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STRATEGY AND EFFECTS OF EARLY INTERVENTION IN SURDOLOGY

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SUMMARY

Congenital hearing loss presents important handicap in development of a child because it considerably impacts, not only listening, but speech-language development, cognition and education as well. Hearing impairment is the most frequent congenital sensory defect that affects 1-3 in 1000 newborn babies. Regardless of dynamic development of genetics and other diagnostic procedures etiology of congenital hearing loss still remains unknown in 50% of cases. Early intervention in surdology tends to minimize consequences of congenital deafness. Surdologists insist on early intervention for congenital hearing loss considering the fact that early amplification and rehabilitation should be applied during first 3.5 years of life, while plasticity of central nervous system is maximal. During previous five-year period, from 2006 to 2010, the authors have observed very low rate of early intervention and therefore advocate the need for change of strategy in everyday clinical practice. Strategy for minimizing deleterious effects of congenital hearing loss is based on Early Hearing Detection and Intervention (EHDI) program which will be explained in details. The objective of the study is to evaluate actual strategy of early intervention in surdology. Effects of early intervention for congenital hearing loss in children treated in Audiology Rehabilitation Department of Clinic for ENT and HNS over a period 2006-2010 were analyzed.

Key words: deafness, hearing loss, early intervention, children

INTRODUCTION

Strategy for maximal diminishing consequences of congenital hearing loss is based on early intervention program (EHDI – Early Hearing Detection and Intervention) which consists of following elements: 1. Neonatal screening, immediately after birth up to one month, 2. Diagnosis confirmation up to 3 months of age and 3. Early intervention through amplification and choice of appropriate habilitation strategy, up to 6 months of age. That is the way to achieve the best boost throughout the period of maximal central nervous system plasticity in order to minimize deleterious effect of auditory deprivation. Early intervention is effective system which can prevent or diminish negative effect of congenital hearing loss on speech and language development in children. (Calderon, Naidu, 1999; Kennedy et al., 2006; Moeller, 2000). Congenital hearing loss is a major handicap because it affects not only hearing, but cognition and education as well. It is the most frequent congenital sensory defect affecting 1 to 3 ‰ of newborns (Finitzo, Albright, O'Neal, 1998; Van Naarden, Decoufle, Caldwell, 1999). Despite of progress in genetic testing and other diagnostic studies etiology in 50% of congenital hearing loss is still unknown (Mikić, 2006). The goal of early intervention

for congenital hearing loss is improvement of speech and language as well as overall communication capacity (Callow-Heusser, 2011). Intervention in first 3.5 years after birth enables the best results due to maximal plasticity of developing brain (Sharma et al., 2004). That is the rationale for early intervention for congenital hearing loss.

Introduction of neonatal hearing screening had decreased the age of diagnosis of congenital hearing loss considerably. Confirmation of diagnosis of congenital HL by the age of 3 months enables the start of intervention before 6 months of age, thus improving the outcomes. Early intervention with the use of hearing aids and /or cochlear implants improves auditory perception and speech-language development helps congenitally deaf kids to almost achieve milestones of their hearing peers (Mikić et al., 2011). Although neonatal hearing screening is the first step in diagnosis of congenital deafness it is necessary to follow-up all the babies who have failed NHS in order to diagnose those with hearing loss and commence surdological intervention in due time. Without the comprehensive procedure and follow-up it is impossible to achieve optimal results.

The objective of our study was a survey of current state in program of early surdological intervention for congenital hearing loss in Belgrade. Effects of early intervention during the period from 2010 till 2016 have been analyzed using patient files from Audiology Rehabilitation Department in Clinic for Otorhinolaryngology and Maxillo-facial Surgery of Clinical Center of Serbia.

History of early intervention for congenital deafness

Directors of Speech and Hearing Program in State Health and Welfare Agencies (DSHPSHWA) have decided to introduce national program of neonatal hearing screening in early eighties of XX century (Curry, Gaffney, 2010). Joint Committee on Infant Hearing (JCIH) has defined the goals in 1early detection of congenital HL in 1995.

Coverage of NHS in 46 states of North America, Europe, Asia, Middle East, Oceania and Africa during the period 2004-2006 has been 46% despite the recommendation of JCIH of more than 95% (Wolf et al., 2007). The data from 32 countries with local or regional UNHS revealed the coverage of 64%.

"The American Academy of Pediatrics (AAP, 2010) and the Joint Committee on Infant Hearing (JCIH, 2007) have recommended "1-3-6" benchmarks for follow up: (1) complete newborn hearing screening (NHS) before 1 month of age, (2) diagnose HL before 3 months, and (3) enrol those identified with HL in early intervention before 6 months of age (Holte et al., 2012). According to CDC (Centers for Disease Control and Prevention) in 2006, 91.2% of newborns were screened for hearing loss and 2.1 % did not pass the screening (Gaffney, Green & Gaffney, 2010). By 2009, the latest year for which data are available (U.S. Centers for Disease Control, 2011), national statistics improved: 98% of newborns were screened and 1.6% did not pass. (Holte et al., 2012). The major problem in USA is still very high rate of lost to follow-up after failed screening.

Neonatal hearing screening in Europe was implemented in Europe at about same time as in USA. Nowadays, the same timeline of EHDI is administered in Europe as in America: screening before 1 month, diagnosis before 3 months and intervention before 6 months of age. European consensus on neonatal hearing screening required a lot of logistics and legislation, which has been implemented as follows (Grandori, 2002):

- National Institutes of Health Consensus Statement 1993.
- EC-Biomedical & Health Program – Project on Otoacoustic Emissions (CA-OAE) 1993-95.
- World Health Organisation 1995.
- EC-Biomedical & Health Program – Project AHEAD 1996-99.
- European Consensus Development Conference 1998.
- American Association of Pediatrics Statement 1999.
- 1st International NHS Conference, Milan, Italy, 2000.

First decade of EHDI program implementation in Europe (2000-2010) has revealed a lot of factors and obstacles which have caused delays and setbacks. Major problem has been considerable percentage of children who were lost to follow-up (LTFU) after failed screening in maternity wards. Early intervention programs were compromised by concomitant medical conditions in hearing impaired babies, poor parental knowledge regarding hearing loss impact, accessibility of audiological and rehabilitation facilities, long waiting lists, etc. (Holte et al., 2012).

Table 1 *Implementation of NHS and EHDI in Europe in 2006*

EHDI %	Country
> 90%	Austria, Belgium, Croatia Denmark, UK Luxemburg, Netherlands, Poland
20% – 90%	Germany, Italy, Lithuania, Malta, Slovenia, Spain, Sweden, Switzerland, Cyprus
≤ 20%	Czech Republic, Estonia, Finland, Greece, Hungary, Latvia, Norway, Portugal, Romania Slovakia, Turkey

Early detection and rehabilitation of congenital hearing loss in Serbia started in Serbia in late sixties, but were widely implemented in eighties of XX century. Hospital based neonatal hearing screening first started in year 2000. in several maternity wards in Beograd, Novi Sad, Pančevo, Subotica, Sremska Mitrovica, Niš, Užice, but there is still no national UNHS program, despite numerous initiatives. Babies with risk factors for congenital hearing loss were usually referred for audiological assessment, whereas well babies did not have any hearing test. They were not referred for hearing testing unless they had speech delay, between ages 2 and 3 years.

EHDI (Early Hearing Detection and Intervention) in USA has shown progress from 32% of screened babies in 2000 to 95% u 2007, but percentage of LTFU is still extremely high (47%). Percentage of screened babies in EU in 2007 was 45%, with extremely low LTFU rate of 5%. The best national hearing screening program in our region is in Croatia with screening rate of 96% of all new born babies and excellent follow-up and early intervention system (Grandori, 2010).

Table 2 *EHDI protocol following NHS*

OAE screening in hospital				
Pass		Fail		
Risk factors	No risk	Repeat screening using OAE		
Rescreening in 6 months	No further screening	Pass		Fail
		RF	No RF	Rescreening in 3 weeks
		Rescreening in 6 months	No further screening	Audiological and surdological evaluation in 2 weeks

Table 2. shows contemporary protocol for hearing screening procedures. It is adjusted to EHDI principles defined by American Academy of Pediatrics in 2010. It covers the 1-3-6 rule for congenital HL, meaning that every baby should be screened before age of 1 month, diagnosed with hearing loss before age of 3 months and enrolled in early intervention before age of 6 months. Same protocol should be applied for acquired pre-lingual HL with timeline adjustment according to hearing impairment onset.

Indications for early intervention for congenital hearing loss

Hearing function is developing prenatally (Niparko, Tobey, Eisenberg, 2010). In 26th gestational week foetus reacts to both internal and external sounds. Listening to steady heart beat in utero is an early engram which is a base for subsequent speech development and rhythm of syllables. (Bojanin, 1986). Prenatal reaction to sound is atypical and primitive mostly expressed through acoustic-motor reflex. Listening in foetus is considerably different to that in older babies and infants due to immature auditory system (Warrier et al., 2004).

Hearing is a capability to receive, conduct and process acoustic signals to the brain and store them in the memory to increase knowledge. Development and interactive processes of auditory perception are induced through processing of auditory processing. Auditory perception is closely related to other systems and functions such as attention, memory, emotions and speech.

Learning process develops the ability to differentiate various auditory stimuli. Sensory stimulation leads to classification of the information in appropriate class or impression. Those classes are based on experience. Learning process is affected by development of perception. The process is not static, but could be modified through learning.

Cochlea is fully functional at birth (Kisilevsky, Hains, Jacquet, Granier-Deferre, Lecanuet, 2004). Auditory system is immature: the sound is perceived, but auditory processing is undeveloped. It takes years to achieve full maturation of auditory system. Latencies of cortically evoked potentials and P1 wave, generated in thalamic and cortical structures, are gradually decreasing over time (Sharma et al., 1997; Cunningham et al., 2000; Ponton et al., 2000). Wave P1 latency could be treated as index of maturity of auditory system and is especially useful for studies in auditory derived population (Ponton et al., 2000; Sharma et al., 2002).

The study conducted in Auditory Rehabilitation Department in Belgrade in 1999 has shown that age of diagnosis of congenital hearing loss is affected by the degree of hearing impairment, so that children with profound hearing loss (practical deafness) were diagnosed between ages 1 and 2 y, whereas mild to moderate hearing loss was usually discovered between ages 4.5 and 5.5 y (Ostojić, 1999). There is critical time frame for development of auditory as well as all other sensory functions. Numerous studies have shown the importance of early intervention in order to affect cortical reorganization and prevent cross modal plasticity. Central nervous system plasticity is maximal in the first three years of life. Early intervention is essential for prevention of irreversible changes in CNS induced by long-standing auditory deprivation. There are numerous studies regarding different organization of sensory cortex and takeover of the cortical regions by other sensory modalities such as vision and touch in case of

congenital deafness, the phenomenon known as cross modal plasticity (Lee et al., 2001; Neville, Bavelier, 2002; Roder et al., 2002). That could be the explanation for traditional belief that blind people have better hearing acuity, while deaf are more sensitive to visual stimuli. Sensitive period for therapeutic intervention is limited to the first seven years of life (Lee et al., 2001). The results of intervention for congenital deafness are the best before 3 years of life and some impact is achieved in the first 7 years, but in case of late intervention achievement is limited and full maturation of auditory function could never be fully reached.

According to AAP congenital hearing loss of various aetiology is found in 1–3 ‰ of newborns, whereas the percentage of hearing impaired babies is considerably higher in neonatal intensive care units (NICU) reaching 2–4%. In population of newborns with risk factors for hearing loss according to JCIH the rate of congenital hearing loss is 17 times higher than in well baby population according to FDA survey at the age 5-8 years.

Table 3 *Auditory maturation from birth to 24 months (Northern, Downs, 2002)*

Month	Sound dB (SPL)	Speech dB (HL)	Reaction
0 – 1.5	90 in noise 50 – 70 in quiet	40 – 60	Newborn awakes, gross motor reaction
1.5 – 4	50 – 60	45	Eye movements, head shake
4 – 7	40 – 50	20	Head turning, begins to listen
7 – 9	30 – 40	15	Direct horizontal localization
9 – 13	25 – 30	10	Direct localization downwards
13 – 16	25 – 30	5	Direct localization upwards
16 – 21	25 – 30	5	Direct localization sideways, downwards, upwards
21 – 24	25	5	Precise localization in all directions

Discrimination of speech in babies is based on prosody. Young babies can listen to any language with undivided attention. Babies are born with auditory capacity to learn any language. In two month olds they begin to prefer their native language. Ability to recognize contrast in foreign language decreases over time. Baby ignores acoustic details irrelevant for phonetic features of mother tongue. They tend to listen to phonetic details in speech perception rather than words. Babies could discriminate intonation, basic frequency, intensity and duration of acoustic signals at the age 1 to 4 months. Ability to spot fine prosodic information, metric features of speech, such as syllables and accent develops over time. Infants could discriminate emotional connotation of verbal stimuli making difference between angry and tender voice. Four month olds could make connection between auditory and visual presentation of speech. Speech understanding is based on link between auditory perception and visual and tactile perception (Nelson, 1995). Results of numerous studies have proved that early intervention for profound congenital HL (1-3-6 months) enables those children to achieve auditory capacity of their hearing peers by the age of 6 years (Yoshinaga-Itano et al., 1994; Yoshinaga-Itano, Mah-rya, 1998).

That implies the necessity of early surdological intervention. Period of intensive auditory development (perception, processing and language acquisition) is limited. Early intervention for congenital HL could minimize consequences of auditory deprivation. EHDI principles are based on central nervous system plasticity and reorganization

which is maximal in the first 3 years after birth. A lot of studies have shown that EHDI for congenital HL leads to speech and language development approaching the development curve for hearing children by the age of 4 years (Yoshinaga-Itano et al., 1998; Mikić, 2006; Ching et al., 2013).

Sample and methodology

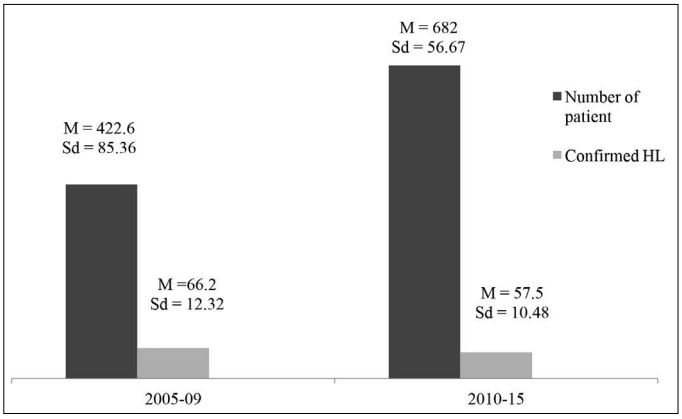
Maternity ward in Clinical Centre of Serbia applies two stage neonatal hearing screening using TEOAE. Babies who failed NHS twice are referred for further audiological testing in Audiology Rehabilitation Department. Test battery consists of Behavior Observation Audiometry (BOA), otoacoustic emissions (TEOAE and DPOAE), tympanometry, acoustic reflex measurements and Brainstem Evoked Response Audiometry (BERA). Audiological assessment is done by multidisciplinary team consisting of audiology physician, surdologist and psychologist. First assessment is done at the age of 1 month and repeated at 3 and 6 months in well babies. In babies with risk factors for HL according to JCIH follow-up period is extended to 12 months and in babies with family deafness to 3 years and more. The study has encompassed 331 baby who were referred for audiological assessment because of failed screening in the period 2010-2015.

RESULTS AND DISCUSSION

Table 4 *Incidence of hearing loss diagnosis among the patients assessed in Audiology Rehabilitation Department of Clinic for ENT and HNS, Clinical Centre of Serbia, in the period 2005-2015*

Year	Number of patients (n)	Confirmed HL (n, %)
2005.	327	80 24.4
2006.	348	69 19.8
2007.	423	74 17.5
2008.	510	49 9.6
2009.	505	59 11.7
2010.	738	61 8.3
2011.	720	76 10.5
2012.	690	53 7.7
2013.	586	51 8.7
2014.	646	58 9
2015.	712	46 6.5

The survey of medical records has revealed ever increasing number of patients referred for audiological assessment. Average number of patients per year has considerably increased in five year period 2010-2015 (682) as compared to previous five year period from 2005-2009 (422.6) The increase in number of patients referred for hearing tests could be explained by introduction of NHS in biggest maternity ward in Belgrade as well as the raised awareness of pediatricians about the importance of early diagnosis and intervention for congenital HL. The public awareness of hearing loss impact possibilities for problem solving has increased lately. Parents, doctors and teachers recognize the problem earlier and react promptly in case of suspected HL or risk factors. Outcomes in children with congenital HL who were involved in EHDI have proved that impact of congenital deafness could be minimized by early intervention. Study by Yoshinaga-Itano, (2003) has shown that children diagnosed with congenital HL before 6 months of age had significantly better vocabulary, general language abilities, speech intelligibility and phoneme repertoires, syntax as measured by mean length of utterance, social-emotional development, parental bonding, and parental grief resolution.



Graph 1 Average number of patients referred for audiology testing and confirmed HL in periods 2005-09 and 2010-15 y, in Audiology Rehabilitation Department

Data shown in *Graph 1* reflect increase of overall number of patients referred for audiological assessment whereas the number of confirmed hearing loss is stable. Incidence of congenital HL remains 1 – 3‰ of live births (WHO, 1995, 2010). The total number of patients with confirmed hearing loss per year in first period 2005-2009 is slightly higher (M=66.2, SD=12.32) than from 2010-2015 (M=57.5, SD=10.48). That could be caused by two possible reasons: slightly lower number of newborns and increased number of diagnostic centres for audiology.

Table 5 *Children without risk factors for hearing loss who failed NHS in Maternity ward of CCS in Belgrade from 2010-2015*

Audiological assessment	Age	N	∑
I	≤ 3 months	82	131
I	> 3 months	49	
II	≤ 6 months	31	36
II	> 6 months	5	
HL diagnosed	< 12 months	3	
Overall HL		4	

Table 6 *Children with risk factors for hearing loss who failed NHS in Maternity ward of CCS in Belgrade from 2010-2015*

Audiological assessment	Age	N	∑
I	≤ 3 months	112	200
I	> 3 months	88	
II	≤ 6 months	65	106
II	> 6 months	41	
III	< 12 months	64	
IV	< 24 months	47	
HL diagnosed < 12 months			
Sensorineural		29	44
Conductive		9	
ANSD*		6	

*ANSD(auditory neuropathy spectrum disorder)

Tables 5 and 6 display the incidence of HL in babies who failed NHS in Maternity ward of CCS in Belgrade with and without risk factors for hearing loss according to JCIH. Joint Committee for Infant Hearing had defined the list of ten risk factors which could possibly cause HL, such as prolonged NICU stay, low APGAR score (>5 days), low birth weight < 1500g, APGAR score 1-3, hyperbilirubinemia, mechanical ventilation > 5 days, intrauterine infection (TORCH), neonatal bacterial meningitis, cranio-facial malformations, ototoxic medication and familial deafness (American Academy of Pediatrics, 2007). Regardless the fact that risk factors for HL could not always be prevented or avoided, it is of utmost importance to register all babies with known risk factors and apply appropriate screening and diagnostic procedures through careful follow-up (Nikolić, Ostojić, 2016).

Table 5 depict the data of audiological assessment in babies without risk factors who failed initial NHS. Audiological assessment was done before the age of 3 months in 82 babies and in 49 babies after that age. Three babies were diagnosed before 12 months of age. Sensorineural hearing loss was diagnosed in 4 children (3%) in population of babies without risk factors who failed NHS.

Table 6 shows the data for babies with known risk factors for HL who failed NHS. First audiological assessment was done in 112 babies before 3 month of age and in 88 after that. Hearing loss was diagnosed before the age of 12 months in 44 babies. Sensorineural HL was diagnosed 29(14.5%) babies. There were also babies with conductive hearing loss (9) and auditory neuropathy spectrum disorder – ANSD (6). There is considerable prevalence of HL in group of babies with known risk factors. Average number of babies with congenital HL in general newborn population is 1.4 per babies screened (CDC, 2009; AAP 2007).

Early diagnosis and intervention for congenital HL have been implemented in Serbia for decades, but there is still no universal neonatal hearing screening or appropriate legislation for introducing national EHDI program. NHS has been implemented over last 12 years in several hospitals, so that percentage of screened newborns is estimated between 12-15%. Lack of national data base results in high percentage of lost to follow-up, so that the number of babies diagnosed with HL in first 12 months of age is still not satisfactory. Successful EHDI program requires legislation, accessible diagnostic equipment, affordable hearing aids and cochlear implants, steady financial resources

and well educated and equipped multidisciplinary teams for early identification of HL and intervention. Public awareness and information on congenital HL and importance of early diagnosis and treatment is still insufficient. It has been improved in the last five years, but it is not enough. Coordination of neonatal teams involved in primary NHS and secondary and tertiary audiological facilities is essential. Central national database is required in order to obtain proper follow-up of babies who failed initial screening.

CONCLUSION

The study has analyzed the NHS data from a biggest maternity hospital in Belgrade (GAC CCS) and subsequent audiological assessment in babies who failed NHS in Audiological Rehabilitation Department of CCS in Belgrade from 2010-2015. The results have shown considerable increase in number of patients who were audiological assessment (M=682), in comparison with previous five year period from 2005-2009 (M=422.6). It could be attributed to wider implementation of NHS as well as to increased public awareness of importance and consequences of congenital hearing loss. The results have shown considerably higher prevalence of diagnosed HL in children with known risk factors (29 or 14.5%) as compared to babies without risk factors (4 or 3%). All of those babies were referred for further audiological assessment due to failed NHS. It is in accordance with data that congenital hearing loss is found in 1.4 per 1000 babies screened (CDC, 2009; AAP 2007).

National EHDI strategy in Serbia is compromised at the moment because of lack of universal hearing screening. There is ongoing public initiative for obtaining OAE screeners for all 56 maternity wards in Serbia organized by Ministry of Health and media called “Heart for children”. National program requires more than just screeners. Future parents should be well informed on importance of hearing screening and early detection of congenital hearing loss. National data base and IT network is necessary in order to provide proper follow-up of babies screened. Good network of diagnostic audiology facilities as well as rehabilitation units is needed for continuous EHDI program. It starts on the second day after birth with OAE screening and continues afterwards in case of failed screening according to EHDI protocol and results of audiological assessment and potential risk factors for HL (Table 2).

Effects of early intervention for congenital HL are demonstrated in speech-language outcomes at the age of 5 years comparable with those of hearing peers. EHDI program has decreased age of diagnosis of congenital HL from 30 months before it has been implemented to 3-6 months nowadays.

Multidisciplinary team of Audiology Rehabilitation Department of Clinic for EN and HNS of Clinical Center of Serbia has been dedicated to early detection and rehabilitation of children congenital hearing loss and/or complex needs for decades. The results were presented in several international and national projects as well as on numerous international conferences, published in international and national journals and books. The achievements of our team were highly regarded both abroad and in Serbia. Principles of early diagnosis and treatment of congenital HL have been established in 1967 through international collaboration with experts from USA, and developed

ever since. Hospital based NHS introduced in 2000 has improved early diagnosis and introduction of digital hearing aids and cochlear implantation for profound hearing loss in 2003 has improved rehabilitation options and outcomes. Audiological assessment is done periodically every 3 to 6 months in order to monitor hearing thre sholds and maturation. Rehabilitation is tailored to the child abilities and progress and is provided 5 days a week, twice a day for older children. Rehabilitation program for each child is individually designed and progress is monitored and discussed on regular team meetings. Percentage of mainstreamed children with HL is steadily increasing. Inclusive system of education of deaf children in mainstream schools is still to be improved providing individual curriculum and education assistant in order to stimulate maximal achievement for each child with hearing loss.

Families of deaf children have some financial support through Ministry of health and Ministry of Social Affairs, but it is usually not enough to cover real cost of rehabilitation for years. Accessibility to hearing aids and cochlear implants is relatively good but it should be even better in future.

Continuous education of all professionals on the diagnostic and rehabilitation team is mandatory as well as improvement of technical support for diagnostic and rehabilitative equipment. Strategy for early detection and intervention for congenital hearing loss should be national priority because the results of early treated hearing loss were proved to be cost effective for the society and last, but not least, a huge improvement in quality of life of a deaf child and his family.

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