Project Number: 733352

Project Acronym: EUSCREEN

Project title: Implementation of cost-optimized childhood vision and hearing screening programmes in middle-income countries in Europe

Periodic Technical Report

Part B

Period covered by the report: from January 1st, 2017 to June 30th, 2018

Periodic report: 1st

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History of changes

HISTORY OF CHANGES		
VERSION	PUBLICATION DATE	CHANGE
1.0	August 30th, 2018	Initial version

1. Explanation of the work carried out by the beneficiaries and Overview of the progress

1.1 Objectives

The following objectives are described in section 1.1 of the Description of Action:

- Objective 1: Screening experts from all 41 EU-countries will report on demography, circumstances for screening, existing screening programmes and health systems, uptake, screening tests, diagnostics, treatment options, envisaged health benefits, societal costs and adverse effects in their country.
- Objective 2: From these data and from the literature, the current provision of childhood screening, the types of screening programmes used and the key features of vision and hearing screening programmes will be identified across Europe.
- Objective 3: A decision-analytic, cost-effectiveness modelling framework of repeated screening will be prepared and populated with the reported data and with data from the literature.
- Objective 4: Current VAHSPs will be evaluated for their impact on health outcomes, cost-effectiveness and compliance with WHO-criteria for screening.
- Objective 5: Two model-developed VAHSPs will be tested in two large-scale implementation studies: In the county of Cluj in Romania for vision screening, and in three counties in Albania for hearing screening.
- Objective 6: A strategy for implementation will be developed from detailed tracking of the implementation studies, from identified requirements, facilitators and barriers, and from good-practice guidelines for existing VAHSPs.
- Objective 7: The decision-analytic modelling framework and the strategy for implementation will be packed into a transferable TOOLKIT that will assist healthcare providers and policy makers in Europe and beyond in their decisions about introduction or modification of VAHSPs in their country.

The work carried out by the consortium during the reporting period towards the achievement of the project objectives is described in section 1.2 of this report.

1.2 Explanation of the work carried per work package

1.2.1 Work package 1: Project management

Task 1.1 Coordination and monitoring of the EUSCREEN research programme

Since the start of the EUSCREEN project the Erasmus MC has ensured the oversight, coordination and monitoring of the study in order to enable and support the participants and maintain the consortium structure and procedures, to achieve the objectives set, to complete the milestones in time and to complete the deliverables. Eight deliverables were successfully submitted during the first reporting period. Four milestones were achieved timely. The external ethical advisor was appointed to monitor the quality of the project, look into consequences for the research participants and oversee how researchers in the project deal with ethical issues. An inventory was made of the risks foreseen in the Grant Agreement.

Task 1.2 Work Plan Definition

The work plan was defined in de proposal before the start of the project and was meant to ensure that all partners were informed about the work to be carried out, the deadlines, the required budget in human resources, etc.. The work plan was presented by:

- The work packages' ("WP" in further text) scheme providing information about the leading partners and partners responsible for carrying out tasks within each WP. A hierarchical structure of the scheme shows the relationships and the directions of information flows among the WP.
- The Gantt chart determines the duration of the whole project, each WP, each task within it and the reporting moments.
- The description of WP.
- The partners' estimated budgets and efforts.
- The deliverables and milestones schedule.
- The list of potential risks and the mitigation measures description.

Task 1.3 Project administration, consortium contract financial and legal management

The project administration, consortium financial and legal management were carried out by the study coordinator and the project manager in close collaboration with the financial division at the Department of Public Health in the Erasmus MC. Regular communication regarding financial and administrative aspects was running via the project manager. The consortium partners were consulted on such topics as personnel costs, time recording, eligibility of costs, acknowledgement of the subsidy provider, etc.. The internal Erasmus MC experts, the EU project officer and the EU legal officer were involved in solving the administrative and legal issues when their expertise and advise were needed.

Task 1.4 Prepare six-monthly internal progress reports, prepare progress reports to the European Commission (every 18 months) and coordinate the final report with recommendations

The consortium arrangement to submit the internal 6 months reports (scientific and financial) was significant for the study as it ensured a consistent flow of information at previously agreed time points. This enabled the management teams to make pivotal decisions immediately. It also allowed the study coordinator to follow the

project progress carefully and thus anticipate and track potential financial or human risks. In case of a problem, the study coordinator endeavoured to find and propose a solution or a rescue plan.

Two internal reports (after 6 and 12 months of the project) were submitted within the consortium. The study coordinator and the project manager checked whether the objectives, deliverables, milestones were reached and whether the individual financial statements from each partner explained the use of resources satisfactorily. The partners' reports were consolidated, reviewed by the coordinator and the reported work progress and achieved results were discussed with the WPs' leaders during the web meetings. The decision to approve the next payment (in accordance with the payment scheme in Art. 7.3.2 of the Consortium Agreement) to a partner depended on the following aspects:

- completeness of the internal reports;
- whether the agreed activities have been actually carried out during the reporting period;
- adherence to the reporting deadlines.

At the 12 months internal report the WP6 had prepared insufficiently for screening the 30% children living in rural areas in the county of Cluj and measures were taken and implemented to give additional screening courses to the family doctors and their nurses from rural areas.

The project coordinator was responsible for preparation of the first18 months project periodic report. The information required for continuous, technical and financial reports was collected from the partners. The coordinator provided them with clear instructions and templates of reports and set reasonable internal deadlines to make sure there will be enough time left for consolidation and internal discussion before the final submission to the European Commission.

Task 1.5 Foundation "Stichting Country-Committees Joint-Partnership of EUS€REEN Study Consortium"

The foundation "Stichting Country-Committees Joint-Partnership of EUS€REEN Study Consortium" was created in 2014. The foundation subcontracts members of all 41 countries (Israel included) within Europe, willing to participate in the study. The foundation acts as a separate partner in the EUSCREEN study and subcontracts all representatives who participate in the study by filling out the extensive questionnaire on the project website. The 41 countries (three Country Representatives per country: vision, hearing and general screening) constitute an advisory board of the EUSCREEN study. The foundation pays remuneration to Country Representatives who have completed an extensive questionnaire. Two Country Representatives have been already paid.

Task 1.6 Organising meetings

During the last 18 months, two consortium meetings were organised: one in Tirana (January 27th, 2017) and one in Cluj-Napoca (March 16th, 2018). In between these meetings, several conference calls were organised. Our aim was to organise at least one conference call per month, inviting all consortium partners. Some meetings only included a subgroup. The dates of the conference calls were: 17-05-2017, 24-05-2017, 05-07-2017, 02-08-2017, 09-08-2017, 06-09-2017, 05-10-2017, 17-10-2017, 23-10-2017, 21-11-2017, 20-12-2017, 08-01-2018, 12-02-2018, 11-04-2018, 30-05-2018, 19-06-2018.

When visiting conferences to present the study, small gatherings were organised to update Country Representatives. These were held at the congress of the EFAS 2017, EUSUHM 2017, ORL-HNS 2017, ESPO

2018, HEAL 2018 for hearing. For vision, these were organised at the Bielschowsky-Gesellschaft 2016 and 2017, ESA 2016, ESA 2017, CVRS 2017, ARVO 2017, ARVO 2018.

Within Rotterdam the team aimed to meet every two weeks.

Task 1.7 Risk Management

Thanks to regular communication with the WPs' leaders via conference calls, meetings and internal reporting tool, the project coordinator was able to recognise (potential) risks and to take action to avoid them or to minimalize the possible negative impact on time.

As a part of risk management, the consortium has forecasted a number of potential risks before the beginning of the project and has identified them in the Annex 1 providing the description of the corresponding mitigation measures. A few of these risks have materialised during the first reporting period. Their state of play has been described in the project continuous report.

No unforeseen risks have arisen after the project start.

Two Interim implementation study reports on Hearing screening in Albania and on Vision screening in Cluj County are attached to this report (Appendix 1 and Appendix 2).

1.2.2 Work package 2: Network, data collection, database, stakeholder analysis & dissemination

Task 2.1 Data collection with the questionnaire

In the first months of 2017 the extensive questionnaire for inventory of vision, hearing and general screening programmes in 41 states in Europe has been developed starting from preliminary versions made in 2014 for a first and in 2016 for a second pilot-study, by a focus group from EMC, USFD, KI and READ. Questions were structured as multiple-choice with comments in 9 domains: demography and epidemiology, administration and general background, existing screening systems, coverage and attendance, tests, follow-up and diagnosis, treatment availability, cost and benefit, adverse effects. This resulted in a very extensive questionnaire that was made available on the EUSCREEN study website www.euscreen.org.

Answers from Country Representatives (CRs) were collected and stored in a secure database. The spreadsheet containing data about screening programmes across Europe contained some errors in copying information. These problems were fixed by our database manager.

To comply with EU regulation (Art. 10 of the Horizon 2020 Annotated Model Grant Agreement: V2.1.1 – 1 July 2016) and to ensure the quality of the answers filled, a tender procedure was added on the website. This way CRs could be selected based on their knowledge, experience and position. It must be said, however, that we had great difficulty finding competent CRs who were willing to fill out this very long and detailed questionnaire, even with the €2.000 reimbursement for the administrative effort in sight. It occurred very rarely that more than one person registered as candidate-CR and only in Denmark three candidate-CRs for Vision Screening registered, only one of whom became active and filled out the questionnaire in August 2017.

Candidate CRs had to register on the website and filled out the following information from the tender procedure:

1. Name, address, email, affiliation.
2. A short description of their experience and / or involvement in screening.
3. Examples of sources that they will consult to get the data.
4. Their first impression of how much of the questionnaire can be filled out, given the state of screening in their country, the availability of data etcetera.
5. Those who would assist them in getting the data or filling out the questionnaire. Collaboration with others to fill out the questionnaire is a sign of quality and reduces the workload, but the remuneration also had to be split.
6. Conflicts of interest statement: It they were, for instance, involved in selling an automated screening device or optotype charts, this should be specified.
7. CRs had to agree that the EU has the right to carry out checks, reviews, audits and investigations on their work.

After registering on the website, a profile with contact details was made for each user. This profile contained an inbox for private communications. CRs were able to fill out the questionnaire, save and alter their answers whenever they want the enter new information.

Task 2.2 Maintaining the network

When available, three Candidate CRs, ophthalmologists, orthoptists, otolaryngologists, pediatricians and audiologists, from 41 European countries were then personally invited by e-mail to register as CR on the website and to start filling out the extensive questionnaire. When no candidate-CRs had registered for vision, hearing or general screening in a country – something which often occurred - new contacts were found through other CRs, national societies, experts who published papers about vision and hearing screening, etc. This was a lot of work, took a lot of time and caused a considerable delay.

Some problems occurred while using the website (www.euscreen.org) for the collection of data about vision, hearing and general paediatric screening programmes in the last six months. A few CRs lost the answers provided on the website. These problems were addressed. We managed to solve all of these problems.

In many cases, filling out the questionnaire stagnated. For practising ophthalmologists, for instance, it was often too difficult or too time-consuming to get all the data, for instances on salaries of professional who could carry out the screening, or the regional differences were so large, that only good data about a specific region could be provided. At the Annual Meeting of the EUSCREEN Study in Cluj-Napoca on March 16th, 2018 we decided that the cost-effectiveness model that is being developed, profits more from good data from a region than from bad data from a nation and we would be content with the former. This minor change in policy was communicated to the CRs, released the tension in some cases and more questionnaires were completed.

Facing a delay in the Spring of 2018, we started to improve the relationship with our CRs further by cultivating a personal relationship through detailed emails discussing their partial filled-out questionnaire with them. This worked but, again, was very time consuming. We remained in contact with them and tried to arrange personal encounters on congresses.

Task 2.3 Survey on vision, hearing and general paediatric screening

Participating countries in the EUS€REEN Foundation were: Albania, Austria, Belgium, Bulgaria, Croatia, Cyprus, Czech Republic, Denmark, Estonia, Finland, France, Germany, Greece, Hungary, Iceland, Ireland, Israel, Italy, Kosovo, Latvia, Liechtenstein, Lithuania, Luxembourg, Malta, Macedonia, Moldova, Montenegro, Netherlands, Northern Ireland, Norway, Poland, Portugal, Romania, Serbia, Slovakia, Slovenia, Spain, Sweden, Switzerland, Turkey and United Kingdom.

Other countries, outside the EU (e.g. Russia, Rwanda, India) also registered on the website and filled out the questionnaire, because they were keenly interested in participating in the EUSCREEN Study. If part of the funds in the EUSCREEN Foundation remains unused, we will remunerate these CRs in countries outside Europe when they have filled out the questionnaire completely. For the development of the cost-effectiveness model in the EUSCREEN Study their participation is immensely profitable, as Low Income Countries are lacking in Europe and we want to develop a model that can also predict the best and cheapest vision and hearing screening programme in Low Income Countries outside Europe.

In many cases it was necessary to search for new and/or additional CRs as some of our CRs did not continue their started work, could only report for a region or were unable to fill out the entire questionnaire. We looked through personal networks of the consortium partners, national societies, participants in conferences and our already existing CRs. This again was very time consuming and needed a lot of hard work.

Because of the enormous, disproportional effort in getting the data from the 41 countries in Europe and several countries, we now have the largest data set about vision and hearing screening ever and created a solid basis for development of the model.

Task 2.4 Database management

Several meeting took place between EMC and the web designer to develop the website and make it user friendly for CRs to fill out the questionnaire on the website. The updated questionnaire was added to the new website www.euscreen.org.

Submitted questionnaires were stored on a secure SQL server database and backed up once a day using a backup server. Answers were entered into a spreadsheet by the administrator of the website. This spreadsheet can be accessed by the partners involved in data collection and validation (EMC, USFD, READ and KI).

On July 1st, 2018, Representatives registered for 41 countries for Vision screening, 39 for Hearing screening and for 36 General paediatric screening. Complete questionnaires were submitted for 32 countries for Vision screening, 28 for Hearing screening and for 28 General paediatric screening. Out of the submitted questionnaires, Country Reports have been made for 11 countries for Vision screening and 16 countries for Hearing screening. The CRs from Bosnia for Vision screening and from Malta for Hearing screening both received the remuneration of €2.000.

Task 2.5 Analysis and review of the development of vision and hearing screening programmes in EU-countries, the reasons for their large diversity and country-specific stakeholder analysis

Data about vision and hearing screening programmes will be collected until December 2018. On March 8th, 2018, the Annual Meeting of the EUSCREEN Study will be held in Poznan and be open for all CRs. We will report back to them the results of all countries, and the analysis and review of the vision and hearing screening programmes in all countries in Europe.

Task 2.6 Dissemination

The dissemination of the project is realised through the CRs, three per country who are members of the Country Committees, through scientific media and through dissemination of the TOOLKIT to the stakeholders.

In the EUSCREEN project seven partners in six countries in Europe participate but also, and more importantly, CRs from 41 countries in Europe and 13 countries outside Europe participate. Part of the project was asking these CRs about the screening programmes in their countries with the extensive questionnaire and in doing so, good communication has been maintained with all CRs. Most CRs were highly motivated to supply data about screening programmes and fill out the questionnaire.

Dissemination by scientific media to inform the scientific community has until now mainly taken place through conferences. When consortium members attended a conference they always organised a meeting between CRs. At least six presentations were given by consortium members.

The final and most important part of dissemination will be the cost-effectiveness model wrapped in the TOOLKIT that will be made in the coming two years.

1.2.3 Work package 3: Verification and analysis of existing vision screening programmes

Task 3.1 Mapping and documenting existing vision screening provision

USFD

Mapping and documenting existing vision screening provision has involved several stages. Firstly, an updated literature search was conducted (first 6 months) to identify any potential data sources that could be useful for the model (WP5). The literature search replicated that undertaken by Carlton et al (Carlton J, Karnon J, Czoski-Murray C, Smith KJ, Marr J. The clinical effectiveness and cost-effectiveness of screening programmes for amblyopia and strabismus in children up to the ages of 4-5 years: a systematic review and economic evaluation. Health Technology Assessment 2008;12:25). All potential papers were then disseminated to WP2 and WP5 for consideration for inclusion in the model development and calibration.

Analysis of the questionnaire data (co-ordinated by WP2) has been implemented. Progress on the analysis has been influenced by when country representatives submit data, but also by the amount of data they submit. Initially a numerical value was attributed to data reliability (i.e. 0: Missing data; 1: No data available; 2: I do not know; 3: Rough estimate; 4: Real estimate; 5: Actual data with source and year). This was useful to identify which countries country reports could be created. It became apparent that those countries that had submitted responses to the questionnaire were not able to complete the questionnaire in its entirety. The responses considered as part of WP3 only related to vision, and within this set of questions there was a high proportion of missing responses, or uncertainty in the responses given.

A template country report was compiled and significant progress has been made in populating this for countries that have submitted final responses to the questionnaire. The draft report contains sections relating to: the population and healthcare system; vision screening commissioning and guidance; vision screening (pre-term babies, birth to 3 months, 3 months to 36 months, 36 months to 7 years); automated screening; provision for visually impaired; knowledge of existing screening programme (prevalence, diagnosis; coverage and treatment success; screening evaluation); and cost of vision screening in children (cost of treatment for amblyopia, cost of treatment for strabismus, cost of treatment for cataract).

The draft reports are then sent back to the relevant country representative (by WP2) for content validation checks. This step ensures that interpretation of the initial submitted responses to the questionnaire is considered. Further questions (for clarification purposes) are included within the draft report for the country representative to answer. The additional questions have been informed by WP5, and relate to data required to populate and calibrate the model. Some questions specifically relate to photoscreening. These include: information on what photoscreener was used; whether other tests are also used (such as visual acuity), or whether it is a stand-alone test; questions on comparisons between regions or areas comparing photoscreening versus vision screening; whether photoscreening is conducted on all children (or only on selected groups); age of children having photoscreening; and referral criteria (including risk factor referral). Once a response has been received from the country representative (via WP2), the additional information, if given, is incorporated into the report, and this is finalized.

To date (18/7/18) eleven draft country reports have been compiled and sent to WP2. A further 11 draft reports that are in progress and will shortly be forwarded to WP2. Six draft reports have been received back from country representatives following content validation checks, and these will be finalised in due course. The completion of final reports will commence during September 2018 - December 2018. By this time it is

anticipated that all draft reports will have been completed, and validation checks received. Only following the finalization of all country reports has been completed can the mapping of vision screening across all 41 countries can begin.

Synthesis of United Kingdom (UK) screening data has been undertaken. Two UK specific reports have been compiled using data submitted to the British and Irish Orthoptic Society (BIOS) Special Interest Group. Permission has been received from BIOS to provide raw data to WP5 for the model development and calibration. The 2017 report is available at:

https://figshare.com/articles/BIOS_Screening_Audit_report_2015-2016/5532910/1

The 2018 report will be uploaded shortly.

UREAD

The input from Reading was always planned to be advisory, with no specific deliverables. Reading's contribution has been Prof. Horwood's expertise is in relation to photoscreening for risk factors as opposed to screening for direct markers of the target condition i.e. amblyopia and poor acuity. Prof. Horwood had significant input into the country questionnaire development, and now that data from the country representatives is coming in, has more recently refined additional information to be requested from countries where photoscreening is used. She has contributed to a paper being prepared for publication on photoscreening in Belgium and Iran.

Prof. Horwood attended consortium meetings in Tirana, Albania in January 2017 and in Cluj, Romania in March 2018. She was also able to attend consortium meetings attached to conferences in Northern Ireland in June 2017 and Porto, Portugal in September 2017.

Task 3.2 Literature review of the impact of vision screening

USFD

The literature review of the acceptability of childhood screening is underway. The aim of the review is to identify synthesize and critically appraise all methods that evaluate "acceptability" of childhood screening programmes to inform the content of the TOOLKIT to be developed in WP8. The review will consider any condition screened in childhood, with consideration of factors that may influence acceptability (such as age, gender, ethnicity, screening test, and so on). The review has been registered with PROSPERO, the international prospective register of systematic reviews (registered on 3rd July 2018).

https://www.crd.york.ac.uk/PROSPERO/display_record.php?RecordID=99763

Significant progress has been made with the review. Five databases (Medline, Embase, PsycINFO via Ovid, CINAHL, and the Cochrane Library) have been searched using a search strategy designed in consultation with an Information Specialist at the University of Sheffield. Search terms included: i) the population of interest, that is "children" and derivatives; ii) the programme of interest, that is "screening" and derivatives; iii) methods of assessing acceptability of screening programmes; iv) A search strategy was used to combine search terms (i) AND (ii) AND (iii) to identify all articles assessing the acceptability of childhood screening programmes from the child and/or parent perspective. No restrictions on time was applied to the search strategy.

All relevant studies (including, but not limited to randomised controlled trials, cross-sectional studies, case-control studies or cohort studies) assessing the acceptability of childhood screening programmes that are published as a full-text original article (i.e. not abstracts, editorials, or reviews) in English have been included. An inclusion and exclusion criteria have been established. All studies will be assessed using an appropriate Critical Appraisal Skills Programme (CASP) checklist. Data will be analysed based upon method of assessment of acceptability. When and where appropriate, subgroup analyses will be carried out to determine the effect of the type of screening programme, and/or target condition. To minimize bias, all titles and abstracts have been independently reviewed by two researchers for consideration for inclusion in the review. Where there was disagreement, this was resolved by a third reviewer.

A narrative synthesis will be performed. Studies will be assessed for methodological quality. The following information will be extracted using a standardised data abstraction sheet: level of evidence scored according to the GRADE system; study purpose; study design (e.g. cohort, case-series, clinical trial); subjects (sample size, demographics, location of screening); measure of acceptability; and study limitations.

4696 papers were retrieved, a further 4 were identified through other sources. After removal of duplicates 4257 papers remained and were screened. 3370 papers were excluded at title. 769 were excluded at abstract. 118 full text articles will be assessed for eligibility in the coming weeks.

UREAD

A literature review and field orientation study carried out by a member of the Rotterdam team in Flanders, Belgium and Iran, has highlighted that photoscreening is being used in very different ways. The preliminary literature review carried out (above) was restricted to the use of the Plusoptix device.

1.2.4 Work package 4: Verification and analysis of existing hearing screening programmes

Task 4.1 Mapping and documenting existing hearing screening provision

Answers to the HEARING SCREENING section of the EUSCREEN questionnaire have been submitted by a portion of the targeted number of countries and regions. Partners in WP2 have coordinated with Country Representatives to submit answers to the questionnaire and have coordinated with partners in WP4 when answers are submitted.

Answers submitted to the HEARING SCREENING questions of the EUSCREEN questionnaire have undergone continuous scrutinization by WP4.

Countries/regions that have provided the most complete set of answers to the HEARING SCREENING section of the questionnaire have been thoroughly evaluated and undergone cross-checking / validation procedures (described below in Task 4.2). Answers that are considered poor in clarity where the validation procedure (described in Task 4.2) resulted in inconclusive data are marked for follow-up.

During validation, an initial draft of the Country Report is completed for internal review.

Along with this first draft of the Country Report, a list of clarification questions is created. This list contains clarification questions relating to answers that were marked poor in clarity during the initial validation process (Task 4.2).

Clarification questions contain two parts. Each question first explains the discrepancy or missing source information to the Country Representatives, and then asks for clarification or confirmation of the correct answer. Clarification questions and the first draft of the Country Report are sent to WP2, who is the central point of contact for Country Representatives.

- Answers have been scrutinized according to the procedure in Task 4.2 for 15 countries/regions.
- Country Reports (first draft) and associated clarification questions have been completed for these 15 countries/regions.
- Clarification answers have been returned by 9 countries/regions.

Once clarification answers have been returned to WP2, information is transferred to WP4 for continued analysis. Clarification answers undergo the same cross-checking /validation method in Task 4.2. A second draft of the Country Report is completed based on all data aggregated. Second-draft Country Reports are sent to the Country Representative for final confirmation.

- Clarification answers returned from 8 countries/regions have been cross-checked and validated using the procedure in Task 4.2.
- One Country Report (draft 2) has been completed and was sent to the Country Representative for confirmation.

Dissemination of research relating to Task 4.1 up to June 30th, 2018:

- 13-15th March 2018: Tema Hörsel in Örebro, Sweden. Presented preliminary methods and findings to delegates across Sweden.

- 6-9th June 2018: Hearing Across the Lifespan in Cernobbio, Italy. Presented descriptive findings of childhood hearing screening across submitted findings.

Work on Task 4.1 will continue as follows:

- Writing the individual Country Reports will continue. Second-draft versions will be released to the Country Representatives. Country Representatives will have the opportunity to make final comments or corrections, and revisions will be made when determined.
- Information from final Country Reports will be conglomerated into one Hearing Screening Report outlining the descriptive, outcome and cost data accumulated from all participating countries.
- Quality and outcome data will be analysed across all participating regions, with respect to programme variables and regional variation.
- Engagement will be maintained with WP2 and WP5 on data collection and data planning for informing the microsimulation model.
- Findings of quality assurance procedures across participating countries will be presented at the World Congress of Audiology in October 2018.
- Findings will be presented in Poznań, Poland to Country Representatives and EUSCREEN partners in March 2019.

Task 4.2 Reliability and validity of hearing screening data

In order to ensure accurate input to the model, an extensive validation procedure was created and carried out on all HEARING SCREENING data provided by each Country Representative to the questionnaire for the 15 countries/regions evaluated thus far.

The procedure has been as follows:

1. Each answer was classified post-hoc into descriptive, outcome, or cost.
2. With respect to the Reliability Score provided by the Country Representative, each corresponding answer was qualified post-hoc as being an approximation (i.e., roughly estimated, guessed) or determination (i.e., a known fact or a calculation based on a source).
3. Each answer was internally cross-checked to repeated or similar questions to ensure internal answer validity.
4. When two Country Representatives have responded to the questionnaire, each answer was cross-checked between County Representatives.
5. A literature review was performed, including grey literature, specific to the country or region under evaluation. Any and all external material relating to the hearing screening programme was accumulated. Any literature published in a language other than English was translated via Google Translate when necessary and reviewed.
6. All source material provided from the Country Representatives on the screening programmes was translated via Google Translate when necessary and reviewed.
7. All answers with corresponding source material were cross-checked with the information provided in that source.
8. Answers marked poor in clarity were those that were,
    a. discrepant with other answers provided.
    b. discrepant with answers provided by a secondary Country Representative
    c. discrepant from their source material

d. discrepant from externally acquired material
e. classified as outcome or cost and the reliability was classified as determination (calculated or acquired from a source), but where that source is neither provided or fully documented

Dissemination of research relating to Task 4.1 up to June 30th, 2018:

- May 18th, 2018: STELLA meeting in Stockholm, Sweden. Presented methods and preliminary findings of quality assurance procedures and outcome measurements to a network of researchers within the field of hearing impairment and deafness in children.

Work on Task 4.1 will continue as follows:

- Evaluation of data provided in answers to the EUSCREEN questionnaire will continue using the specified procedure for each country/region that has submitted HEARING SCREENING answers.

Task 4.3 Literature review of acceptability of and adaptiveness to hearing screening programs and treatment/management

A literature review was performed on key principles described in the draft of the model for implementation in Albania

- Findings from a literature summary, including tables, were communicated to WP5 included prevalence rates, age of identification, protocols for screening, sensitivity, specificity and pass/ refer rates.
- Review, comments and feedback were provided to the manuscript in preparation for submission to publication.

A systematic literature review is planned for identifying referral rates and loss-to-follow-up rates across single- and multi-step neonatal hearing screening programme protocols for both well and at-risk infants.

- A study plan describing the methods of the literature review was submitted to the PhD program at CLINTEC, Karolinska Institutet
- Preliminary search has been performed with guidance from the Karolinska Institutet library and initial evaluation of articles has begun.
- Results of this preliminary search will be used for modification of study question, search criteria, inclusion/exclusion factors and outcome variables.

Work on the systematic review will continue as follows:

- Two Karolinska Institute librarians specializing in systematic reviews will be engaged to perform a comprehensive literature search.
- The study will be entered into Prospero. Search strategy and reporting will follow the PRISMA statement.
- Results from the database searches will be reviewed via title and abstract and sorted by at least two reviewers
- Extracted data from studies will be grouped into programme variables. Referral rates and loss-to-follow-up rates will be compared across screening programmes and populations.
- Engagement with WP5 to deliver findings to inform the microsimulation model and toolkit.

1.2.5 Work package 5: Development of a decision-analytic, cost-effectiveness modelling framework

Task 5.1 Development and validation of the vision and hearing screening models

WP5 partners have been working on the development of a decision-analytic, cost- effectiveness modelling framework of repeated screening. Consortium partners were consulted to build the appropriate model structure. Micro-simulation models were developed for hearing and vision screening, using the MIcrosimulation SCreening ANalysis (MISCAN) model structure. MISCAN is extensively used to evaluate cancer screening programs. It is programmed in Delphi and aims to simulate a situation with and without screening. MISCAN follows the individual life events of a large birth cohort. Typical events in the model are birth, disease onset, disease detection and death. One feature of the simulation is that a current (health) state might depend on the directly preceding (health) state of the individual but cannot depend on earlier states, which is a key assumption of a Markov model. Additionally, transition from one state to another may occur between time sloths and the probabilities for a transition may also depend on duration (dwelling time) of presence in a certain state. This semi-Markov modelling approach enhances flexibility of our model and Monte Carlo simulations (random application of the probabilities distributions) allow variation in the input parameters to reflect uncertainty as observed in real life. These techniques suit the unknown factors of the natural history of hearing and vision impairment in children and the random behaviour of individuals related to screening attendance, treatment adherence and effectiveness of screening programs (Figure 1). Both models enable to perform cost-utility analysis over a life-time horizon.

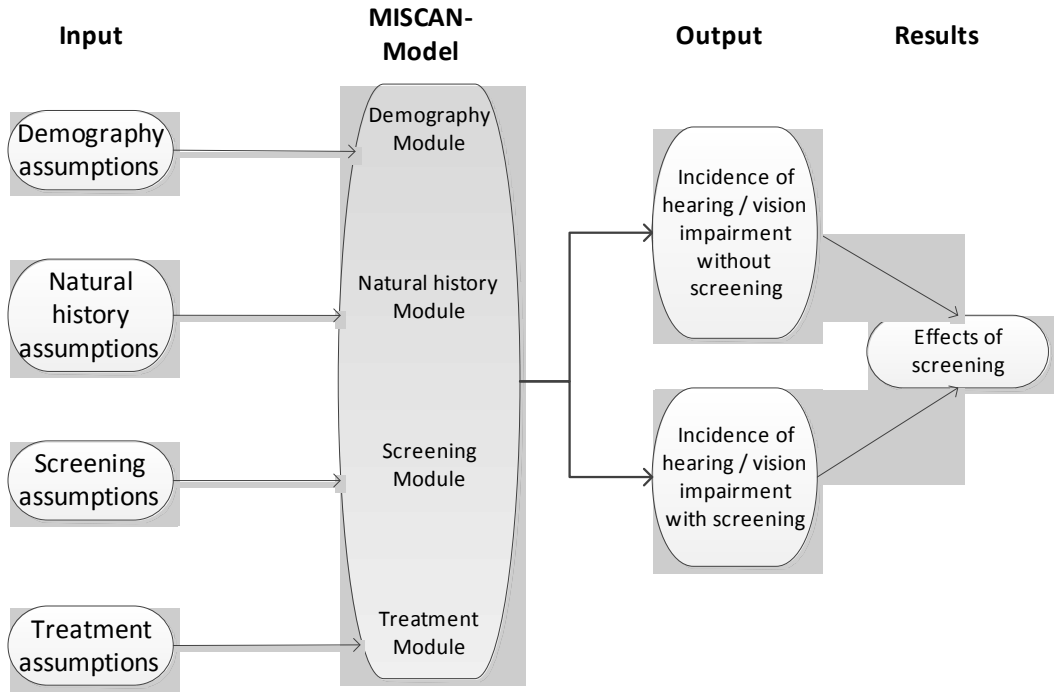

Figure 1: MISCAN-Model overview

Input data

As country-specific data is still being collected (WP2) and validated (WP 3 and 4), current model input is mainly collected from available evidence from the literature. Parameter values were obtained from observed data, literature review and expert discussions. This evidence was carefully discussed and reviewed by all hearing- and vision-experts from all WPs. EUSCREEN partners in Albania and Romania were contacted directly for model input on country-specific costs (for screening and treatment) and local circumstances that might influence screening test characteristics.

Both models will be extended in 2018/2019. The natural history module will become more detailed, for example by adding transient hearing loss, refractive error and levels of visual acuity. Additionally, with help from the EUSCREEN questionnaire data, a modelling framework will be further developed to take into account local circumstances, specifically for low-income and middle-income countries.

Comparison of outcome with existing country-specific screening

Both models were calibrated. The hearing-screening-model was calibrated on prevalence of congenital hearing loss and incidence at birth and severity of hearing loss. The vision-screening-model was calibrated on cumulative incidence of amblyopia and vision screening test characteristics, using the simplex optimization method of Nelder and Mead. We used published data from large prospective-cohort studies from the United Kingdom for the hearing-model and data from a large prospective-cohort study from the Netherlands for the vision-model input (Ramses-study).

Validation

As data from other countries is currently becoming available, model validation will be performed in the second half of 2018 and 2019.

Both micro-simulation models were reported in detail in Deliverable 5.1, uploaded December, 2017.

Task 5.2 Calculating cost-effectiveness in MLIC

Model predictions for the county of Cluj and the three counties in Albania were based on the initial model (as demonstrated in Deliverable 5.1). Input parameters on demography (life tables), attendance rates for screening and life time costs were adjusted appropriately for the specific settings of Cluj (Romania) and Albania. First model predictions show that vision screening in Cluj and hearing screening in Albania can be cost-effective. All model predictions were reported in Deliverable 5.2, uploaded December, 2017.

Since many country-specific input parameters were uncertain, several assumptions had to be made to inform the model. As both vision and hearing screening commenced January, 2018 in Cluj (Romania) and Albania respectively, more accurate data will be collected throughout these pilot-studies. These data will enable further refinement of the model in preparation for the final model version as part of the TOOLKIT (WP8).

Task 5.3 Determining the optimal screening programmes

Preliminary predictions of the most optimal screening programmes for Cluj and Albania were provided in Deliverable 5.2. For hearing screening the most optimal screening program in Albania is most likely a two-staged test sequence of OAE and aABR testing for new-born babies while still admitted at the maternity ward. For vision screening in Romania, measuring visual acuity targeting to detect amblyopia is most optimal at age

6. If further expansion of the screening program is considered, a combination of screening at either age 6 and 7 or age 5 and 6 are possible scenarios. Further extensions of the program (3 times screening) may be considered, but different costs of screening between kindergarten, preschool and primary school must be taken into account.

Sensitivity analyses

One-way sensitivity analyses was performed for both models. For hearing screening, model input was varied for the following parameters; the distribution of severity categories of hearing loss; the age of clinical detection; screening attendance rates; costs of diagnosis and quality of life. Altering these parameters showed that the two-stage OAE-aABR (bedside) screening strategy is still preferred over all other strategies tested in the model, even when using these different assumptions.

For the vision screening model, input was varied for the following parameters; test sensitivity, treatment success latest optimal age, quality of life loss of treatment and utility of life time unilateral visual impairment. The results of these analyses showed that although the number of QALYs gained and the costs varied, the efficient strategies were mostly the same, except for the assumption that treatment is only (optimal) successful until age 5. In the latter, screening at younger ages is more cost-effective. If no utility loss for living with unilateral visual impairment is assumed, the ICER exceeds the willingness-to-pay threshold for Romania.

1.2.6 Work package 6: Implementation study of vision screening in Romania

Task 6.1 Development of a regional programme for increasing the capacity of the Health Care System of early detection of amblyopia among children from the target group (age 4-5 years): prestart – M12

Goethe

Goethe University has a small share in the total project (1.8%). The reason for participation of Prof. Fronius is her expertise concerning diagnostic assessments for amblyopia and experience with amblyopia treatment. In addition, she has some knowledge of the situation in Romania. She proposed the UMF as EUSCREEN partner and participated in the preparatory meeting in Cluj-Napoca in January 2016. Her main role in the project is to provide vision expertise to the University in Cluj-Napoca. She was involved in the discussion about refinement of the cost-effectiveness model for vision screening, providing literature and comments concerning assumptions of the model. Prof. Fronius was also involved in checking translations of the database forms from English to Romanian. She participated in the meeting in Northern Ireland in 2017 and the group meeting in Cluj-Napoca in March 2018.

Task 6.1.1. Development of the required structures for the implementation of the screening programme

UMF

In order to complete the necessary tasks, the following arrangements were made:

- Setting up the project team. Establishing the attributions sheet for each member of the project team. Hiring the project team members.
- Identifying the personnel that will be screening the children: kindergarten nurses in urban areas and family doctor's nurses in the rural areas.
- Acquisition plan for all the needed materials in the project.

DASM

The DASM core project team members were appointed on December 22nd, 2016 prior to the commencement of the project through the DASM director's decisions, in line with the applicable national legislation.

The initial core project team was comprised of six appointed members: one responsible for the project (manager), one responsible for stakeholder engagement, one for human resources, one medical coordinator, one responsible for public procurement and one financial manager. This structure was designed in order to ensure a proportionate division of tasks that correlated with the administrative and scientific activities that fall under DASM remit.

Observing general requirements of the Horizon 2020 programme with regards to gender balance, the appointed members of the team were three men and three women. They are not newly hired persons but DASM employees who were entrusted with EUSCREEN project responsibilities. Tasks were assigned to team members by the project responsible and included in each of team members' job descriptions.

DASM also identified its personnel (nurses) eligible to be involved in the EUSCREEN project in Cluj-Napoca. A database with potentially eligible nurses employed by DASM (at kindergarten level) was

developed. There are 45 nurses currently working in 48 units (47 kindergartens and one daycare centre). 44 consented to be involved in the project. Subsequently, director's decisions were issued for the nurses involved and their EUSCREEN responsibilities were included in their job descriptions.

Task 6.1.2. Notifying the community, doctors and nurses involved in the project about the commencement of the screening

UMF

A number of oral presentations about the EUSCREEN project were given during scientific medical conferences:

- Family Medicine Conference - Medic.ro - February 23-24 2017, Cluj Napoca - EUSCREEN – European project of early detection of amblyopia in pre-school children - Dr Mihai Mara.
- Family Medicine Conference - Medical Forum Cluj Napoca - June 12-13, 2017 - EUSCREEN – European project of early detection of amblyopia in pre-school children - Dr Mihai Mara.
- The VIII-th National Congress of University and School Medicine, April 28-29, 2017, Cluj-Napoca. SIMPOZION - EUSCREEN – European project of early detection of amblyopia in pre-school children – Prof. dr. Cristina Vlăduțiu, Dr. Simona Căinap, Dr. Mihai Mara, Dr. Daniela Rajka, Aurel Mocan, Monica Ghițiu.
- The VIII-th National Congress of University and School Medicine, May 18-19, 2018, Bucharest. EUSCREEN – program of early detection of amblyopia in children aged 4 to 6. – Lecturer Dr. Simona Căinap – UMF Cluj-Napoca.
- The XVI-th National Congress Of Ophthalmology with international participation, Sinaia 27th-30th September 2017 - How to avoid late diagnosis in amblyopia. Prof. Dr Cristina Vladutiu.

Letters with information detailing the screening project were sent out to:

- The Ministry of Health and The Ministry of Education;
- Cluj County School Inspectorate;
- Rural mayor offices (they have been sent both letters and emails);
- All family doctors in the rural areas.

Face-to-face meetings were organized with representatives of local communities (mayors, vice-mayors, representatives of the social services department, school doctors and nurses) in Câmpia Turzii, Turda, Dej, Gherla, Huedin and Floresti.

The project was presented at a meeting with the school doctors and nurses from Cluj-Napoca.

Each family doctor's office in the rural areas has received at least one informational email about the project and has been contacted via telephone (at least 3 times).

There were four meetings with the rural family doctors and their nurses: two in Cluj-Napoca, one in Huedin and one in Gherla.

DASM

The DASM nurses to be involved in the project were informed about the project during a joint meeting organized together with UMF on May 29th, 2017.

Building upon its prior good relationship, DASM faciltated UMF's contact with the other towns in the county and with the Floresti commune. DASM contacted (phone calls) and formally requested meetings (written requests) with the local public authorities of one commune (Floresti) and five towns (Huedin, Turda, Campia Turzii, Gherla, Dej).

All six municipalities expressed interest in the EUSCREEN project. Eventually six meetings were held on May 29, 30 and 31, 2017 across six locations with a wide variety of local stakeholders. These stakeholders were relevant for either the implementation of the project or for supporting its implementation at political level: mayor, deputy mayor, town manager, school and kindergarten doctors as well as school and kindergarten nurses. The objectives of these meetings were presenting the EUSCREEN project, safeguarding the collaboration of the relevant local authorities, formalizing the collaboration (cooperation protocols), estimating the number of children and professionals to be involved, discussing the implementation and possible bottlenecks.

Several ideas emerged from the discussions during these meetings:

- The importance of consent was highlighted.
- Generally, the participants in meetings were positive that the parents will be open to screening. They also emphasized the great responsibility attached to carrying out the screening.
- In certain locations there are children from marginalized communities who are not enrolled in education. UMF and DASM plan to provide screening for this category as well. To this end, professionals working with marginalized communities (local Roma experts, community facilitators) would be involved in mobilizing the parents to allow access to their children.

Task 6.1.3. Foundation of a monitoring and evaluation system of activities in the study, which could be subsequently extended

UMF

A system for monitoring and evaluation of activities was established:

- Weekly meetings of the UMF Cluj team: establishing directions for actions, employing the members of the team, determining the plan for purchases.
- Regular meetings with local partners.
- The activity of the hired personnel is documented via timesheets.
- Creating a database containing personnel eligible for the project.
- The forms that the nurses fill out are collected and verified by project team members.
- Project team members have actively contributed to the optimisation and the functionality testing of the database.
- The national coordinator, the project manager and the project team members are monitoring the data input in the database.
- Keeping track of the children that have been referred to an ophthalmologist and keeping track of the forms received from the ophthalmologist.

DASM

During the first 18 months of the project several M&E actions were put in place:

- Regular and exceptional meetings of the DASM core team.
- Participation of the DASM project manager in the UMF's project team meetings when needed/upon request.
- Regular meetings and contact (via WhatsApp) with the DASM nurses.
- Visits to kindergartens were the screening will take place.
- Monthly timesheets for the nurses and core project team.
- The forms that the nurses fill in are collected and verified by the DASM's medical coordinator.
- The project manager is monitoring the data input in the database.
- Questionnaires were applied to the nurses.
- A small-scale impact assessment using a qualitative method (interviews) was carried out with several DASM nurses in order to see what worked and what didn't in the first year of the screening.

Task 6.2 Increasing the competences of nurses and doctors from kindergartens and general practitioner offices in vision screening: M9-M12

Goethe

Prof. Fronius visited Cluj-Napoca in November 2017 together with Prof. Horwood from the UK to participate and give support during a course for nurses and doctors; subsequently they provided a report about the experience during the courses, the progress that the Romanian team made as well as the problems the group faced preparing the implementation of screening, especially in rural areas.

Despite lots of problems such as delayed predictions from the model concerning vision screening, the necessity to change the provider of the VA charts or protracted agreement on the VA testing procedures to be taught, the courses were delivered in time so that screening could start as scheduled in January 2018.

UMF and DASM

In 2017 we organized three courses to train the medical personnel how to measure the visual acuity in children. The courses were attended by kindergarten nurses working in the urban area, school doctors, rural family doctors and their nurses.

Furthermore, in March 2018 we organized a new series of training courses for better coverage of the rural areas, taking into consideration the insufficient attendance rate of the rural medical personnel at the 2017 courses.

To a limited extent DASM was involved in facilitating the access of its own personnel to the courses mentioned above.

Task 6.2.1 Designing the curriculum and layout of the Continuing Medical Education courses meant to improve competences in the evaluation of vision and visual acuity

Goethe

Many telephone calls and telephone conferences as well as Skype sessions were held with Prof. Cristina Vladutiu and Anna Horwood and also Herb Simonsz for discussing and preparing VA assessments in Romania, type of VA charts and notation of VA values. Prof. Fronius commented on and made suggestions for the PowerPoint presentations prepared by the Romanian team and by Prof. Horwood for the courses in Cluj-Napoca. She prepared and contributed material and literature for explaining log versus linear scaling of VA charts and correct VA notation.

UMF

The course curriculum was developed by the national coordinator of the project, Prof. Doctor Cristina Vladutiu.

The course curriculum included theory on visual problems in children as well as four hours of practical workshops and a one hour debriefing. The theory was presented in a simple, synthetic and highly didactical manner so as to be as accessible as possible to course participants. The main aim was to establish an optimal ratio between the theory and the practical workshops.

In order to ensure an appropriate level of knowledge for screening children, the courses have been credited by the Romanian College of Physicians and by the Romanian Order of Nurses with twelve points of continuous medical education.

Task 6.2.2 Distribution of the course participants into groups

UMF

March 3rd -4th, 2018 -	17 nurses (11 rural areas, 6 private kindergarten Cluj-Napoca)
	12 doctors (8 rural areas, 4 private kindergarten Cluj-Napoca)
November 4th -5th, 2017 -	47 nurses (2 rural areas, 45 kindergarten Cluj-Napoca)
	19 doctors (1 rural areas, 1 small town, 17 Cluj-Napoca)
November 18th -19th, 2017 -	37 nurses (28 rural areas, 4 small town, 5 Cluj-Napoca)
	27 doctors (21 rural areas, 3 small town, 3 Cluj-Napoca)
October 21st -22nd, 2017 -	50 nurses (16 rural areas, 23 small town, 11 Cluj-Napoca)
	17 doctors (11 rural areas, 5 small town, 1 Cluj-Napoca)

DASM

DASM facilitated the participation of its own personnel in the courses mentioned above. The training organized on November 18th-19th, 2017 where 37 nurses and 27 doctors attended was specifically designated

for the DASM nurses. Nevertheless, both DASM and UMF were flexible and catered to the needs of the participants. Whenever needed other dates were arranged for the participants.

Task 6.2.3 Delivering the schedule for the programme of the training courses

UMF

The course schedule was comprised of the following elements:

First day:

1. Detailed presentation of the EUSCREEN project.
2. Course: visual problems in children.
3. Course: the principles and technique of measuring visual acuity in children; when to refer a child to the ophthalmologist?
4. Practical workshop: the technique of measuring visual acuity in children.

Second day:

1. Practical workshop: the technique of measuring visual acuity in children (continued).
2. Debriefing.
3. How to fill out the child's forms.
4. Official test.
5. Discussions.

Task 6.2.4 Ensuring the quality standards for the activity of early detection of amblyopia at the level of schools and general practitioners

UMF

a) Each course lasted for two six-hour days, meaning a total of twelve hours of theory and practical training for all participants. During the practical workshops each participant has performed on his or her own at least one visual acuity examination.

The courses were concluded with a multiple choice test to evaluate the participants' knowledge of the course subjects. The exam consisted of ten multiple choice questions. By passing the exam the participants received twelve points of continuous medical education.

The participants who passed the test received a diploma that will allow them to be hired to measure the visual acuity of children for the EUSCREEN project.

b) Since the screening started in January 2018, the project team visited 31 family doctors' offices in rural areas to offer support and guidance to ensure the best possible outcome of the screening activity. Family doctors' offices were visited in Viisoara, Tureni, Moldovenesti, Valea Draganului, Poieni, Sancraiu, Sacuieu, Rachitele, Izvorul Crisului, Marisel, Belis, Alunis, Bontida, Mica, Caseiu, Catcau, Baciu, Garbau, Aghires Fabrici, Fizesu Gherlii, Iclod, Dabaca, Bobalna, Tritenii de Jos, Cojocna, Apahida, Jucu, Ceanu Mare, Unguras and Vultureni.

The team members also visited the medical offices at kindergartens in the small cities of Turda, Campia Turzii, Gherla, Dej, Huedin and Floresti, in order to check the progress of the screening and to offer support, advice and to address possible queries.

In 2018 we visited several kindergartens in Cluj-Napoca, Floresti, Huedin, Turda and Campia Turzii, as well as seven family doctors' offices in the rural area of Cluj County, together with the international coordinator. There were discussions with the medical personnel involved, about difficulties, measurement techniques and expectations.

c) Each month when collecting and verifying the forms, there were also discussions with the medical personnel aimed at clarifying issues that were identified during the screening activity.

Goethe

Prof. Fronius participated in the preparation of the training courses by providing vision testing expertise. She provided information during the training courses for nurses in Cluj-Napoca and feedback to individual trainees.

Task 6.3 Implementing the actions of early recognition of amblyopia in children: M13-M36

UMF

1. Contracts and job descriptions for nurses in the urban area (cities in Cluj County) and the rural areas were elaborated upon.
2. Procedures for the employment of nurses from urban and rural areas commenced.
3. A list of ophthalmologists who will examine the referred children was prepared. A letter containing details about the project, about how to examine the children and how to fill out Form 5V was sent to the ophthalmologists on the aforementioned list.

Task 6.3.1 Mapping the target group at the level of Cluj County

UMF and DASM

An inventory was made of the number of children eligible for inclusion in the screening program, based on the reports required from each nurse who would participate in the screening project. Each nurse or doctor who attended the course was asked to report the number of children eligible for inclusion in the screening program from the kindergarten or rural family doctor’s office where he /she works.

Attempts were also made to establish the number of eligible children at each location by personally asking the rural family doctors either when we telephoned them or when the project team visited their offices.

Data regarding the number of children born in Cluj-Napoca between 2012-2015 was provided to DASM by the National Statistical Office and by the Population Registry Directorate within Cluj-Napoca Municipality.

Task 6.3.2 Notifying the parents about the implications of the research study and obtaining the informed parental consent for the project

UMF and DASM

Each parent received an informed consent form, accompanied by a letter with information about the project as well as a colourful and attractive leaflet. Only the children that presented the informed consent form signed by one of their parents have been included in the project.

Where needed (vulnerable communities), additional information were provided to the parents by the nurse. As an approach the whole process of obtaining the informed consent took place at the kindergarten in the presence of the nurse prior to the screening.

Task 6.3.3 Dissemination of information about the project among the staff from kindergartens in urban areas and general practitioners' offices in the countryside and convincing them to join in the study

UMF

1. Details regarding the project have been presented at scientific conferences – organized with DASM - that were attended by family doctors, school doctors, family doctors' nurses and kindergarten nurses:
   - Family Medicine Conference - Medic.ro - February 23-24, 2017, Cluj Napoca - EUSCREEN – European project of early detection of amblyopia in pre-school children - Dr Mihai Mara.
   - Family Medicine Conference - Medical Forum Cluj Napoca - June 12-13, 2017 - EUSCREEN – European project of early detection of amblyopia in pre-school children - Dr Mihai Mara.
   - VIII-th National Congress of University and School Medicine, April 28-29, 2017, Cluj-Napoca. SIMPOZION - EUSCREEN – European project of early detection of amblyopia in pre-school children – Prof. Dr. Cristina Vlăduțiu, Dr. Simona Căinap, Dr. Mihai Mara, Dr. Daniela Rajka, Aurel Mocan, Monica Ghițiu.
   - VIII-th National Congress of University and School Medicine, May 18-19 2018, Bucharest. EUSCREEN – program of early detection of amblyopia in children aged 4-6. – Lecturer Dr. Simona Căinap – UMF Cluj-Napoca.
2. Several articles on the EUSCREEN project have been published:
   - School and University Medicine Magazine, Vol IV, Nr. 1, January 2017 - EUSCREEN. European project of early detection of vision and hearing impairment in children. Prof. Dr. Cristina Vlăduțiu, Dr. Simona Căinap, Dr. Simona Sevan, Dr. Daniela Rajka, Dr. Mihai Mara.1

[1] http://www.medicinascolara.ro/download/revista/vol4_nr1_2017/7%20EUSCREEN.pdf

- School and University Medicine Magazine, Vol IV, Nr. 3, July 2017 - Amblyopia - Etiopathogenesis and current treatment options. - Dr.Simona Sevan, Dr.Oana Teodosescu.2
- School and University Medicine Magazine, Vol V, Nr. 1, January 2018 - Practical concepts on the measurement of visual activity within the EUSCREEN, European project of early detection of vision and hearing impairment in children. Prof. Dr. Cristina Vlăduţiu, Dr. Simona Sevan, Dr. Oana Teodosescu, Dr. Raluca-Maria Ursu.3

3. Each family doctor's office from the rural area has received at least one email with information about the project and the course details and has also been contacted via telephone at least three times.

During each contact we emphasised the importance of vision screening in children for the early detection of amblyopia and we offered details about the project. Moreover we mentioned each time that the training courses were free of charge, that graduating the courses would mean twelve points of continuous medical education and that remuneration will be available for work on behalf of the screening project.

DASM

1. Two meetings (one prior to the screening, the other one prior to the training) were organized for the DASM nurses.
2. Kindergartens were visited, where discussions with directors took place as needed.
3. Regular individual meetings with the nurses took place every month at DASM headquarters.

Task 6.3.4 Supplying the necessary equipment for the development of the screening

UMF and DASM

Packages were prepared and distributed to the participating nurses, containing educational and other materials:

- Movie - visual acuity evaluation technique in children.
- Animated movie - teddy bear at the ophthalmologist.
- Written informed consent forms for the parents.
- Forms 1, 2, 3V, 4V.
- Letter to the ophthalmologists.
- Glasses for examination (one lens removed, the other one patched).
- A reward for the children: teddy bear sticker.
- Optotypes (eye charts).

Goethe

Prof. Fronius was involved in developing the specifications of the test charts to be used for VA screening.

[2] http://www.medicinascolara.ro/download/revista/vol4_nr3_2017/4%20Ambliopia.pdf

[3] http://www.medicinascolara.ro/download/revista/vol5_nr1_2018/3%20RMS%20NOTIUNI%20PRACTICE.pdf

Task 6.3.5 Visual acuity measurement in children from the target group

UMF

In the first six months of 2018 (from January 1st to June 30th) the following numbers of children were examined:

- 3385 children from Cluj-Napoca
- 645 children from the rural areas
- 1740 children from the small cities from Cluj County (Campia Turzii, Turda, Gherla, Dej, Huedin).

From a total of 5770 examined children, 828 children have been referred to an ophthalmologist.

It is important to note that part of the children from the rural areas attend a kindergarten in a nearby city, hence they are included here in the urban part of the screening, even if they are from a rural village.

Task 6.4 Assessment of the data obtained from the screening: M25 - M42

Not applicable for this reporting period.

Task 6.5 Long term dissemination of expertise: M37-M48

Not applicable for this reporting period.

1.2.7 Work package 7: Implementation study of a neonatal hearing screening programme in Albania

Task 7.1 Wide-scale implementation study of an evidence-based, cost-optimised neonatal hearing screening programme in the regions of Tirana, Kukës, and Pogradec in Albania

The 12 first months of the project's implementation were dedicated to the preparation of the personnel, equipment, protocols and the general conditions for a smooth and proper screening and diagnosis of the hearing loss.

During the first months of the project's implementation a lot of effort has been spent on preparing and signing various documents and contracts. These included the working contracts with each institution involved as well as with individuals. In this stage we had several discussions and meetings in order to select the best screening staff in the institutions. The staff was proposed by the institution where the EUSCREEN-study is managed and cross-checked and verified by the project staff.

The preparation of the training calendar and curricula was also object of discussion not only within the Albanian staff but especially with experts from other WPs. The work resulted in various sessions of training during the Autumn of 2017.

Regarding the equipment, the list of equipment has been revised and discussed several times together with WP1, looking at the technical specifications and the screening protocols. The screening and some diagnostic device were purchased in Summer – Autumn of 2017. They were used during the training of the personnel for the practical sessions and afterwards delivered at each site together with the respective manuals and documents in Albanian. A PC and internet also has been provided for registering the tests in the database at each screening location.

Some basic furniture was also delivered at the sites for creating a proper room for testing. A lot of work was done for preparation of a diagnostic booth at Tirana University medical center. A store room was converted into an acoustically insulated room with silent ventilation. A screen OAE and a diagnostic ABR were put there for testing. This is the best audiologic booth and also the first experience with such a booth in Albania. It was very difficult to find the proper personnel and people to complete the job. Once the rest of the devices will be provided in 2018, this will be the best equipped booth for the diagnosis of the childhood hearing loss in Albania to date.

From January until now EUSCREEN is screening in all locations. 4803 babies have been screened from January to June, comprising 97.5% of all babies born during these months. Apart from the first tests, 765 second tests, 73 third tests and 10 diagnostics test have been completed. There is increasing number of babies missing at the second-stage testing but some of the babies are born in Tirana but live in other cities: If they do not come for second-stage testing, we do not follow them up in the city they live. The lack of information of the population regarding issues of early detection of hearing impairment and the financial and logistic problems of some families in certain geographic areas, are causing 'lost to follow up' cases. This requires a long and consistent information campaign in order to change the attitude of the population.

The sequence of screening, paper work, documentation, database, tracing the babies who fail, has been going on simultaneously as a part of monitoring and for improving the process of screening. The needs for training and technical assistance of the staff are continuously evaluated and addressed.

Two more training sessions took place in the Spring of 2018, one for technical audiological staff and another for speech therapy for children with hearing loss. The first one had a theoretical part and a practical part in the new booth.

Both trainings and several group-discussions helped to create a pathway for children with various levels of hearing loss and to fill the gaps from identification to rehabilitation.

A number of technical difficulties occurred in the first months of screening. First of all there was a delay in the development of the database forcing the nurses to fill out all data on paper forms. Even now there are still some issues related to the proper functioning of the database: data is not exported correctly into excel-files with shifting columns (for instance, the result of screening is listed under the postcode heading).

In addition, there were some technical difficulties with the equipment. A few devices stopped working due to software malfunctions. Probes broke without obvious reason. We were told that also in other countries probes and other sensitive devices broke frequently when neonatal hearing screening was first implemented.

The number of failed measurement initially was high but decreased over months. Each screening nurse has her own learning curve and the first months of screening appeared to have a high rate of 'repeat' cases which subsequently reduced in the following months.

During the second part of 2017 there were some issues related to a partially overlapping screening programme by an Italian NGO. Sorting out and coordinating the two programs required many hours of reporting, meetings and negotiating, especially in the political situation of that specific period in Albania and the difficulty governmental structures had to take adequate decisions.

Implementation of neonatal hearing screening in Albania by a Horizon 2020 study presents many administrative and technical problems as it progresses. This is partly related to the lack of experience of various bodies with such research programmes and partly to the bureaucracy and the level of corruption of other bodies. This produces various obstacles and delays and requires sometimes the intervention of various administrative levels for sorting out the problems. For example, the finalisation of the audiometric booth, and the transfer of funds from Albanian bank to the University bank account needed months a lot of human effort to be realised.

Task 7.2 Increasing the competences of nurses in performing the hearing screening tests by theoretical and practical training, assessment of competence, and accreditation

The training of nurses/midwifes took place from end October to early November 2017 and was carried out at two levels.

The first level consisted of a theoretical curriculum, both for nurses, GPs or other interested healthcare personnel. The objective of this one day theoretical course was to inform the health staff regarding the hearing screening program in the area we are working and also to deliver basic information about the screening programs, the necessity of hearing screening, respective tests, diagnostics, rehabilitation services and the benefits of such a program. The training days were very much welcomed, accredited with CMEs and certified by the Centre of Medical Accreditation in Tirana.

Kukës, 30-9-2017	49 participants.
Pogradec 14-10-2017	38 participants

Tirana 21-10-2017 31 participants.

Part of the training days was a pre- and a post-test as well as an individual evaluation form for the training and lectors. The Public Health team has analysed the tests and the evaluation forms and the conclusion is that there are many gaps in the theoretical knowledge of the health personnel, especially regarding the hearing pathways and their evaluation in children.

The training curriculum was delivered by our team in cooperation with some lectors from the Faculty of Medicine, UMT. They were hired and paid on a daily basis by the project: Prof. Pjerin Radovani, ORL, Dr. Aida Bushati, neuro-paediatrician, Dr. Migena Bedeni Kika, Head of the Centre of Child Rehabilitation (CCR) and neuro-paediatrician.

The only objective that was not met, was the participation of the paediatricians and midwifes from the Child Consultancy centres in Tirana, we had planned. We have agreed with MOH to select one paediatrician and one nurse from each centre (10 centres in Tirana) but the notification sent by them was wrong.

The second level of training was focused in delivering a theoretical foundation and some practical testing skills for our project staff. 6 midwifes and 1 administrative person from each maternity hospital in Tirana, 8 staff and 2 administrative persons from Pogradec and Kukës were brought in Tirana to participate in this training. The venue was the UMT in Tirana. The program was a combination of the theoretical topics with practical sections. The Interacoustics (IAs) and Natus representatives in Albania sent their engineers to teach and supervise (6 extra personnel all together) the use of the devices and their databases. The midwifes used the devices on each other.

The communication with parents and its importance was also discussed in a group work discussion session. We agreed on a paragraph to explain the test and present the test results to the parents. The paragraph was written down and sent to each team to memorize and use in a standardized way.

In December we felt the need to do some more practicing with devices and tests. Therefore we coordinated with the CCR for testing hearing in their patients. The midwifes formed groups of 3-4 persons and used both OAE and aABR there, continuing with testing each other and downloading the data in the databases. The tests and PC work was assisted again by the IAs and Natus companies staff and supervised by Dr. Qirjazi and Dr. Toci.

In Kukës just before Xmas we arranged a testing day in the paediatric ward of the Kukës hospital were all the children were tested with aABR and later the data were loaded on the database. The whole practical day lasted 6 h. In Pogradec, there were 2 days of 6 h each were both OAE and aABR were used for testing hearing in a kindergarten in children 3-5 years old (around 26) and then the data was downloaded in the respective database. Both Kukës and Pogradec sections were supervised by Dr. Qirjazi.

We think that there is still need for in-job training of our staff during the project duration. Also, we have to plan to test the knowledge and the practical skills of our staff regarding the hearing screening procedures.

Task 7.3 Improve the awareness in parents of the importance of early detection and treatment of hearing loss in young children

The philosophy of the program is to consider parents as partners along the process of the identification and the intervention of hearing loss. This is especially true when we talk about the rehabilitation process where the role of the expert is to guide and teach the parents to work with their children.

Increasing the awareness of the parents is a long and difficult process. In order to achieve this we have prepared leaflets, posters, TV programs, website information.

The 2 types of leaflets aim to give information about the importance of hearing screening and facts about the incidence of hearing problems and the types of tests and prerequisites for a better testing of the baby. The written information is supplementing the information given by the screening nurse.

The second leaflet is targeting the parents whose babies have to be referred for a diagnose work up in the hospital because they have failed all the screening tests. This also gives info regarding the importance of the early hearing loss identifications and the types of tests performed for the confirmation of the hearing status and what type of intervention follows.

The posters also reinforce the necessity of early hearing testing for the proper language development and are hung in various parts of the maternity buildings. In the main maternity hospital in Tirana, the information about hearing screening test and the opportunity to have it done in that institution, has been advertised all the day for months in the main hall big screen.

The importance of the second, third or the diagnostics tests for the babies who fail the first one, has been the main topic in two TV programs,

https://www.youtube.com/watch?v=zA85yW9_bvU,

https://www.youtube.co m/watch?v=ljpRR7aUJSs,

both very popular and with a high attendance rate, not only in Tirana area, but in all Albania. This information would reach not only the parents who have babies but also all other parents and is the best way of mass communication, whereas thee websites;

http://www.dritare.net/2018/01/23/doktoreshe-qirjazi-apel-prinderve-mos-i-denoni-femijet-me-shurdheri-ja-cfare-duhet-te-beni/1

https://konica.al/2018/04/prinder-kujdes-si-te-zbuloni-sa-me-heret-problemet-qe-lidhen-me-degjimin-tek-femijet-tuaj/

and the articles about the screening program actually running in Albania and the importance of the early identification if hearing loss, targeted a different category of parents, possibly with a higher level of education who receive the majority on information via portals, and other internet resources.

Task 7.4 Creation of a monitoring and evaluating system of activities in the study, which could be expanded to a national level after the study.

All the tests done together with the results, risk factors and other epidemiologic data are filled in the database, where each subject has a different consecutive code. Through the database all the subjects who have failed 1, 2 or 3 tests can be identified. The codes in the testing device and the database, coupled with the registers in each institution, secure the identification and tracking of the subject if necessary.

Task 7.5 Creation of a registry of cases of failed screening tests, that could be expanded to a national level after the study

The registry of failed cases must correspond with the ability of the database to filter the data as well as transfer them into an Excel-file, i.e. make them editable and workable. The register could not be made since there were still some issues related to the proper functioning of the database (output files have test results listed under “postcode”, for instance), but these are being resolved. The task will be reported in the next project periodic report.

1.2.8 Work package 8: Development of a TOOLKIT comprising of a cost-effectiveness modelling framework and strategy plan for implementation

The WP8 is not applicable for this reporting period.

1.2.9 Work package 9: Ethics requirements

Task 9.1 H - Requirement No. 2 - Informed consent templates and data authorizations

Deliverable 9.1 Informed consent templates and data authorizations is submitted to the European Commission by the project coordinator on June 27th, 2018. The deliverable describes and demonstrates the informed consent forms and the information sheets for parents of the children to screen (i.e. brochures, posters and films) used by the beneficiaries performing vision and neonatal hearing screening by children (UMF, DASM, UMT). The data authorizations are described in the second part of the deliverable report.

Task 9.2 H - Requirement No. 6 - Copies of ethical approvals

Deliverable 9.2 Copies of ethical approvals is submitted to the European Commission by the project coordinator on June 28th, 2018. The deliverable contains the copies of and the explanations regarding the ethical approval by the beneficiaries performing vision and neonatal hearing screening by children (UMF, DASM, UMT).

Task 9.3 POPD - Requirement No. 9 - Data protection confirmation

Deliverable 9.3 Data protection officer is submitted to the European Commission by the project coordinator on December 22nd, 2017. The deliverable is presented by the letter from the Erasmus MC Data Protection Officer stating that the project proposal is conducted according to Regulation (EU) 2016/679.

Task 9.4 OEI - Requirement No. 20 - Appoint ethical advisor

Deliverable 9.4 Appoint ethical advisor is submitted to the European Commission by the project coordinator on December 22nd, 2017. The external independent ethical advisor was appointed for the EUSCREEN project to oversee with impartiality the ethical concerns involved in this research. This person is meant to monitor the quality of the project, look into consequences for the research participants and oversee how researchers in the project deal with ethical issues.

The ethical project review written by the project ethical advisor is included by this report (Appendix 3).

1.3 Impact

The contribution of the project to the expected impacts described in section 2.1 of the Description of Action is still relevant.

1.4 Access provisions to Research Infrastructures

This part is not applicable for the EUSCREEN project.

1.5 Resources used to provide access to Research Infrastructures

This part is not applicable for the EUSCREEN project.

2. Update of the plan for exploitation and dissemination of result

The plan for exploitation and dissemination of results does not need to be updated.

3. Update of the data management plan

This part is not applicable for the EUSCREEN project.

4. Follow-up of recommendations and comments from previous review(s)

This part is not applicable for the 1st reporting period of the EUSCREEN project .

5. Deviations from Annex 1 and Annex 2

5.1 Tasks

Work package 1: Project management

None.

Work package 2: Network, data collection, database, stakeholder analysis & dissemination

A database was developed to follow up screening in Albania and Romania. In the Grant Agreement, the development of this database was anticipated to take place in Sheffield but development took place in Rotterdam. Since Rotterdam is studying the implementation it seemed logical, that all partners involved in the implementation and implementation study, were involved in the development of the database. Since WP2 is responsible for gathering data and the contacts with Country Representatives were precious, but also difficult and laborious, it was best to have one contact point at WP2, hence the database in Rotterdam.

Data collection from all 41 countries in Europe has been delayed. This has been caused by a delay in the development of the questionnaire and a delay in filling out and submission of the questionnaire by the Country Representatives. However, the data collection is expected to be finalised in the Autumn of 2018, so that the work progress in other WPs will not suffer from this delay. WP3 and WP4 have to make country reports and other before July, 2019.

Deviations foreseen in the second reporting period

A Vision and Hearing screening Cost-effectiveness Conference will be organised on March 8th, 2019. We plan to allocate some of the budget to reimburse CRs for their travel costs and lodging.

An invaluable consultant on hearing screening (previous head of the screening programme in the UK) has been involved in planning the implementation of hearing screening in Albania, training the diagnostic team, evaluating reports etc. Initially she did not want to be reimbursed for the time spent on the project.

Work package 3: Verification and analysis of existing vision screening programmes

USFD

Country reports have been delayed in completion due to the delayed collection of data caused by delayed questionnaire submission (December 31st, 2018). This will delay the final summary report which was due for completion by December 31st, 2018.

UREAD

Now that data is coming in from the country representatives, it appears that the quality of the data on screening for risk factors in countries where it is used in Europe may not be available in fine enough grain for meaningful analysis.

Work package 4: Verification and analysis of existing hearing screening programmes

Analysis of the hearing part of the extensive questionnaire is behind schedule for two reasons. The first reason is delayed collection of data caused by delayed questionnaire submission.

Furthermore, the validation process has shown that answers provided are often poor in clarity or validity. Answers received from Country Representatives are often unclear, or discrepant with internal or external sources. The poor quality of answers requires a more intense validation procedure and a high number of clarification questions.

These two factors have resulted in fewer number of completed Country Reports than expected by June 30th 2018.

Work package 5: Development of a decision-analytic, cost-effectiveness modelling framework

None.

Work package 6: Implementation study of vision screening in Romania

Goethe

The VA charts envisaged to be used for screening turned out to be inappropriate. The charts that would be sponsored by OCULUS GmbH turned out to be scaled not logarithmically. The only available correct VA carts were donated by Good-Lite Inc. to the study. Under considerable time pressure decisions on different charts and their provision had to be taken. These problems could be solved in a concerted action between Profs. Vladutiu, Fronius, Horwood and Simonsz. However due to delayed delivery the appropriate VA charts could not be demonstrated during the first course for nurses. Despite these problems the courses in Cluj-Napoca took place according to the project schedule, so that vision screening could start as planned in January 2018.

UMF Cluj

Obstacles registered during screening in rural areas in Romania

Screening in the rural area is unfolding with difficulty because of a number of reasons.

Most rural offices have only one nurse and it is impossible for her to neglect work related obligations in favour of the visual screening. This is the reason why the courses have taken place on weekends and not during workdays.

The communication with the nurses was not an easy one, it was done mainly via telephone as many of them do not have an email address (nor feel the need to have one).

Attending the course, a mandatory condition for participating in the screening project, implied travelling from their villages to Cluj-Napoca. For some this was an impediment , due to the distance, the infrastructure, the lack of means of transportation or of family related duties. Travel by nurses from rural areas to the UMF-Cluj was not supplied or reimbursed by UMF Cluj.

Some rural nurses are either close to retirement or are already retired but still working, and have no interest of developing new professional skills (such as measuring the visual activity in children)

Two other screening programs have been implemented as trials in Cluj county: cervical cancer screening program and breast cancer screening program, both by the Cluj Oncology Institute. Only the cervical cancer program has continued after the trial period, but still the response rate, and the involvement of the doctors is not according to expectations. Maybe the fact that these two programs didn't go as expected made the doctors' interest in screening programs diminish.

In Cluj county there is a very rich offer of Continuous Medical Education (CME) courses organised by either UMF Cluj or by the pharma industry. That is why the 12 CME points per EUSCREEN course were not as motivating as we thought.

It is difficult for the family doctors to adequately financially motivate their nurses, considering the huge wage-gap between the private sector (family doctors) and the public sector (hospitals). It is no surprise that at times there is an increase in the turnover of medical staff. For example: the nurse form the village of Bobalna attended the course but did not sign the work contract and didn't even answer her phone. On one of the rural visits Dr Mara and Dr Cainap went to Bobalna where they met with the doctor from the village. It was then they found out that the nurse quit her job at the rural office for a better paid job in the nearby city.

Some of the nurses and doctors who attended the free course (and received 12 points of CME), afterwards didn't wish to participate in the screening project.

For the rural offices with a small number of eligible children (at most 10), the financial part is not considered motivating enough.

The bureaucracy is considered by some nurses and doctors to be unappealing (filling in forms, delivering them once a month to UMF).

The parents are reluctant to the request of taking the children to the doctor's office to get screened. The nurses are faced with the same attitude even when calling the parents for their children's immunizations. In opposition, in the urban kindergartens the nurse has all eligible children in the kindergarten every day of the week and the screening is running smoothly, she has already a trust relationship with the parents and with the children and can be helped, if needed, by the teachers.

The real number of children living in the rural areas is smaller than first estimated based on the official reports of the Romanian National Institute of Statistics. The latest numbers offered by Cluj County Health Insurance House state that there are approx. 5200 children enlisted at rural family doctors. Still, a part of these children are attending a kindergarten from a city nearby and hence have already been screened in the urban area. (We are keeping a separate evidence of these children). In the villages near Cluj-Napoca we are faced with a different situation: children actually living in the rural area but having the official address in Cluj-Napoca only so that the kindergarten in Cluj-Napoca allow them to attend.

The main objectives, dissemination of project activity and informing the local communities about the importance of early detection of amblyopia, have been reached.

From the 5246 eligible children from the rural area (both 4- and 5-year-old children), in the first 6 months of 2018 a total of 637 have been screened by rural nurses and doctors. Another 500 children that live in rural area have been examined in urban kindergartens.

DASM

Following negotiations, in the original budget 125,068.75 euro were allocated to DASM. 100,055 represented direct personnel costs whilst 25,013.75 euro represented indirect costs. During implementation the need for purchasing certain goods and services emerged in the view of ensuring smooth implementation of the project. As a consequence 7,303.23 euro were spent on purchasing certain goods and services. This amount would correctly fall under the category other direct costs. Details regarding these expenditures are provided in the REPORT ON EXPLANATIONS ON THE USE OF RESOURCES.

Work package 7: Implementation study of a neonatal hearing screening programme in Albania

Screening protocol

In the Grant Agreement, the protocol for hearing screening in Albania was expected to be OAE, OAE, aABR for well babies and aABR aABR for NICU babies in order to identify cases of auditory neuropathy in infants with risk factors. This protocol was expected to be used in all three maternity hospitals. Before the start of implementation, it was decided that in Kukës, a region more poor and mountainous, the protocol for both well babies and infants with risk factors would be aABR aABR.

It was assumed that a team would be trained especially to perform aABR testing. They would visit all four maternities to perform aABR testing. During training the protocol could still have changed based on the model predictions, so nurses were trained to be able to perform both OAE and aABR. This made a separate team to perform aABR unnecessary.

Finally, when infants fail the first test, it was expected that the second test would take place at a mother consultancy centre or at home one week after the initial test. Since all screeners are working in the hospital, it seemed more practical to have parents return to the hospital for follow up testing. To date no screeners were trained in the mother consultancy centres.

Screening facilities

As already described in the grant agreement, the babies who failed all the stages of screening had to be sent to the University Hospital of Tirana for full audiological diagnosis. Unfortunately until now the ENT department in this hospital has had scarce resources for the assessing the hearing loss in children and especially a proper room for ABR or VRA testing. Furthermore, in Albania there is not much expertise in sound treating rooms for hearing tests as well. So after extended discussions with various types of engineers, Institute of Public Health (IPH) specialists and the Audiological team in Rotterdam, we managed to turn a store room into an audiometric booth.

Three offers were taken from three companies, with considerable differences in mounts (reflecting different concepts of sound treatment indeed), we have chosen the one which did not include the use of lead (a metal not recommended by the EU) and for a good value of money. The personnel of IPH did the measurements afterwards and we are happy with the insulation of the room. Part of this, was setting up a silence ventilation system of the room, given the fact that babies might need a lot of time to fall asleep. That was a subject of another contract .

Both contracts made possible the creation of an audiometric booth which is fully functional and has been successfully used for the diagnose of hearing loss since January 2018.

Work package 8: Development of a TOOLKIT comprising of a cost-effectiveness modelling framework and strategy plan for implementation

The WP8 is not applicable for this reporting period.

Work package 9: Ethics requirements

None.

5.2 Use of resources

Work package 1: Project management

None.

Work package 2: Network, data collection, database, stakeholder analysis & dissemination

The hours worked are generally in line with those anticipated but the personnel assigned to the project changed during the first 18 months. During the pregnancy leave from one of the employees, a substitute was assigned to the project to not cause any additional delays in data collection.

Foreseen budget reallocations in the second reporting period

The development of the database was anticipated to take place in Sheffield. Because the Rotterdam partners needed to have access to the database, the costs of the database were partially funded by Rotterdam instead of Sheffield. Funds were made available within the Rotterdam budget to fund the database. We expect a budget shift from Albania and Romania of €10.000 to Rotterdam to fund the development of the database.

To be able to reimburse travel expenses and lodging for CRs who are attending the Vision and Hearing screening Cost-Effectiveness Conference we allocated €23.000 within the Rotterdam budget. Sheffield will contribute the money they were planning to use for the database to this cause.

We expect our consultant on hearing screening to charge around €20.000 for the time spent on the project. This amount will be split equally amongst the partners in Rotterdam, Sweden and Albania.

Work package 3: Verification and analysis of existing vision screening programmes

USFD

Hours worked are in line with those anticipated. As the EUSCREEN questionnaire was not redesigned and recirculated (as outlined in the application) there has been no requirement for arranging online data collection and storage via epiGenesys. The budget for this will be reallocated to WP2 for a future meeting with country representatives in 2019.

There is an underspend on expenses due to the low number of face-to-face meetings. Meetings have been mainly via web conferencing.

UREAD

Hours worked are generally in line with those anticipated, and at some time points there was little to do as we waited for data to come in. Prof. Horwood has used the time to contribute to the development of the vision screening programme in Cluj and start planning the literature review.

There is a significant underspend on expenses, because at the time of budgeting we did not know the frequency, format and location of face-to-face meetings. We have made more efficient use of conference calls than anticipated, and taken the opportunity to meet at conferences where Prof. Horwood's expenses were partly or wholly covered from other sources of funding. Meetings have been held in countries where accommodation and expenses were cheap, and airports served by low-cost airlines.

Work package 4: Verification and analysis of existing hearing screening programmes

The process of finding a good postdoc/ PhD student to this interesting project was harder than expected, using advertising and personal networks. After the internal procedure at KI with an Admission seminar in October, Allison Mackey started her position as a fulltime doctoral student on November 1st, 2017. Thus, the allocated resources were not used from January to November 1st, 2017. The position is for four years extending through 2021, requiring the resources allocated in the budget.

Work package 5: Development of a decision-analytic, cost-effectiveness modelling framework

None.

Work package 6: Implementation study of vision screening in Romania

UMF Cluj

Obstacles registered during screening in rural areas in Romania

A part of the children from the rural areas will not be included in the visual screening at this time.

We requested permission (through an amendment) to organize an extra course for the rural medical personnel to get trained and hired in the project. Unfortunately, the participation of doctors and nurses from rural areas was low although efforts have been made to persuade doctors and nurses from rural areas to participate in the project.

We requested permission (through an amendment) to be allowed to hire doctors for the screening in the rural areas (if the nurses did not wish to get involved, and with the same financial benefits)

Team project members organized regional meetings in order to better explain the project's aim and to answer possible questions. At these meetings there were present kindergarten nurses, rural family doctors and their nurses and local authorities.

Team project members visited medical offices from 31 communes in order to offer support for those who already started the screening, or to have face-to-face discussions and to try to convince those who initially, when contacted via telephone or email, refused to participate in the screening.

After receiving feedback from rural nurses and doctors we advised them to screen the children directly in the kindergartens (rural kindergartens do not have nurses, like the ones from the cities have).

We wish to organize one more course with a duration of only one day.

It must be taken into account that all difficulties that were reported during the unfolding of the project will most probably appear, maybe in a more pronounced way, when trying to implement the screening at a national level.

Work package 7: Implementation study of a neonatal hearing screening programme in Albania

Screening protocol

All screeners were trained to be able to perform both OAE and aABR testing. Allowing them to perform the complete screening protocol themselves instead of a special team performing only aABR screening. The number of screeners and the location where screening takes place, the maternity hospitals, did not change.

Work package 8: Development of a TOOLKIT comprising of a cost-effectiveness modelling framework and strategy plan for implementation

The WP8 is not applicable for this reporting period.

Work package 9: Ethics requirements

None.

5.2.1 Unforeseen subcontracting

Work package 1: Project management

None.

Work package 2: Network, data collection, database, stakeholder analysis & dissemination

We are delighted that representatives from several countries outside Europe have expressed their interest to participate in our comparison of cost-effectiveness. In particular countries that have no formal screening programme yet, that wish to implement vision or hearing screening programmes and want to spend their budget as efficiently as possible. Russia, India, South Africa, Malawi and Rwanda are willing to fill out the questionnaire we developed. The data they are supplying are of the utmost importance for our project (to include in our model) as the EU has no low-income countries. Therefore, we want to reimburse these representatives, who meet the same requirements we have set for the 41 representatives from EU-countries, for their time as well.

The CRs within Europe are entitled to a €2.000 remuneration for the administrative effort provided the questionnaire is filled out completely or at least contains the data that is available for that country. In principle the EU can only reimburse participants from EU countries and countries that participate in the Horizon 2020 research programme. However, not all of the money available for the 41 countries in Europe will be spent as quite some CRs have filled out 2 domains instead of one and cannot be paid in full for a second domain. The remainder will be split among the participants outside Europe.

Work package 3: Verification and analysis of existing vision screening programmes

None.

Work package 4: Verification and analysis of existing hearing screening programmes

None.

Work package 5: Development of a decision-analytic, cost-effectiveness modelling framework

None.

Work package 6: Implementation study of vision screening in Romania

None.

Work package 7: Implementation study of a neonatal hearing screening programme in Albania

None.

Work package 8: Development of a TOOLKIT comprising of a cost-effectiveness modelling framework and strategy plan for implementation

The WP8 is not applicable for this reporting period.

Work package 9: Ethics requirements

None.

5.2.2 Unforeseen use of in kind contribution from third party against payment or free of charges

Work package 1: Project management

None.

Work package 2: Network, data collection, database, stakeholder analysis & dissemination

None.

Work package 3: Verification and analysis of existing vision screening programmes

None.

Work package 4: Verification and analysis of existing hearing screening programmes

None.

Work package 5: Development of a decision-analytic, cost-effectiveness modelling framework

None.

Work package 6: Implementation study of vision screening in Romania

UMF Cluj

The VA charts envisaged to be used for screening turned out to be inappropriate. The charts that would be sponsored by OCULUS GmbH turned out to be scaled not logarithmically. The only available correct VA carts were donated (100) by Good-Lite Inc. to the study. They had to be used at 3 meters and had illiterate "E" on one side as planed in the Description of Action, and LEA children pictures on the back. They were very appropriate and readily adopted by all screening nurses and doctors.

Work package 7: Implementation study of a neonatal hearing screening programme in Albania

Two broken OAE probes were replaced by NATUS free of charge. These probes are a necessary part of the hearing screening device.

Work package 8: Development of a TOOLKIT comprising of a cost-effectiveness modelling framework and strategy plan for implementation

The WP8 is not applicable for this reporting period.

Work package 9: Ethics requirements

None.

Appendix

Appendix 1. Interim implementation study report on vision screening in Cluj County

1.1 On-site observations in Cluj County during January and March 2018

From January 1st, 2018 onwards, a program for vision screening for children aged 4 – 5 is being implemented in Cluj County. In the implementation study the caregivers are the study objects, not the children.

Children living in the municipalities Cluj-Napoca, Câmpia Turzii, Dej, Gherla, Turda and Huedin are now being screened in kindergarten by nurses working for DASM (Cluj-Napoca) and UMF (small cities), most often by the nurses who are taking care of the children at the kindergarten. Some of these nurses and some family doctors have followed one of the courses for vision screening in Cluj-Napoca, given in the Autumn of 2017.

Children living in rural areas of Cluj County are being screened at the family doctor's practice by the doctor or his/her nurse. These nurses have also followed the vision screening course given by UMF. After this they were contracted by UMF to perform the vision screenings at the practice of the family doctor.

Unfortunately attendance at the courses is lower for rural screeners in comparison with the nurses working in the kindergarten in Cluj Napoca (working for DASM) or small cities (Turda, Campia Turzii, Gherla, Dej, Huedin, etcetera).

The following observations and discussions are divided in three different parts, one for each area where the vision screenings takes place: Cluj-Napoca, five small cities and rural areas. This is followed by overall observations and discussion with conclusions.

Cluj-Napoca (DASM)

Observations

- All nurses we met in Cluj-Napoca were enthusiastic about the project
- The DASM-team has all nurses fill out paper forms. These are then entered in an online database by one person for the DASM, using the database account of the nurse who examined the child. The packages with instructions for the screeners and information material for the parents of the children were being hand out in person by DASM to the screeners
- One of the members of the DASM-team initiated a WhatsApp-group with all screening nurses in it so they can discuss difficulties with each other and with the DASM
- Overall we got the impression the nurses want to know exactly how to perform the screening and they take this task of performing vision screenings very seriously
- The nurses had started screening children and seemed secure in their task
- The start of the screening supervised by DASM was timely and started mid-January 2018 at full speed

Five small cities

Observations

- All urban nurses we met were enthusiastic about the project
- The UMF-team has all nurses fill out paper forms. These are then entered in an online database by one person for the UMF (Daniela Rajka or one of the secretaries), using the database account of the nurse who examined the child
- The packages with instructions for screeners and information for the parents were being hand out in person by UMF to the screeners. In this way Daniela Rajka could give the latest updates about the screening to the nurses
- Overall we got the impression the nurses want to know exactly how to perform the screening and they take this task of performing vision screenings very seriously
- Besides Cluj-Napoca, there are also ophthalmologists working in Gerla, Dej, Huedin, Turda and Campia Turzii, to which the parents of referred children can go with their child
- Some nurses had started screening children mid-January 2018 and some nurses were about to start at the moment we visited them. They seemed a bit less secure in comparison to the nurses at the DASM

Rural areas

Observations

- All rural nurses and doctors we met were enthusiastic about the project
- The Cluj-team has all nurses and doctors fill out paper forms. These are sent to the UMF-Cluj and entered in an online database by one person from UMF team (Mara Mihai or one of the secretaries), using the database account of the nurse who examined the child
- The packages with instructions for the screeners and information material for the parents were being handed out in person by UMF to the screeners. In this way Mara Mihai can give the latest updates about the screening to the nurses
- Overall we got the impression the nurses want to know exactly how to perform the screening and they take this task of doing vision screenings very seriously
- The nurses had not started screening children in mid-January 2018 and not all practices had received the information materials and VA charts in the third and fourth week of January 2018
- By May 2018 only five children from rural areas had been entered in the database. This was because the UMF found it unethical to create email addresses for them in the case that the screener did not have an email address yet

Discussion

- Some children from Viisoara go to a kindergarten at Campia Turzii, so they might be screened twice: once at the GP-office in Viisoara (rural - UMF) and once at the kindergarten in Campia Turzii . Maybe this is something that can happen more often in other surrounding villages near small cities as well
- Besides Cluj-Napoca, there are also ophthalmologists working in Gerla, Dej, Huedin, Turda and Campia Turzii, to which the parents of referred children can go with their child.
- There are also kindergartens in rural areas, yet these kindergartens do not have nurses. A possible solution could be a screening nurse who travels between the kindergartens in the rural villages

- The nurses in the rural areas are too busy – they have to deal with everything medical, because rural kindergartens do not have nurses – and therefore possibly less enthusiastic about vision screening children
- Overall the screening started too late in comparison with Cluj-Napoca and the five small cities. The low attendance rate at the courses of doctors and nurses from rural areas is a possible explanation for this

Overall observations

- The Cluj-team had paid a lot of attention to the instructions for screeners and the information materials for the parents. These are of high quality, which is of great importance
- Two amendments were applied for and approved in the reporting period: one for doctors to also be able to screen in rural areas (instead of only nurses) and one for an extra course to be held in March 2018, mainly for the nurses from rural areas
- The attendance rate was very low at this course: 50 doctors and nurses signed up, only 29 actually attended at the course (17 nurses and 12 doctors)
- One of the reasons for not attending the course was the fact that the course was held on the weekend and doctors did not want to spend the whole weekend working after a busy working week. Another reason was the lack of reimbursement for travel expenses and the fact lodging was not provided
- A short explanation letter was drafted to accompany form 5 in case a child is being referred to an ophthalmologist. The letter asks to cooperate with the EUSCREEN project. Form 5 is the referral form that nurses give to the parents of children who did not pass the screening test and take with them to the ophthalmologist
- A list with names of all these ophthalmologists is included in the aforementioned referral letter. This way the parents can decide for themselves to which ophthalmologist they will take the child for further examination. There are ten ophthalmologists in Cluj-Napoca and also ophthalmologists in Gerla, Dej, Huedin, Turda and Campia Turzii

Overall discussion

In November 2017 Anna Horwood and Maria Fronius visited and assisted at the courses for vision screeners, given by UMF in Cluj-Napoca. They noticed that it was difficult to motivate personnel from rural areas to participate in the courses and the project. Since then it seems little has changed as far as this is concerned, although there was a plan made in December 2017 to increase the participation of nurses and doctors from rural areas. Also the last free course for nurses and doctors was promoted via the councils in the villages and transport to and from UMF should have been provided.

Overall, in the rural areas the motivation to participate in the project appears to be lower than in the urban areas. We are not sure if this only goes for the family doctors and nurses or for the parents of the children as well.

The VA charts envisaged to be used for screening turned out to be inadequate. The charts that would be sponsored by OCULUS GmbH turned out to be scaled not logarithmically. Good-Lite Inc. donated the only available correct VA carts to the study. Under considerable time pressure decisions on different charts and their procurement had to be taken. These problems could be solved in a concerted action between Profs. Vladutiu, Fronius, Horwood and Simonsz. However due to delayed delivery the adequate VA charts could not

be demonstrated during the first course for nurses. Despite these problems the courses in Cluj-Napoca took place according to schedule, so that vision screening could start as planned in January 2018.

All materials were in Cluj on time and are being distributed among the kindergartens in rural areas. Not all rural family doctors' offices already have the VA charts and other equipment on the first day screening was supposed to be start. Daniela however again explains to the nurses how to perform the screening and updates them about the latest changes very well.

Improvements in performing the tests are noticeable and referral rates are decreasing.

It is conspicuous that screening was well organized and had a high coverage rate in Cluj-Napoca and in the five small cities, by the nurses that are working in the kindergartens.

In the remaining period of 2018 and 2019 the possibility to have children screened by a travelling nurse to kindergartens in rural areas should be examined.

No data is available on eligible children, i.e. all children in Cluj County age 4 and 5. The nurses we met at the kindergartens and family doctor's offices make a list of all children aged 4 and 5 that are in that specific kindergarten or practice.

According to family doctors who filled out a questionnaire for us, the percentage of Roma children in their practices is remarkably high: 17.9% on average stated by rural, compared to 3.5% of Cluj County's population belonging to this ethnic group (according to official figures). Due to associated stigma Roma do not always self-identify as such; they would rather state they are Romanian or Hungarian. The percentages of Roma living in rural areas are far higher than in urban areas. This could explain part of the difference between official data and estimations from the field.

The Roma children are also screened by DASM, by nurses at kindergartens and/or at a Roma Day Care Center. In case of referral, it is not sure the children will visit an ophthalmologist according to the nurses that we spoke. The parents might not go by themselves due to uncertainty about the screening procedure or out of fear of possible costs for the examination by an ophthalmologist and the possible treatment (patches/glasses).

In the case a child does not understand the first test, the Tumbling E-chart, the test on the other side of the chart, the Lea-test, should be done immediately. At the moment we visited the nurses they were told to repeat the test one month later. It would be best, however, to optimize the testing moment as much as possible and keep the rate of repeated tests as low as possible. Nevertheless, if both tests are insufficient, a second consult to test the visual acuity should be done.

Overall conclusions

The first priority of vision screening is to detect amblyopia, but after that there has to be a plan on how to finance patches and glasses. In principle medical treatment is guaranteed by the state.

The price of patches and/or glasses can be too high for low-income parents. The cheapest glasses cost around RON 140,- (€ 30,-) and one patch costs RON 2,70 (€ 0,60) while the average Romanian salary is around RON 2300 (€ 495,-) per month. A cheaper alternative for a patch could be an eye-pad that is normally used for adult ophthalmologic patients.

Patches and glasses are not funded by the project; the parents have to rely on reimbursement via their health insurance.

To make the treatment of amblyopia cheaper, it might be a good idea to train orthoptists in Romania in the future. To this date there are no orthoptists working in Romania, so all amblyopia is treated by ophthalmologists.

1.2 Survey among family doctors in rural areas in Cluj County

From January 1st, 2018 onwards, a program for vision screening for children aged 4 – 5 is being implemented in Cluj County. Vision screening is an eye examination primarily to detect amblyopia at an age that amblyopia can be treated effectively by patching the healthy eye for several hours per day. In the implementation study the caregivers are the study objects, not the children.

Children living in the municipalities Cluj-Napoca, Câmpia Turzii, Dej, Gherla, Turda and Huedin are now being screened in kindergartens, most often by the nurses who are taking care of the children. These nurses have followed a course for vision screening in Cluj-Napoca in the autumn of 2017. The vision screening has been organized effectively by DASM in Cluj-Napoca and by UMF and similar organizations in the smaller cities and rural areas.

It has become clear that vision screening is more difficult to realize in rural areas. Family doctors and their nurses have been invited to follow eye examination courses that were given at the UMF-Cluj in the autumn of 2017, but only a minority of family doctors and nurses from rural areas attended these instruction courses. An additional course was organized on March 3-4 2018, but only seventeen nurses and twelve doctors attended. The course was free, but travel expenses and sleeping facilities for the Saturday night in between these days ware not reimbursed or provided.

To gain more insight into the difficulties encountered when attempting to introduce vision screening of all children in rural areas in Cluj County, one hundred family doctors in rural areas in Cluj County were sent a short questionnaire on March 28, 2018.

Until now, eleven completed questionnaires have been returned. Even when eleven completed questionnaires do not constitute a large amount, these nevertheless provide helpful insights. Respondents were from eleven different communes . Here the results of the questionnaire are presented.

Table 1: how respondents heard about the study (multiple answers possible)

Email	7
Telephone call	4
Announcement at meeting	2
Course	2
Not heard about it before	0
Total	**15**

The first question was how the respondents had heard of the study concerning vision screening of children in Cluj County. Based on the answers to this question it would appear that the most effective way of reaching these doctors is email, followed by phone. Although none of the respondents said they had not heard about the study before, during an on-site visit on January 26, 2018 we met a family doctor in a poor rural village who had not been informed about the study.

The next question dealt with the perceived importance of vision screening and eye examination for children aged 4 – 5. All respondents deemed this important; a large majority even very important.

Table 2: importance of vision screening for children aged 4 – 5

Very important	8
Important	3
Total	**11**

Subsequently, the question was posed what age group is most in need of vision screening. All respondents answered children aged 4 – 5. Only three considered the screening of school children equally as important as the screening of preschool children aged 4 – 5.

Table 3: what children's age group is most in need of more screening examinations? (multiple answers possible)

School children	3
Preschool children aged 4-5	11
Children aged 2-3	0
Infants aged 0-1	0
Total	**14**

The fourth question dealt with the relative importance of various types of screening for preschool children. Respondents were asked to assign a number to each of eight types of screening, with one being the most important and eight the least.

Table 4: screening examinations of preschool children aged 4-5 in sequence of importance by adding numbers from 1 – 8 (1 being the most important). Shown are the average numbers assigned.

Vaccination	4.1
Cognitive/mental development	5.4
Motoric development	5.4
Growth	5.6
Vision	5.7
Hearing	6.0
Speech	6.8
Teeth	6.9

On average, as can be seen in table 4, vaccination was considering the most important while dental screening ended up being the least important. However, there were large differences between the individual respondents and not all respondents interpreted this question in the same way. Six assigned numbers 1 to 8 to each of the options, while the other five sometimes assigned the same number several times (two even assigned the number 8 to all available options).

Also note that hearing screening here does not mean neonatal hearing screening, that is already being implemented in Romania.

The next questions dealt with the predicted acceptance of parents and children to accept the free examination and willingness to purchase and wear glasses if prescribed. As shown in tables 5-7, the respondents are confident that parents and children will comply with what is deemed necessary.

Table 5: will parents accept free examination of their children's eyes?

Yes	10
I don't know	1
Total	**11**

Table 6: will parents buy the glasses prescribed by an ophthalmologist?

Yes	11
Total	**11**

Table 7: will the children wear the glasses?

Yes	10
I don't know	1
Total	**11**

The fact that all family doctors who filled out the questionnaire are confident that the parents will buy the glasses may seem surprising, but it must be said that income levels in each of the communities have not been taken into account here.

The following question dealt with who could or should perform vision screening. Answers were more or less split between either a family doctor or a nurse (be it a family doctor's nurse or a kindergarten nurse). Several respondents, however, chose more than one option here.

Table 8: who could best perform the eye examination of preschool children aged 4-5 in rural areas (multiple answers possible)

Family doctor	6
Family doctor's nurse	4
Kindergarten nurse	2
No answer	1
Total	**13**

It is important to note that only family doctors in rural areas were queried and most kindergartens in rural areas do not employ a nurse. Considering the difficulties with implementation of vision screening in rural areas, where 30% of all children live, alternative ways of screening, like screening by travelling nurses in kindergartens in rural villages, could be considered.

Respondents were also asked to estimate the approximate costs of the different options, but only one respondent – who had chosen the option family doctor's nurse – answered this question and estimated the cost at € 5 (presumably per screening per child). Another – who noted the examination could be performed by all –

elaborated here: “the family doctor option would not represent additional costs on the yearly balance, just an extension of the medical treatment; the costs of the family doctor’s nurse option would have to be discussed.”

The ninth question asked how training courses should be organized. Should these be one or two days and should these be given on weekdays or during the weekend? As shown in table 9, the majority of respondents appears to prefer a two-day course during the weekend. One hour of a course is equal to one point of continuous medical education, meaning a full weekend course is equal to twelve points (two six-hour days).

Table 9: how free training courses for eye examination should be organized (for family doctor or nurse):

2 days (Saturday and Sunday)	3
No answer	2
2 days	2
Weekdays	2
1 day (Saturday)	1
1 day	1
Total	**11**

Subsequently respondents were asked who should pay for vision screening for preschool children: the Ministry of Health or the Ministry of Education? Almost all chose the Ministry of Health, with one respondent saying both should cover the costs. Vision screening is one of the few forms of paediatric screening (see table 4) that would be eligible for financing by the Ministry of Education, as it is performed at the age of 4- 5 only.

Table 10: should vision screening and eye examination of preschool children aged 4-5 be financed by the Ministry of Health – or by the Ministry of Education (school medicine)?

Ministry of Health	10
Both	1
Ministry of Education	0
Total	**11**

In Austria, children are only admitted to school (at age six, to learn to read and write) when the parents have the ‘eye examination’ checked in a ‘screening passport’ with all compulsory screening exams. Respondents were asked whether this would be a good idea for Romania too. As can be seen in table 11, almost all agreed.

Table 11: should children only be admitted to school at age six when ‘eye examination’ is checked in a ‘screening passport’?

Yes	9
Not necessarily compulsory	1
No	1
Total	**11**

Respondents were then asked about the most important problems affecting their practice. Noteworthy is that this question received no less than 37 answers from eleven respondents; significantly more than any other question where more than one answer was possible.

Table 12: the most important problems affecting the respondents' practice (multiple answers possible)

Too few tests are reimbursed	5
Travelling to remote areas to see patients	5
Other problems	5
Amount of patients in practice	4
Low payments by insurance	4
Rise of personnel costs	4
Health care reform not yet applied	4
Too much work	3
Too much administrative work	2
Too many patients treated for free	1
Total	**37**

A variety of different issues were all mentioned several times as most important, but it appears that cost-related problems are generally considered the most important issue. Next to these, five respondents mentioned travelling to remote areas to see patients as the most important problem, either together with other issues or as the single most important problem.

Four respondents chose the number of patients in their practice as an important problem and also mentioned the number of patients. These ranged from 1100 to 2600 with the average number being 1725. It should be noted, however, that patients go to the family doctors with all of their medical problems.

Only four doctors considered the health care reform not being carried out an important problem and just one family doctor mentioned too many patients being treated for free as an important problem, but like with several earlier questions (see tables 5-7) it should be noted that income levels in the communities have not been taken into account here.

Respondents who (also) answered 'other problems' were given the option to elaborate. Mentioned several times was lacking education where screening is concerned, both among medical professionals and among the general population. Also mentioned were low pay for doctors and lack of assistance and the fact that it is difficult for doctors to be present when they need to be in isolated areas.

The next question dealt with the percentage of Roma children in the respondents' practice. This could be perceived as somewhat of a delicate question since, in general, the number of Roma in Romania is disputed. An official 2011 census for Romania put the number at 621.473 , but the Council of Europe estimates that approximately 1.85 million Roma live in the country (8.3% of the population) . According to the aforementioned census, the population of Cluj County numbers 691.106 people of which only 22.531 are declared Roma (3.5%).

Table 13 shows the estimates of the respondents of the percentage of Roma children in their practice. The highest figure given was 85% while the lowest was 1% (mentioned twice). More than half the respondents estimate the figure to be below 25%. In fact only three respondents gave a figure above 10%; the average being 17.9%. However, as far as could be determined by cross-referencing with data from the 2011 census, almost all respondents gave a figure that was – often significantly – higher than the figure for their commune.

Table 13: estimate of the percentage of Roma children in the respondents' practice?

More than 75%	1
50-74%	0
25-49%	2
0-24%	7
Total	**10**[4]

Question number fourteen asked whether the respondents are content about their work. As shown in table 14, most replied affirmatively.

Table 14: are the respondents content in their work?

Content	6
Most of the time	4
Sometimes	1
Sometimes discontent	0
Total	**11**

Finally, the respondents were given the opportunity to add suggestions they might have. One mentioned the necessity of better education, for both doctors and parents, while another offered the suggestion that screening in remote areas could most efficiently be carried out in a mobile way by employing a bus. Should it turn out that some remote rural areas are severely underrepresented in vision screening, screening by travelling nurses at kindergartens in rural villages could be considered.

While it is obvious that caution needs to be exerted when drawing conclusions from a small number of submitted questionnaires, there are nevertheless several interesting observations to be made:

- Most if not all family doctors in rural areas in Cluj County consider vision screening for children aged 4 – 5 to be (very) important. They also consider this the age group that is most in need of screening examinations.
- However, vision screening is not necessarily seen as the most important form of children's screening: vaccination is justly considered as most important. Vision screening ends up somewhere in the middle as far as importance is concerned.
- The doctors are positive that parents will accept free examinations and, if necessary, parents will purchase glasses and the children will wear these. This would of course mean that vision screening, once implemented, would be highly effective since its results would be followed up on. However, income levels in the communities have not been taken into account here.
- When it comes to who should perform vision screening, family doctors as well as nurses (family doctor's or kindergarten nurses) are seen as viable options, though nurses might need (more) additional training. As far as this additional training is concerned, most seem to prefer to take the course over two days during weekends.

[4] One respondent did not answer this question.

- Almost no one gave an answer when asked to estimate the costs of screening though; apparently it is not an easy task to do so. All respondents are in agreement when it comes to who should cover the costs, though: the Ministry of Health. This while vision screening is one of the few forms of paediatric screening that would be eligible for financing by the Ministry of Education.
- The respondents identify several problems are the most important ones affecting their practice. Most often mentioned were cost-related problems (too few tests reimbursed, personnel costs, low payments by insurance) followed by travelling to remote areas to see patients. Only four doctors mentioned the health care reform not being carried out.
- The percentage of Roma children in the respondents' practices is remarkably high: 17.9% on average, compared to 3.5% of Cluj County's population belonging to this ethnic group (according to official figures).

In conclusion, this small survey appears to indicate that implementing vision screening for all children in Cluj County will not be an altogether easy task. Key problems are lack of sufficient funds, less than optimal education, travel to remote areas, number of patients and the health care reform not being carried out.

There are, however, definitely indications that, if implemented in the right way, Cluj County would be fertile soil for a vision screening program: all family doctors agree vision screening is (very) important and they generally see the possibility of making a screening program work.

Appendix 2. Interim implementation study report on hearing screening in Albania

Neonatal hearing screening (NHS) started on January first, 2018 in four maternity hospital in three provinces in Albania: Tirana, Pogradec and Kukës. The screening programme has been implemented in two maternity hospitals in Tirana, one in Pogradec and one in Kukës. Both maternity hospitals in Tirana have a neonatal intensive care unit (NICU). Both Pogradec and Kukës are more rural and mountainous. Inhabitants live in very remote areas, they are more spread out across the country and parents have to travel much further to reach the maternity hospital.

Most deliveries (>97%) take place in a maternity hospital. The yearly amount of births in the selected maternity hospitals is expected to be around 11.500. Most of these deliveries take place in the two maternities in Tirana (10.000). About 700-800 infants are born in the maternity hospitals in Pogradec and Kukës each year. In general, mothers are discharged from the maternity hospital within 24 to 48 hours after giving birth. After leaving the maternity hospital, they do not return for any follow up appointments so it was decided that the first phase of screening takes place in the maternity hospital before discharge to increase coverage in the first phase.

Infants born healthy are tested using a three stage protocol, OAE-OAE-aABR, for infants admitted to NICU a two-stage sequence of aABR-aABR was chosen to be able to identify potential cases of auditory neuropathy. These protocols were selected for both maternity hospitals in Tirana and the maternity hospital in Pogradec. A different protocol has been selected for rural and mountainous Kukës, to limit repeat and referral, a test sequence of aABR-aABR was chosen.

Nurses and midwifes participating in the programme were trained. After training they were assessed by a test. Several training rounds were organised in order for the nurses to be taught about general knowledge concerning hearing and ears, to get accustomed with the screening devices and to practice communication skills. During screening they were supervised by a team from the University of Tirana to whom they can also direct all of their questions.

The awareness of parents about the importance of the screening programme and their acceptance is of great importance for the success of the programme. To inform parents about hearing impairment and the effect on the development of their infants, leaflets and posters were developed.

Each maternity hospital equipped a separate room for screening. This room contained a bed for the neonates to rest on while they were being screened as well as all devices needed for screening, consumables and a computer with an internet connection to upload all screening data and fill out the database. It was impossible to make acoustical improvements to these screening rooms. Therefore, quiet rooms were chosen, away from the busy corridors of the maternity hospital.

During the first weeks of screening, most screeners experienced difficulties performing the screening tests. They needed some time and had to gain experience. Many tests were paused and restarted. The referral rate during these first weeks of screening was very high. In some maternities up to 80% of infants were referred at the commencement of screening. The referral rates decreased to <10% as experience was gained.

During the first months some problems with the equipment occurred such as malfunctioning devices and broken probes. These probes and devices were sent back to the manufacturer for replacement but it took several weeks for the equipment to be returned. This did not delay screening. Screeners were instructed to be more careful handling the probes when transporting the devices.

A multidisciplinary team was trained, they will provide therapy to hearing impaired infants, help in the diagnostic assessment, fitting hearing aids and they will guide parents through the care pathway.

Appendix 3. Ethics report 'EUSCREEN'

By independent ethics advisor dr. Eline Bunnik, Erasmus MC, 25 June 2018

In this study, screening practices for visual acuity and hearing across 41 European countries are being compared with regard to cost-effectiveness. Two implementation studies are being conducted to set up screening programmes and help build capacity in Romania and Albania. A cost-optimization model will be developed to assist policy makers in low- and middle-income countries (LMICs) to decide what (type of) screening programme will be best and most cost-effective in the context of their health care systems to implement. This project thus contributes to an important aim: the introduction of screening programmes for vision and hearing in European countries that have so far lacked such programmes and the improvement and cost-optimization of existing programmes. The research project involves children aged 4-5 years (in Romania) and newborn babies (in Albania), but entails very little (if not negligible) risk for these research participants. Ethical issues will most likely pertain to the feasibility and social value of the research project and the sustainability of the programmes upon completion of the implementation studies.

I was asked to serve as an independent ethics advisor for this study. I am an assistant professor at the Department of Medical Ethics at Erasmus MC and a member of the research ethics review board, and am not involved in this study. I have read the ethics assessment of July 2016, the grant application of December 2016, and a deliverable detailing the informed consent and assent procedures for the implementation studies. I had an extensive meeting with Prof. Dr. H.J. Simonsz, who is Principal Investigator and main applicant of EUSCREEN and Tanya Bovenkamp-Ivanova, who is the project manager. We agreed that I will attend one or more research meetings in the period 2018-2020 to be able to monitor more closely any ethical issues arising within the project.

In the ethics assessment consensus report (CR) of July 2016, a range of ethics issues were identified and it was felt that details were lacking with regard to the implementation studies, informed consent, child assent, ethics approval, data protection and benefit sharing. Also, it was felt that the researchers were unclear about how children's health would be improved in case of detection of vision or hearing problems. In the Grant Agreement of December 2016, these issues were - in my view - adequately addressed.

In Romania and Albania, where the two implementation studies are conducted, informed consent processes have been carefully set up. Parents are given written information about the study and are referred to a video explaining the research in the local language and in a for parents intelligible fashion. Whereas in these countries, other screening tests are conducted on the basis of oral consent, parents are asked to provide written consent for the implementation studies. To the extent that this is possible, the 4- or 5-year old children are involved in decision-making about research participation. The partners in Romania have developed a video for the children, to explain to them the nature of the research study in which they are taking part and the research procedures they will be exposed to. This is a good and supererogatory way to ensure that assent from children is sought and obtained.

The implementation study in Romania is proceeding according to plan. Romania has a system of nurses employed at kindergartens who perform the visual acuity screening, and send the coded data to the central database. Nurses have now been trained to detect and treat vision problems (notably amblyopia) in children, which will help to prevent long-term visual impairment in Romania. However, the researchers have encountered difficulties with the implementation study in Romania. Most notably, children in rural regions of the country are not being reached and/or invited to participate in the screening. Reaching out to children in rural areas will - and must - be a priority for the researchers over the next couple of months.

In Albania, technicians were not accustomed to fitting hearing aids in very young children, as - because of lack of screening - most hearing problems have commonly been detected only later in children's lives. These technicians have received training as part of the EUSCREEN implementation study and are now competent in fitting hearing aids in newborns. Cochlear implants cannot be offered within the Albania health care system because of its low numbers of eligible patients. Though patients are sometimes referred for cochlear implants to surrounding countries, hearing aids are effective in most children. Screening and early treatment of hearing problems (with hearing aids) will help prevent delays in language and speech development and increase social participation for children in Albania. However, when newborns who do participate in screening but do not pass the hearing test, they are not always successfully referred to the central audiological centre in Tirana. Keeping doctors in Albania involved and motivated to take part in the implementation study.

Documentation related to ethics approvals has been submitted to the Commission. Data on percentage of children tested, acceptance and percentages of pass/no-pass results, are collected. The data are coded – and personally identifying information is removed - and transferred to a central database. This is addressed in the revised grant agreement and mentioned in the informed consent forms.

The researchers have established Country Committees for EUSCREEN in 41 countries, many of which are dedicated and motivated to contribute to the research in which details are gathered on screening practices in these countries. This requires a lot of work from respondents: the researchers estimate that it takes a full work-week to answer all questions in the survey. The Principal Investigator is committed to gathering the information in all 41 countries, and data collection is proceeding according to plan.