률은 모두 하위질병들의 연령관련 특성들과 더불어 인구지리학적 요인, 경제적 요인에 의해 영향을 받은 것으로 나타났다. 보다 큰 도시에 살거나 보다 높은 경제적 지위를 가지고 있는 영유아가 그렇지 않은 영유아에 비해 발달장애가 더 많이 선별되었으며, 이러한 결과는 보다 작은 도시에 살거나 경제적 지위가 낮은 영유아가

발달장애에 취약하다는 사실을 보여주고 있다.

결론적으로 발달장애 유병률과 발생률이 지난 15년 동안 꾸준히 증가하고 있는 가운데 영유아건강검진 프로그램은 발달장애를 선별하는데 별다른 효과를 보이지 못하고 있다. 빠르게 증가하고 있는 발달장애 유병률을 저지하기 위해 보다 효과적인 발달장애 선별 시스템을 개발함과 더불어 발달장애 고위험 영유아를 대상으로 한 관련 정책을 개발하고 입안하는 데 더 많은 노력을 기울여야 할 것이다.

주제어 : 발달장애, 선별검사, 어린이, 영아, 역학연구, 전국민 기반 코호트 연구

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