**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 July 1st, 2018 to December 31st, 2019

**Periodic report:** 2nd

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# History of changes

| HISTORY OF CHANGES | | |
|---|---|---|
| VERSION | PUBLICATION DATE | CHANGE |
| 1.0 | February 28th, 2020 | Initial version |
| 1.1 | May 27th, 2020 | Delays caused by Covid-19 summarised as addition to Chapter 5 |
| 1.2 | June 12th, 2020 | Months for re-examination of children in Cluj shifted for Covid-19 |
| 1.3 | June 18th, 2020 | Months for re-examination shifted, Int Orth Assoc congress postponed |

# 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 WP

## 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.

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' scheme providing information about the leading partners and partners responsible for carrying out tasks within each work package. A hierarchical structure of the scheme shows the relationships and the directions of information flows among the work packages.
- The Gantt chart determines the duration of the whole project, each work package, each task within it and the reporting moments.
- The description of work packages.
- 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 advice 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 24 and 30 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 work packages' 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.

The project coordinator was responsible for preparation of the first 36 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.

**Task 1.6: Organising meetings**

During the last 18 months, one consortium meeting was organised in Poznan (March 8th, 2019). 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: Aug 7, 2018, Sep 11, 2018, Oct 3, 2018, Nov 26, 2018, Dec 17, 2018, Feb 2, 2019, Apr 24, 2019, May 8, 2019, June 12, 2019, July 10, 2019, Aug 14, 2019, Sep 4, 2019, Sep 18, 2019, Oct 30, 2019, Dec 4, 2019, Dec 18, 2019.

When visiting conferences to present the study, small gatherings were organised to update Country Representatives.

Within Rotterdam the team aimed to meet every two weeks.

**Task 1.7: Risk Management**

Thanks to regular communication with the work packages' 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.

One unforeseen risk has arisen: in the County of Cluj most of 104 nurses who screened children have done so very well, but 4 nurses have together examined 799 children, each of them more than 100 children, but not referred any children, with a prevalence of amblyopia of 3.25%. The chance that this occurs naturally is (BINOMIAL_CDF), p = 0.0005, well below alpha = 0.05. A solid plan of corrective measures has been made by the Cluj Team (UMF and DASM) that is described in detail in 5.1 Deviations.

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

**Task 2.1: Data collection with the questionnaire**

In the second half of 2018 and first half of 2019, the data collection continued with the extensive questionnaire for inventory of vision, hearing and general screening programmes in 41 EU member, (potential) candidate and associate countries.

Data collection took longer than initially planned. It proved difficult to find competent Country Representatives (CRs) who were willing and able to fill out the long and detailed questionnaire about the current screening programmes in their countries or regions. This time-consuming task stagnated in many cases, leading to a delay in the data collection process. Faced with this delay, EMC improved the relationship with the CRs by cultivating a personal relationship through detailed emails discussing their partially filled-out questionnaires. This worked well, but was also time-consuming. Additionally, personal encounters at congresses were arranged whenever possible.

After thus intensifying the relationship with the CRs and – in some cases – recruiting new or additional ones to provide missing data, most of the required data were nevertheless collected by the end of 2018. The final data were collected in the first half of 2019. The data submitted by the CRs were collected and stored in a secure database (see 2.4).

**Task 2.2: Maintaining the network**

When available, three candidate CRs, ophthalmologists, orthoptists, otolaryngologists, paediatricians, audiologists and other experts, from 41 EU member, (potential) candidate and associate countries were personally invited to register as CR on the website and to fill out the extensive questionnaire. When no candidates had registered for vision, hearing or general screening in a country – something that often occurred - new contacts were found through existing CRs, national societies, experts who published papers about vision and hearing screening, and so forth. This proved a labour intensive task that took much time and caused a considerable delay.

Also in many cases, filling out the questionnaire stagnated. For practising ophthalmologists and otorhinolaryngologists, for example, it was often too difficult or too time-consuming to get all the data, for instance on salaries of professionals who (could) perform screening.

The Annual Meeting in 2019 in Poznan (see 2.5) also substantially contributed to the maintenance of the network of CRs by boosting their enthusiasm for the project and reaffirming their commitment.

**Task 2.3: Survey on vision, hearing and general paediatric screening**

Participating countries in the EUS€REEN Foundation were 41 EU member, (potential) candidate and associate countries: Albania, Austria, Belgium, Bosnia and Herzegovina, Bulgaria, Croatia, Cyprus, Czech Republic, Denmark, Estonia, Faroe Islands, Finland, France, Germany, Greece, Hungary, Iceland, Ireland, Israel, Italy, Kosovo, Latvia, Lithuania, Luxembourg, Malta, Moldova, Montenegro, the Netherlands, North Macedonia, Norway, Poland, Portugal, Romania, Serbia, Slovakia, Slovenia, Spain, Sweden, Switzerland, Turkey and the United Kingdom.

CRs from several other countries also registered on the website and filled out the questionnaire, because they were keenly interested in participating in the EUSCREEN Study. Five of these countries managed to submit complete data for at least one of the domains: China, India, Malawi, Russia and Rwanda. Funds in the EUS€REEN Foundation that had remained unused, were used to remunerate the CRs in these countries outside Europe who completely filled out the questionnaire. The remuneration of all eligible CRs was completed in the first half of 2019. CRs were paid

€2,000,- if they completely filled out the questionnaire and after validation of the submitted data. In cases where more than one CR filled out a questionnaire, the remuneration was split proportionate to the amount of work done by each CR. This was arranged in consultation with the CRs involved and did not lead to any problems.

For the development of the cost-effectiveness model in the EUSCREEN Study the participation of CRs from countries outside Europe was immensely profitable, as there are no Low Income Countries in Europe and the project aims to develop a model that can also predict the most cost-effective vision and hearing screening programmes in Low Income Countries (LICs) and Lower Middle Income Countries (LMICs).

Thanks to the enormous effort involved in collecting data from all the aforementioned countries, the project has succeeded in amassing the largest data set about vision and hearing screening ever. This data set serves as a solid basis for further development of the cost-effectiveness model.

**Task 2.4: Database management**

Submitted questionnaires were stored on a secure SQL database server and backed up once a day on a backup server. The answers were exported to a spreadsheet by the website administrator. This spreadsheet could be accessed by all partners involved in data collection and validation (EMC, USFD, READ and KI).

Occasional technical problems occurred during data collection. Some CRs encountered problems trying to log in on the website while others lost answers they had submitted. These problems were addressed and solved.

**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 were collected until the summer of 2019. By the summer of 2019, complete questionnaires had been submitted by 45 countries for vision screening, 45 for hearing screening and 42 for general paediatric screening. Based on the submitted questionnaires, Country Reports were completed for 45 countries/regions for vision screening, 47 for hearing screening (for two countries, two reports were completed for different regions) and 40 for general paediatric screening.

On March 8th, 2019, the Annual Meeting of the EUSCREEN Study was held in Poznan. The Annual Meeting was open to all CRs. We reported back to them the results of all countries, and the analysis and review of the vision and hearing screening programmes in all countries across Europe.

The meeting started with plenary morning and afternoon session with presentations from all Consortium Partners of the EUSCREEN study as well as four presentations by CRs. All presentations contained a lot of new and interesting information for all attendees and led to interesting talks. People from different countries and domains (hearing, vision and general paediatric screening) interacted and exchanged thoughts on the subject. Thereby the meeting contributed to cross-border communication on the subject of screening.

The meeting concluded with simultaneous sessions on the cost-effectiveness model: a demonstration and try-out sessions with practical exercise for both hearing and vision screening. These sessions were very informative and effective for everyone involved. Valuable feedback from CRs was collected to improve the model. A full report of the meeting can be found here.

The day before, on March 7th, 2019, a meeting of the consortium partners took place. All partners presented the preliminary results of their respective work packages and the project's progress was discussed. This meeting was combined with the project's mid-term review meeting and was therefore also attended by the EU project officer and an independent expert.

**Task 2.6: Dissemination**

The dissemination of the project was realised through the CRs (three per country who are members of their respective Country Committees) and through scientific media. In the EUSCREEN project seven partners in six European countries participate but also, and more importantly, CRs from 46 different countries.

Dissemination through 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 ten presentations were given by EMC at various conferences. Preliminary results of the project were also disseminated at the meeting in Poznan (see 2.5).

Additional dissemination was realised through the publication of project news on the website. Also, all general and vision screening country reports were published on the website. The hearing screening country reports will follow shortly.

The final and most important part of dissemination will be the cost-effectiveness model, wrapped in the TOOLKIT that is currently being developed (see WP5 and WP8). A preliminary version of the model has been made available for testing by CRs in October 2019. Since then, 46 CRs have registered on the model website, tested the preliminary version and provided useful feedback, that will be used to further develop the model in 2020.

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

**Task 3.1: Mapping and documenting existing vision screening provision**

University of Sheffield

Mapping and documenting existing vision screening provision has been completed. Final country reports were submitted to WP2 for the following countries:

1. Albania
2. Austria
3. Belgium
4. Bosnia
5. Bulgaria
6. Croatia
7. Cyprus
8. Czech Republic
9. Denmark
10. England and Wales
11. Estonia
12. Faroe Islands
13. Finland
14. France
15. Germany
16. Greece
17. Hungary
18. Iceland
19. Israel
20. Italy
21. Kosovo
22. Latvia
23. Lithuania
24. Luxembourg
25. Malta
26. Moldova
27. Montenegro
28. Netherlands

29. North-Macedonia
30. Northern Ireland
31. Norway
32. Poland
33. Republic of Ireland
34. Romania
35. Scotland
36. Serbia
37. Slovakia
38. Slovenia
39. Spain
40. Sweden
41. Switzerland
42. Turkey

Further country reports were completed for countries outside of the EU (China, Russia, India, South Africa, Rwanda, and Malawi). This work was not anticipated as part of the EUSCREEN project, however the results will be useful to inform the model (developed by WP5). The reports have been uploaded to the EUSCREEN website (https://www.EUSCREEN.org/country-reports/) and in the White Rose repository. (http://eprints.whiterose.ac.uk/).

Three UK specific reports have been compiled using data submitted to the British and Irish Orthoptic Society (BIOS) Special Interest Group. Permission was sought and approved from BIOS to provide raw data to WP5 for the model development and calibration. The reports are available from:

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

https://figshare.com/articles/BIOS_Screening_Audit_report_2016-2017/6839813

https://figshare.com/articles/BIOS_VISION_SCREENING_AUDIT_Academic_Year_2017-2018/10282781

A summary report of the EU screening data has been completed and was submitted to WP2 on 28th June 2019. This was an abridged version to avoid publication of the full dataset before peer review and formal publication.

An abstract has been submitted to the International Orthoptic Congress (IOC) led by University of Sheffield. Details are provided below.

**Title:** Frequency of childhood visual acuity screening in Europe

**Authors:** Griffiths HJ[1], Carlton J[1], Mazzone P[1], Horwood AM[2], Nordmann M[3], Simonsz HJ[3].

**Institutions:**

1. University of Sheffield, United Kingdom

2. University of Reading, United Kingdom

3. Erasmus Medical Centre, Rotterdam, The Netherlands

**Task 3.2: Literature review of the impact of vision screening**

University of Sheffield

Due to the complicated process of data collection and validation, the window for data collection and validation required extension. There was a delay in aggregating and validating sufficient data from all countries. Therefore, there was a 6-month delay for Work Package 3 to deliver Country Reports. All Country Reports were submitted before end of December 2018. An overall summary report on vision screening has since been completed and submitted in June 2019, to fulfil the requirements of the deliverable for WP3 (Deliverable 3).

Subsequent progress on literature review of the acceptability of childhood screening programmes (Milestone 5) was significantly affected by delays experienced in collecting and validating data from the Country Representatives. The unexpected number of reports (including countries outside of the EU) further impacted upon the completion of the review. A draft manuscript has been circulated internally for comment (with a view for submission for consideration for publication in 2020) to *Journal of Medical Screening,* and Milestone 5 is expected to be reached in the Summer of 2020.

An abstract has been submitted to the International Orthoptic Congress (IOC) led by University of Sheffield. Details are provided below.

**Title:** Acceptability of childhood screening programmes

**Authors:** Carlton J[1], Griffiths HJ[1], Mazzone P[1], on behalf of The EUSCREEN Foundation[2]

**Institution:**

1. University of Sheffield, United Kingdom (Corresponding author: j.carlton@sheffield.ac.uk)
2. Erasmus Medical Centre, Rotterdam, The Netherlands

University of Reading

The input from Professor Horwood at the University of Reading was primarily in respect of her expertise in photoscreening. There are plans to add photoscreening to the MISCAN model, so that the costs of photoscreening can be compared with other methods. The main input of Reading has been a systematic review of the photoscreening literature, specifically addressing the quality of evidence of the scope of use, costs and cost-effectiveness of photoscreening in relation to other screening modalities such as visual acuity screening which is being adopted in Romania, and which is established in many countries. This literature review has shown that although photoscreening is being widely marketed and used, and there are many papers describing research and community projects of different sizes, evidence of its relative costs and cost-effectiveness is weak. The EUSCREEN country reports have shown that the impression gained from the literature does not reflect the actual adoption of photoscreening in most of the 25 countries where it is used. It is usually used as an adjunct to visual acuity screening, rather than as a stand-alone test as suggested by the literature. Comparative costs are very rarely reported. This has confirmed the potential value and novelty of the MISCAN model.

A paper has been submitted to “Eye” and is under review.

**Title** Scope and costs of autorefraction and photoscreening for childhood amblyopia – a systematic narrative review in relation to the EUSCREEN project data

**Authors** Anna M Horwood, Helen J Griffiths, Jill Carlton, Paolo Mazzone, Arinder Channa, Mandy Nordmann, Huibert Simonsz, on behalf of The EUSCREEN Foundation.

A presentation was made of the work at the European Strabismological Association Conference in Helsinki in June 2019, and an update has been submitted for presentation at the final Vision and Hearing Screening Conference for all Country Representatives and Consortium Partners (Deliverable 1 & Milestone 11), scheduled for November 13-14th, but to take place on May 21-22nd if a 6-months extension of the EUSCREEN Study for the Covid-19 pandemic will be granted.

Professor Horwood has been collaborating with members of WPs 5 and 6 in the development of the vision screening programme in Romania and with developments of the model. She went to Romania in January 2019 as part of the central project monitoring and support process for the Cluj team. She has also been working on the planning and preparatory stages of the Manual with a Strategy for Implementation.

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

**Task 4.1: Mapping and documenting existing hearing screening provision**

- Responses to the questionnaire were aggregated by WP2 and delivered to WP4 for analysis. As of July 1, 2018, responses from 15 countries or regions had been delivered and scrutinized, with 9 countries/regions having returned clarification questions after verification/validation.
- Since July 1, 2018, the questionnaire was sufficiently completed by experts in an additional 32 countries/regions. A grand total of 40 of the 41 originally selected countries responded, plus 5 additional countries.
- When it was not possible to collect data on a national level (due to decentralisation of the screening programme), regional data were provided. In two countries, multiple regional experts filled out the questionnaire. Plus, five additional countries outside the original 41 selected countries responded to the questionnaire. In total, 47 countries/regions responded.
- We validated data for the remaining countries/regions that responded since July 1, 2018 (Task 4.2)
- An part-time audiologist was engaged to help with the process during the maternity leave of Allison Mackey (September 2018-March 2019).
- Data from a total of 47 countries/regions were entered into a database and scored based on the level of accompanying evidence provided (Task 4.2).
- We analyzed newborn hearing screening data using key methods:
    - Descriptive reports and tables
    - Descriptive statistics
    - An agglomerative hierarchical cluster analysis across 5 key elements of protocol design
    - Risk ratios/chi-square analyses of referral and follow-up rates across programmes by types of protocol and programme parameters.
- Two manuscripts are in final stages of preparation.
- A combination of factors contributing to a prolonged data collection period resulted in a subsequent delay in the analysis and dissemination of data. The 30-month deliverable of report synthesizing the cross-EU findings of existing hearing screening programmes was submitted. This was an abridged version to avoid publication of the full dataset before peer review and formal publication.
- In the meantime, a detailed description of the methodology performed and basic information about the results received, in addition to detailed country reports from 47 countries and regions, were delivered before the 30-month deliverable deadline.
- A Summary Report of cross-EU findings (according the deliverable description) was submitted on June 27, 2019, before the deliverable deadline, with details of the data collection and validation of hearing screening data. Preliminary findings were also described, as well as a discussion of the implications.
- A Supplement to the Summary Report was delivered to add a description of the more detailed findings of the hearing screening results from the EUSCREEN questionnaire for both newborn and preschool hearing screening.

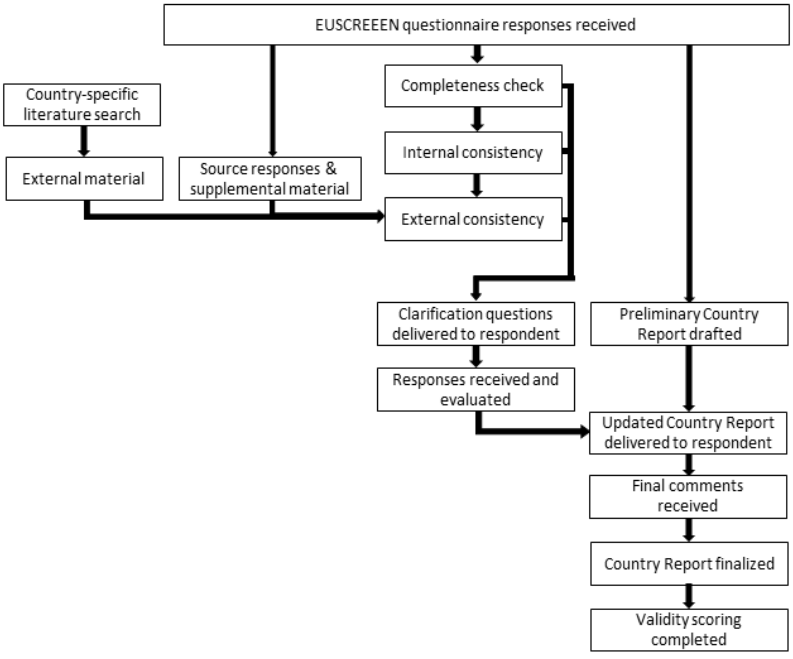
Dissemination relating specifically to Task 4.1 since July 1, 2018

- March 2019 (Poznan, Poland): A meeting held for Country Representatives to present and discuss interim findings. WP4 presented on the protocols used across participating countries/regions, in addition to a presentation relating to Task 4.2.
- May 2019 (Lisbon, Portugal): At the European Federation of Audiological Societies, the EUSCREEN project held a spot in the pre-conference programme in which hundreds of delegates were present. WP4 held three presentations, two of which relate to Task 4.1. (a) a description of the existing hearing screening programmes across countries according to country-specific information such as infant health care, income and spending; (b) a summary and analysis of the key outcome measures and relationships to protocol design.

**Task 4.2: Reliability and validity of hearing screening data**

A description of the verification and validation process is illustrated in the following figure. Specifically, responses received by WP2 were delivered to WP4. We checked each response for completeness, as well as for internal and external consistency. We used two sources for performing an external consistency check. The first was from the sources provided by the Country Representative. The second was from a comprehensive search of the literature (including grey literature). We translated all relevant material via Google Translate that was used for cross-checking with responses provided.

We drafted a preliminary version of the Country Report and a list of clarification questions for each country/region from the consistency check, that was delivered back to the Country Representative (via WP2). Once responses were received on the clarification questions, the Country Report was updated and sent to the Country Representative for final confirmation or additional remarks if needed.




Once all Country Reports were finalized, data were transferred from the reports to a master database. Data were divided into descriptive information (protocol design, programme parameters, etc.) and outcome/results (coverage rates, referral rates, compliance rates, prevalence, etc).

- Descriptive information was determined valid once the cross-checking/clarification/confirmation procedure in the flow-chart was considered completed. In addition, the vetting process of the Country Representative by WP2 was vital to assuring descriptive information was considered valid.

- Outcome/results were more difficult to validate. We implemented a scoring method on outcome data based on information provided about the data and the source used to verify. The scoring method is presented in the table below. Data that were not considered valid were not included in the final analyses; however, descriptions of the percentage of programmes providing valid/non-valid data were disseminated, as this is informative to the issues surrounding quality assurance and the use of outcomes for monitoring the performance of screening.

| Minimum criteria for valid values | Examples of values considered not valid |
|---|---|
| ✓ Actual data / calculation | × Rough estimations |
| ✓ Representative of target population | × Pilot study or data from before 2014† |
| ✓ Large sample size* | × Extrapolated from other population |
| ✓ Recent (data from 2014 or later)† | × Small sample size* |
| ✓ Consistent with literature / source | × Data inconsistent with literature or source and could not be clarified |

* Sample size criteria varied based on the outcome measure (i.e., coverage rates included despite the sample size; referral rates were not included if sample size was < 1000; detection rates were not included when the sample size was <5000).

† Date of data collection was not included as a validity criterion for detection rates, so long as data were reflective of the NHS programme.

Dissemination relating specifically to Task 4.2 since July 1, 2018

- October 2018 (Cape Town, South Africa). At the World Congress of Audiology, one presentation was held by WP4 on the findings related to the issue of successful quality assurance and outcome monitoring of newborn hearing screening programmes.
- March 2019 (Poznan, Poland): At the EUSCREEN conference for Country Representatives, WP4 presented on the barriers of quality assurance and monitoring of screening programmes.
- May 2019 (Lisbon, Portugal): At the European Federation of Audiological Societies, WP4 presented a synopsis of the available quality outcomes measures available by Country Representatives, in addition to discussion topics on improvements in which data are collected and used for monitoring performance.

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

Since July 1, 2018, we have started a systematic literature review on the outcomes of newborn hearing screening by programme and protocol variables, for both well- and at-risk infants. The literature review will aim on the performance of newborn hearing screening, and identify factors contributing to variability in its success. Specific outcomes targeted are referral rates (false positives) and follow-up rates. We will look at variations in these outcomes from an international perspective. The results of the systematic review will be inputted as default values to the cost-effectiveness model and used to inform the TOOLKIT.

- We have consulted with librarians at Karolinska Institutet Library and an extensive literature search was performed across 5 databases, with no restrictions on language or date.
- A medical resident, Valeria Del Vecchio was engaged to help with screening and sorting articles.

- Over 11,000 articles were found, many being duplicates. We screened a resulting 5500 articles by title and abstract. Over 2000 articles were included based on title/abstract. Any article pertaining to newborn hearing screening was included, as the targeted outcomes may be only part of the results in the full text.
- We read and sorted over 2000 articles based on detailed pre-determined inclusion/exclusion criteria.
- After sorting was completed, a data extraction template was drafted and data extraction has commenced.
- The systematic review has been submitted to Prospero (October 2019) and is awaiting publication.

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

**EMC Department of Public Health**

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

Deliverable 5.1 has been achieved in December 2017. Micro-simulation semi-Markov models were developed using input parameters on demography, natural history of amblyopia, screening characteristics and treatment. Parameter values were obtained from observed data (for vision screening), literature review and expert discussions. Model calibration was performed using a Nelder and Mead optimization process.

**Task 5.2: Calculating cost-effectiveness in MLIC**

Deliverable 5.2 has been achieved in December 2017. 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 and the cost-effectiveness for various programmes was calculated.

**Task 5.3: Determining the optimal screening programmes**

Overview

After December 2017, further refinements to the models were made as a continuous process. We further developed the microsimulation model as a generic tool for policymakers. This deliverable is in line with the activities of WP8 (development of TOOLKIT comprising of a cost-effectiveness modelling framework and a strategy plan for implementation).

The two previously developed micro-simulation models for vision and hearing screening were transformed into a user-friendly web based version to be used by local experts. During multiple working group sessions we developed a conceptual framework, resulting in a pre-module and input modules to support the MISCAN core model. Figure 1 shows the general structure of the first version of the webtool.

A software company has developed the interface of the webtool. The webtool is available at miscan.EUSCREEN.org. People can only use the tool after registration. About 50 people have used this webtool.

Content of the first version of the webtool

A pre-module has been developed to evaluate the appropriateness, acceptability and sustainability of the implementation of a vision or hearing screening program, which is especially useful for low-income countries. In this pre-module users had to enter the values of 12 WHO indicators (for example the prevalence of undernourishment and the DTP 3 immunization coverage among 1-year olds) for their country. Based on their answers an advice was given to whether or not screening may be appropriate, acceptable and sustainable.

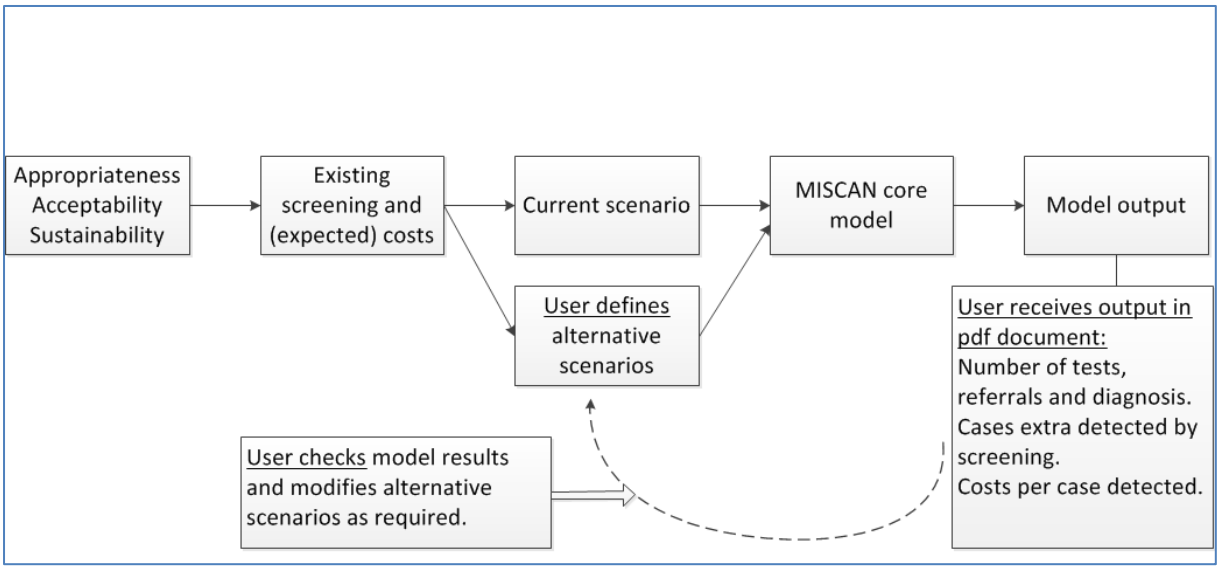



**Figure 1. EUSCREEN webtool model overview**

If the user continued, he/she could enter the country, number of children born each year and existing screening program, current general pediatric health care and educational systems. Next, the user defined alternative scenarios, including the expected (or monitored) attendance and referral rates and costs of salary, equipment, training, start-up and diagnosis. The input was restricted as much as possible to positive numbers, percentages and drop down menus for text to prevent errors in the calculations. After the input was completed, the model was running for about one minute per screening scenario and then presented the outcomes of the tool for the current scenario as well as the alternatives. These outcomes were the annual number of tests, referrals to diagnosis, detected by screening, number of persistent amblyopia prevented, and costs of screening and diagnosis. These results were shown on the screen as well as in a PDF and the user received an email that the run was ready. The PFD's of the results were stored under the user's account and could be easily accessed again later by the participants.

Technically, the input users entered was copied to csv files. These csv files were read by an R-script, which generated the input files (one for each scenario) for the MISCAN model and started the model runs. The results of the MISCAN model were read by another R-script which also used the input by the users to generate the final results in a csv file. This csv file is presented on the screen and copied into a PDF.

The first version of the webtool was ready in February 2019 and was tested at the workshop in Poznan in March 2019 by a group of Country Representatives during three separate workshop sessions. In total, 39 people practiced with the tool themselves during the workshop (22 for hearing and 17 for vision). The other participants attended a demonstration session. A total of 15 out of the 39 (38%) participants filled out an evaluation form of the tool (6 for vision and 9 for hearing). The evaluations of these first experiences of working with the cost-effectiveness model are compiled in Appendix 5.

Overall, the session was rated 4.2 on a scale from 1 (strongly disagree/bad) to 5 (strongly agree/very good). The usefulness for various stakeholders (professionals, policy makers, coordinators and researchers) was rated 4.0. The lay-out of the tool was logical and user-friendly was rated 4.0. The module for entering the costs was seen as most difficult and was rated 3.4. Further feedback on the tool was received by open questions and verbally during the sessions. This feedback has been considered in developing the second version of the webtool.

Content of the second version of the webtool

The second version of the webtool has been developed in September-December 2019. The pre-module has been simplified by reducing the number of questions from 12 to 9. Most simplifications were in the cost module. Because in this version of the tool the user first choses the screening program to be simulated and then enters the costs, only the salaries of the professionals involved in the chosen screening program have to be filled-out. Training costs were omitted in this version. In addition, only one scenario per run was allowed. The test sensitivity and specificity per test were added to the input. In the previous tool the sensitivity was included in the tool as fixed values by age and the referral was used instead of specificity. Also the attendance at diagnostics was added (this was 100% in the previous tool).

The preliminary second version of the input of the model was ready October 31, 2019 and the link to the tool has been send around to all consortium members and country representatives to provide comments. One week later the webtool was also running. The webtool is available here. People can only use the tool after registration. So far, 60 people (including 46 CRs) have registered and many have (extensively) tested the model and provided valuable feedback for the further development of the model. The comments they provided were discussed weekly, frequently leading to updates of the tool. The questions and answers were added to a log file on the EUSCREEN website.

Furthermore, a paper describing the cost-effectiveness analysis for hearing screening in Albania has been submitted and a revision is being drafted.

# 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):**

**DASM and UMF**

This was undertaken in the previous reporting period.

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

**DASM and UMF**

This was undertaken in the previous reporting period.

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

**DASM and UMF**

This was undertaken in the previous reporting period.

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

**DASM**

1. Regular and exceptional meetings of the DASM core team
2. Participation of the DASM project manager to the UMF's project team meetings when needed/ upon request.
3. Regular meetings & contact (via What's App) with the DASM nurses
4. Monthly timesheets of the nurses and core project team
5. The forms that the nurses fill in are collected and verified by the DASM medical coordinator
6. The project manager is monitoring the data input in the database.
7. EUSCREEN webmeetings.
8. The screened cases were uploaded to the database.

**UMF**

1. Weekly meetings of the UMF Cluj team: establishing directions for actions, employing the members of the team, fixing the purchases plan

2. Regular meetings with local partners

3. The activity of the hired personnel is documented via timesheets.

4. Maintaining a database containing the eligible personnel for the project.

5. The forms that the nurses fill in are collected and verified by project team members.

6. Project team members have actively contributed to the optimisation and the functionality testing of the database.

7. The national coordinator, the project manager and the project team members are monitoring the data input in the database.

8. Keeping evidence of the children that have been referred to an ophthalmologist and keeping track of the forms received from the ophthalmologist.

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

**DASM, Goethe and DASM**

This was undertaken in the previous reporting period.

### 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 and UMF**

This was undertaken in the previous reporting period.

### 6.2.2 Distribution of the course participants into groups

This was undertaken in the previous reporting period.

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

This was undertaken in the previous reporting period.

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

**UMF**

Prof Vladutiu and the researchers taught the travelling nurses all the needed notions for measuring the visual acuity in children. During the teaching sessions a great importance was given to the practical skills of the nurses.

The researchers employed in the project (Dr O. Teodosescu, Dr R. Ursu) have constantly offered their support to rural healthcare professionals for the good unfolding of the screening (during rural visits together with the nurses they have examined children, they offered telephone advice whenever requested).

Furthermore, Dr Teodosescu and Dr Ursu accompanied the two travelling nurses in their first visits in rural kindergartens in order to offer their support and expertise and to make sure the nurses apply correctly what they've been taught and that the examinations are done flawlessly.

From January 16 to January 23, 2019 the EUSCREEN Cluj team together with the international partners (Maria Fronius, Anna Horwood, Mandy Nordmann and Jan Kik) organized several visits in different locations from Cluj county, in urban area and in rural area: urban kindergartens, rural kindergartens and rural family doctor's offices. Also, there were extensive talks with DASM and UMF professionals responsible for the implementation. These visits were carried out in the county seat Cluj-Napoca, the two smaller cities Campia Thurzii and Gherla and twenty rural communes throughout the county.

From October 9th to October 18th 2019 EUSCREEN Cluj team together with the international partners (Mandy Nordmann and Jan Kik) organized several visits in 21 different locations from Cluj county, in urban area and in rural area: urban kindergartens, rural kindergartens and rural family doctor's offices (Huedin, Campia Turzii, Turda, Dej, Negreni, Tureni, Viisoara, Luna, Apahida, Alunis, Vad, Vultureni, Aschileu Mare, Aschileu Mic, Cristorel, Iara, Sacel, Tritenii de Jos, Camarasu, Cojocna, Fizesu Gherlii). During the trips to Turda and Dej Mandy and the Cluj team met and discussed with two of the ophthalmologists from these towns that examined children referred by the nurses in the EUSCREEN project. Meetings have been arranged with representatives of the Cluj county Public Health Institute, representatives of the Cluj-Napoca Mayor's Office and of UMF Cluj. On October 14th 2019 a focus group has been organized at UMF offices with 12 nurses involved in the screening project attending; several aspects regarding the difficulties of screening have been discussed. On October 18th, 2019 a meeting with the travelling nurse was organized.

Also, each month when collecting and verifying the forms, there are discussions with the medical personnel aimed at clarifying issues that have been identified during the screening activity.

**Goethe**

During a visit to Cluj county in Jan. 2019 Prof. Fronius gave a presentation on appropriate visual acuity testing. Questions of correct vision testing were discussed during visits to general practitioners' offices in rural areas.

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

**DASM**

The directors of the schools and kindergartens continued to organise meetings with the remaining parents of the children involved in the study. They offered information regarding the benefits of the study, what the study involves, the need and the importance of the parental consent. All the parents were informed about the location where the screening will take place, about the persons who will be involved in these actions.

Each parent was provided by the DASM nurses with the informed consent and an info sheet containing details about the project as well as a leaflet. In cases when 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.

The children from each kindergarten were screened in the medical office, with the materials which were made available by the UMF and DASM, following a planning which was carried out, by age groups. Results were then included in a data base and kept strictly confidential.

Regular individual meetings take place every month when the nurses came at DASM headquarters.

**UMF**

1. The project team kept in touch with the medical staff from the rural area. Discussions with the nurses in order for them to examine the children from the nearby villages where the medical staff working there did not wish to get involved in the EUSCREEN project (example: the nurse from Jucu went to the communes of Bontida and Rascruci and examined children).

2. In order to boost visual screening in rural areas we hired 2 travelling nurses with the aim of increasing the number of screened children from the rural areas. The first travelling nurse was hired in March 2019 and the second one in

May 2019. The second travelling nurse, that we hired at the beginning of May 2019, quit her job on June 3rd. She explained in her resignation letter that she found the job too difficult, and the fact that there are serious discrepancies between the estimated number of children from a village and the actual number of children attending the kindergarten is for her a major obstacle in ensuring a decent income.

3. The project team contacted rural kindergartens to ask for permission and support in order to organize the screening in the kindergartens.

4. The project team (especially Dr Mara - responsible of the rural screening) has made numerous visits to rural medical practices and to kindergartens to supply the materials needed for screening and to boost the screening activity.

5. At the proposal and with the agreement of the study coordinator it was decided to increase the payment per examination to 10 euro starting September 2019 for the children examined in the rural area by the travelling nurse. Starting with November 2019 the payment per examination was increased to € 14 for the communes in which no screening had been done up until that moment. These changes were proposed in order to increase the number of rural children examined in the project and to increase the rural coverage.

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

**DASM and UMF**

This was undertaken in the previous reporting period.

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

**DASM and UMF**

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

### 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**

a. The National Congress of School Medicine, Cluj-Napoca, April 2019: " ***The school doctor's approach in the main ophthalmological conditions of the child***– Prof. Dr. Cristina Vlăduțiu – *Amblyopia. Early detection. EUSCREEN* ***Study***– Prof. Dr. Cristina Vlăduțiu, Senior Lecturer Dr. Simona Căinap, Dr. Mihai Mara, Dr. Daniela Rajka, Dr. Simona Sevan, Dr. Oana Teodosescu, Dr. Raluca Ursu – UMF Cluj-Napoca. The presentation was held in front of 200 school doctors and 200 school nurses from 30 counties from Romania.

b. School Medicine Forum - November 27th 2019, Timisoara - EUSCREEN project of early detection of visual problems in children - Prof. Dr. Cristina Vlăduțiu, Senior Lecturer Dr. Simona Căinap, Dr. Oana Teodosescu, Dr. Raluca Ursu, Dr. Daniela Rajka

**Goethe**

Jan. 2019: As it was difficult to recruit vision screeners mainly in rural areas, Prof. Fronius together with the UMF Cluj team and EUSCREEN members from the Netherlands and Prof. Horwood (Univ. Reading) visited kindergarten nurses and general practitioners' offices in the countryside to convince them to join in the study. A report about the special challenges in the rural areas was written together with the colleagues (Appendix 1.1).

### 6.3.4. Supplying the necessary equipment for the development of the screening

**DASM, UMF and Goethe**

This was undertaken in the previous reporting period.

### 6.3.5. Visual acuity measurement in children from the target group

**DASM and UMF**

From January 1st 2018 to December 31st 2019 a total number of 12906 children have been examined:
- 6475 in Cluj-Napoca by DASM
- 3181 in the smaller cities
- 3250 in the rural area

Seeing that not all the forms have been introduced in the database yet, minor differences might appear when the database in completely filled.

Important mention: an important number of children that live in the rural area attend kindergartens in the urban area. Up until June 30th, 2019 the Cluj project team identified a number of 951 children from the rural area that were examined in the urban kindergartens (723 examined in kindergartens from Cluj-Napoca, 175 in the smaller cities and 53 in private kindergartens). In 2020, after all forms will be introduced in the database, these numbers will be updated.

To sum it up, the total number of children from the rural area that have been examined in the EUSCREEN project is 4201.

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

a. A total of 1466 children were referred to an ophthalmologist, as follows:
- 867 children from Cluj-Napoca by DASM
- 263 children from the smaller cities.
- 299 children from the rural area

b. Up until 13.08.2019 a total number of 217 forms were received from the ophthalmologists, as follows:
- 173 in Cluj-Napoca
- 34 in the smaller cities
- 10 in the rural area

c. At the end of each month the project team collects the screening forms from the medical staff involved in the screening process.

d. Each form is carefully verified by the project team and only afterwards introduced in the database.

**Goethe**

Prof. Fronius was involved in monitoring of the vision screening process, following the data analyses and proposing actions for solving difficulties. She participated in writing a report (Appendix 1.1) and preparing a manuscript for a publication about the results of the first year of vision screening.

**EMCawss**

EMC has carried out an implementation study concerning the vision screening programme in Cluj. In the reporting period, two extensive visits were made to Cluj in order to make on-site observations for this implementation study, in January 2019 (together with Goethe and UREAD) and in October 2019 (these visits are also described in 6.2.4 by UMF). Based on the on-site observations, as well as analysis of the project database, EMC is finalising a first article on the implementation study and preparing a second, in cooperation with partners DASM, Goethe, UMF and UREAD. Full reports of the aforementioned visits are attached as Appendix 1.

Analysis of the data of all screened children in the project database by EMC shows that the target (screening two birth years in 2018-2019) was reached in both Cluj-Napoca and the small cities (107% and 102% of the target, the theoretical maximum was 150% as three birth years could have been screened in a period of two years).

In the rural areas, 63% of the target was reached. It should be noted, though, that many children were screened in the rural areas in 2019. Initially children were to be screened by family doctors' nurses at the doctors' offices. This did not work, according to the nurses because parents would not bring their children to the doctor's office for screening. In August 2018, nurses were therefore advised to visit the rural kindergartens to screen the children there instead. Although this worked better, after the first year of the implementation still only 24% of the target had been screened in the rural areas (both Cluj-Napoca and the small cities were at 74% at that point). In March 2019 therefore a travelling nurse was hired to visit rural kindergartens and screen children there. The travelling nurse managed to screen 805 children in the rural areas. A full report of the data analysis is attached as Appendix 2.

The on-site visits focused on the rural areas. Through numerous interviews with doctors, nurses, kindergarten staff and other professionals, barriers and facilitators were identified. The main findings of these visits were that screening went well in Cluj-Napoca and the small cities, but implementation was difficult in the rural areas. This has various reasons: nurses working for family doctors in rural areas lacked time to screen and do the necessary paperwork and parents were unwilling to bring their children to the doctor's office for screening. Screening by the nurses at the rural kindergartens went better, although the numbers of eligible children attending the kindergartens were low (many children were said to be abroad with their parents or not to attend often even when enrolled).

A few issues require attention not only in the rural areas but also in the cities: it is not possible to say much about the quality of the screening, largely due to a lack of data on follow-up. Of only 15% of referred children, a diagnostic report of an ophthalmologist who examined the child was entered in the database. Of the other 85% of referred children, it is not known whether the results of their examinations were not reported back or whether they did not see an ophthalmologist.

Data analysis did lead to the discovery of an issue with four nurses who screened more than 100 children each and did not refer any children. One of these nurses examined 86 children in 2018 and, according to the database, all these children had the exact same vision in both eyes. These results are highly unlikely. A detailed description of this issue can be found in Appendix 3.

### 6.5. Long term dissemination of expertise: M37-M48

UMF

Not applicable for this reporting period.

**Goethe**

The talk that Prof. Fronius gave in Cluj-Napoca in January 2019 about appropriate visual acuity testing as well as the information provided during the visits in rural areas to family doctors and their nurses about amblyopia and its diagnosis were supposed to disseminate long term knowledge and expertise. The talk at the Annual Meeting in Poznan in March 2019 for the Country Representatives may also lead to long term dissemination of expertise. Prof. Fronius was involved in planning of the Toolkit handbook and wrote the chapter about testing of visual acuity. She participated in testing the EUSCREEN cost-effectiveness model and provided feedback for its development and completion.

**Discussion group for strategy for implementation of vision screening in all of Romania**

UMF and DASM will participate in the discussion group started by Jan Kik (EMC) to work out a strategy for future implementation of vision screening in all of Romania, specifically for screening in the rural communes.

According to Jan both nurses of family doctors and travelling screening nurses will be needed, and according to Maria Fronius (Goethe) it would be good when travelling screening nurses not only screen in remote areas but also teach the nurses of the family doctors how to screen and thereby carry and guard the screening expertise. To limit travelling time, they could best be stationed in the smaller cities, leaning on the expertise carried and guarded by the DASM-like organisations in smaller cities. Question then is how they should be paid, when stationed in the smaller cities and screening and teaching in the surrounding rural communes.

Cristina has prepared a letter for the Ministry of Health in Bucharest to make screening compulsory for family doctors and their nurses and securing budget for this purpose, but the Direcția de Sănătate Publică a Județului Cluj has advised Cristina to wait until the implementation study produces solid results and then first discuss the proposal with them before forwarding the proposal to Bucharest.

Mandy has emphasized that the kindergarten teachers also could be instructed by the travelling screening nurse and screen the children they care for. Again the question arises how they could be paid for screening.

Finally, the Romanian Paediatric Ophthalmic Society could be asked whether the paediatric ophthalmologists in Romania would favour a paramedical training for orthoptists in, for instance, Bucharest and Cluj-Napoca. Most countries in Europe have orthoptists and schools for orthoptists. Orthoptists could then be stationed in smaller cities in Romania and facilitate low-threshold access of care for children from rural areas treated for amblyopia.

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

### 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

During last 18 months WP7 has continued with screening per stage, diagnosing and referring to the CRC the babies who are suspected or confirmed to have a hearing problem. Preliminary screening results show that from January 2018 until December 2019, 18.715 babies have been screened in the four Maternity Hospitals. The average coverage rate with the first screening test during 2019 is 96.2%. The number of the parents who refused the tests has been really low and only occasionally families have left the hospital very early without the test having been performed.

During the two years of screening 22 babies have been identified with various degrees of hearing loss, 6 are under evaluation and some more are expected to be evaluated. Three of these babies, born on 2018, are on the waiting list for Cochlear implantation. Some changes in the staff structure were also needed. One screening nurse has left therefore substituted in the Maternity Hospital no. 2 of Tirana; the Public Health Coordinator Enver Roshi resigned for political reasons. With his recommendation the position was filled by prof. Genc Burazeri, general health EUSCREEN coordinator, who is dealing at the moment with the Ministry of Health and the preparation of the strategy document.

One more nurse and one nurse for substitution started in the Maternity Hospital no.1 in Tirana on January 2019 and onwards. The reason for these internal changes is that the number of babies delivered in the Maternity Hospital no 1 is already twice the number of the deliveries in the Maternity Hospital no 2. While 6 midwifes screen at maternity 2, at Maternity 1 since September only 5 midwifes have been screening, which made it necessary to add two more staff. The site administrator has proposed some new midwives, who worked alongside our staff for already two months. The local study coordinator followed the way they tested the babies, their communication with parents and their paper work several times, and in the end two were chosen to work with EUSCREEN program.

Also in Kukës the program administrator has left. Given the low number of births, her tasks were taken over by one of the team members.

**EMC**

EMC made several contributions to the implementation of the hearing screening in Albania. Support was provided for the set-up and monitoring of the screening, as well as for the training of hearing professionals in the follow-up programme. In addition, an implementation study has been performed. For this aim several visits were made to all four screening locations during different stages of the implementation process. During these visits, the screening process and the screeners were observed, furthermore, screeners were asked to fill out a questionnaire that asked about their opinion on the screening programme implemented. The results of screening were registered in a database which was analyzed. Based on the on-site observation of the first visits, as well as analysis of the project database, EMC has finalised a first article on the implementation study that has been submitted to an international peer reviewed journal. The visits as well as the reporting has been performed in close cooperation with the project partners in Albania.

The main findings of the implementation of NHS in Albania were that most parents were willing to participate in screening which lead to high attendance in the first screen. However, for all maternities, parents experienced difficulties to return to follow up screening. This resulted in an attendance rate of 67,4% for the second screen and 65,6% for the third screen. Reasons for not showing up were: parents were convinced their child could hear, parents did not think repeat screening was necessary, parents lived too far away from the location where screening took place, parents were unable to return, or the child had another illness. An extensive description of the obtained results, the difficulties experienced and the successes achieved is given in Appendix 4.

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

The teams of nurses in the Maternity Hospitals are regularly supervised by the core staff and the results of screening are collected both in database and monthly reports. If unclear cases are seen, we go back to the site and the nurse for further clarification and to follow up the subjects.

On November 4th 2018 in Pogradec, EUSCREEN organized a round table with the teams from all the four sites. The objective was to report on the work done and to identify problems and solutions. The meeting was extremely successful, a lot of brainstorming took place and many things were clarified, especially regarding the reporting and the data collection.

This meeting was followed by a similar one with the screening staff and administrators on October 6th, 2019 in Kukes. Apart from reports from each team, the administrators of the program discussed some issues to be addressed differently (database, codes and follow up etc), or weak points that needed reinforcement. Several discussions about organization scheme, problems and solutions took place.

This meeting, the same as the one in Pogradec a year ago, proved to be a good way of exchanging ideas and finding solutions.

Both meetings showed it is imperative to record the results of the tests in a document that can be accessed by various medical staff, if needed, in the future. The baby book appears as the best option. Unfortunately there has been a shortage of books in other districts apart from Tirana, so the info about the test has to be hand-written on the discharge letter issued by the doctor. The latter requires a good cooperation with the pediatricians in the Maternity Hospitals. This cooperation was also an issue raised during the meeting, because the proper interest of the pediatricians in the test results is lacking sometimes. This in turn, can influence the rate of non-attendance at the second and third testing. Also, coordination between the visits with pediatrician and the following hearing test can improve the rate of follow up.

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

The rate of families who do not show up for the further screening tests is still high. In order to understand what are the reasons for not showing up for further testing, Public Health team conducted a survey in the Maternity Hospitals of Tirana. 282 parents were contacted via phone and according to their responses the main reason for not coming back for further testing was negligence, their belief that the baby could hear and 4.3% of them have visited a private facility. The responses showed the need for more input on raising the awareness about early identification of hearing loss.

Although we do not have many parents who refuse to do the first tests, there are a few who do not give the consent and also there is also still a high rate of loss of follow up cases (LTFU). This figure is quite ambiguous, because about 1/3 of the babies born in Tirana do not live there. This means that, although the date and time are given for further testing they are not monitored by our staff in the process. Also, winter and the distance from Tirana, could be reasons for not showing up for further testing.

Furthermore, there have been baby transfers from site to site which makes the calculations sometimes difficult. Independently, the relatively high LTFU figure means that the information the parents have regarding the early identification and intervention of HL issues, is not sufficient.

In order to enhance this information WP7 has organized one presentation day at the University Medical Center of Tirana (UMCT), with doctors, medical students, professors of the University of Medicine and UMCT on October the 10th, 2018 .

The presentation day was attended by 201 participants. During the day were given various information regarding different aspects of hearing screening and rehab and also was presented the progress of EUSCREEN in Albanian. The activity was CME accredited.

During 2019, the hearing screening issues and EUSCREEN program appeared in two TV channels, one blog and several posts on University social media.

Within these lines, WP7 in cooperation with the Albanian Ministry of Health organized another informative day with the staff (doctors and midwives) from the Child Consultancy Centers in Tirana. The informative day took place at the UMCT premises on July 20th and all the different aspects of the hearing screening program were discusse as well as the role of the Child Consultancy Centers to support it. This activity was also CME accredited.

**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.**

The incidence of hearing loss is much lower that the incidence of amblyopia in children therefore thousands of tests are needed in order to be able to discern a pattern in each screeners work. This threshold number unfortunately is not reached neither in Kukes nor in Pogradec, but is sufficient for the majority of nurses in Tirana to make some comparisons.

Independently, as the data quality monitoring is quite important, an evaluation of the work completed by each nurse took place in December 2019. All the screening tests completed during 2019 were analysed and each nurse's results were also checked.

Taken into consideration that hearing loss occurs in around 1-2 cases in 1000 well babies (and Pogradec and Kukes have only well babies) and the small number of subjects screened by some nurses (less than 300/year) it seems that the data in the database are reliable. All nurses in Kukes and Pogradec have done second and third tests and also the majority of them have referred at least one baby for diagnostic assessment. The only identified problem in Kukes is that one nurse is not capable to use the computer, therefore her tests are filed under the tests done by other nurses. The number of tests done by each member of the team in Kukes was therefore requested. The analyses of the paper modules filled showed that her data are similar to the others nurses' data.

The situation in Tirana appears somehow different because the number of babies screened by each nurse is higher and we have NICU babies also. Based on 2019 screening tests each nurse has done second and third tests and also sent babies for diagnostic assessment. The number of second and third tests varies considerably per nurse. Therefore the project administrator has been asked to compare device results with the database results for four nurses in the Tirana teams and report back.

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

The register of high risk babies in Tirana Maternity 1, which is indeed the biggest site, for some time have been unsuitable for statistical analyses and not yet as detailed as needed. So, after several meetings with the staff, from July 2019 and on the register has been kept in a more practical way. This site's registers are quite important because the only way to identify a baby for follow-up is to couple these registers with the subjects' database code (personally identifiable information is not registered in the database).

The registering systems in the other three sites were well organised earlier on.

WP7 is preparing a new database system to be suggested to the Ministry of Health for the future screening at the Maternity Hospitals.

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

**EMC Department of Public Health**

**Task 8.1: Development of a TOOLKIT for policymakers**

We started the development of the toolkit for vision and hearing screening. The toolkit will consist of two parts: the webtool as developed in WP 5 and a manual with information for policymakers describing aspects as health care systems, finance and cost-reimbursement systems, ethical and societal considerations, informed decision making and sustainable implementation.

In monthly calls the content of the manual and the leadership by chapter has been discussed. We defined the end user as an expert who has to implement screening. Our aim is to write a concise manual (about three pages per chapter) that will be easy accessible on the internet with cross references to relating paragraphs and definitions. We will provide references to existing materials as much as possible instead of rewriting or repeating existing protocols or methods as much as possible. Detailed information will be included in appendices when needed.

The manual will consist of four parts: background, planning and decision making, practical implementation guide and conclusions and recommendations. The following content is set to be included:

**Part I: Background**

**1 Introduction**

1a The EUSCREEN study

1b History of medical screening

1c Child hearing and vision screening

1d Development and purpose of the TOOLKIT

1e Outline and target audiences

1f Key messages

**2 General insights on hearing and vision screening**

2a Criteria for responsible screening

2b Effectiveness and cost-effectiveness of hearing and vision screening programmes

2c Benefits versus harms of hearing and vision screening programmes

**Part II: Planning and decision making**

**3 Planning and decision making**

3a Appropriateness, acceptability, feasibility and sustainability

3b Minimum resources needed for a screening programme

7a Introduction

7b Implementing vision screening

7c Programme objectives and targets

7d Coordination

7e Communication

7f Screening personnel

7g Training screening personnel

7h Screening locations

7i Screening pathways

7j Protocol

7k Communicating results to parents

7l Monitoring

7m Adapting an existing programme

7n Overcoming barriers

**8 Preschool vision screening**

8a Implementing a new programme

8b Programme objectives and targets

8c Coordination

8d Communication

8e Screening personnel

8f Training screening personnel

8g Screening locations

8h Pathways

8i Protocol

8j Communicating results to parents

8k Monitoring

8l Adapting an existing programme

8m Overcoming barriers

**9 Public awareness and communication**

9a Public awareness barriers and facilitators

9b Communication plan and materials

**10: Monitoring: database, quality assurance, evaluation and reporting**

10a Database

10b Quality assurance

10c Evaluation

10d Reporting

**Part IV: Summary and recommendations**

**Appendices**

Most chapters have been drafted already. These drafts will be finalised after review by all consortium partners.

**Task 8.2: Support capacity building in other EU countries**

The webtool was used by country representatives during the workshop on March 2019 in Poznan. Currently the webtool is available on the internet and users can ask questions and provide feedback to the developers. During the final conference in November 2020 (or May 2021 if the 6-months extension because of the Covid-19 pandemic is granted) the final TOOLKIT (webtool and manual) will be presented.

**Task 8.3: Development of a generic tool for policymakers**

See WP 5 for a description of the webtool. In this webtool users can enter their child preventive health care programme and can look for possibilities to combine contact moments to minimize the burden for parents. The model code is flexible, therefore by changing input parameters the model can be used for other diseases as well. We will assess the applicability of the model for neonatal vision screening, which has different target diseases than amblyopia.

## 1.2.9 Work package 9: Ethics requirements

In the 18-months Report, several ethical requirements had to be met: Informed consent templates and data authorizations, copies of ethical approvals, data protection confirmation and the appointment of an ethical advisor.

In this report, no formal Ethics requirements have to be addressed, but in an Ethics Check session on October 23-25 2019 about the EUSCREEN project, the following questions were asked:

Database

1. To implement appropriate anonymization/pseudonymisation procedures to ensure protection of the personal data of children participating in the screening studies and provide information to the EC on these procedures and on the measures used to prevent unauthorized access to this personal data/equipment.

2. To provide ethics approval and additional information regarding the consent procedures for participants in the professional network /online survey.

Vulnerability of Roma Minority

3. To submit the risk mitigation strategy implemented to protect the vulnerable Roma population from any stigmatization or enhanced vulnerability.

4. To confirm that the export of data relating to Roma communities from Albania and Romania into the EU is permitted under national law as well as that any national legal obligations covering the processing of Roma data are adhered to.

5. To check and confirm whether the processing or export of data relating to Roma populations require a specific declaration to be made to the applicable Data Protection Authorities of the countries involved (Albania and Romania).

ad. 1 All partners in the EUSCREEN Consortium adhere to the EU's regulation 2016/679 (GDPR) for data handling in the electronic database by pseudonymisation: "processing of personal data in such a manner that personal data can no longer be attributed to a specific data subject without the use of additional information, provided that such additional information is kept separately and is subject to technical and organisational measures to ensure that the personal data are not attributed to an identified or identifiable natural person."

When introduced in the EUSCREEN database each child is assigned an anonymous code. The examining nurse and her secretary keep a list on paper to be able to match the children examined by that nurse to their codes, in case diagnostic information has to be added to the child's record in the database. These lists are kept separately and are not accessible to anyone else than the examining nurse or her secretary. Months later, when the Form filled out by the ophthalmologist has been received, the secretary or the examining nurse searched in her list on paper the name of the child, the assigned code is found, the code is found in the EUSCREEN database and the Form filled out by the ophthalmologist is entered into the database. The authorized secretarial personnel has signed confidentiality clauses and GDPR clauses in their work contracts.

ad 2. The Country Representatives were subcontractors to the EUS€REEN Foundation who were recruited by a tender procedure, and they reported about the screening programmes in their country, with demographic background data.

ad 3. Concerning our risk mitigation strategy to protect the vulnerable Roma population from stigmatization or enhanced vulnerability, it is correct, as the Ethics Committee states that "studies are being carried out on patients from the Roma population which is considered a vulnerable group. Specific measures to address this vulnerability and

associated risks from participation are not provided", but the fact that our study covered all areas of Cluj-Napoca including areas with Roma communities, covered all smaller cities including a city with a large Roma community and covered all rural communes including all communes with Roma communities, implicitly avoided stigmatization and avoided enhanced vulnerability. As additional measures, in Cluj-Napoca, the screening nurse from the Roma Daycare Center has made home visits to explain to parents the necessity of vision screening, escorted children to the ophthalmologist when suspected of amblyopia and even screened some children at home.

In the four maternity clinics in Albania, the study has made no distinction between different ethnic groups and treated the newborn Roma babies like all other children. Screening was offered to the parents of all babies and no questions about ethnicity were asked. Therefore, the strategy against possible stigmatisation is implicit in our study design.

ad 4 and 5: Data relating to Roma communities from Albania and Romania was not collected in the database. We had no possibility in our database to enter ethnicity. Only anonymous data were entered into the database.

# 1.3 Impact

**Data collected in network of professionals: Requisites, barriers and facilitators identified**

By our collaborative effort we now have the largest set of data about vision and hearing screening ever. As suspected, vision and hearing screening programmes differed tremendously among countries in Europe.

A very important obstacle to efficient screening identified by the EUSCREEN Study thus far is lack of monitoring, quality control, data collection and evaluation. Even in HIC countries with highly developed nation-wide screening programmes this is lacking or insufficient. This may be caused, in part, by the relatively low degree of competition in preventive health care as compared to curative health care: Screening programmes are funded by the state, province or council and very few parents will think that screening is better elsewhere or seek second opinion. The impression that monitoring, quality control, data collection and evaluation could be improved arose from the data gathered in the network of professionals but also became evident in the implementation studies.

Another disturbing issue was the high percentage of children who did not return for a second screen in neonatal hearing screening in Albania and the high percentage of children who were referred for diagnostic assessment of vision but are not (yet) backreported by an ophthalmologist in County Cluj. There are many possible reasons for this and these are currently being assessed.

**Development of the cost-effectiveness model and its impact**

At the end of the reporting period (months 19-36) the cost-effectiveness model (available at miscan.EUSCREEN.org) had been developed to the point that test users anywhere in the world can enter their choice of screening programme, like the number of neonatal hearing tests and the salary of the screening technician, or the number of visual acuity measurements and the age of the tested child, to calculate the total costs of the screening programme, the cost per screen and the cost per detected case. As the user of the cost-effectiveness model himself enters data on the availability and the salary of professionals who could screen in that country and several other parameters reflecting the region and its organisational and resource requirements, a custom-made prediction of cost-effectiveness of a screening programmes is calculated for that country or region.

In long discussions about the model and in exercising with the model during its development, the great advantage of combining vision and hearing screening with other high-attendance events was stressed, ranging from being born (neonatal hearing screening in maternity clinics) to immunization and heel prick blood test (hearing screening), immunization boosters (vision screening), both for better coverage and for higher cost-effectiveness.

In our study, the participation of countries outside Europe, including Russia, Malawi, Ruanda, South-Africa, India and China is very important for the development of the model, because Europe has no low-income or lower middle-income countries and data from such countries is essential to construct a cost-effectiveness model that works for all countries in the whole world. This again is important because several lower middle-income countries like India would like to implement vision and hearing screening nation-wide and need objective calculations to get the best value for money in screening. It proved necessary to add a pre-module to the cost-effectiveness model with questions to determine whether the introduction of vision or hearing screening would be inacceptable or inappropriate, for instance in case of more urgent health issues like famine or high infant mortality.

**Manual with a Strategy for Implementation and TOOLKIT**

A manual with a strategy for implementation is being developed from the results of the implementation studies, from identified requirements, facilitators and barriers, and from good-practice guidelines for existing screening programmes.

Finally, the TOOLKIT, consisting of the cost-effectiveness model and the Manual with the Strategy for Implementation, will be made. It will assist healthcare providers and policy makers in their decisions to introduce or modify screening programs and offer many advantages over the present situation where each country has expert committees to develop guidelines, a screening committee to approve and coordinate guidelines and commissioners to decide whether to follow these guidelines. It is an innovative solution to the problem of inefficiency in the delivery of preventive health care to children and will confer greater cost-effectiveness, improved health outcome, greater health equity.

In this way, this study has increased EU lead and competitiveness in expertise in preventive youth health care further. Europe traditionally has very good expertise on youth preventive health care, whereas, for example, in the USA only 36% of the children get vision screening. In LMIC countries outside Europe cost-effective screening programmes will be introduced, assisted by the TOOLKIT. Within Europe, the TOOLKIT can be applied for other kinds of screening programmes with repeated screening in children.

**Direct impact: Continuation of hearing screening in Albania**

The transition of the implementation study of neonatal hearing screening to state-paid neonatal hearing screening is a reality now that the Albanian government has included neonatal hearing screening in the 2020 budget. A national plan is now being written by our local EUSCREEN Study coordinator to include the other parts of Albania in neonatal hearing screening in the course of 2020.

**Direct impact: Continuation of vision screening in Romania**

In Romania the largest problem with implementation of vision screening has been the rural communes. Of all people in Romania 46% lives in rural communes. Vision screening by the Family Doctor's nurses was only partly successful, more so when they started to visit the local kindergartens in the villages for screening. However, full coverage was only reached after a travelling screening nurse had been appointed, who screened 805 children in underserved rural areas in 6 months. It seems that full coverage for all rural communes in Romania could be reached by a dual solution of Family Doctor's nurses screening in kindergartens in most of the rural communes and travelling screening nurses to (i) screen children in remote and underserved rural communes, (ii) train and monitor the Family Doctor's nurses and (iii) carry and guard the expertise of quality vision screening. To limit travelling distances – a problem identified in the implementation study – they could be stationed in the smaller cities and larger cities and be employed by the local council administration organisations like the DASM in Cluj-Napoca, that have been so successful in screening in Cluj-Napoca and the 5 smaller cities in County Cluj. In addition, the teachers in kindergartens in rural communes could be instructed by the travelling screening nurse and screen the children they care for. Finally, a training for orthoptists, paramedics who treat children with amblyopia, could be started in, for instance, Bucharest and Cluj-Napoca. Most countries in Europe have orthoptists and schools or university degree courses for orthoptists. Orthoptists could then be stationed in smaller cities in Romania and facilitate low-threshold access of care for children from rural areas treated for amblyopia. This would enhance availability and cost-effectiveness of treatment of children with amblyopia.

# 1.4 Access provisions to Research Infrastructures

not applicable

# 1.5 Resources used to provide access to Research Infrastructures

not applicable

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

Dissemination of the results of the study is via (1) the Country Representatives, (2) scientific media and (3) by the TOOLKIT.

1. In our study, data on screening programmes, demography, administration, general screening, screening professions, uptake and treatment availability has been collected in a network of country Representatives in 40 European countries and several countries outside Europe, including Russia, Malawi, Ruanda, South-Africa, India and China with questionnaire with domains 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. By our collaborative effort we now have the largest set of data about vision and hearing screening ever.

On March 9th, 2019, a Study Meeting of the EUSCREEN Study was held in Poznan, for the Consortium Partners but also for 60 of the approximately 100 Country Representatives. They were first instructed on the use of the cost-effectiveness model with lectures about its working and practical exercises, both for vision screening and for hearing screening. There is a high degree of commitment of the members of the Country Committees and the need felt to exchanging expertise on vision and hearing screening across Europe and resolve the large differences between screening programmes that exist.

Many more Country Representatives will attend the Final EUSCREEN Study Meeting for all Consortium Partners and the Country Representatives in Rotterdam on November 13th-14th, 2020 (or May13th – 14th, 2021 if the 6-months extension for the Covid-19 pandemic is approved) where the final analysis will be presented on the data gathered with extensive the questionnaires the Country Representatives filled out, a complete analysis and review of the vision and hearing screening programmes in Europe will be presented and the Cost-Effectiveness model will be presented officially, together with the Manual with Strategy for Implementation of Vision and Hearing Screening Programmes, these two constituting the TOOLKIT.

2. Results are being disseminated in the scientific community via scientific publications and congresses. Presentations are given by all Consortium Partners, but also Country Representatives present the results of the inventory of screening in their country. It has enhanced the awareness of the need for comparing screening programmes in Europe. These publications are made available in the online repository of participating universities, whereas open access has also been enabled by payment by the university pay of a fixed amount per article to the publisher. Apart from dissemination to the scientific community via scientific publications and congresses, presentations have been given by all members of the Consortium.

Also the Country Representatives within the EUSCREEN Study have reported on the state of vision and hearing screening in their country on the basis of data they had collected themselves. In that way they advertise the results of the EUSCREEN study and stress that the diversity in vision and hearing screening, and its resulting inefficient use of public health resources, can and should be resolved.

3. The TOOLKIT made in WP8 will contain the cost-effectiveness model made in WP5 and populated with data gathered in WP3-4. It will provide evidence for introduction or modification of screening programmes to stakeholders described above and to health policy makers. The cost-effectiveness model is now available for all Consortium Partners, but also for all 100 Country Representatives but also available in the public domain (miscan.EUSCREEN.org) and, hence, available to healthcare providers and policymakers worldwide.

We will encourage the the primary stakeholders in countries with nation-wide vision and hearing programmes to use the TOOLKIT: (i) expert committees that develop guidelines, (ii) screening committees that approve and coordinate

guidelines and (iii) commissioners who decide whether to follow these guidelines. These may reside in different public services, ministries, health insurers etc.

**Website**

Communication and exchange of expertise within the study is primarily via the study's website www.EUSCREEN.org. It hosts communications like announcements, the study's logo, image, and masthead design, announcement of meetings, important updates, group announcements, open, group and private conversations, private messages, newsletters, webinars, blogs, documents like study reports for the EU-Commission, manuscripts to be submitted, publications, all to foster communication and exchange of expertise among all participants, including the members of the 40 Country Committees. It bundles expert-opinion within the consortium and is directly linked to the cost-effectiveness model at miscan.EUSCREEN.org.

# 3. Update of the data management plan (if applicable)

This part is not applicable for the EUSCREEN project.

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

Midterm Review recommended to follow up the agreements achieved during the MTR meeting on March 7, 2019:

The delay in visual screening of children from rural areas of the Cluj County, Romania.

• The Romanian beneficiary (UMF-Cluj) committed to do their best to screen a total of at least 3,000 children living in rural area of the Cluj County. Vacancies for up to four nurses (or other professionals who could be trained to screen) will be published ASAP.

• Reimbursement of travel costs for screening nurses in rural areas will be considered.

• The period of screening could be extended by six months as this would still be within the duration of the project and hence no amendment would be necessary.

• To avoid other delays it is recommendable to keep the project implementation within the current GA and avoid requests for amendments, in particular those related to extension of the implementation period.

These have proven to be very valuable recommendations and all of them have been implemented. In summary:

Despite the fact that 34% in County Cluj lives in rural communes (46% nation wide) vision screening was initially hardly offered to children from rural communes. This was caused by the way screening had been planned, e.g. by the family doctor's nurse in the doctor's office. It went much better when these nurse went to the local kindergartens to screen children there and, for rural communes without screening family doctor's nurses, when a travelling screening nurse was put on the job. Due to the implementation of these risk-mitigation measures and of the recommendations from the Mid Term Review, vision screening has been offered in all rural communes in County Cluj before December 31st, 2019. This issue is described in Part B in great detail under WP6 and in Appendices 1 and 2.

The second and third Recommendations of the Mid Term Review, on data quality and on effectiveness and acceptability (for future work) have been followed up only partly until December 31th, 2019.

“Mixed methods study on data quality” has not yet been performed. The fact that “a mathematical model, to be applied in the same countries of the study and further transferred to additional countries, is only as good as the data it is based on” is not yet an issue in the cost-effectiveness model we currently develop, as the user has to enter his own data for his country and, yes, the predictions of the model are only as good as the input data, but this is a problem that cannot be solved easily and, in most cases, not by use of mixed methods study. For instance, the most important parameters are sensitivity and specificity of screening tests and specificity very much depends on the professional, state of training, experience, age of the child and the type of test. This problem is so difficult to solve that some in our team even advocate the abolition of specificity in the model altogether, and use “expected referral rate” instead, as is used in cancer screening models. Instead, Goethe and Reading have been asked for experts’ opinion on sensitivity and specificity of vision screening tests and Karolinska on those of hearing screening tests, in relation to professional state of training, experience, age of the child and the type of test.

We agree that “additional qualitative sub-study on the acceptability, and other social factors relating to the effectiveness of the intervention (for example, mobility, residence patterns, access to care, acceptability of proposed

solutions) could be a positive asset for the study." but, really, it is beyond the scope of the study and we would have to apply for extension of the EUSCREEN Study and for extra budget to fit these additional issues in.

# 5. Deviations from Annex 1 and Annex 2 (if applicable)

**5.1 Tasks**

***Addition 27.5.2: Deviations from Annex 1 related to the Covid-19 pandemic, after the initital submission of this report (26.2.2020) but before its resubmission (27.5.2020)***

*This page in Italics has been added to this report on its resubmission on May 27th 2020 as the Covid-19 pandemic has drastically changed the course of the EUSCREEN Study. As a corrective measure we will apply for an amendment for a budget-neutral extension of the EUSCREEN Study by 6 months until June 30th, 2021, based on GA Art. 51. The consortium partners spoke unanimously in favour of a budget-neutral extension of the study in April and May 2020 , to cope with the delays. The application for an amendment for an extension of the EUSCREEN Study by six months with postponement of the pending Deliverables and Milestones by six months will include: which tasks are delayed and why, who will work on these tasks between January and June 2021 and what will be the projected working hours of these people. Below the delays as of 27.5.2020 are listed. They are also specified in Italics with each WP.*

*The vision screening implementation study in Romania ended on December 31st, 2019, but re-examination of the 799 children in Romania who were purportedly examined by 4 of the 104 screening nurses but without any referral at all, has been delayed due to the Covid-19 pandemic. Accordingly, the report on the screening in Romania (Deliverables 9, May 2020) is delayed.*

*The hearing screening implementation study in Albania ended on December 31st, 2019, but the transition to government-paid continuation of screening in the four maternities that was decided upon by the Albanian government in January 2020, is delayed due to the Covid-19 pandemic. Accordingly, the report on the screening in Albania (Deliverable 10, May 2020) is delayed.*

*The development of the screening cost-effectiveness model software (miscan.euscreen.org, Deliverable 11 & Milestone 12, November 2020) and the Manual with the Strategy for Implementation of Vision and Hearing Screening Programmes (Deliverable 11 & Milestone 12, November 2020) that together constitute the EUSCREEN Toolkit, the main produce of the EUSCREEN Study are delayed caused, in part, by the Covid-19 pandemic.*

*The final Vision and Hearing Screening Conference for all Country Representatives and Consortium Partners for broad dissemination (Deliverable 1 & Milestone 11, November 2020) could best be moved from November 2020 to May 2021.*

**Work package 1: Project management**

None

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

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, most of the required data were nevertheless collected by the end of 2018 and the final data were collected in the first half of 2019, so that the work progress in other work packages did not suffer from this delay.

***Addition 27.5.2020:*** *the final Vision and Hearing Screening Conference for all Country Representatives and Consortium Partners (Deliverable 1 & Milestone 11, November 2020) could best be moved from November 2020 to May 2021in the face of the Covid-19 pandemic.*

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

**USFD**

Due to the complicated process of data collection and validation, the window for data collection and validation required extension. There was a delay in aggregating and validating sufficient data from all countries. Therefore, there was a 6-month delay for Work Package 3 to deliver Country Reports. All Country Reports were submitted before end of December 2018. An overall summary report on vision screening has since been completed and submitted in June 2019 (Deliverable 2), to fulfil the requirements of the deliverable for WP3 (Deliverable 3).

Due to the delays described in producing the Country Reports, progress of the review of acceptability of childhood screening programmes was affected (Milestone 5). A draft manuscript has been circulated internally for comment (with a view for submission for consideration for publication in 2020) to Journal of Medical Screening, and Milestone 5 is expected to be reached in the Summer of 2020.

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

Due to the complicated process of data collection and validation, the window for data collection and validation required extension. There was a delay in aggregating and validating sufficient data from all countries. Therefore, there was a 6-month delay for Work Package 4 to deliver Country Reports from 47 countries and regions. The final country submitted data in June 2019. All Country Reports were delivered before June 30, 2019.

A Summary Report of Findings was delivered fulfilling the requirements of the deliverable D 4.1.

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

***Addition 27.5.2020:*** *Development of the screening cost-effectiveness model software (miscan.euscreen.org, Deliverable 11 & Milestone 12, November 2020) and the Manual with the Strategy for Implementation of Vision and Hearing Screening Programmes (Deliverable 11 & Milestone 12, November 2020) that together constitute the EUSCREEN Toolkit, the main produce of the EUSCREEN Study, is delayed, partly due to the Covid-19 pandemic.*

None

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

***Addition 27.5.2020:*** *The vision screening implementation study in Romania ended on December 31st, 2019, but re-examination of the 799 children in Romania who were purportedly examined by 4 of the 104 screening nurses but with no referrals at all, is delayed due to the Covid-19 pandemic. Accordingly, the report on the screening in Romania (Deliverables 9, May 2020) is delayed.*

**UMF**

Obstacles during screening in rural areas in Cluj.

**Problem definition**

Screening in rural areas in Cluj was hampered by low coverage. This was caused in part by the initially chosen way of screening, by the family doctors' nurses at the doctors' offices. The number of rural nurses who wanted to get involved in the project was small (only 24 nurses started examining children in 2018), due to their heavy workload and a lack of time, and parents often did not bring their children to the medical office for screening, even when asked to do so several times.
The nurses were therefore advised to screen the children at the kindergartens instead. Here an additional difficulty encountered was the fact that the numbers of children attending the rural kindergartens were lower than expected. Reasons for this are that there are children that are registered at the kindergarten but do not or only rarely attend; many young families moving to cities or moving abroad; attendance fluctuates through the year and parents who live in villages that are close to cities prefer to take their children to the extended program urban kindergartens (the rural kindergartens close at 12:00, the urban kindergartens at 16:00 or 18:00).
UMF identified in more than 900 rural children that that were actually examined in urban kindergartens.
A more detailed analysis of the problems encountered in the rural areas can be found in Appendix 1.

**Corrective measures taken:**

In August 2018, UMF Cluj team advised the nurses to examine the children in the rural kindergartens and encouraged 5 nurses to examine children from communes other than their own as well. In the summer of 2018, in order to increase the rural coverage , UMF Cluj team had visited many rural medical offices in order to convince more nurses and doctors to get involved in the project and to examine children, but these visits had only modest results. In 2018 only 890 children were examined in the rural area in a third of the communes in Cluj county.
The need for better rural coverage and more children examined led to the idea of hiring a nurse who would travel full time to the communes where there was no nurse to examine children. In March 2019 UMF hired one travelling nurse who managed to examine in 2019 a total of 806 children in the rural area. A second travelling nurse was hired in May 2019 but she resigned three weeks later due to the discrepancy between the estimated number of children (as indicated by the Health Insurance House and from the National Institute of Statistics) and the actual number of eligible children

attending the rural kindergarten, a problem that was identified and reported by UMF earlier in 2018. The travelling nurse highlighted in her reports the low attendance at rural kindergartens, the bad roads she had to travel and the need for at least two or three visits to the same village in order to examine the majority of the children. She also mentioned in her reports a poor collaboration with some rural teachers regarding the screening.
It was decided by the project coordinator that from September 2019 onwards the travelling nurse would be paid €10 per child (instead of €7) in order to boost the rural screening in communes with few children and far away from Cluj and also to be able to cover some of the expenses regarding the daily rural travels.
At the end of 2019 there were still a few communes where no children were examined. In order to cover these communes as well UMF Cluj team visited the kindergartens, talked to principals and teachers, set up Screening Day and then sent one nurse (*who already examined children from five different communes*) to examine the children in these "zero communes": Vad, Sanmartin, Jichis, Rasca, Buza, Palatca, Alunis and Recea Cristur. It was decided by the project coordinator that this nurse would be paid €14 per child in order to be motivated to examine the children from the above mentioned communes in less than two months.
Thus at the end of 2019 in all communes of Cluj county (*except Belis and Marisel where children were already examined by the Lyons foundation)* children have been examined in the EUSCREEN project.
A total of 3200 children from the rural area were examined in two years: 890 in 2018 and 2310 in 2019 (note that not all these children had been introduced in the database yet by January 10, which is why these numbers do not match the numbers in Appendix 2).

**UMF and DASM**

**Problem definition**

An analysis of the project database at the end of 2019 identified four nurses who examined more than 100 children and who referred zero children to the ophthalmologist. These nurses attended the UMF courses in 2017 and were visited in 2018-2019 by the UMF team, which made sure their examination technique is good and the criteria for a failed test were understood. These four nurses examined 799 children that were entered in the database by January 10, 2020 (see Appendix 3 for a more detailed description of this issue).

**Corrective measures planned**:

In order to verify the possible problems that could have appeared in the examinations of these four nurses, the following measures will be taken:

1). 05.01.2020 - 20.01.2020 - All children examined by the four nurses with zero referrals are to be identified in the project's database.

2). 05.01.2020 - 20.01.2020 - All the consent forms are to be checked again in order to verify the identity of the children and the signature of the parents. This will prove that the children exist and that they were examined. All the consent forms are in the project's archive.

3). 20.01.2020 - 30.09.2020 - The kindergartens in which the children were examined will be visited in 2020. The nurses' examination technique shall be verified. All the children present in the kindergarten during our team's visit will be re-examined.

Re-examinations schedule (June – October 2020, postponed because of the Covid-19 pandemic):

- Campia Turzii
- Vad and Bogata
- DASM
- Bontida
- Iclod

- Livada
- DASM
- Cojocna
- Gilau
- Jucu
- DASM

A good collaboration of the children will mean that they were indeed accustomed with the tumbling E's and the LEA symbols and that they have been examined in 2018-2019.

4). 15.09.2020 - 15.10.2020 - Reports of the kindergarten visits will be made, containing a detailed situation regarding the children:

- how many children were re-examined
- how many children were OK
- how many children were referred after the re-examination
- how many children were absent during our visits
- how many children are now attending school;
- how many children got transferred to another school (in another village, another city or another county).

5). 15.10.2020 - 31.10.2020 - The results of the re-examinations will be introduced in the database. A second Form 4 will be added in the database for each re-examined child. The results of the re-examinations will be summarized in Deliverable 9, postponed from May 1st to November 1st, 2020, concomitant with the 6-months extension that is applied for by an amendment, because of the Covid-19 pandemic

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

***Addition 27.5.2020:*** *The hearing screening implementation study in Albania ended on December 31st, 2019, but the transition to government-paid continuation of screening in the four maternities that was decided upon by the Albanian government in January 2020, is delayed due to the Covid-19 pandemic. Accordingly, the report on the screening in Albania (Deliverable 10, May 2020) is delayed.*

Because of the irregularities found in the implementation study in Cluj (WP6), an evaluation of the work completed by each nurse took place in December 2019. All the screening tests completed during 2019 were taken into analyses and each nurse's results were also checked.

Taken into consideration that the hearing loss is around 1-2 cases in 1000 well babies (and Pogradec and Kukes have only well babies) and the small number of subjects screened by some nurses (less than 300/year) it seems that the data on the database are reliable. All nurses in Kukes and Pogradec have done second and third tests and also the majority of them have referred at least 1 baby for diagnostic assessment. The only identified problem in Kukes is that one nurse is not capable to use the computer, therefore her tests are filed under the tests done by other nurses. The number of tests done by each member of the team in Kukes was therefore requested. The analyses of the paper modules filled showed that her data go along the pattern of the others nurses' data.

The situation in Tirana appears somehow different because the number of babies screened by each nurse is higher and we have NICU babies also. Based on 2019 screening tests each nurse has done second and third tests and also sent babies for diagnostic assessment. The number of second and third tests varies considerably per nurse. Therefore the project administrator has been asked to compare device results with the database results for 4 nurses in the Tirana teams and report back.

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

***Addition 27.5.2020:*** *Development of the screening cost-effectiveness model software (miscan.euscreen.org, Deliverable 11 & Milestone 12, November 2020) and the Manual with the Strategy for Implementation of Vision and Hearing Screening Programmes (Deliverable 11 & Milestone 12, November 2020) that together constitute the EUSCREEN Toolkit, the main produce of the EUSCREEN Study, are delayed, partly due to the Covid-19 pandemic.*

None

**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 work effort of beneficiary 9 (EUS€REEN Foundation) stated in Annex 1 Part A, 0.1 project-month, should be corrected. This will be done during a future amendment regarding the 6-months project extension because of the COVID-19 pandemic.

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

**USFD**

Hours worked are in line with those anticipated. 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 have been as anticipated, but towards the end of 2019 Professor Horwood worked some more hours on the project during the writing phase of the paper submitted to the journal Eye. There is still some underspend on the Reading budget because of significant efficiencies in the running of the project since the projections made in the grant application in 2016. Conference calls have become the norm instead of some of the face-to-face meetings anticipated, and Professor Horwood has often managed to combine a EUSCREEN meeting with collaborators during conferences funded from elsewhere. The meetings we have had have been able to be in cities served by very low-cost airlines and inexpensive hotels. Professor Horwood has increased her hours to cover the writing stage of the Handbook.

**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**

None

**DASM**

The work effort of beneficiary 9 (EUS€REEN Foundation) stated in Annex 1 Part A, 0.1 project-month, should be corrected. This will be done during a future amendment regarding the 6-months project extension because of the COVID-19 pandemic.

**Goethe:**

From the Goethe budget € 2500 was reallocated for the Poznan meeting with the country representatives March 2019.

Costs for journeys could be kept somewhat lower than anticipated (lower number of face-to-face meetings, web conferences instead; meetings held in places with inexpensive hotels and accessible via low-cost flights). However, more working hours have been necessary, e.g. for supporting solutions for the unforeseen problems with vision screening in rural areas in Cluj county. A lot of communication in the project and writing a report was necessary concerning the obstacles for vision screening activities, especially in rural areas. Prof. Fronius has increased her hours to cover the writing stage of the Manual with the Strategy for Implementation. Therefore some resources of Goethe University may need to be shifted from travel to personnel in the coming period.

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

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

None

**Work package 9: Ethics requirements**

None

### 5.2.1 Unforeseen subcontracting (if applicable)

**Work package 1: Project management**

None

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

Country representatives from several countries outside Europe, Russia, India, South Africa, Malawi, Rwanda and China, filled out the questionnaire we developed. The data they supplied are of the utmost importance for our project (to include in our model) as the EU has no low-income countries. We have reimbursed these representatives, who meet the same requirements we have set for the 41 representatives from European countries, with half the remuneration for the Country Representatives within Europe who were entitled to a €2000 remuneration for the administrative effort provided the questionnaire is filled out completely or at least contains the data that is available for that country.

**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**

None

**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**

None

**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**

None

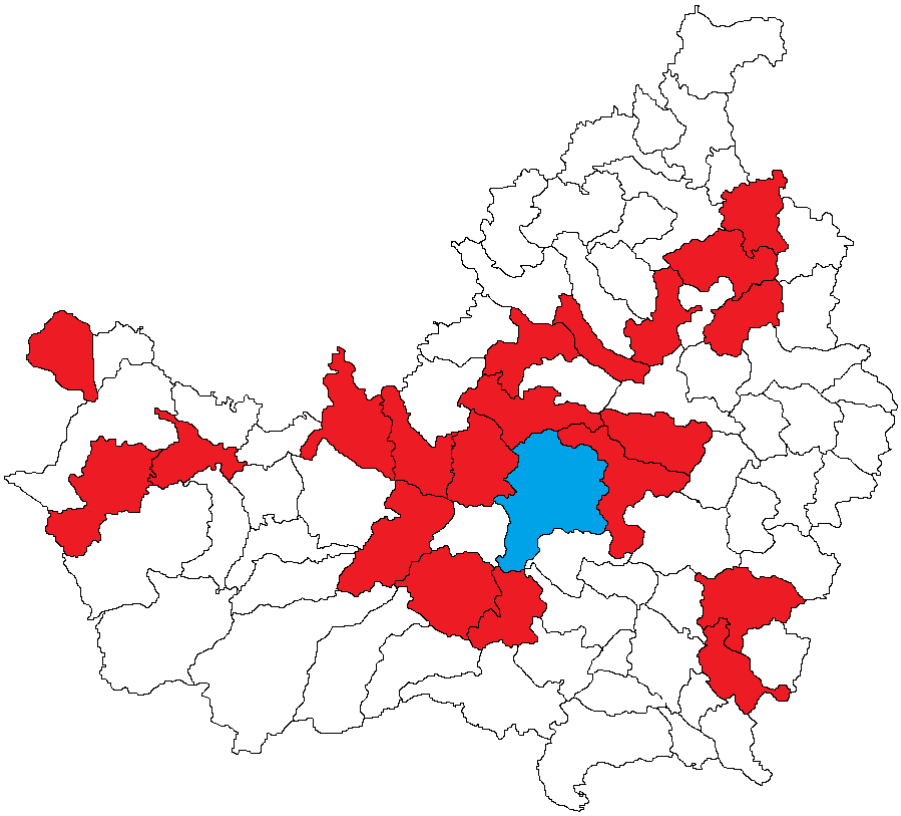
**Work package 9: Ethics requirements**

None

**Appendix 1. Reports on on-site observations in Cluj County in January and October 2019**

## 1.1 On-site observations of the implementation of vision screening in Cluj County by Mandy Nordmann, Jan Kik, Anna Horwood and Maria Fronius (January 2019)

In January 2019, the implementation of vision screening in Cluj County was observed on-site. The observation included visits to kindergartens (in cities and rural areas) and family doctors and their nurses (in rural areas) as well as extensive talks with DASM and UMF professionals responsible for the implementation. These visits were carried out in the county seat Cluj-Napoca, the two smaller cities Campia Turzii and Gherla and twenty rural communes throughout the county.



*Map of Cluj County with the communes visited marked red. Cluj-Napoca is marked blue.*

The most important observation made in the course of these visits is that there are huge differences in Cluj County that complicate the implementation of vision screening. These concern differences between the urban and rural areas, but also differences among the various rural areas as well as differences between the professionals involved in screening.

Screening appears to be going well in the urban areas. This seems to be the general opinion among the people involved. Children are screened in kindergartens by the resident nurses. The urban kindergartens are usually attended by many children, meaning the nurses are able to develop screening proficiency relatively quickly.

It should be noted, however, that while the organisation of screening in the urban areas seems to be going very well, it is not possible, based on these visits, to say much about the quality of the screening and referrals.

In the rural areas, the organisation of screening is much more difficult, but not so to the same extent everywhere: some communes that are rural in name are, in reality, more like suburbs. Others are indeed located remotely and inhabited by small populations spread over a relatively large area, with only small numbers of children eligible for screening (four- and five-year-olds).

There are kindergartens in rural areas, but these generally do not have resident nurses. And while quite some caretakers at the kindergartens did express their willingness to assist, for example by informing parents, a big problem is that the number of children actually attending these kindergartens tends to be lower than the number of children officially enrolled there. In winter this discrepancy is larger because more children stay at home because of the road conditions and illness.

Originally, the children in rural areas were to be screened by the family doctors' nurses at the family doctors' offices. Since it became clear at an early stage that this was not working well due to a variety of reasons, different strategies have been attempted on a small scale in the second half of 2018, such as a family doctors' nurse screening the children at the kindergarten or a family doctors' nurse travelling to one or more nearby villages to screen children at a family doctors' office or the kindergarten there. This approach, however, was hampered by a lack of reimbursement of travel expenses for travelling nurses, making it hardly worth their while to put in a lot of effort to screen a few children. In addition, travelling to other locations is difficult for nurses who do not have a car since there is often little or no public transport.

There are several reasons screening was not working too well at the family doctors' offices. First of all the family doctor and his/her nurse – usually very busy already – had to agree to follow a course to be able and allowed to screen, after signing a contract with UMF Cluj. It appears the paperwork involved was a barrier for some doctors and nurses. In rare cases, a family doctor could not screen at all when the contract with UMF Cluj implied that a minimum wage would have to be paid to the family doctor which would be equivalent to screening 60 children.

For the doctors and nurses as well as for the parents, their level of information about screening played an important part in their interest to participate in the project. Some doctors seemed unconvinced or simply unaware of the benefits of vision screening. Other doctors were very interested in screening. A few had already been screening before the project out of their own accord and some mentioned they found their newly acquired screening knowledge useful in their general practice too. But others plainly did not want to have anything to do with screening, for reasons that were not always clear.

We were told that parents often had other priorities than having their children screened, sometimes because they did not understand or sometimes because they did not care. It does seem that parents' attitude towards screening was also strongly influenced by the way it was explained and presented to them: it made a big difference whether they were just given a leaflet and a consent form to sign or whether everything was clearly explained to them in person, by someone they had a good relationship with. The enthusiasm of that person seems to be of importance as well.

Cultural differences sometimes play a part here as well. There are several communes in Cluj County where a large part – in some cases even the majority – of the population speaks Hungarian as their first language and others where a substantial part of the population are Roma, who sometimes are illiterate. A leaflet in Romanian is unlikely to be of much use to either group. The Roma population appears less receptive to preventive healthcare in general.

Motivation is also an important factor: a motivated doctor is much more likely to go through the trouble of arranging all the paperwork, for example and a motivated nurse is more likely to spend time to convince parents of the benefits of screening.

These issues occurred in different combinations. No two communes were visited where circumstances were exactly the same.

By November 15, 2018, children had been screened in less than a third of the rural communes (19 out of 75). The importance of screening in rural areas should not be overlooked. While in Cluj County a minority of the population lives in rural areas (34%), in the whole of Romania almost half the population lives in rural areas (46%). In some counties this is as much as two-thirds. When considering nationwide implementation of vision screening in Romania, finding a feasible way to screen in rural areas is therefore imperative.

Most seem convinced that the way to deal with many if not most of the aforementioned problems could be a combination of several methods of operation. A travelling nurse, whose travel expenses are reimbursed, could be employed to visit the underserved rural areas and screen the children there. In order to ensure this travelling nurse is able to efficiently screen children, local contacts would need to be appointed in every commune. A local contact could be the local kindergarten caretaker, a local kindergarten nurse if present or the local primary school teacher; someone with a network in the community who knows parents and children and is aware of specific local circumstances. Since the local contact would also need to be able to inform parents and answer their questions, some education may be required.

The local contact could do all the preparatory work: contacting and informing parents, obtaining their consent (only during the course of the study) and scheduling dates for screening. This way when the travelling nurse visits, he/she can focus on the actual screening since all the other work has already been taken care of. An additional benefit of this way of working would be that the travelling nurse would be able to screen many children and therefore gain experience quickly.

In order to ensure this runs smoothly and as efficiently as possible, someone at the county level should coordinate the efforts of the local contacts and the travelling nurse and serve as knowledge base for both.

There is not a lot of experience with follow-up in rural areas yet, because so far few children have been screened, but views among doctors and nurses differ as to whether parents will actually take referred children to the ophthalmologist – always requiring travel to a nearby city – and, if necessary, buy patches or glasses. This mostly seems to depend on how affluent the parents are, though awareness of the necessity was also mentioned as a relevant factor.

It should be noted, however, that thus far parents have not been study objects and therefore all information on parents' attitudes towards screening in the different communities was provided by others (nurses, doctors, kindergarten caretakers).

## 1.2 On-site observations of the implementation of vision screening in Cluj County by Mandy Nordmann and Jan Kik (October 2019)

**Background**

A vision screening programme is being implemented in 2018 and 2019 in Cluj County in north-western Romania. Cluj-Napoca is the county seat with a population of 324,276. The county's urban areas consist of the municipalities of Turda (population 55,907), Dej (38,250), Câmpia Turzii (27,745), Gherla (23,002) and the town of Huedin (9,564). In the county's rural areas 251,481 people reside, spread across 75 communes (groups of villages).

All children aged four and five in Cluj County were eligible for screening, meaning that all children born in Cluj County in 2013, 2014 and 2015 were eligible for screening during the two years of the implementation. Accordingly, although the target was set a two birth years, the total number of eligible children comprised three birth years. This is the reason why the DASM in Clij-Napoca will probably surpass two birth years. Altogether these were 19,752 children: 9,381 in Cluj-Napoca, 3,495 in the small cities and 6,876 in the rural areas.

The vision screening programme is being implemented in Cluj-Napoca by the Directia de Asistenta Sociala si Medicala (DASM) and in urban and rural areas by the Universitatea de Medicină şi Farmacie (UMF-Cluj). In the cities, children were screened at kindergartens by the resident nurses. Children living in Cluj-Napoca were screened at kindergartens by nurses working for the DASM, who had screening included in their job descriptions. In the small cities children were also screened at kindergartens by the nurses working there, who were hired specifically for this task by the UMF-Cluj. In the rural areas the children were to be screened by the family doctors' nurses at the doctors' offices, because there generally are no nurses at the rural kindergartens (save in some larger communes). All nurses received a payment of €7,- gross (around €4 net) per child screened. The nurses had to attend a special screening course and pass an exam in order to be allowed to screen children.

When it became clear during the first year of implementation that hardly any children were being screened in the rural areas, in June and July 2018 the possible causes were discussed. One of the obstacles was that the family doctors demanded at least €1,50 for use of their practice room during the screening examinations by their nurse. In August 2018 the family doctors' nurses were advised to screen the children at the kindergartens instead of the doctors' offices. This did not result in many more children being screened: after the first year of implementation, 957 children had been screened in rural areas, in 23 of 75 communes.

In December 2018, as only a fraction of children had been screened of the eligible rural children, the UMF-Cluj's budget was cut accordingly, regarding the variable portion of the budget. This amount has been transferred to the DASM, who have been very successful in screening and will screen more than the target in the second half of 2019.

On the basis of the calculations of the cost-effectiveness model, that showed it would be cheaper for a travelling nurse to visit kindergartens in the villages, in March 2019, a travelling screening nurse was hired by the UMF-Cluj who visited the rural kindergartens to screen children. By August 14th, the number of children screened in rural areas in the database had increased to 2,036, or 30% of the total number of eligible children in rural areas (at this point, Cluj-Napoca was at 62% and the small cities were at 61%). Per September 1st, 2019 the payment for the travelling nurse was increased to €10,- gross (€5,50 net) per child screened.

By September 16th, 2019, according to the project database, 5,863 children had been screened in Cluj-Napoca (63% of the total number of eligible children; equal to three birth years), 2,182 in the small cities (62%) and 2,256 in the rural areas (33%). Children were screened in 56 of the 75 rural communes.

In October 2019, Cluj County was visited for the fourth time to determine the adoption of and adherence to the programme. Interviews were held with screeners on several locations across the county that were visited, as well as with principals and teachers at rural kindergartens. In addition, screenings were observed and interviews were held with other professionals involved in the programme such as ophthalmologists, data entry personnel, financial and legal professionals and public servants. On October 14th, a focus group was organised with twelve nurses from Cluj-Napoca, small cities and rural areas where they discussed their experiences with the screening programme.

**Screening in rural areas**

Because the numbers show that screening in rural areas is not going as well as in the cities, the different ways screening has been attempted in the rural areas were a focal point of the on-site observation.

According to eight of the family doctors' nurses who attempted to screen children at the doctor's office, this was unsuccessful because when parents were invited specifically for screening the majority did not come. This was also mentioned in the focus group.

The main reasons for their unwillingness to come to the doctor's office for screening were the extra time it takes for the parents and the fact that it was not a priority for them, according to five nurses. This was confirmed in the focus group.

Consensus in the focus group was that combining vision screening with something else is not possible. There is a vaccination at age five but when children have to come in for a shot they may be afraid and most likely not in the right mind set for VA testing (and not all parents bring the children for vaccinations either). The nurses also think that that it would be too much for the children and therefore they would be less attentive. Two nurses who were interviewed individually dissented from this opinion, though, and said screening can be combined with something else. One of these said this will depend on the individual child.

Two nurses who initially tried to screen at the family doctor's office, reported they themselves decided to switch to screening at the kindergarten when it became clear that very few or no parents would bring their children to the doctor's office for screening. Two nurses said they persisted in screening at the family doctor's office, because there was no suitable space for screening at the kindergarten, but one admitted to having problems getting the parents to bring the children for screening.

Another problem is that not all family doctors' nurses in rural areas were willing to screen children, meaning that for the communes where this was the case another solution had to be found. One nurse was interviewed who cited a lack of time as the reason she did not want to screen, even though she said she thought screening is important. However, as the only nurse at a doctor's office with 3,000 patients, there is no way she could fit screening into her routine.

In August 2018, family doctor's nurses were advised to screen children at the kindergartens instead of the doctor's office. Two interviewed nurses said they had already begun doing this of their own accord, as mentioned above.

The nurses initially went to the kindergarten to explain the screening to the principal and/or the teachers. The teachers then informed the parents and distributed the informed consent forms (necessary because the implementation study is a research project). Subsequently a date for screening was set.

While the nurses in the focus group were positive about this way of screening, one nurse who tried to screen at a kindergarten was initially unsuccessful because the kindergarten teachers refused to cooperate. In this case, this could be solved by contacting the principal, but if the principal had also refused to cooperate nothing could have been done because screening is not mandatory and therefore voluntary cooperation of the kindergarten staff is required. According to the nurse who encountered this refusal to cooperate, the reason given by the teachers was a lack of time to accommodate the screening programme.

A downside of screening by the family doctor's nurse is that, if she only screens children in her own commune, in many small communes she will screen only a small number of children and therefore may not achieve the required level of screening proficiency.

Obviously, screening at the kindergarten by the family doctor's nurse is also not an option in communes where the nurse refuses to screen in the first place.

**Travelling nurse**

In March 2019, a travelling nurse was employed by the UMF-Cluj who visits kindergartens in communes where the local family doctor's nurse does not want to screen. The travelling nurse worked in the same way as the family doctors' nurses who screened at the kindergartens: she first visited a kindergarten to explain the screening and hand out consent forms, kept in touch with the kindergarten teachers by phone and once the consent forms were signed, a date was set for screening and the travelling nurse visited the kindergarten again to screen the children.

Of course, like the family doctors' nurses, the travelling nurse is dependent on the voluntary cooperation of kindergarten staff. Although the number of kindergartens that adamantly refuse to cooperate is small (estimated by the UMF-Cluj at no more than ten in the entire county) this is an issue that needs to be addressed if full coverage is the goal. It is important to realize that the kindergarten staff are not reimbursed in any way for their cooperation.

The travelling nurse has been successful in screening many children. According to the database, she screened 486 children between March and September 16$^{th}$, 2019. She screened enough children to be self-proficient. This however is based on very limited reimbursement: only the used petrol is reimbursed, whereas all other car costs are ignored by the UMF-Cluj. She also had a relevant car repair due to damage because of bad roads in rural areas with potholes, but the UMF-Cluj declined to participate in these costs.

The concept is not without its drawbacks: the travelling nurse requires a lot of (travelling) time to screen children, because in many rural kindergartens the number of eligible children is small. Each kindergarten has to be visited twice: once to hand out the informed consent forms and once to screen the children.

The number of children present in rural kindergartens is, in most cases, also (much) smaller than the number of children supposed to be there. This was observed at every kindergarten visited. According to four kindergarten teachers, this is because many parents who work in nearby cities take their children to the kindergartens there, because the kindergartens in the cities are usually open until five PM while the rural kindergartens close at one PM at the latest.

Also, the number of attending children that is actually present tends to fluctuate throughout the year, according to one teacher, mainly because fewer children attend in winter because of illness and weather conditions, while in January more children are present because the parents receive vouchers and at Christmas more children attend because then they receive presents. When attendance is low when the travelling nurse visits the kindergarten to screen, she needs to travel to the kindergarten again at a later date to screen the children who were not present the first time.

Other issues the travelling nurse recounted that make planning difficult are personnel changes at kindergartens, meaning that sometimes the travelling nurse arrives at a kindergarten on the day she was supposed to screen but finds other teachers there who are unaware of the arrangements she made with the previous ones, meaning she has to start over again. It also happens that teachers cancel a scheduled visit on short notice.

According to the travelling nurse, the job is not very attractive. The main reason for this is the effort required per child screened and the low numbers of children she encounters at rural kindergartens, meaning low pay. Additionally she mentioned the uncertainty of how many children she will be able to screen, meaning no stable income and the fact that she has to use her own car and her travelling expenses, other than gas, are not reimbursed (for example car damage sustained because of poor road conditions). By and large, to screen the children in the remaining 19 communes that are motivated for screening as mentioned by two principals, the travelling expenses must be reimbursed to a reasonable level per kilometre driven.

**Refusal to consent**

A specific issue encountered not only in the rural areas, but also in the cities, was parents' refusal to consent to screening, even when most nurses said that parents' initial response to the screening programme was positive.

It should be noted, however, that parents' informed consent is only needed in the EUSCREEN implementation study and will, in the future, not be required when screening takes place as an integrated component of preventive health care.

Nurses (both those individually interviewed and in the focus group) and kindergarten teachers mentioned several reasons why parents refuse consent:

- they do not want to fill out the ID number on the consent form
- they are afraid to sign something
- the child has already been diagnosed with an eye condition
- lack of awareness of the importance of screening

Exactly how many parents refuse consent is difficult to assess. Nurses (both individually and in the focus group) and kindergarten teachers all gave very different accounts: some said all parents consented without any trouble whatsoever (6) or only a few refused (1), while others reported almost all parents refused consent (2). One nurse mentioned it was very difficult to get the parents to consent, even when they did not explicitly refuse. According to her, this is because

screening is just not a priority for them so they do not take the trouble or forget about it. She said she had to hand out the forms three times before they were returned.

According to nurses and kindergarten teachers who work in areas where a substantial part of the population is Roma (2), refusal of consent is common among Roma communities, though not among all of them. They also mentioned that kindergarten attendance is also lower among Roma communities, although, again, not everywhere.

Parents' refusal to consent is an issue that requires further analysis. It is, however, difficult to definitively comment on parents' attitudes towards screening because parents were not a study object and therefore all information on parents' attitudes was provided by others (nurses, doctors, kindergarten teachers).

**Quality and follow-up**

Quality and follow-up are points of concern everywhere, not just in the rural areas: based on the available information (the project database and the on-site observations) there is insufficient insight in the quality of the screening and there is a lack of (data on) follow-up.

It would appear that the lack of (data on) follow-up is a bigger problem than establishing full coverage, also because this makes it impossible to draw conclusions on the quality of the screening. There are some indications, though, that the quality of screening is not up to standard: the overall referral rate is still higher (12%) than to be expected and referral rates vary enormously between different nurses. This suggests some nurses have high rates of false positives and others high rates of false negatives. Of the 104 screeners who had screened children according to the database by September 16th, 2019, 42 had a referral rate between 5% and 15%, 26 a referral rate above 15% and 36 a referral rate below 5% (sixteen did not refer a single child, even though they screened 58 children on average and four of these screened more than 100 children). This means that the majority of screeners had a referral rate outside of the 5-15% window.

If children already wear glasses, in most cases they are not screened and of course not referred either. This might lower the number of referred children. During the observation at least one child per classroom was wearing glasses.

Several nurses mentioned that four-year-old children are too young to screen properly (4).

Observation of eighteen screenings in October 2019 indicated that most nurses' screening performance has improved. The average time for screening was around five minutes, compared to 8.3 minutes at the beginning of screening.

The nurses knew to which line they had to measure the visual acuity (VA) per age, they instructed the children well and seemed comfortable in performing the screening. Not all nurses were aware of the referral criteria (refer when there is a difference of two or more lines in VA between both eyes). This was most important for children aged four that were measured up to VA 0.2 LogMAR and no further. Children with a two line difference of 0.0 LogMAR and 0.2 LogMAR will not be detected in this way.

It was observed that screening in rural areas, at the doctor's office by the nurse, took a lot of time and effort. Because the child was not used to the nurse and in an unfamiliar setting, screening was difficult, even though a parent was present.

The database that keeps tracks of all the children's examinations appears to function adequately. According to personnel responsible for data entry (2), the database is intuitive and easy to work with. Entering a single form takes about five to ten minutes, depending on how completely it is filled out.

Analysis of the database shows that most of the data are entered correctly in the database, with the exception of some errors. These do raise concerns about the quality of the data as well as the quality of the database itself. Had more dropdown fields been employed in the database instead of text fields, for example, many of these mistakes could not have occurred. A bigger issue, however, is that once a record has been submitted, it can no longer be edited, so mistakes cannot be corrected.

There are children who are examined by an ophthalmologist but whose diagnosis (the so-called form 5) is never received. Although a few of those involved (nurses, teachers) say most or even all children bring back the form (3), the majority say no or very few children bring it back (8). In some cases, a prescription is received instead (2).

According to two nurses, some ophthalmologists refuse to fill out the form because they consider it too much trouble. Also, not all ophthalmologists in the county were willing to participate in the project. Parents were free to choose an ophthalmologist for examination and ophthalmologists not willing to participate are unlikely to fill out an extra paper form. One nurse in the focus group suggested giving parents the name of just one ophthalmologist instead of a list, but this is not legally possible because parents have to be able to choose.

According to several nurses (both individually and in the focus group) and teachers, it also happens that the ophthalmologist fills out the form, but the parents never return it, especially when nothing is found wrong with the child (1) or that the parents bring the form to the family doctor instead of the screening nurse (1).

One nurse stated she specifically stresses to the parents the importance of returning the form.

In the focus group, it was mentioned that sometimes children leave the kindergarten shortly after being screened (because their parents move or because they turn six and go to school) and the teachers no longer are in contact with the parents and never receive the form.

In any case, the number of forms entered in the database is small. For 1,300 children referred after either the first (1,009) or second (291) screening, 210 diagnostic forms were entered it the database by September 16$^{th}$, 2019 (16%). A possible solution for the lack of forms being returned, suggested by one ophthalmologist, could be to give ophthalmologists the option the fill out the forms digitally.

While the issue of forms not being returned needs to be addressed, it appears many of the referred children are simply never examined. It is, however, amazing how opinions among nurses and kindergarten teachers varied on the subject of whether parents of referred children will take the children to an ophthalmologist, but many say most or all parents will not do so (6). On the other hand, many others say most or all parents will take the children to be examined (6), while yet others say about half the parents will do so (2) and yet others say they simply they do not know (3).

**Unsuccessful referrals**

Two kindergarten teachers specifically mentioned that it is a flaw of the current project that it provides screening, but not diagnosis and treatment. One nurse mentioned that

some parents also expected the screener to take the referred children to the ophthalmologist herself.

Based on the accounts of nurses and teachers, there seem to be large differences between locations when it comes to this issue, but it does appear this is a bigger problem in rural areas.

The following barriers to follow-up were mentioned by nurses (both individually and in the focus group) and teachers:

- when a child is referred by the screening nurse, the parents cannot go to an ophthalmologist straight away, but have to go to the family doctor for an official referral first
- in rural areas there are no ophthalmologists so travel to a city is necessary. Some communes are close to a nearby city, but others are not
- some parents lack awareness and do not see the importance of screening
- some parents have other priorities and/or lack the time
- some parents cannot afford travel and/or treatment
- some parents refuse to accept the child has a problem because as far as they can tell the child can see, they do not observe that something is wrong and it does not hurt

**Continuation of screening**

When asked, all involved say they think screening should be continued (7). A few said it would be best if the project were simply continued (2), but this is not a possibility. Several said it would be best if screening were implemented nationwide and paid for by the Ministry of Health (4). All agreed, though, that this is unlikely to happen, because the health budget is low and there are many other priorities. Prevention is usually not a priority.

If the Ministry were to be convinced, according to public servants (3), this will have to be based on the results of the current study. Several screening projects in Romania were discontinued over the past years after the pilot phase because the results were unsatisfactory. A model simulation showing that screening can be cost-effective may also help, but the results of the study will be more important.

Another problem is that even if screening were to be financed by the Ministry, the cost of treatment would still have to be covered by the health insurance and it is deemed unlikely this will happen (2).

The nurses in the focus group, as well as two individually interviewed nurses insisted that they will continue screening no matter what. They have had the training, they now have the equipment and they find that screening enhances their status in the eyes of the parents and the community.

All involved agree that the Ministry should at least make screening mandatory even if it does not finance it. This would at least mean that screening is no longer fully dependent on voluntary cooperation from parties involved. If screening would be mandatory, outside of a research project, informed consent would also not be required, thus removing one of the barriers.

**Conclusions**

Coverage and attendance are not a problem in Cluj-Napoca and the small cities, but are an issue in the rural areas. Of the options investigated during the implementation, screening by the family doctor's nurse at the kindergarten or by the travelling nurse are the kindergarten appear to be the only feasible ones. Both options have advantages and disadvantages, though there appear to be differences in what works between different locations.

A hybrid solution could be to have family doctors' nurses screen in their own commune and several surrounding communes, as 'semi-travelling' nurses. This way they would be able to screen more children and develop and maintain their skills more efficiently. Some family doctors' nurses are already doing this, of their own accord, and they say this is working well (2).

The option to train the kindergarten teachers to screen has not been explored in this study, because a medical examination by teachers who do not have any paramedical training was excluded from the outset, for it is unlikely that this would be permitted for an eye examination whereas all the other examinations for preventive health care are done by the family doctors or their nurses. Nevertheless, a few of those interviewed mentioned this as a possibility (2) and one kindergarten teacher said she would be willing to learn to screen. It would have a lot of practical advantages over the aforementioned options: the teachers already have a relationship with the parents, they are present every day so children's attendance is less of a problem and if one teacher screens all children at a kindergarten he/she will be able to screen a relatively large number of children.

There are indications that the quality of screening is not up to standard (persistent high overall referral rate, large differences in referral rates between screeners). However, because of a lack of available data on follow-up it is not possible to assess the quality of the screening more precisely. When nurses are observed when performing screening, they do appear confident and able to motivate the children.

Another point of concern is a lack of follow-up in general. It appears many parents do no take referred children to an ophthalmologist, though there is no consensus among nurses and kindergarten teachers about this and an exact number cannot be given because it is not known how many children were examined without their diagnosis ever being brought back.

While there is widespread support for continuation of screening after the end of the project among those involved, most consider it unlikely the Romanian government will be willing to finance nationwide implementation, because of a lack of funds and many competing priorities.

However, screening will require some funding. Even when nurses continue screening in spite of no longer receiving additional pay for it, there will be nurses who retire, leave for other jobs and so forth. These will have to be replaced with other nurses who will need training. Also, since screening is still in its infancy in Romania, some monitoring will be necessary to assess the quality of the screening. This will also incur costs. Charts will have to be replaced from time to time and a travelling nurse is of course also not an option without funding.

**Appendix 2: overview vision screening in rural areas Cluj County 2018-2019**

Here the data entered in the project database by January 10th, 2020 are presented. Note that these numbers are not definitive, because not all data pertaining to 2019 had been entered in the database by then.

*Table 1: children screened in Cluj-Napoca, small cities and rural areas, related to the total number of eligible children*

| | **Children eligible (3 birth years)** | **Children screened 2018-2019** | **Percentage of eligible children (3 birth years)** |
|---|---|---|---|
| Cluj-Napoca | 9382 | 6707 | 71% |
| Small cities | 3495 | 2372 | 68% |
| Rural areas | 6877 | 2870 | 42% |
| **Total** | **19754** | **11949** | **60%** |

Almost two-thirds of the children who could have been screened (all children born in Cluj County in 2013-2015) have been screened, according to the database. However, the target was not three but two birth years.

*Table 2: children screened related to the target*

| | **Target (2 birth years)** | **Children screened 2018-2019** | **Percentage of target (2 birth years)** |
|---|---|---|---|
| Cluj-Napoca | 6254 | 6707 | 107% |
| Small cities | 2330 | 2372 | 102% |
| Rural areas | 4584 | 2870 | 63% |
| **Total** | **13068** | **11949** | **91%** |

As can be seen in table 2, the target has been achieved in Cluj-Napoca and in the small cities. In the rural areas, close to two-thirds of the target has been achieved. According to the database, 2,870 children were screened in rural areas, in 65 out of 75 rural communes. Note that in the last two months of 2019, children were screened in most of the 10 remaining communes as well, but the data pertaining to these were not yet introduced in the database when it was exported on January 10, 2020.

The map below shows the communes and cities where, according to the database, children have been screened in 2018 (red), 2019 (green) and both 2018 and 2019 (blue for communes, purple for cities). In two communes children were screened by the Lion's Club (grey). White communes are communes where, according to the database, no screening has taken place.

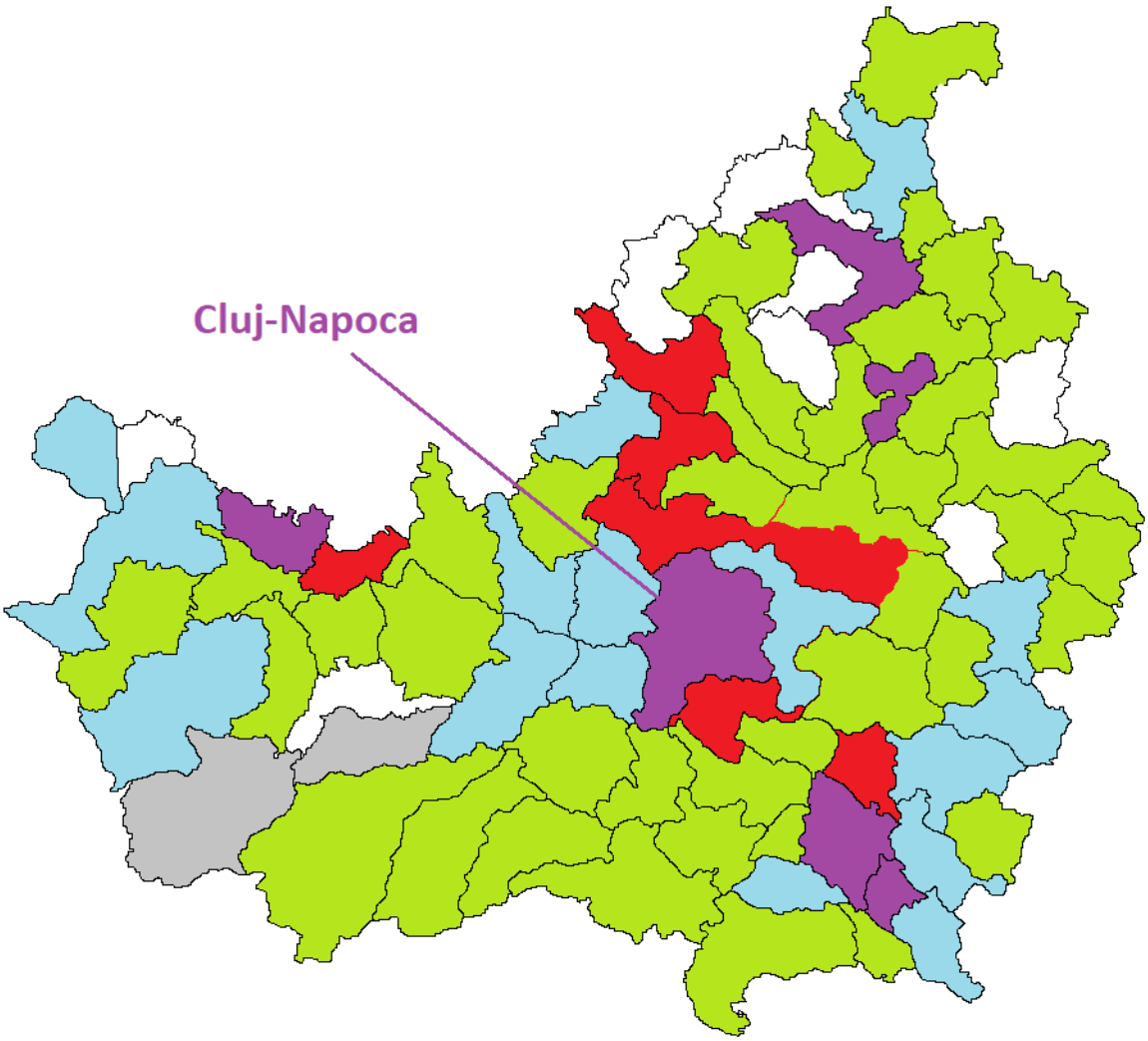



*Table 3: screening results (first screening)*

| | | |
|---|---|---|
| OK | 10297 | 86% |
| Repeat | 320 | 3% |
| Referral | 1317 | 11% |
| Unknown | 15 | 0% |
| **Total** | **11949** | **100%** |

*Table 4: screening results (second screening)*

| | | |
|---|---|---|
| OK | 147 | 46% |
| Referral | 125 | 39% |
| Unknown | 48 | 15% |
| **Total** | **320** | **100%** |

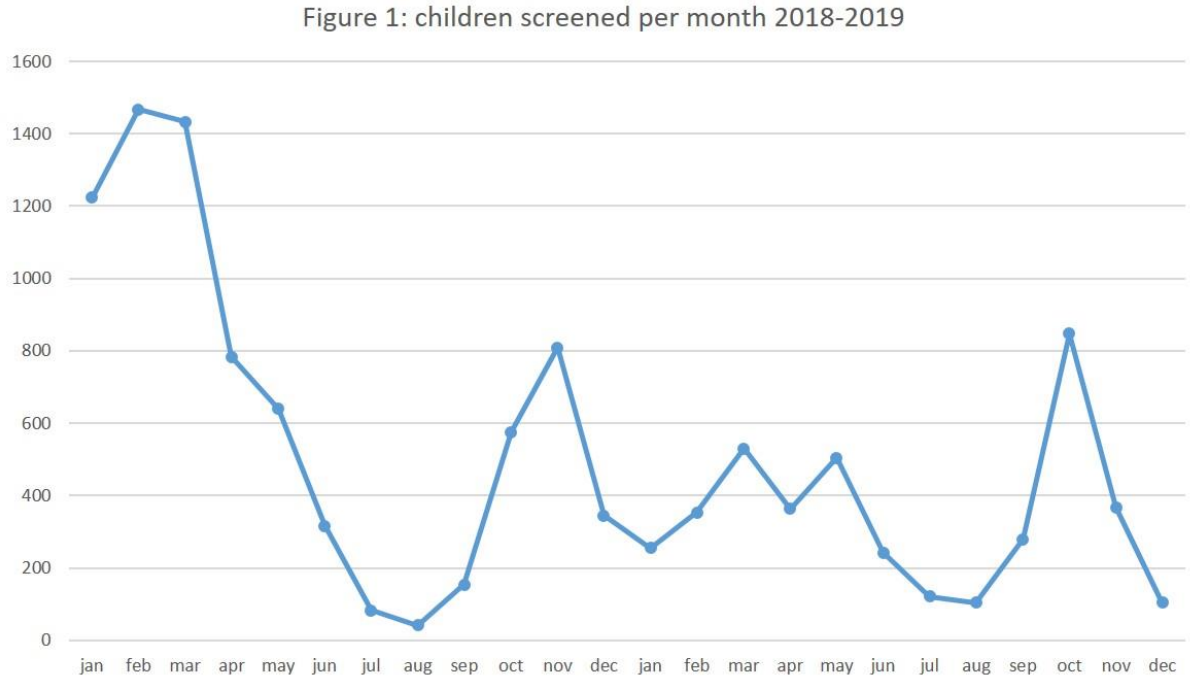
As expected, referral rates went down during the project. In the first quarter of 2018 the overall referral rate was 15% while by the last quarter of 2019 the rate had dropped to 7%. There were fluctuations, though, and there were also large differences in referral rates between the 104 screeners, as can be seen in table 5. A large number of screeners had a referral rate of 5% or lower (41%). Of these, sixteen did not refer a single child even though they examined, on average, 68 children (four screened more than 100 children, of which two screened more than 200 children). This suggests high rates of false negatives.
Also of note is the fact that five-year-olds were referred more than twice as often as four-year-olds (15% compared to 7%).

*Table 5: screeners' referral rates*

| | |
|---|---|
| >40% | 4 |
| 31-40% | 2 |
| 21-30% | 10 |
| 11-20% | 20 |
| 6-10% | 25 |
| 1-5% | 27 |
| 0% | 16 |

There were also differences between areas: in Cluj-Napoca the referral rate was 13% while in both the small cities and the rural areas it was 8%. Altogether 1,442 children were referred (12%). Diagnostic reports were entered for 218 children (15%). In 83 of these 218 cases, glasses were prescribed (39%). In 16 cases, occlusion was prescribed (8%).

Figure 1 shows when the children were screened. The largest number of children was screened in the first three months of 2018, the first quarter of the project (35% of all children screened during the two years of the project).



The table below shows how old the children were when they were screened.

*Table 6: age of children screened*

| **Age** | **Cluj-Napoca** | | **Small cities** | | **Rural areas** | | **Total** | |
|---|---|---|---|---|---|---|---|---|
| 3 | 14 | 0% | 7 | 0% | 8 | 0% | 29 | 0% |
| 4 | 3754 | 56% | 1306 | 55% | 1394 | 49% | 6454 | 54% |
| 5 | 2218 | 33% | 1028 | 43% | 1434 | 50% | 4680 | 39% |
| 6 | 10 | 0% | 14 | 1% | 18 | 1% | 42 | 0% |
| Unknown | 711 | 11% | 17 | 1% | 16 | 1% | 744 | 6% |
| **Total** | **6707** | **100%** | **2372** | **100%** | **2870** | **100%** | **11949** | **100%** |

In Cluj-Napoca and the small cities, noticeably more four- than five-year-olds were screened. In the rural areas the numbers were about the same. Also noticeable is the fact the in Cluj-Napoca, in a substantial number of cases, the age of the child was not entered in the database (11%). The number of ineligible children (three- and six-year-olds) that were screened was negligible.

*Table 7: children screened in the small cities, related to the total number of eligible children*

| | **Children eligible (3 birth years)** | **Children screened 2018-2019** | **Percentage of eligible children (3 birth years)** |
|---|---|---|---|
| Campia Turzii | 617 | 548 | 89% |
| Dej | 852 | 459 | 54% |
| Gherla | 568 | 457 | 80% |
| Huedin | 278 | 162 | 58% |
| Turda | 1180 | 746 | 63% |
| **Total** | **3495** | **2372** | **68%** |

*Table 8: children screened in the small cities, related to the target*

| | **Target (2 birth years)** | **Children screened 2018-2019** | **Percentage of target (2 birth years)** |
|---|---|---|---|
| Campia Turzii | 411 | 548 | 133% |
| Dej | 568 | 459 | 81% |
| Gherla | 379 | 457 | 121% |
| Huedin | 185 | 162 | 88% |
| Turda | 787 | 746 | 95% |
| **Total** | **2330** | **2372** | **102%** |

As table 8 shows, the target has been reached in Campia Turzii and Gherla, with the other three small cities not being far from the target.

*Table 9: children screened in rural communes in 2018-2019 (and entered in the database by January 10, 2020)*

| Commune | Total eligible (3 birth years) | Children screened | |
|---|---|---|---|
| Aghiresu | 157 | 74 | 47% |
| Aiton | 16 | 4 | 25% |
| Alunis | 26 | 0 | 0% |
| Apahida | 424 | 185 | 44% |
| Aschileu | 41 | 30 | 73% |
| Baciu | 366 | 108 | 30% |
| Baisoara | 36 | 19 | 53% |
| Belis | 37 | 0 | 0% |
| Bobalna | 38 | 20 | 53% |
| Bontida | 146 | 85 | 58% |
| Borsa | 22 | 11 | 50% |
| Buza | 25 | 9 | 36% |
| Caianu | 63 | 32 | 51% |
| Calarasi | 46 | 24 | 52% |
| Calatele | 66 | 15 | 23% |
| Camarasu | 114 | 41 | 36% |
| Capusu Mare | 62 | 18 | 29% |
| Caseiu | 138 | 46 | 33% |
| Catcau | 67 | 14 | 21% |
| Catina | 40 | 14 | 35% |
| Ceanu Mare | 74 | 101 | 136% |
| Chinteni | 92 | 11 | 12% |
| Chiuiesti | 68 | 40 | 59% |
| Ciucea | 25 | 0 | 0% |
| Ciurila | 45 | 7 | 16% |
| Cojocna | 147 | 78 | 53% |
| Cornesti | 25 | 9 | 36% |
| Cuzdrioara | 70 | 9 | 13% |
| Dabaca | 26 | 5 | 19% |
| Feleacu | 90 | 25 | 28% |

| | | | |
|---|---|---|---|
| Fizesu Gherlii | 75 | 11 | 15% |
| Floresti | 1514 | 688 | 45% |
| Frata | 100 | 68 | 68% |
| Garbau | 52 | 10 | 19% |
| Gheaca | 32 | 13 | 41% |
| Gilau | 239 | 231 | 97% |
| Iara | 75 | 9 | 12% |
| Iclod | 100 | 53 | 53% |
| Izvoru Crisului | 20 | 7 | 35% |
| Jichisu de Jos | 25 | 0 | 0% |
| Jucu | 102 | 59 | 58% |
| Luna | 97 | 32 | 33% |
| Maguri Racatau | 61 | 28 | 46% |
| Manastireni | 30 | 7 | 23% |
| Margau | 39 | 37 | 95% |
| Marisel | 38 | 0 | 0% |
| Mica | 108 | 19 | 18% |
| Mihai Viteazu | 91 | 59 | 65% |
| Mintiu Gherlii | 102 | 19 | 19% |
| Mociu | 82 | 56 | 68% |
| Moldovenesti | 69 | 31 | 45% |
| Negreni | 41 | 37 | 90% |
| Palatca | 25 | 0 | 0% |
| Panticeu | 63 | 17 | 27% |
| Petrestii de Jos | 15 | 7 | 47% |
| Ploscos | 11 | 3 | 27% |
| Poieni | 104 | 36 | 35% |
| Recea-Cristur | 55 | 0 | 0% |
| Risca | 17 | 0 | 0% |
| Sacuieu | 46 | 5 | 11% |
| Sancraiu | 23 | 15 | 65% |
| Sandulesti | 48 | 9 | 19% |
| Sanmartin | 24 | 0 | 0% |
| Sanpaul | 87 | 17 | 20% |
| Savadisla | 93 | 33 | 35% |

| | | | |
|---|---|---|---|
| Sic | 48 | 26 | 54% |
| Suatu | 39 | 14 | 36% |
| Taga | 33 | 12 | 36% |
| Tritenii de Jos | 103 | 33 | 32% |
| Tureni | 47 | 27 | 57% |
| Unguras | 59 | 22 | 37% |
| Vad | 56 | 0 | 0% |
| Valea Ierii | 14 | 5 | 36% |
| Viisoara | 161 | 65 | 40% |
| Vultureni | 22 | 16 | 73% |
| **Total** | **6877** | **2870** | **42%** |

**Appendix 3: issue identified with nurses who screened many children but did not refer any**

It was discovered, through analysis of the project database, that four nurses who each screened more than 100 children (and two of which who screened more than 200) did not refer a single child. This is shown in the table below (data exported on January 10, 2020). Assuming this is not a case of database errors (and there is no reason to assume it is) these are extremely unlikely results. The prevalence of amblyopia is approximately 3.2%, in fully developed screening programmes like in the Netherlands about 8% of children is referred for diagnostic evaluation by an orthoptist and ophthalmologist.

Assuming a low referral rate of 3.2%, the chance of a nurse not referring any children is less than 5% as soon as the number of examined children exceeds 92, according to the formula (HP Prime) BINOMIAL_CDF (93, 0.032, 0) = 0.0486, which is slightly less than alpha = 0.05.

| | Screened 2018 | Screened 2019 | Total screened |
|---|---|---|---|
| Nurse A (UMF rural) | 86 | 210 | 296 |
| Nurse B (UMF rural) | 96 | 119 | 215 |
| Nurse C (DASM) | 160 | 0 | 160 |
| Nurse D (UMF urban) | 93 | 35 | 128 |
| **Total** | **435** | **364** | **799** |

Further analysis revealed that the 86 children screened by nurse A in 2018 all had the same vision in both eyes, according to the database. Nurse A was hired in November 2019 as the second travelling nurse to visit rural kindergartens.

Altogether, there were sixteen screeners (out of a 104) who did not refer any children, as can be seen in the referral rates table below.

| | |
|---|---|
| >40% | 4 |
| 31-40% | 2 |
| 21-30% | 10 |
| 11-20% | 20 |
| 6-10% | 25 |
| 1-5% | 27 |
| 0% | 16 |

Of the other twelve nurses who did not refer any children, though, none screened more than 74. On average these twelve nurses screened 24 children.

**Appendix 4: Contribution of the Erasmus MC to the implementation study in Albania**

EUSCREEN hearing experts from the Erasmus MC studied the implementation of neonatal hearing screening (NHS) in three provinces in Albania. The screening process was observed during several visits. These visits took place in July 2017 during the preparation period, and in January 2018, April 2018, January 2019 and October 2019, during the implementation of NHS. Based on the on-site observations, as well as analysis of the project database, EMC is finalising a first article on the implementation study and preparing a second, in cooperation with the Albanian team.

Four locations participated in the study, two maternity clinics in Tirana, one in Pogradec and one in Kukës. The screening process and the screeners were observed, questionnaires were distributed amongst screeners, and interviews with screeners and parents were conducted, to identify barriers, facilitators and requisites. The results of the screening process were monitored for all locations using a database in which the screening outcomes are collected. In April 2018, a multidisciplinary team from the Child Centre for Rehabilitation was trained by hearing experts of the Erasmus MC and an external expert. This team has an important task in the diagnostic assessment and early intervention of infants with a hearing impairment. Infants and their parents can stay up to two weeks in this Child Centre for daily therapy.

During the first months, equipment failures such as malfunctioning OAE devices and broken OAE probes disturbed the screening process. The broken probes and devices were sent back to the manufacturer for replacement but it took several weeks for the equipment to be repaired. This did not delay screening since back-up screening devices were available. Screeners were instructed to be more careful when handling the probes and transporting the devices. Initially, it was difficult for the screeners to place the probe and make sure all conditions were right for screening. Many tests were paused and restarted when screeners experienced difficulties placing the probe or when the infant was restless during the test. Initially "Fail" results occurred often when screeners had not yet gained sufficient experience in screening. These problems resulted in high failure rates (50-80%) for the first screen. In Pogradec, 78% of infants were referred to the second screen in the first month of screening. In all maternity hospitals the failure rates for the first screen decreased to less than 10% after eight months as experience was gained. The more neonates were being screened, the better the test seemed to be executed and the lower the failure rate was. During the later visits equipment problems played a minor role. The presence of spare parts/systems was however crucial to keep the screening on going in case of (small) technical failures. This has to be taken in account when considering the costs of a new screening programme.

Screeners were very motivated to perform the hearing screening. The answers to the initially distributed questionnaire identified a positive attitude from the screeners towards the screening program. They indicated to believe screening should be provided to all Albanian newborns. Screeners feel confident and able to participate in the program, they are ready to take on extra screening tasks because they strongly believe in this screening program. They found the training course to be very informative but would like to have more follow up training. The answers provided by the screeners after one year of screening were generally similar, but also showed some differences: after one year the screeners felt more confident, did not have to spend as much time preparing for screening and considered screening to be an obvious part of their daily work. Screeners spend more time on informing the parents about hearing screening and noticed parents understood the aim of the screening program better because of their explanation. The screeners indicated to be interested to expand their knowledge on hearing and screening even further. As described above, the rate of 'fail' results for the first screen was high initially but a steep learning curve among the screeners was seen; the more neonates screened, the better the test was executed and the lower the referral rate. In the largest maternity hospital in Tirana, the number of infants born each day was so high, additional screeners were hired in 2019.

During the last visit in October 2019 we noticed that in Maternity 1 in Albania tests were repeated a number of times in case of initial failure of the test. This may be an important reason for the remarkably low referral rates at this location. According to the local protocol, the test may only be repeated two times. This issue was discussed with the screeners, they indicated to repeat the test more than two times when the conditions were unfavourable, for example a noisy environment or a restless infant. Another reason was that they wanted to be able to give the parents good news. A failure of the test may disappoint the parents and the family, which can be a motivation for the screeners to repeat the test until they have achieved a "good result". However, too many repetitions may lead to a "false negative result", which means that infants with hearing loss may be missed during the initial screening stage. After our observation, the screeners at this location were instructed to limit the number of repetitions to a maximum of two.

Parents were eager to participate in screening, which lead to high coverage rates across participating maternity hospitals in the first test round. However, after failing the first test, a substantial number of parents did not return for follow up screening. In the first year of screening, 67.4% attended the second screen and 65,6% the third screen. Despite the expectation that parents would be able to return more easily in more urban areas, a high number of non-attendance were recorded in the maternity hospitals in Tirana. Mothers from all areas of Albania give birth in the maternity hospitals in Tirana. After the parents returned home, they often perceived it to be too difficult or to be too costly to travel back to Tirana for repeat screening. Other important risk factors for lost to follow up, as reported by the screeners, were a low educational level and a strong religious background of parents. In Pogradec and Kukës parents were expected to have more difficulties returning for follow up screening when they live in rural areas and the maternity hospital could not be reached easily, especially in winter. Notwithstanding these difficulties, about 80% of parents attended follow up screening in Pogradec and about 90% attended follow up screening in Kukës. The screeners explained that parents were very motivated to return, Travel times were limited to a maximum of two hours, which appeared to be acceptable for parents.

Because of the expected high lost to follow up in the rural areas, we decided to implement only two screening steps in Kukës, using aABR instead of OAE. Based on the present results, such a two-step protocol may also be considered for the locations in Tirana. Another point of concern is that a protocol using aABR as third step after two steps OAE has the disadvantage that only few infants will be tested with aABR, especially in the two rural areas with low birth rates. With only a few aABR measurements per year, it is difficult to maintain the expertise of the screeners at the required level. Screeners in Pogradec confirmed that they did not always feel confident with the aABR screening. This might also be a reason to decrease the number of steps to two instead of three. When the definitive outcomes of the screening programme will become available during the finalisation of the project later this year, optimal strategies for hearing screening in Albania will be reconsidered using the complete data set in combination with the EUSCREEN cost-effectiveness model.

Analysis of the data in the project database by EMC showed that the coverage was high in all participating maternity hospitals, in 2019 more than 95% of the infants born in the maternities were screened. Out of all infants screened, 38 underwent a diagnostic test. The majority of these referred infants were diagnosed with permanent hearing loss. This means that hearing screening in Albania was implemented successfully, and despite continuing challenges, NHS will be continued by the local team after the EUSCREEN project has ended.

## Appendix 5: Evaluation of first use of cost-effectiveness model in workshop in Poznan

During the EUSCREEN study meeting in Poznan, a webtool version of the EUSCREEN vision and hearing screening evaluation model was presented. Participants (both Country Representatives and Consortium partners) had the choice between attending a demonstration of the prototype or participating in a practical exercise with the model prototype. The objectives of the workshop were a) to introduce the EUSCREEN webtool, b) to facilitate a practical exercise and c) to create a moment of feedback for further improvements of the tool.

All participants had received an email in preparation of the workshop. This included registration instructions and preparatory questions. There were approximately 20 participants in each exercise group (vision and hearing). A total of 15 participants filled out an evaluation form (6 for vision and 9 for hearing). This report provides mean responses and highlights some open comments. The evaluation form is attached to this report.

Overall, the workshop was rated 4.2 on a scale from 1 (strongly disagree) to 5 (strongly agree). The meeting of expectations by the webtool was rated 3.7. On average, the usefulness for various stakeholders (professionals, policy makers, coordinators and researchers) was rated 3.95.Table 1 shows the overall rating of the webtool content. With the first three items rating the overall usability of the webtool and the next five items rating each module of the webtool.

**Table 1: Mean scores of webtool evaluation**

| **Overall rating:** | **Mean score (scale 1 – 5)** |
|---|---|
| The lay-out was logic and user friendly | 4.0 |
| It was clear what information was required | 3.7 |
| The information/indicators required was relevant | 3.7 |
| **Module rating:** | |
| Priorities for public spending | 3.9 |
| Existing contact moments | 3.9 |
| Costs | 3.4 |
| Scenarios | 4.2 |
| Results | 3.8 |

Most responses were similar for both vision and hearing groups, with a maximum difference in average response of 0.6 points between the two groups. Except for the following three questions. Sufficient time for individual support during the workshop was rated 4.7 in the vision group and 3.9 in the hearing group. The meeting of expectations by the webtool was rated 3.3 in the vision group and 4.1 in the hearing group. The usefulness of the webtool for policy makers was rated 3.5 in the vision group and 4.2 in the hearing group.

If a variable is considered to be relevant in the webtool could be answered by either ‘yes’, ‘maybe’ or ‘no’. On average, all respondents found all variables relevant for the webtool. The number of screening locations and the number of screening devices were both found least relevant for the vision group (>50% of respondents answered no/maybe). The hearing group respondents found the number of tests failed/rejected the least relevant (six respondents answered ‘maybe’).

In general, it is found to be quite difficult to obtain empirical data on model parameters such as disease incidence; target condition; screening coverage and attendance rates; number of tests failed/rejected; training costs and treatment success rates.

**Open questions**

Not all respondents answered all open questions. It is therefore difficult to draw general conclusions from these answers. We will quote some responses that are provided, either written on the evaluation forms or verbally as feedback during the workshop sessions.

- Costs data is often difficult to provide and it should be made more clear what is exactly required.

- A help text could be useful, not only for costs but also for definitions of attendance and referrals.
- Some countries have vision screening and diagnostics within one consultation.
- Some countries perform both OAE and aABR tests for all well babies in their first hearing screen.
- In some countries, medical specialists have private offices where they perform their exams. This option should be made available.
- It would be useful to have a graphical overview of final results.

Additional output parameters that users would like to evaluate:

- Number of children with a refractive error
- Costs for false positive and false negative cases
- Number needed to screen per 1 case of hearing loss
-

Overall, the workshop was very well received by participants. It is noted that there was only limited time to get acquainted with the webtool. For that reason, the webtool remains available for all participants to practice. It is still possible to register as user at https://miscan.euscreen.org/login
All written and verbal feedback will be discussed within the EUSCREEN consortium and – where applicable and possible – be taken into account when improving the webtool.

**Questionnaires used**

| **Workshop experience** | **Strongly Disagree** | **Disagree** | **Undecided** | **Agree** | **Strongly agree** |
|---|---|---|---|---|---|
| Information was clearly presented. | 1 | 2 | 3 | 4 | 5 |
| There was enough time to practice. | 1 | 2 | 3 | 4 | 5 |
| There was enough time for individual support. | 1 | 2 | 3 | 4 | 5 |

| **Overall webtool experience** | **Strongly Disagree** | **Disagree** | **Undecided** | **Agree** | **Strongly agree** |
|---|---|---|---|---|---|
| The webtool met my expectations | 1 | 2 | 3 | 4 | 5 |
| This approach will be useful to policy makers in my country. | 1 | 2 | 3 | 4 | 5 |
| This approach will be useful to screening professionals in my country. | 1 | 2 | 3 | 4 | 5 |
| This approach will be useful to screening programme coordinators in my country. | 1 | 2 | 3 | 4 | 5 |
| This approach will be useful to screening researchers in my country. | 1 | 2 | 3 | 4 | 5 |

| **Webtool content** | **Strongly Disagree** | **Disagree** | **Undecided** | **Agree** | **Strongly agree** |
|---|---|---|---|---|---|
| **Priorities for public spending** | | | | | |
| The lay-out was logic and user friendly | 1 | 2 | 3 | 4 | 5 |
| It was clear what information was required | 1 | 2 | 3 | 4 | 5 |
| The indicators are relevant for countries with no screening programme | 1 | 2 | 3 | 4 | 5 |
| **Existing contact moments** | | | | | |
| The lay-out was logic and user friendly | 1 | 2 | 3 | 4 | 5 |
| It was clear what information was required | 1 | 2 | 3 | 4 | 5 |
| The information required was relevant | 1 | 2 | 3 | 4 | 5 |
| **Costs** | | | | | |
| The lay-out was logic and user friendly | 1 | 2 | 3 | 4 | 5 |
| It was clear what information was required | 1 | 2 | 3 | 4 | 5 |

| | | | | | |
|---|---|---|---|---|---|
| The information required was relevant | 1 | 2 | 3 | 4 | 5 |
| **Scenarios** | | | | | |
| The lay-out was logic and user friendly | 1 | 2 | 3 | 4 | 5 |
| It was clear what information was required | 1 | 2 | 3 | 4 | 5 |
| The information required was relevant | 1 | 2 | 3 | 4 | 5 |
| Adding 4 extra scenarios is sufficient | 1 | 2 | 3 | 4 | 5 |
| **Results** | | | | | |
| The lay-out was logic and user friendly | 1 | 2 | 3 | 4 | 5 |
| The information required were clear and understandable | 1 | 2 | 3 | 4 | 5 |

**Model input parameters**

The future toolkit will most likely contain a broader set of input parameters to vary. We would like to know your opinion on all parameters

| **Parameter** | **Relevant variable in the webtool** | **Too difficult to obtain data for my country** |
|---|---|---|
| Disease prevalence | Yes / Maybe / No | Yes / Maybe / No |
| Disease incidence by age | Yes / Maybe / No | Yes / Maybe / No |
| Target condition (for vision screening: refractive error, amblyogenic risk factors, etc.) | Yes / Maybe / No | Yes / Maybe / No |
| Referral criteria | Yes / Maybe / No | Yes / Maybe / No |
| Coverage | Yes / Maybe / No | Yes / Maybe / No |
| Attendance | Yes / Maybe / No | Yes / Maybe / No |
| Referral rate | Yes / Maybe / No | Yes / Maybe / No |
| Test sensitivity | Yes / Maybe / No | Yes / Maybe / No |
| Test specificity | Yes / Maybe / No | Yes / Maybe / No |
| Number of tests failed / rejected | Yes / Maybe / No | Yes / Maybe / No |
| Number of screening locations (current/new) | Yes / Maybe / No | Yes / Maybe / No |
| Types of screening professionals | Yes / Maybe / No | Yes / Maybe / No |
| Number of screening professionals (current/new) | Yes / Maybe / No | Yes / Maybe / No |
| Types of screening devices | Yes / Maybe / No | Yes / Maybe / No |
| Number of screening devices (in use / needed) | Yes / Maybe / No | Yes / Maybe / No |
| Salary costs of screening professionals | Yes / Maybe / No | Yes / Maybe / No |
| Costs of training current / new professionals | Yes / Maybe / No | Yes / Maybe / No |
| Startup costs in case of new programme | Yes / Maybe / No | Yes / Maybe / No |
| Types of treatment / early intervention | Yes / Maybe / No | Yes / Maybe / No |
| Success rate of treatment / early intervention | Yes / Maybe / No | Yes / Maybe / No |

Do you miss other important parameters in the current Webtool? If yes, please specify.

_____
_____
_____

1. Do you think this Webtool adequately captures most of the issues that you experienced for vision/hearing screening in your country?

   _____
   _____
   _____

2. What features of the Webtool do you think work well, and why?

   _____
   _____
   _____

3. What features of the Webtool do you think need changing? Do you have suggestions for improvement?

   _____
   _____
   _____

4. Do you think the indicators of 'acceptability, appropriateness and sustainability' adequately capture the priorities in public health care spending?

   _____
   _____
   _____

5. The simulation results shows an overview of your simulations and the output. Do you miss any information in this overview and are there other output parameters you would like to evaluate?

   _____
   _____
   _____

6. Do you wish to make any further comments or suggestions?

   _____
   _____
   _____