





Summary: Hearing Screening

Estonia

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1. Glossary of Terms: Hearing Screening

Abnormal test result	A test result where a normal "pass" response could not be detected under good conditions. The result on screening equipment may indicate "no response," "fail," or "refer."			
Attendance rate	The proportion of all those <u>invited for screening</u> that are <u>tested and receive</u> <u>a result,</u> • <u>Invited for screening</u> includes all those that are offered the screening test. • <u>Tested and receive a result</u> could be a "pass" or "fail". Attendance rate provides information on the willingness of families to participate in screening.			
Attendance rate in first year of life	See definition of Attendance rate. The calculation cut-off is after <u>one year of life</u> .			
Compliance with	The percentage of those who are <u>referred from screening</u> to a diagnostic assessment that actually <u>attend</u> the first diagnostic assessment.			
referral (percentage)	Percentage of compliance provides information on the willingness of families to attend the diagnostic assessment after referral from screening.			
Coverage	 The proportion of those eligible for screening that are tested and receive a result within a specific time. Eligible for screening includes those within the population that are covered under the screening or health care program. Tested and receive a result could be a "pass" or "refer to diagnostic assessment". Specific time can be defined, such as 1 month after birth, 3 months after birth, etc. Coverage provides information on the overall effectiveness and timeliness of a complete screening programme. Factors such as being offered screening, willingness to participate, missed screening, ability to complete the screen, and ability to document the screening results will influence the coverage. 			
Coverage in first year of life	See definition of Coverage. The <u>specific time</u> is pre-defined as within the first year of life. In other words, the coverage is the proportion of those eligible for screening that complete the screening sequence to a final result within the first year of life.			
False negatives	The percentage of <u>infants/children with a hearing loss</u> (defined by the target condition) that <u>receive a result of "pass"</u> during screening.			



	Example: If 100 infants with hearing loss are screened, and 1 infant passes the screening, the percentage of false negatives is 1%.			
False positives	The percentage of <u>infants/children</u> with <u>normal hearing</u> that <u>receive a result of "fail"</u> from the final screening test. Example: If 100 infants with normal hearing are screened, and 3 infants fail the screening and are referred for diagnostic assessment, the percentage of false positives is 3%.			
Guidelines	Recommendations or instructions provided by an authoritative body on the practice of screening in the country or region.			
Hearing screening professional	A person qualified to perform hearing screening, according to the practice in your country or region.			
Inconclusive test result	A test result where a normal "pass" response could not be detected due to poor test conditions.			
Invited for screening	Offered screening.			
Outcome of hearing screening	An indication of the effectiveness or performance of screening, such as a measurement of coverage rate, referral rate, number of infants detected, etc.			
Permanent hearing loss	A hearing impairment that is <i>not</i> due to a temporary or transient condition such as middle ear fluid. Permanent hearing loss can be either sensorineural or permanent conductive.			
Positive predictive value	The percentage of infants/children referred from screening who have a confirmed hearing loss , as described by your protocol or guideline and indicated in the Target Condition (see definition). For example, if 100 babies are referred from screening for diagnostic assessment and 90 have normal hearing while 10 have a confirmed hearing loss, the positive predictive value would be 10%.			
Preschool or (pre)school children	All children between 3-6 years of age.			
Preschool or (pre)school screening	Screening that takes place during the time children are between 3-6 years of age. This refers to <i>any</i> hearing screening during this age. The location of the screening is irrelevant to the definition.			



Prevalence	The number or percentage of individuals with a specific disease or condition. Prevalence can either be expressed as a percentage, proportion, or as the value per 1000 individuals within the same demographic.				
Programme	An organized system for screening, which could be based nationally, regionally or locally.				
Protocol	Documented procedure or sequence for screening, which could include which tests are performed, when tests are performed, procedures for passing and referring, and so forth.				
Quality assurance	A method for checking and ensuring that screening is functioning adequately and meeting set goals and benchmarks.				
Referral criteria	A pre-determined cut-off boundary for when an infant/child should be retested or seen for a diagnostic assessment.				
	For example, referral criteria may be "no response" at 35 dB nHL.				
Risk babies / Babies	All infants that are considered to be at-risk or have risk factors for hearing loss according to the screening programme.				
at-risk	Two common risk factors are admission to the neonatal-intensive care unit (NICU) or born prematurely. However, other risk factors for hearing loss may also be indicated in the screening programme.				
	The percentage of infants/children with hearing loss that are identified via the screening program.				
Sensitivity	For example, if 100 babies with hearing loss are tested, and 98 of these babies are referred for diagnostic assessment while 2 pass the screening, the sensitivity is 98%.				
	The percentage of infants/children with normal hearing that pass the screening.				
Specificity	For example, if 100 babies with normal hearing are tested, and 10 of these babies are referred for diagnostic assessment and 90 pass the screening, the specificity is 90%.				
Target condition	 The hearing loss condition you are aiming to detect via your screening programme. This includes: The <u>laterality of the condition</u>, whether the program aims to detect both unilateral and bilateral hearing loss or just bilateral hearing loss. The <u>severity of the condition</u>, whether the program aims to detect hearing loss ≥ 30 dB HL, ≥ 35 dB HL, ≥ 40 dB HL or ≥ 45 dB HL 				
Well, healthy babies	Infants who are <i>not</i> admitted into the NICU or born prematurely. Well, healthy babies may or may not have additional risk factors for hearing loss, according to the procedures indicated in the specific screening programme.				



2. Abbreviations

ABR – auditory brainstem response

aABR – automatic auditory brainstem response

ANSD – auditory neuropathy spectrum disorder

ASSR – auditory steady-state response

CI – cochlear implant

CMV - cytomegalovirus

dB HL – decibel hearing level

dB nHL - decibel normalized hearing level

dB SNR - decibel signal-to-noise ratio

DPOAE – distortion product otoacoustic emissions

HA – hearing aid

NICU – neonatal intensive care unit

OAE – otoacoustic emissions

TEOAE – transient-evoked otoacoustic emissions



3. Background

In Estonia, hearing screening is performed nationally and also organized nationally. The following report contains information with regards to hearing screening across the <u>entire country of Estonia</u>.

3.1. General

The country of Estonia has a total area of 43 432 km² and a population of 1 315 635 as of January 2017 (Statistics Estonia, 2018). In Estonia, each birth is registered through the Estonian Birth Registry. The number of live births in Estonia in 2016 was either 13 861 as cited by the National Institute for Health Development Database (National Institute for Health Development, 2018) sourced from the Estonian Birth Registry, or 14 053 as cited by Statistics Estonia (2018).

The World Bank income classification categorizes Estonia as a high-income country (The World Bank, 2018). The gross domestic product (GDP) was €16 035 per capita per year in 2016 (Statistics Estonia, 2018).

From the World Health Organization (WHO) Global Health Expenditure Database, health expenditure in Estonia in 2015 was 1 112 USD or €952 per capita (World Health Organization, 2018).

Infant mortality rate in the country of Estonia was 2.5 and 2.3 per live 1000 births in 2015 and 2016, respectively (National Institute for Health Development, 2018; United Nations Statistics Division, 2016)

3.2. Neonatal hearing screening

In the country of Estonia, neonatal hearing screening is conducted universally, with all babies in the country having access to hearing screening, though screening is not obligatory for parents. The universal program for well and at-risk babies was first implemented in 2004, and by 2014, neonatal hearing screening was implemented across the country.

Neonatal hearing screening is now embedded in the Preventive Child Health Care screening system in combination with phenylketonuria (PKU) and congenital hypothyroidism (KsHT) screening. Prior to 2015, neonatal hearing screening was organized as an independent programme; however, since 2015, it became financed together as part of general maternity care. Screening is funded through Estonian Health Insurance Fund. which is the case for both at-risk and well-baby screening.

In Estonia, the same hearing screening protocol is followed across the country.

3.3. Preschool hearing screening

In Estonia, there is no regulated preschool hearing screening programme. Family doctors perform a whisper test as part of general preventative testing when children are 6-8 years of age.

It is unknown when doctors began performing the whisper test as part of pre-school control testing; but this type of testing has been in effect since at least 2009. Hearing screening, as part of the general preventive programme, is funded by Estonian Health Insurance.

The whisper test is performed across the country.



4. Guidelines & Quality Control

National guidelines and protocol for neonatal hearing screening were originally published together in 2008. (Kruustük & Luht, 2008).

The content of the original programme was decided on by a project steering group, in cooperation with the University of Manchester. The hearing screening programme was initially implemented by the Estonian Health Insurance Fund in collaboration with Tartu University Hospital. The Estonian Ear, Nose and Throat and Head and Neck Surgeons Society had initial responsibility in managing the programme with leadership from Katrin Kruustük and Liina Luht.

The protocols documented in the screening guidelines have not changed since implementation. However, since 2015, hearing screening has become embedded into the maternity/newborn medical services, and there are no future plans for revision of the programme.

Quality assurance of hearing screening programmes is not imposed by the government. Prior to 2015, hearing screening was managed independently by the Estonian Ear, Nose and Throat and Head and Neck Surgeons Society. During this period data were collected manually.

Since the integration of newborn hearing screening in general maternal and newborn care, quality assurance, data collection, and outcome monitoring are no longer performed. Data are collected on the medical claims made to the Estonian Health Insurance Fund. Because information is available regarding the number of claims filed, the number of infants screened can be estimated. Furthermore, the type of test performed and basic diagnosis by the International Statistical Classification of Diseases and Related Health Problems can be estimated. However, data regarding screening results and follow-up are not available.

Annual reports were available internally from 2004 to 2014: however, some data were missing during these reviews. An audit was published in 2013 with data reported from 2004 to 2012. Since 2014, annual reports are not available.

It is unknown whether research has been performed on hearing screening in Estonia apart from auditing prior to 2015. Research has not been performed since 2015.



5. Process: Screening, Diagnosis, Intervention

5.1. Neonatal hearing screening

Well-babies are screened in the maternity hospital, and at-risk babies are screened in the hospital, either in the maternity ward, NICU or outpatient centre. Well-baby families and families of infants at-risk are invited for screening directly in person when nurses take consent from the parents to perform screening. It is roughly estimated that 99.7% of births take place in the maternity hospital, where the average length of stay after delivery is 3 days. Only around 0.3% of births take place at home.

There are no set criteria for when hearing screening should be completed. The ultimate goal is the intervention is provided by 6 months of age, so the diagnostic assessment is ideally completed by 3 months and screening by 2-3 months of age.

The target condition for screening for well- and at-risk babies is bilateral or unilateral hearing loss of ≥ 40 dB HL.

There are no data available on the percentage of infants considered to be at risk. At-risk infants are defined in protocol as those born prematurely and with birth weight <1500 g, hyperbilirubinemia, perinatal infections, bacterial meningitis, NICU admissions, craniofacial abnormalities, administration of ototoxic drugs, or family history of congenital hearing loss.

The prevalence of CMV or meningitis in Estonia is unknown.

Infants with various risk factors have unique schedules for screening and surveillance, depending on the specific risk factor presented. Infants at-risk undergo OAE and aABR screening similar to well infants without risk factors but are also followed-up at a later age according to a specific schedule.

5.2. Neonatal diagnostic assessment

The diagnostic assessment tests performed after neonatal hearing screening referral is an ASSR. As indicated diagnostic testing should be performed by 3 months of age for well infants. There is no set benchmark age for diagnostic evaluation among all infants at risk, but for each risk factor, diagnostic follow-up and surveillance ages are independently indicated in protocol.

5.3. Preschool hearing screening

The whisper test is performed in primary health care centres by family doctors. Families are invited to participate via a phone call by the doctor or nurse, or are otherwise informed by the schools to participate.

There is no target condition for the whisper test.

5.4. Intervention approach

In Estonia, treatment options available include hearing aids, bone conductive devices, cochlear implants, FM-systems and CROS systems. The Estonian Health Insurance Fund has also financed auditory brainstem implants operated abroad. Infants are fitted with hearing aids from 6 months of age and cochlear implants from 12 months of age.

The official hearing aid fitting criteria as stipulated by the government in Estonia is a hearing loss of 30 dB HL, though no further information is provided by the government in terms of frequency or



laterality. Professionals choose to fit unilateral hearing loss with amplification on a case-by-case basis, depending on the needs of the child.



6. Protocols

Hearing screening protocols are described for neonatal hearing screening (well and at-risk) as well as for preschool hearing screening when applicable.

- The <u>Test</u> performed is the screening technique used
- The Age of the child is indicated in hours, days, months or years
- <u>Referral criteria</u> may be the lack of an OAE response at specified frequencies, a response-waveform repeatability constant, the absence of an aABR response at a specified intensity, or an absent behavioural response at a specified intensity. Referral criteria may be defined within a protocol or limited based on the device used.
- The Device is the screening device used.
- <u>Unilateral Referrals</u> indicates whether children are referred if only one ear fails screening.
- The <u>Location</u> is where the screening takes place

6.1. Neonatal hearing screening (well)

The neonatal hearing screening protocol in Estonia is described below. The protocol is 3 steps, including two OAEs and one aABR. However, there is some variability in the sequence of the OAE screens. Some hospitals may perform two OAEs with the first few days of birth before discharge from the maternity hospital. If the second OAE fails, a rescreen will occur at 2 weeks of age. Other hospitals will perform only one OAE before discharge and then rescreen at 2-4 weeks of age. The aABR takes place at 2-3 months of age before referral to diagnostic assessment.

Table 1: Screening process for well babies in Estonia (Kruustük & Luht, 2008).

		Referral		Unilateral	
Test	Age	Criteria	Device	Referrals?	Location
OAE1*	24-72 hours	3 dB SNR at 3	Interacoustics	Yes	Maternity hospital
OAE2	2-4 weeks	frequencies		Yes	Maternity hospital
aABR	2-3 months	40 dB nHL		Yes	Tartu University Clinic or
					East Tallinn Central Hospital

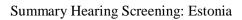
^{*}OAE1 may be performed once or twice before discharge, depending on the hospital. Some hospitals will only perform OAE1 once before discharge, and rescreen 2-4 weeks later. Other hospitals will repeat OAE1 before discharge. If the infant still does not pass, they will rescreen 2 weeks later before referring for aABR at 2-3 months.

6.2. Neonatal hearing screening (at-risk)

As indicted, screening at-risk infants follows the same protocol as well infants, with the exception that infants with risk factors that pass the screening are followed-up at a time that is specific to their risk indicator. For example, infants that are admitted into the NICU are re-examined at 8 months of age with OAE, ABR and VRA, then examined again at 18 and 36 months of age. Infants with a family history of hearing loss are re-tested with OAEs every 6 months from birth to age 18 months and once per year until age 7. A specific schedule is indicated independently for each risk indicator (Kruustük & Luht, 2008).

6.3. Preschool hearing screening

Table 2: Screening process for 6-8-year-old children in Estonia.





Test	Age	Referral criteria	Location
Whisper test	6-8 years	None (physician's discretion)	Health Care Centre



7. Professionals

7.1. Neonatal hearing screening (well)

Screening for well babies is performed by nurses and midwifes.

During the neonatal hearing screening programme prior to 2015, official training was provided and required for any nurse or midwives to perform neonatal hearing screening. Training involved both theoretical and practical components. The practical component included role playing and professional psychologists were involved in teaching how to deliver information to parents. Written course content was also provided to nurses and midwives who participated in the training. Official neonatal hearing screening training is no longer offered.

7.2. Neonatal hearing screening (at-risk)

Screening for at-risk infants is performed by trained intensive care nurses. See section 7.1 for information on training.

7.3. Preschool hearing screening

The whisper test is performed by family physicians.



8. Results: Neonatal Hearing Screening

8.1. Coverage and attendance rates

Calculated by the number medical claims filed to the Estonian Health Insurance Fund, a total of 8837 infants are estimated to have been screened in 2016. The number of live births registered in 2016 was 14 053, and therefore, the coverage rate in Estonia can be estimated to be 63% in 2016; however, this number is inexact. The count of 8837 infants is calculated by summing the number of claims made for OAE testing in a maternity hospital to the mother's ID. It cannot be directly assumed that this is the total number of infants screened.

As indicated, prior to 2015, neonatal hearing screening in Estonia was managed independently by the Estonian Ear, Nose and Throat and Head and Neck Surgeons Society. In 2013, an audit report showed that the coverage rate was 93, 91, and 98% for 2010, 2011, and 2012, respectively (Paat-Ahi, Laarmann, & Sikkut, 2013). At that point, one hospital in Estonia still did not have neonatal screening fully implemented. Furthermore, the number of infants screened that were born at home were not documented.

The number of infants that missed being *offered* screening is not specified, and therefore, attendance rate is not known. However, in the 2013 audit report, the number of refusals per year were recorded to be less than 5 cases per year (Paat-Ahi, Laarmann, & Sikkut, 2013). Data are not available since 2013.

The percentage of infants that attend OAE2 out of those that are referred from OAE1 is not available, nor is the percentage of infants that attend the aABR out of those that are referred from OAE2.

8.2. Referral rates

Referral rates for each stage of the screening process are unavailable.

Based on data derived from codes filed in the Estonian Health Insurance Fund database in 2014, 6% of infants have a claim filed for "brain bioelectric potential. This could be the percentage of infants screened with aABR; however, it is not indicated whether these are only infants that are referred from screening or whether this figure also includes those infants who pass the screening but are followed-up for monitoring of delayed-onset hearing loss due to the presence of risk indicators. In these cases, aABR may also be performed. This claim may also be when an ASSR or neurological ABR is performed. Sometimes the same code is used for adults with non-organic hearing loss. Therefore, the percentage may be even lower.

Referral rate was also not indicated in the 2013 audit report, but instead false positive rates were published. These are presented below in section 8.6. and show an average rate of 5 to 7%, with variation from year to year and across maternity hospitals (Paat-Ahi, Laarmann, & Sikkut, 2013). It is not indicated at which stage in the screening process is applicable to false positive rate of 5-7% (i.e., OAE1, OAE2, or aABR). Therefore, an estimation of a final referral rate is not possible.

8.3. Diagnostic assessment attendance

The compliance rate to diagnostic assessment is unknown.

8.4. Prevalence / Diagnosis

The prevalence of permanent hearing loss among neonates in Estonia can be estimated in several ways. First, in the 2013 audit report, it was calculated that one case of hearing loss was be detected in

about 700 to 800 screened infants from the years 2008 to 2011 (Paat-Ahi, Sikkut, & Laarmann, 2014; Paat-Ahi, Laarmann, & Sikkut, 2013). This would amount to an overall prevalence rate of 1.25 to 1.43 per 1000 neonates.

Second, the Estonian Health Insurance Fund Database indicates that 12 infants were detected with bilateral sensorineural hearing loss in 2016. Out of a total screening coverage of 8837 infants, this would result in a calculated prevalence of 1.36 per 1000 neonates. The Estonian Health Insurance Fund Database also indicates that 3 children were detected with unilateral sensorineural or mixed hearing loss in 2016. Out of a total estimated screening coverage of 8837 infants, this would result in a prevalence of 0.34 per 1000 neonates. These figures are not exact, as they were calculated from insurance claims that cited a diagnosis code of bilateral sensorineural hearing loss, or unilateral sensorineural and mixed hearing loss.

Prevalence values of permanent childhood hearing loss in Estonia prior to the implementation of neonatal hearing screening are presented in Table 3 according to a study by Uus and Davis (2000). These data were collected among children ages 7 to 13.

Table 3: Prevalence of permanent hearing loss among children in Estonia without being screened (Uus & Davis, 2000).

	Bilateral			Unilateral	
	\geq 40 dB HL	\geq 70 dB HL	\geq 95 dB HL	\geq 40 dB HL	\geq 80 dB HL
Prevalence per 1000 (Uus & Davis, 2000)	1.72	0.98	0.46		

Data on the prevalence of bilateral auditory neuropathy are unavailable in Estonia.

8.5. Treatment success

In Estonia, it is unknown how many infants with neonatal hearing impairment are fitted with hearing aids per year. In the 2013 audit report, it is indicated that 4 children were implanted with cochlear implants each year from 2010 to 2012. More recent figures are not provided.

8.6. Screening evaluation

Data are unavailable regarding screening evaluation in Estonia since the responsibility of management of the neonatal hearing screening programme was transferred. Prior to 2014, the false positive rate ranged from about 5 to 7% from 2008 to 2012, according to the 2013 audit report (Paat-Ahi, Laarmann, & Sikkut, 2013). In 2006 and 2007, the false positive rate was higher at around 9 to 9.5%. The 2013 audit report noted considerable variation between hospitals, ranging from 2% to 27% false positive rates (Paat-Ahi, Laarmann, & Sikkut, 2013).



9. Results: Preschool Hearing Screening

9.1. Coverage and attendance rates

In 2016, the Estonian Health Insurance Fund received claims for the general preventive testing of 16 313 children ages 6-8 years old. However, it is difficult to determine exact coverage across an age range. Furthermore, this figure is for general preventive testing within an age range, and not specifically for the whisper test that is generally performed prior to school entry.

9.2. Referral rates

Data are unavailable.

9.3. Diagnostic assessment attendance

Data are unavailable.

9.4. Screening evaluation

Data are unavailable.



10. Costs: Neonatal Hearing Screening

Neonatal hearing screening in Estonia is free of charge for parents. There is no financial reward when parents attend hearing screening nor is there a penalty for non-attendance. Participation in screening is not obligatory.

There has not been a cost effectiveness analysis completed in Estonia; however, the 2013 audit included an economic analysis of the costs of screening and costs of detecting one hearing loss case (Paat-Ahi, Laarmann, & Sikkut, 2013). The audit highlights the issues relative to modeling cost-effectiveness, namely that long-term effects of neonatal hearing screening are unknown and that a cost-effectiveness model requires high quality input data, such as screening participation, even detection, callback and forwarding rates, etc.

10.1. Screening costs

In Estonia, the total cost of hearing screening is \in 18.85 per child or \in 216 105 annually. For at-risk infants, the total cost of screening is \in 10 243.57, which amounts to is \in 75.30 per child covering 136 infants. It is assumed these costs are separate; however, this is not explicitly stated in the database. These data are based on the number of services claimed in the Estonian Health Insurance Fund; however, the prices of these services are under continuous revision and elevation.

The 2013 audit indicates that the cost for detecting one hearing loss cases was €16,063 in 2011. This figure ranged from €10,820 to €18,000 from 2007 to 2011 (Paat-Ahi, Laarmann, & Sikkut, 2013).

10.2. Equipment costs

Information on equipment costs or associated costs to maintain equipment is unavailable.

10.3. Staff costs

The salary of a nurse or midwife performing hearing screening is €12 156 per year or €6.03 per hour (National Agreement, 2017-04-01, *source unverified*). One factor that influences staff costs is the price of the outpatient visit, which becomes more expensive each year. It is roughly estimated that there are approximately 15 hearing screening professionals per 1 million people in Estonia. The cost of training hearing screening professionals is not indicated, as it is free of charge for the students.

10.4. Diagnostic costs

The cost for a diagnostic assessment is as follows: $\in 9$ for tympanometry, $\in 27$ for play audiometry or VRA, $\in 56$ for ABR/ASSR, $\in 11$ for pure-tone audiometry, and $\in 10$ for speech-audiometry (*source unknown*).

10.5. Amplification costs

In Estonia, all children are treated for hearing loss, except when children of deaf parents refuse cochlear implants.

The cost of each hearing aid is around €440 to €490, each earmold costs €42, and each specialist appointment for reprogramming costs around €56. Children in their first year of intervention typical require around 4-5 appointments per year. Earmolds may be replaced up to 3-4 times per year, depending on the age of the child. Older children require replacement 2-3 times per year.



The initial cost of fitting a cochlear implant is \in 21 621, which includes both the implant and speech-processor as well as the cost of the operation. The treatment costs for the first year of intervention is \in 6310, for the second year of intervention is \in 2932, for the third year of intervention is \in 1466 and every other year the cost is \in 244. Intervention costs include only re-programming of the CI, and do not include diagnostic testing or speech-language therapy (*source unknown*).

10.6. Social costs

In Estonia, there are 4 special education schools for the deaf, with a total of 79 children (ages 6-17) enrolled in the school. The cost per child in a special school for the deaf varies from school to school, ranging from $\[\in \] 2985$ to $\[\in \] 8291$ per student.

Special support is also available for children with hearing loss attending mainstream schools. The cost per child (including normal hearing) in a mainstream school is \in 3306, amounting to a total cost of \in 460 511 336 (*source unknown*).



11. Costs: Preschool Hearing Screening

11.1. Screening costs

The cost of the whisper test is embedded to the total costs for general preventive testing and cannot be indicated separately.

11.2. Equipment costs

Information not indicated.

11.3. Staff costs

Information not indicated.



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