Original Article

Early Hearing Detection and Intervention Results in Northeastern of Iran from 2005 to 2019: A Repeated Cross-Sectional Study

Abstract

Background: Hearing loss is one of the most common congenital disorders. The Early Diagnosis and Intervention Process is designed for the early diagnosis and intervention of hearing loss in infants. The present study aimed to examine the results of Early Hearing Detection and Intervention (EHDI) in northeastern Iran from 2005 to 2019. **Setting:** Northeastern Iran. **Methods:** In most cases, the two-stage protocol (otoacoustic emissions [OAE] and automated auditory brainstem response [AABR]) has been used. Infant assessment methods included the use of OAE, ABR, auditory steady-state response, high-frequency tympanometry, and behavioral audiometry. Interventions included medical interventions, hearing rehabilitation, hearing aids, and cochlear implants. **Results:** 1,162,821 infants were screened. The screening coverage increased from less than 1% in 2005 to about 99% in 2018. The referral rate has been about 1%. 2.17 out of every 1000 infants are hearing impaired, and the most common cases are bilateral hearing loss and mild to moderate hearing loss. **Conclusions:** During 2005 to 2019 the coverage rate reached to more than 95% of live births. To improve the EHDI process in this population, better follow-up of diagnosed neonates and expansion of diagnostic and intervention services are needed.

Keywords: *Congenital hearing loss, early hearing detection and intervention, early intervention, hearing loss, neonatal hearing screening*

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Introduction

According to the World Health Organization (WHO), the number of people with hearing loss is increasing worldwide, even in developed countries.[1] In low- and middle-income countries, there is a higher prevalence of hearing loss during infancy due to various infections or paying less attention to maternal and infant health.[2] Hearing loss is one of the most common chronic disorders in children,[3] which can interfere with the development of communication, cognition, academic skills (such as reading), as well as social and emotional skills.[4,5]

Infants with hearing loss who do not receive the necessary interventions will have lower levels of education and employment in adulthood compared to their peers.[4] Even minimal hearing loss, including unilateral or mild bilateral hearing loss, can cause language and speech development disorders, damage academic achievement, and cause behavioral and psychosocial disorders that require diagnosis and intervention.[6,7]

Hearing loss is typically one of the top 10 causes of years lived with disability (YLDs) worldwide.[8] It was the fourth leading cause of YLDs in the 2013 and 2015 surveys.[9] Hearing loss imposes a heavy burden on society and the healthcare system[10] and costs more than $ 750 billion annually on a global scale.[1]

Congenital problems play an important role in this issue. Despite the various effects of congenital hearing loss, it can be diagnosed and treated at birth. Early intervention at an early age can play a very important role in reducing the burden of hearing loss.[2] Moreover, early interventions can lead to expected and desirable growth.[10] Infants who receive early intervention at the right time perform better in many areas than those who receive interventions later.[3,11]

If hearing impaired infants are identified and treated early, they can reach normal language development[4] and perform well in a variety of areas (e.g., vocabulary development, receptive language, expressive language, syntax, speech production, and social and emotional development).[5]

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Moreover, these infants will have a better quality of life at older ages.[3]

The Early Hearing Detection and Intervention (EHDI) program began in the United States in the 1990s.[5] The goal of EHDI is to identify hearing-impaired infants early and resolve their problems.[4,5,12] This program complies with the International Classification of Functioning, Disability and Health: Children and Youth Version of WHO.[13] Khorasan Razavi province is located in the northeastern of Iran and the process of hearing screening in this province was started in 2005. According to the 2016 census, this province had a population of 6,435,000.[14] The present study aimed to evaluate the EHDI process including screening, diagnosis, and intervention from March 2005 to March 2019 in Khorasan Razavi province.

Methods

In this repeated cross-sectional study, all infants born in cities and villages of Khorasan Razavi province from March 2005 to March 2019 (equivalent to 15 solar years) were studied.

The whole process of screening, diagnosis, and intervention has been carried out under the supervision of the Welfare Organization of Khorasan Razavi Province and the universities of Medical Sciences in Khorasan Razavi Province. This study was approved by the Ethics Committee of Mashhad University of Medical Sciences for the city of Mashhad in 2018 (IR.MUMS.REC.1398.030) and the whole province of Khorasan Razavi from 2005 to 2019 (Supplementary IR.MUMS.REC.1398.030).

Part one: Screening

In this part, a two-stage screening was used. The first screening was performed in the early days after birth and included the transient-evoked otoacoustic emission (TEOAE) test for all live births.[5] This stage of screening was performed in hospitals.

The TEOAE test was performed at least twice for each infant and the criterion for pass in screening was the presence of response in both or most tests. The refer infants were entered the second stage of screening.

The risk factors for hearing loss were also collected from screened infants. These risk factors included family history of hearing loss, more than five days of hospitalization in the neonatal intensive care unit (NICU), hyperbilirubinemia and blood transfusions, intrauterine infections, postnatal infections and positive blood culture of the infant, head and face deformity, history of head trauma, birth weight below 1500 g, history of syndromic diseases in the family, premature birth, and consanguineous marriage of parents.[4,5,12] All infants with these risk factors were referred for the second screening.

These infants also underwent periodic hearing assessments until the age of three. In the first five years (2005–2009), information on hearing loss risk factors was not collected and the only criterion for re-screening was the TEOAE test. From 2010 to 2019, the mean proportions of neonates who passed the TEOAE test and had the risk factors for hearing loss were 15.56% of the total infants screened in the first stage.

In the first stage of screening, parents were given written instruction on the effects of hearing loss and the normal development of language, speech, and hearing. Furthermore, the results of the first stage screening were presented to the parents both orally and in written form.

The second screening stage was performed a few days to a week after the first stage. The method of screening in the second stage varied during the past 15 years. In the first five years (2005–2009), all infants were screened using the TEOAE test, while in the following 10 years (2010–2019) the AABR test was used mostly. On average, 31.2% of infants were evaluated by TEOAE in the second phase of screening. It was attempted to only screen the infants with risk factors using the AABR test, and if the screening center did not have the AABR device, they were referred to centers with the device. The second stage screening was mostly performed by audiologists. Only infants referred from second-stage screening were referred to diagnostic evaluation. At this stage, oral and written instructions on hearing loss were provided to the parents.

Over the years, the number of screening centers and used devices has increased to the point that in 2019, there were 52 screening centers (10 governmental and 42 private), some of which covered several hospitals. In addition, in the same year, five mobile screening centers covered several neighboring cities. Besides, 53 screening devices were used this year, 49 of which had TEOAE and AABR devices, and 80% of their devices were from the same brand (Madsen). All screening devices were automatic and calibrated annually.

Part two: Diagnosis

Infants referred from the second stage of screening were referred to hearing clinics in different parts of Khorasan Razavi province, depending on their place of residence. The diagnostic method for all infants was based on TEOAE and auditory brainstem response (ABR) clinical tests with a click stimulus. In some infants, behavioral audiometry, high-frequency tympanometry, ABR with tone-burst stimulus, distortion product otoacoustic emissions (DPOAE), and auditory steady-state response (ASSR) were used as well. Based on these tests, the type and extent of hearing loss were determined and finally, infants with hearing loss were referred for intervention. Infants with conductive lesions, such as otitis media, were referred to an ENT physician for medical intervention, and infants with sensorineural hearing loss were referred to an audiologist for hearing rehabilitation

at the same center or other centers close to the place of residence of the parents.

Infants with conductive lesions associated with sensorineural hearing loss were referred to an ENT physician and followed up by an audiologist for auditory rehabilitation.

Part three: Intervention

Medical interventions, in cases of conductive impairment, included medication or surgery, which was performed by an ENT physician depending on the type of complication or disease and the age of the patient.

In cases of sensorineural hearing loss, auditory rehabilitation was performed, which included prescription of hearing aids, the frequency modulation (FM) system, cochlear implants, and various methods of hearing rehabilitation. In these centers, hearing rehabilitation was performed by various specialists, including audiologists, speech therapists, educators of hearing-impaired children, and with the advice of healthcare professionals and a psychologist of hearing-impaired children. Finally, Children who needed cochlear implants according to rehabilitation protocols were referred to the only cochlear implant center in the capital of the province.

Statistical analysis

Data were analyzed by SPSS 19.0 software. The variables were described by frequency, percentage, and ratio. Charts and figures show a better presentation of data.

Results

Over 15 years, 1,162,821 infants were screened. The frequency of infants participating in the screening, diagnosis, and intervention process in Khorasan Razavi province during 2005-2019 is shown in Figure 1. In this figure, the following items are specified: 1) live birth rate in Khorasan Razavi province, 2) percentage of newborns screened in the first stage compared to the total population of live births, 3) percentage of infants referred for screening in the second stage compared to the total number of newborns screened during the first stage, 4) percentage of the infants referred for diagnosis compared to the total number of neonates screened in the second stage, and 5) percentage of the infants referred for intervention compared to the total number of infants referred for diagnosis. The frequency of the first screening underwent an increase in consecutive years and, in 2018 and 2019, it reached more than 95% of live births.

Figure 2 shows the percentage of individuals participating in the second screening, diagnosis, and intervention. As shown in Figure 2, participation rates have increased over the years.

During the EDHI process, an attempt was made to involve parents in all stages. In Khorasan Razavi province, participation in various stages is completely optional.

Figure 3 shows the percentage of screening in governmental and private as well as urban and rural centers. In 2005–2010, this project was carried out only in the major cities of the province and private centers.

During these 15 years, most of the screening processes were performed in urban areas and private centers. Figure 4 shows the ratio of diagnosed cases of hearing loss to live births and screened infants at the first and second stages.

Based on the chart of the ratio of diagnosis to second screening, it can be said that this ratio has been increasing in consecutive years, which indicates an improvement in the performance of the first stage screening. This chart also shows an increase in the number of hearing loss cases in recent years. In 2018 and 2019, the percentage of cases diagnosed with hearing loss was higher than in the previous years.

Type and severity of hearing loss in three categories of 1) mild and moderate, 2) moderately severe to severe, and 3) profound are summarized in Table 1. This information was not collected between 2005 and 2009.

The frequency of cases of unilateral and bilateral lesions is summarized in Table 1, which shows that most cases are related to bilateral lesions.

Interventions included medical interventions, hearing aids prescription, and hearing rehabilitation. Medical interventions ranged from cerumen removal, treatment of otitis media, and treatment of amniotic fluid in the middle ear to ear surgeries and cochlear implants. In total, 116, 124, 163, 198, 198, 502, 551, 599, 441, and 271 cases received medical interventions in the years 2010 to 2019, respectively. Moreover, 126, 57, 70, 79, 103, 154, 112, 78, 174, and 114 cases received hearing aids and hearing rehabilitation from 2010 to 2019, respectively. This information was not collected for the years 2005 to 2009.

Years 2005 to 2019, 13, 14, 45, 33, 38, 41, 59, 91, 163, 114, 196, 147, 100, 142, and 121 of cases received cochlear implants of these cases received the implant under two years old. In addition to this province, the cochlear implant center of Khorasan provides services to several neighboring provinces as well.

Discussion

Results of the present study show the EHDI process for northeastern Iran during 2005–2019. In the following, the findings are examined in three parts: screening, diagnosis, and intervention.

Screening

Global hearing loss screening is very important for the detection of hearing loss in infants and leads to early diagnosis.[15] In some areas that do not have a screening program, many cases of hearing loss are diagnosed when they are older than three years due to severe impairments

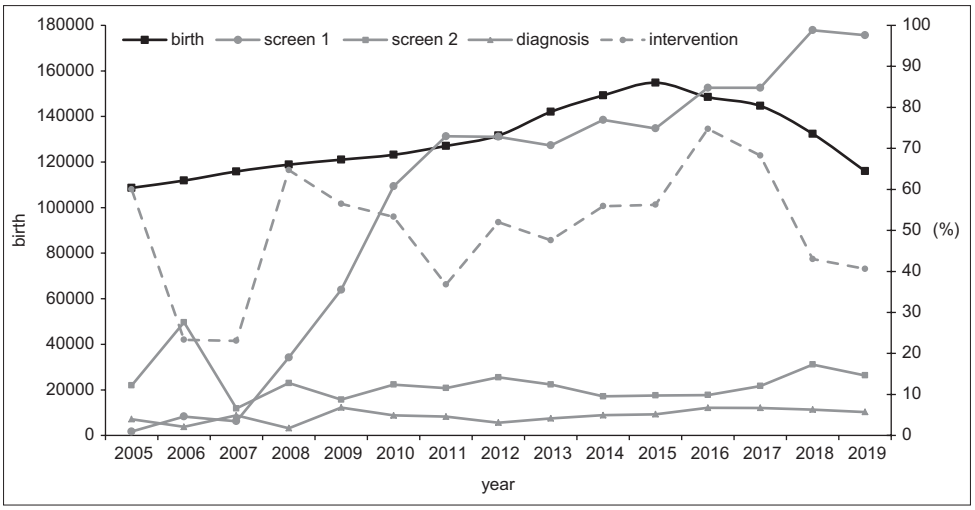

Figure 1: Results of screening, diagnosis, and intervention process in Khorasan Razavi province, Iran during 2005–2019

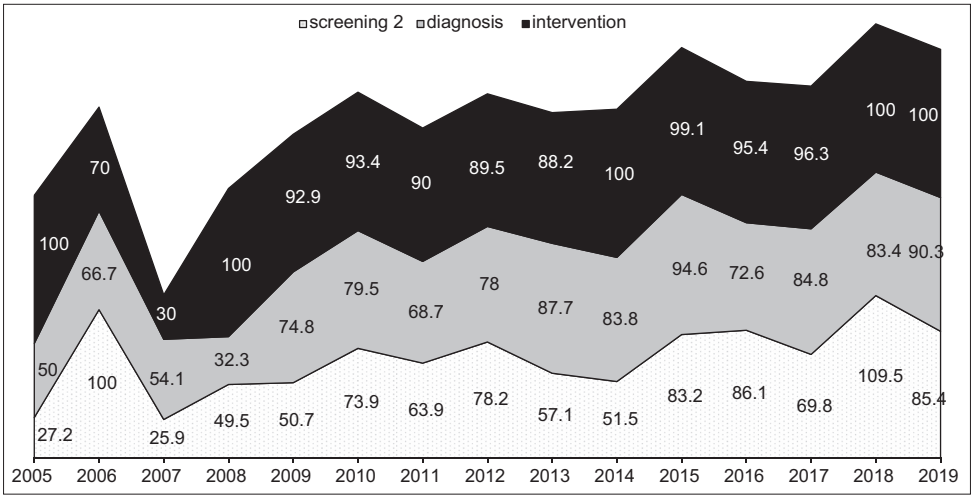

Figure 2: Percentage of participation in the second screening, diagnosis, and intervention in Khorasan Razavi province, Iran during 2005–2019

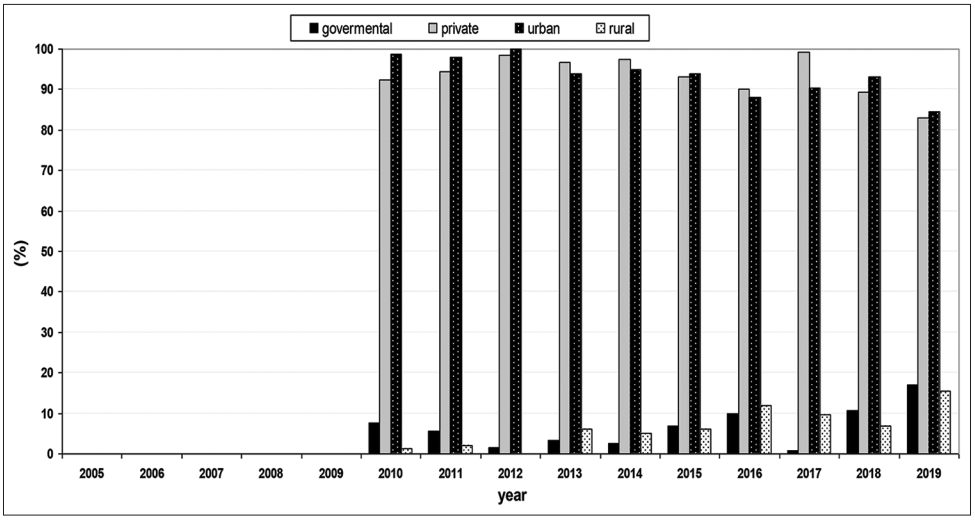

Figure 3: Percentage of screening process in governmental, private, urban, and rural centers in Khorasan Razavi province in the years 2005–2019

of language development and social integration.[16] According to the guidelines of the Joint Committee on Infant Hearing (JCIH), 1) screening should be performed in the first month of birth, 2) screening coverage rate should

Table 1: Type and severity of hearing loss in three categories of hearing loss: 1) mild to moderate, 2) moderately severe to severe, and 3) profound in Khorasan Razavi province, Iran from 2010 to 2019

Year	Diagnosed cases with hearing loss *n* (per 1000)				Level of hearing loss (%)			Type of hearing loss (%)	
	Mild to moderate	Moderately severe to severe	profound	Total	Mild to moderate	Moderately severe to severe	profound	Unilateral	Bilateral
2010	120 (1.6)	1 (0.01)	43 (0.57)	164 (2.19)	73.17%	0.61%	26.22%	17.09%	82.91%
2011	60 (0.65)	16 (0.17)	27 (0.29)	103 (1.1)	58.25%	15.53%	26.21%	29.13%	70.87%
2012	45 (0.47)	23 (0.24)	29 (0.3)	97 (1.01)	46.39%	23.71%	29.9%	22.68%	77.32%
2013	68 (0.68)	31 (0.31)	46 (0.46)	145 (1.44)	46.9%	21.38%	31.72%	12.93%	87.07%
2014	141 (1.23)	36 (0.31)	52 (0.45)	229 (1.99)	61.75%	15.72%	22.71%	20.96%	79.04%
2015	177 (1.53)	51 (0.44)	37 (0.32)	265 (2.29)	66.79%	19.25%	13.96%	47.55%	52.45%
2016	407 (3.23)	50 (0.4)	47 (0.37)	504 (4.00)	80.75%	9.29%	9.33%	33.99%	66.01%
2017	192 (1.57)	30 (0.24)	21 (0.17)	243 (1.98)	79.01%	12.35%	8.64%	4.53%	95.47%
2018	230 (1.76)	112 (0.86)	56 (0.43)	398 (3.04)	57.79%	28.14%	14.06%	35.71%	64.29%
2019	141 (1.25)	38 (0.34)	30 (0.27)	209 (1.85)	67.46%	18.18%	14.35%	35.24%	64.76%
Total	1581 (1.45)	388 (0.36)	388 (0.36)	2357 (2.17)	67.08%	16.46%	16.46%	28.51%	71.49%

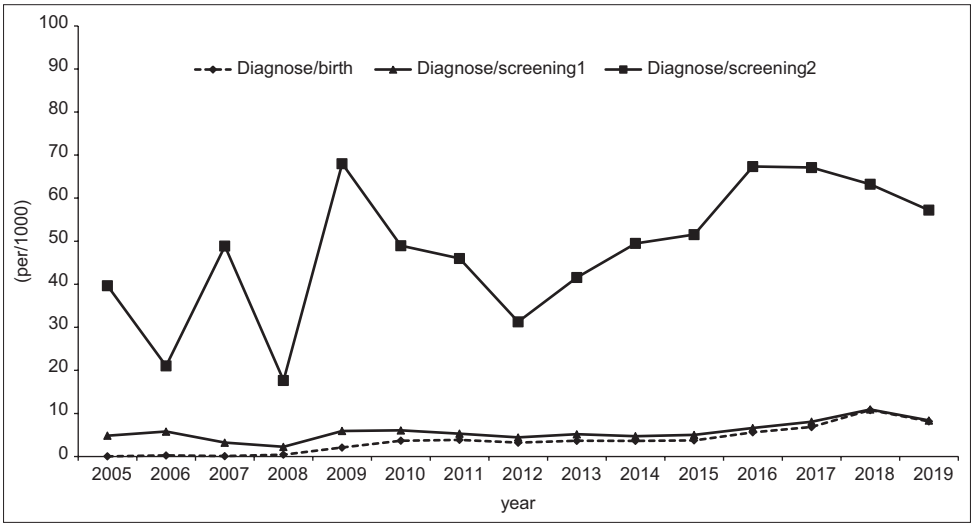

Figure 4: Ratio of cases diagnosed with hearing loss to live births and the number of cases screened in the first and second stages in Khorasan Razavi province, Iran during 2005–2019

be more than 95%, 3) screening referral rate should be below 4%, and 4) follow-up return rate for screening and diagnosis should be more than 95%.[12]

In the studied 15-year period, all the infants screened and born in the hospital underwent hearing screening in the first month of life. Screening coverage was not desirable in the early years; however, in recent years, it was greatly improved and reached the desired level. The referral rate from the screening stage to the diagnosis has been below 1% in all the years. On the one hand, this can indicate the good quality of the screening, but on the other hand, it may indicate the importance of the cases that did not come for their second stage of screening. It should be noted that tracking all infants with hearing loss can be difficult.[4] The patient return ratio indicates the efficiency of the patient follow-up system. Based on the results of previous studies, about one-third of infants who are screened at birth typically do not receive proper follow-up at regular intervals.[5] Nevertheless, the rate of follow-up and participation in Khorasan Razavi province has improved in recent years. The lowest participation was usually seen in the second phase of screening, while the best participation was observed in receiving interventions, and almost all infants diagnosed with hearing loss received the intervention.

In the studied area, OAE and AABR two-stage screening protocols were the most commonly used protocols. Although the pass result of each of the OAE and AABR tests is acceptable for neonatal hearing screening, the OAE and AABR two-stage screening methods reduce the fail rate and the number of neonates in need of follow-up.[5] The second hearing screening was performed in some centers using OAE. This screening was performed only on the group of well-born neonates, and all neonates in the NICU were referred to centers with AABR. Due to the low prevalence of auditory neuropathy in well-born infants, the

JCIH 2019 Declaration authorized re-screening with OAE for this group.[5]

Diagnosis

Based on the results of the present study, there are 2.17 cases of hearing impairment per 1000 infants. The prevalence of hearing loss according to WHO is 0.5–5 cases per 1000 births.[17] The number of cases is lower in some developed countries compared to developing countries; for example, in the United States, it is 1.7.[18] In the present study, most of the diagnosed infants had mild-to-moderate bilateral hearing loss. In other studies, hearing loss in two-thirds of cases has been bilateral as well.[17]

The EHDI aims to identify children with all levels of hearing loss; however, it can only detect hearing loss greater than 35 to 40 dB due to the limitation of screening equipment.[5] In the field of diagnostics, the current process of infant assessment in northeastern Iran has improved over the years. Furthermore, the percentage of people receiving diagnostic services has increased recently. However, there is a distance from the desired level. The follow-up problems exist in other countries as well; for instance, in the United States, only about 51% of newborns whose screening results have been positive were followed for diagnosis, and the procedure was performed for only 70% of them before they were three months old.[19]

Currently, most infant diagnostic evaluations are performed using TEOAE and air-conduction ABR tests with click stimulus. In some infants, high-frequency tympanometry, ABR with tone-burst stimulus, bone-conduction ABR, DPOAE, ASSR, and behavioral evaluation are used; however, this is not the case with all diagnosed infants. According to the global guidelines, proper battery testing, especially ABR with tone-burst stimulus, is necessary for the assessment of the hearing of infants,[20] and ABR with the tone-burst stimulus is the basis for the initial prescription of hearing aids for infants.[5]

Intervention

Early and effective intervention is very important for the achievement of the goals of EHDI, and even cases of unilateral hearing loss should receive intervention as soon as it is possible.[6] According to JCIH guidelines, assessment and intervention should be started as soon as possible when the infant is less than three and six months old, respectively.[12] In some countries, there are some problems in this regard and some intervention information is not properly collected.[21] Regarding the stages of diagnosis and intervention, the official recommendations in the northeastern of Iran are to complete the stages of diagnosis and intervention as soon as possible before the infant is three and six months old, respectively. Audiologists follow up on these steps and collect information about the performed interventions; however, it is not clear exactly at what age each infant receives the diagnosis and intervention. Some children receive their first rehabilitation services when they are more than two years old. This can be due to various reasons, such as financial problems, lack of rehabilitation centers and their inappropriate dispersion, and lack of awareness of the consequences of hearing loss and the importance of early intervention.

Various factors, such as lack of access to local centers, can reduce participation,[22] especially in rural areas.[23] Moreover, a lack of awareness of the consequences of hearing loss can have a significant impact on whether or not the infants receive interventions. Nevertheless, it should be noted that awareness of the effects of hearing loss does not necessarily mean being aware of the importance of early intervention. The level of awareness about these issues can be different and could be desirable[24] or moderate[25] in some areas.

Awareness of the consequences of hearing loss and the importance of early intervention can take time, and sometimes parents need time to learn about hearing loss and its effects.[13] Even in a well-run EHDI program, various factors, such as parental rejection of diagnostic evaluation results, lack of cooperation, and the presence of other illnesses, can delay the onset of intervention.[26]

The new goal of JCIH 2019[5] is to perform the screening, diagnosis, and intervention before the infants are one, two, and three months old, respectively. To achieve this goal, we must be able to follow the different stages of diagnosis and intervention. The presence of follow-up systems improves the effectiveness of the EHDI program[2] and the use of trained patient navigators can improve the follow-up process.[27] Currently, there is no specific follow-up system for medical referrals. Also, genetic testing and assessment of vision or other associated lesions for infants with hearing loss are rarely performed in groups. These are usually performed individually at the request of the family or physician.

According to the JCIH 2000,[12] 2007,[4] and 2019[5] guidelines, real ear measurement is recommended in the process of prescribing hearing aids for infants. During the studied 15-year-period in Khorasan Razavi province, in the hearing aid settings, the estimated mean values of real-ear-to-coupler difference (RECD) were used more than the individually measured RECD. Individually measured RECD is especially important for the verification of hearing aid settings in infants and leads to better results.[28,29] Hearing aids are regularly evaluated electro-acoustically, and the growth and development of the infant in various areas are monitored during hearing rehabilitation.

For the past 15 years, there have been restrictions on data collection. This means that no information was collected about the time of receiving diagnostic services and intervention, while related information was collected about the degree of hearing loss in a categorized manner.

Moreover, the decibel values of hearing loss were not recorded. There was also no complete information on prescribing hearing assistive technologies such as FM systems.

Conclusions

EHDI process in northeastern of Iran had good progress during studied period, especially in screening phase. However, better follow-up and expansion of diagnostic and intervention services are needed.

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Conflicts of interest

There are no conflicts of interest.

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