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Risk factors for non-participation in the Danish universal newborn hearing screening program: a population-based cohort study

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Abstract

Objective: To explore risk factors for non-participation for the Danish universal newborn hearing screening program, including socio-economic, demographic and peripartum conditions. Furthermore, the participation rate between children with medical risk factors for congenital hearing loss and healthy newborns was analyzed.

Methods: The study was register-based and included all live births in Denmark between January 1st, 2008 and December 31st, 2011, in total 251.081 children. Potential risk factors were extracted from eight Danish national registers and analyzed via logistic regression models.

Results: With respect to the participation rate, the strongest predictors of non-participation were increased maternal parity (from OR: 0.85; 95% CI: 0.82-0.89; p<0.0001 to OR: 0.43; 95% CI: 0.38-0.47; P<0.0001), low socio-economic status, including income, (from OR: 1.16; 95% CI: 1.09-1.23; p<0.0001 to OR: 1.46; 95% CI: 1.37-1.56; p<0.0001) and home birth (OR: 0.58; 95% CI: 0.42-0.80; p=0.001). Children with a medical risk factor for congenital hearing loss had a 1.97% lower participation rate. Assisted ventilation and admission to a newborn intensive care unit for > 48 hours were identified as independent risk factors of non-participation for this group (OR: 0.65; 95% CI: 0.52-0.80; p<0.0001 and OR: 0.92; 95% CI: 0.85-0.99; p=0.036, respectively).

Conclusion: In order to improve the participation rate, a national screening database in conjunction with a stronger collaboration between screening units and other health care professionals who are in contact with the family during the newborn period is warranted.

Keywords: Universal newborn hearing screening, Patient participation, Socio-economic status, Demographic factors, Hearing loss, Denmark

1. Introduction

It is well established that an untreated pediatric hearing loss can have devastating effects on language and cognitive development as well as social-emotional skills and academic achievements [1,2,3]. This can not only lead to substantial distress for the child and its family but has also great cost to the society at large. The incidence of congenital hearing loss has been reported as 2-3/1000 in all newborns [4] and 5/100 in children with a risk factor for congenital hearing loss [5]. With the implementation of universal newborn hearing screening (UNHS) programs, newborns with hearing loss can be identified and treated within the first months of life and thereby overcome many of the associated delays and challenges [6,7,8,9]. This is the reason UNHS programs have become standard of care across the world. Many UNHS programs for healthy newborns are setup as a two- or three-stage protocol, where a failed initial screening leads to at least one re-screening before a child is referred to a diagnostic evaluation. However, individual differences are seen across programs, regions and countries [10,11,12]. One difference is the timing of the initial screening. The Joint Committee on Infant Hearing (JCIH) recommends the initial screening to be carried out before discharge from the birth hospital [13] making it an inpatient procedure. This approach has the advantage of a high initial participation rate as reported to be on average > 95% across UNHS programs as evidenced in systematic reviews [11,12]. The disadvantage, however, is that the inpatient protocol often takes place within the first two days of life which increases the risk of false-negative test results [14,15,16,17] This is due to the fact, that many screening programs have adopted OtoAcustic Emissions (OAE) as the measurement of choice for the initial screening [12,14]. OAE measurements are sensitive to obstructions in the ear canal and middle ear, due to birth debris, amniotic fluid and mucus which is most prominent in the first few hours to days of life [17,18]. When the initial screening is not passed a re-screening is performed. As it often will not be possible to complete the re-screening before hospital discharge, this part of the program becomes an

outpatient procedure and studies have shown a shortcoming in compliance. Loss to follow-up is reported to be on average 20-21% [19], although large individual differences can be seen across the different UNHS programs ranging from 0.31-81.25% [11,19,20]. In the effort to avoid a high number of false-positive test results, some screening programs, including Denmark, do not undertake the initial screening until the obstructions in the ear canal and middle ear can be expected to be resolved (e.g. >48 hours of life) [10,21]. However, as most healthy newborns in Denmark are discharged from the birth hospital within 24 hours of birth [22], the initial screening is an outpatient procedure, and thus subject to similar difficulties in compliance as outpatient re-screenings. This is a critical problem as the success of all UNHS programs start with and rely heavily on a high participating rate.

1.1 Settings

Denmark is a small northern European country with approximately 60,000 births per year [23]. The UNHS program has been implemented nationally since 2005. The guidelines for the screening program were developed by the Danish Health Authority and recommend a two-stage screening protocol for healthy newborns. The first stage takes place in screening units primarily based at hospitals with maternity wards after the child's 2nd day of life. The screening professionals are usually nurses, midwives or medical laboratory scientists [21]. If the initial screening is not passed, a re-screening is carried out, typically within a week. If both the initial screening and the re-screening are not passed, the child is referred to a second-stage screening at a local audiology department. If both the first-stage and second-stage screenings are not passed, the child is referred to a full audiological evaluation at an audiology department with a pediatric specialization. For children with congenital risk factors for hearing loss the UNHS program is based on a single-stage protocol as these children will be referred directly to a full audiological evaluation after the first

failed screening. As most families with healthy newborns are discharged within 24 hours of birth, the UNHS in Denmark is primarily an outpatient program.

It is reasonable to expect a very high participation rate for the Danish UNHS program, as health care in the country is free of charge for all citizens [24], and a blood spot screening on the 3rd day of life for metabolic illness has a coverage of nearly 100% [25]. However, the Danish Health Authority reported in their latest evaluation from 2010 a national participation rate for UNHS of only 85% [26], and therefore falls short of the recommended benchmark of a participation rate of at least 95% as stated by the JCIH. In order to improve the Danish participation rate, children at risk for not being screened must be identified. Then, the program can be adapted and its resources allocated in such a way that participation rate for this group of children is enhanced.

1.2 Objectives

The objective of this study was to identify risk-factors for non-participation in the initial screening of the Danish UNHS program.

2. Material and Methods

2.1 Study Design

The present study is a retrospective cohort study. Extractions from 8 Danish national registers, were combined and linked by a unique personal identification number (assigned to all residents in Denmark) to investigate potential risk factors stemming from the following four groups:

1. Socio-economic differences that occur between families whose children have been screened and those whose children have not been screened; included were parental education level, parental affiliation to labor market and household income.

2. Demographic differences that occur between families whose children have been screened and those whose children have not been screened; included were infant gender, ethnicity, parental age, parental cohabitation and maternal parity.

3. Miscellaneous peripartum conditions (defined as events occurring in relation to birth and up to a month after delivery) that might have an impact on non-participation rate; included conditions were type of birth, place of birth, multiple births, number of days admitted to the hospital in relation to birth, readmission of the child in the first month of life and change of residence within the first month of life.

4. Medical risk factors for congenital hearing loss; included were birth weight < 1500g, assisted ventilation, admission to a newborn intensive care unit (NICU) for > 48 hours, Apgar score < 7 after 5 minutes and cranial malformations. The choice of these risk factors was based on the risk factors defined by the JCIH [27,28] and the information available for extraction from the Danish Medical Birth Register.

The study was approved by the Danish Data Protection Agency (J. no. 2012-41-0928). As the study is register-based, in accordance with Danish legislation, no approval from the Biomedical Research Ethics Committee was required.

2.2 Data Sources

The Danish Medical Birth Registry

This registry contains information of all births in Denmark by mothers with a Danish identification number. It provides data on health of the mother before and during pregnancy, complications and interventions during pregnancy and delivery, health of the newborn together with basic demographic information about the parents [29]. This was the source from which the study population was identified and all information regarding the birth, health of the child and parity were extracted. These data are considered to be of high validity [29,30].

The Danish Civil Registration System

Danish residents and all live-born babies are registered with a unique personal identification number for administrative purpose. The validity of the register is considered to be very high as the information is continuously corrected when errors are encountered [31]. From this source, information regarding death, cohabitation and residency were extracted.

The Danish National Patient Registry

This register is considered to be comprehensive, accurate and contains information regarding all diagnoses, procedures and interventions for all somatic and psychiatric in- and outpatients that come in contact with the Danish hospitals [32]. As Denmark does not have a UNHS database, children screened were identified through this register together with the number of readmissions to the hospital. The register has not been validated in relation to the UNHS.

Statistics Denmark

Statistics Denmark is a state-owned institution tasked with collecting, compiling and publishing

statistics about the Danish citizens as well as supplying data for researchers. From here socio-economic information and ethnicity were extracted from 5 sub-registers; BEF (Danish title: Befolkningen; English translation: Population), FAM (Danish title: Familieforhold; English translation: Family status), FAIK (Danish title: Familieindkomster; English translation: Family income), INDH (Danish title: Indkomst; English translation: Income), and UDDA (Danish title: Uddannelser; English transation: Education). These data are generally considered to be of high quality [33,34,35].

For a detailed description and definition of the studied variables see Supplementary Table 1.

2.3 Study population

We performed a population-based cohort study including all live births between January 1st 2008 and December 31st 2011. All children who died within the first month of life were excluded from the study (n=681) due to the assumption that these children's health problems made them ineligible for hearing screening. Thus, the final study population was 251,081 children.

2.4 Statistics

Spearman's rank correlation coefficients were calculated for all pairwise comparisons of variables in order to assess bivariate interrelationships. Parental ethnicity was excluded from further analysis because of a high correlation with the ethnicity of the infant. The same was the case for paternal age and education level as these were strongly correlated with the corresponding information from the mother. In addition, the number of days the mother was admitted to the hospital in relation to birth was excluded because of a strong interrelationship with the comparable information for the child. In a similar vein, the number of children living at home was also omitted due to a strong correlation

with maternal parity. The decision to prioritize the mother over the father is based on a lack of registration of paternity for 4,280 of the children, whereas all mothers where identifiable.

In order to identify risk factors for non-participation, crude and adjusted odds ratios (ORs) with 95% confidence intervals (CIs) were estimated by logistic regression models with screened/not screened as the outcome; $p < 0.05$ was considered statistically significant. All variables deemed relevant and not being strongly correlated in bivariate analyses entered the multivariate logistic regression model. All statistical analyses were performed using Stata/SE 12.1 (StataCorp, Tx, USA).

3. Results

Of the 251,081 children eligible for UNHS, 16,716 did not attend, resulting in a participation rate of 93%. Characteristics of both groups are presented in Tables 1-3.

3.1 Socio-economic risk factors

As presented in Table 1, all the classic measures of low social economic status (SES; brief education, unemployment and low income) were related to non-participation. The largest disparity was found between the groups of employed and unemployed mothers (94.57% vs. 89.10%, respectively) followed by the groups with the highest (largest quintile) and lowest (lowest quintile) household income (95.58% vs. 90.07%, respectively). When controlling for confounders in the multivariate analysis (see Table 4), the socioeconomic variables remained significant risk factors for non-participation. For example the chance of being screened decreased by 22% if the mother was unemployed as compared to if the mother was gainfully employed (OR: 0.78; 95% CI: 0.73-0.82; $p<0.0001$), and the chance of being screened rose with increasing income level with

estimated ORs between 1.16 and 1.46. Notably, as compared to a child from the lowest income group, the chance of screening climbs by 34% in income group 3 (third quintile; OR: 1.34; 95% CI: 1.25-1.42; $p<0.0001$), and to 45% in income group 5 (fifth quintile; OR: 1.45; 95% CI: 1.35-1.55; $p<0.0001$). When comparing mothers whose highest attained education level is basic- or high school in the multivariate analyses, the chance of participation increases with 28% if the mother has completed vocational training (OR: 1.28; 95% CI: 1.22-1.34; $p<0.0001$) and 33% if she has completed higher education (OR: 1.33; 95% CI: 1.26-1.40; $p<0.0001$).

3.2 Demographic risk factors

The multiple logistic regression model revealed no differences in relation to ethnicity, as the chance of being screened was statistically insignificant less in non-Danish than in Danish children (OR: 0.96; 95% CI: 0.91-1.02; $p=0.20$ Table 4). The risk of non-participation in screening was, however, 16% higher when the parents were not living together at the time of birth compared to when they were (OR: 0.84; 95% CI: 0.80 to 0.89; $p<0.0001$).

A strong inverse relationship between maternal parity and participation emerged from the analysis. The risk of non-participation already increased with 15% for the second child (OR: 0.85; 95% CI: 0.82-0.89, $p<0.0001$), 33% for the third child (OR: 0.67; 95% CI: 0.63-0.70, $p<0.0001$) and up to 57% if the mother has given birth more than 4 times (OR: 0.43; 95% CI: 0.38-0.47, $p<0.0001$).

3.3 Miscellaneous peripartum factors

Using the peripartum parameters present in the course of what was considered a “normal” birth as a baseline, we subsequently analyzed the contrasting factors. The strongest predictor of participation was place of birth. The chance of being screened was 42% lower for children born at home versus children born at the hospital (OR: 0.58; 95% CI: 0.42-0.80; $p=0.001$; Table 4). When looking at the

descriptive analysis of the other peripartum factors in Table 2, there did not appear to be an association between number of readmissions to the hospital or type of birth and participation rate. The multivariate analysis in Table 4, however, revealed a significant increase in the likelihood for non-participation with respect to both variables. A decrease of 16% was found for each readmission (OR: 0.84; 95% CI: 0.82-0.86; $p<0.0001$), and, similarly, a decline in participation was seen for planned and emergency caesarian sections with 13% (OR: 0.87; 95% CI: 0.82-0.92, $p<0.0001$) and 10% (OR: 0.90; 95% CI: 0.85-0.95, $p<0.0001$), respectively.

A statistically significant, but clinically negligible difference was observed in the length of admission in relation to birth (OR: 0.996; 95% CI: 0.994-0.999; $p=0.001$), translating into an average increase of the risk of non-participation of 0.004% for every additional day spent in hospital. In contrast to moving out of Denmark within the first month of life, changing residence within Denmark did show a significant decrease in participation (OR: 0.82; 95% CI: 0.74-0.91; $p<0.0001$).

The only peripartum factor that increased the participation rate was multiparous birth (OR: 1.10; 95% CI: 1.01-1.20; $p=0.028$). However, the disparity in participation percent when comparing to singular births is clinically negligible (93.34% vs. 93.44%, respectively; Table 2).

3.4 Medical risk factors for hearing loss

Table 5 summarizes the participation rate for children with identified medical risk factors compared to children with no risk factors. The participation rate between the two groups is in favor of the group with no risk factors with a difference of 1.97%. When looking at each medical risk factor independently, only admission to a NICU > 48 hours (OR: 0.92; CI 95 %: 0.85-0.99; $p=0.036$) and assisted ventilation (OR: 0.65; 95% CI: 0.52-0.80; $p<0.0001$) remained significant after adjusting for the other variables (Table 4). In contrast, the effect of low birth weight and low Apgar score

seen in univariate analyses were not statistically significant in the multivariate analysis. Cranial and other malformations did not in any of the analyses correlate to a higher or lower participation rate. Supplementary Table 2 shows the distribution of ICD-10 diagnoses for the 2,878 children with cranial malformations related to hearing loss.

This study followed the JCIH's definition of low birth weight as <1500g [27].

Due to the fact, that some former studies have used <2500g as the definition for low birth weight in relation to participation in UNHS [36,37], we also conducted an additional multivariate analysis that examined the effect of low birth weight where the cutoff was changed from <1500g to <2500g. Birth weight was the only variable changed in the model from Table 4. This analysis showed a significant decrease of 11% in the likelihood of being screened for the low birth weight group (OR: 0.89; 95% CI: 0.81-0.97; p=0.011). In addition, the effect of NICU stay became statistically none significant (OR: 0.95, 95% CI: 0.88-1.04; p=0.27; results not shown elsewhere).

4. Discussion

This large population-based study found a wide range of significant variables for non-participation of newborn hearing screening among which maternal parity, SES, birth place and assisted ventilation were.

4.1 Socio-economic risk factors

Low SES emerged from the analyses as an especially strong predictor of non-participation, as parental unemployment, low maternal education and low family income all significantly increased the risk of non-participation. This discovery is also supported by previous findings for UNHS [20,38,39,40,41] and other health related screening programs [42,43]. As health care is free in Denmark, it is reasonable to assume that non-participation for families with a lower household

income is due to other circumstances than the family's financial situation alone. One explanation is more likely related to a more complex relationship between low SES and health behavior. The influence of low SES on poor health care choices is not fully understood yet. Low SES has been linked to inadequate health literacy, here defined as: "the cognitive and social skills which determine the motivation and ability of individuals to gain access to, understand and use information in ways which promote and maintain good health" [44]. Low SES is further associated with a decreased sense of self-efficacy and poorer problem-solving skills [45], which may contribute to the difficulty for these individuals in overcoming potential barriers in UNHS participation.

4.2 Demographic risk factors

Lack of parental cohabitation and increasing maternal parity were the only two demographic variables identified as significantly increasing the risk of non-participation.

The decision to look at cohabitation instead of marital status was based on two substantial factors. First, it is not uncommon for couples in Denmark to have children out of wedlock or to live together without ever marrying [46]. Second, it is arguably much easier to coordinate the screening appointment with the resources of two partners in a household compared to that of a single parent. An analysis of marital status does not necessarily provide information on cohabitation. Our study found a significantly increased risk of non-participation if the parents did not cohabitate. To our knowledge, only one other study looked at marital status in relation to the initial hearing screening [38]. This UNHS program is based in Brazil and also organized as an outpatient procedure, but categorized non-married couples living together in the same group as married couples. Their results showed no significant effect of marital/cohabitation status on the participation rate. It is unknown if the discrepancies in the results are caused by categorization or merely cultural differences between

Denmark and Brazil. Ethnicity did not emerge from the regression analysis as a significant predictor of non-participation, in accordance with previous findings from Colorado [37]. Lower participation rates in other preventive screening programs have however been found with respect to immigrants in Denmark [47]. When looking at Table 2, a decrease in participation of 5.65% exists for children of non-Danish origin as compared to those of Danish origin. Even though the lower participation rate is explained by other factors, as seen in the regression model, the discrepancy is of high clinical relevance as this group can more easily be identified when compared to some of the other independent variables. The discrepancy for this group may reflect greater difficulties in navigating the health care system combined with limited access to informational material in the relevant languages.

Decreased participation was further linked to maternal parity with the risk on non-participation rising with an increase in parity. It can be argued that the increased risk is attributed to several factors. First of all, it can be logistically more difficult to participate in a hearing screening, schedule the appointment, organize a trip to the hospital etc., if siblings are involved. Also, the perceived risk of hearing loss is most likely negatively affected by having other children without hearing loss. Finally, if the family feels that the child reacts to environmental sounds, the screening can seem an unnecessary extra test. Therefore, one of the most important aspects in the screening program is to inform the parents of the risk even when hearing loss is not readily apparent in the child or present in other family members. While no other studies, to our knowledge have included this variable in relation to the first stage of UNHS, parity has been shown to negatively affect compliance in both the second-stage screening and in the diagnostic follow up of UNHS [20,48]. The current study further supports previous findings that mother's age [37,38] and infant gender [40,49] are not statistically significant risk factors for non-participation.

4.3 Miscellaneous peripartum factors

Changing residence within 1 month of birth, homebirth, cesarean, single birth, longer duration of infant hospital admission in relation to birth, and higher number of readmissions for the infant in the first month of life did all significantly increase the risk of non-participation.

One of the largest discrepancies, with respect to participation, can be seen in relation to place of birth. Children born at the hospital are 42% more likely to be screened than children born at home. While home births accounted for only approximately 1% of all births, a large 27% of these children were not screened. Due to the fact that home births are getting more popular in Denmark [50], this is an important clinical group to focus on. No relevant literature was found to entail explanations for the lack of participation for this group. However, the main difference between the home and hospital group in Denmark is that when giving birth at the hospital the family already expects to go back to the hospital for both the blood-spot screening and health check-up. This is not the case for home births as a midwife comes to the family's home instead to carry out these procedures. In these instances, the UNHS is the only procedure for which the family has to go to the hospital, and this is a likely reason for screening deselection. Another feasible explanation might be that parents who decide to give birth at home may have personal beliefs which are incongruous with standard medical procedures, such as vaccinations and screening programs. Regardless of the reason, midwives play an important role in ensuring that the parents receive and understand the information regarding UNHS, thus allowing for an informed decision.

Our analysis also showed that there was an increased risk of non-participation that correlated to change of residence shortly after birth. This finding is not surprising, considering that moving is time consuming and that the burden of arranging a screening appointment often falls on the parents. As families with a newborn get regular visits from a specialized nurse (health visitor) during the child's first year of life, engaging these health professionals to follow-up on these children's

screening status would be extremely beneficial. The same applies to families where the child has been readmitted to the hospital during the first month of life. While these families might come in contact with a number of different health-care professionals, they are often focused on other aspects of the child's health. As these admissions can interfere with the screening appointment, the health visitor would be an ideal person to systematically follow-up on these children's hearing screening participation.

It is of interest to note that both type of birth and multiparous birth changed from insignificant in the univariate regression model to significant in the adjusted model. When looking at Table 2, one could postulate that this change can be attributed to the unbalanced sample sizes in the two groups [51].

4.4 Medical risk factors

Of the investigated medical risk factors, birth weight, assisted ventilation and admission to NICU>48 hours were identified as significantly increasing the risk of non-participation.

Even though the participation rate for children with medical risk factors for congenital hearing loss is only slightly lower than for that of healthy newborns, it is an alarming finding. One would generally expect a better participation rate for children with medical risk factors because of the increased incidence of hearing loss in this group. Unfortunately, our study supports previous findings [37,40] that this is not necessarily the case. One could speculate that the reason for this is that it has been difficult to maintain focus on this group after the at-risk screening was changed to the current UNHS program. Also, a possible hearing loss might be neglected when a child is in a critical state, for example newborns admitted to a NICU for a longer period of time or receiving assisted ventilation. When the child's condition is stable, parents might be reluctant to participate on on the grounds that they want to spare the child another test. Furthermore, if there does not exist a

strong collaboration between the screening professionals and the staff at the NICUs, the parents might not have been exposed to the necessary information and subsequently not receive the screening referral. In either case, it is especially important for UNHS programs to be able to track at-risk children at every level in an early and systematic way, to ensure that they all are offered a screening. Even though low Apgar score and low birth weight < 1500g were not statistically significant risk factors in the multivariate analysis, in Table 3 they still pointed to groups which have a lower screening percentage, and therefore may call for increased attention in the clinical setting. This finding is further supported by the fact that when low birth weight in the regression model was changed to <2500g, this variable became significant with relation to a lower participation rate.

4.5 Strengths and limitations

The main strength of this study is the population-based design that makes the study less vulnerable to selection bias. As with all register-based information, we cannot rule out misclassification, which together with missing data could have affected the results. The quality of data used for the analysis is considered reliable, however, uncertainties do exist specifically for screening registrations and admission dates as no validation studies exist. As the study does not include contact with any of the non-participating families, and thus no elaboration exists for the reason behind missed screening, the results are limited to only identifying children at risk of being lost to the program. Therefore, no individual explanations are provided as to why these individuals were not screened. However, the size of the study population did allow for the inclusion of a wide range of different variables for a comprehensive description of the non-participating group of children. This in turn contributed to a greater understanding of whom to target in the improvement of the UNHS program along with an outline of which groups require greater focus in future research.

4.6 Meaning of the study

The current results point to the need for better involvement of the midwives, health visitors and the healthcare professionals in the NICUs. As these individuals often are in direct contact with the children that this study has identified as at risk for non-participation, they are in a position to effectuate an increase in the participation rate. The Danish UNHS program would also benefit from a database that allows for both national and local monitoring of the UNHS program, as seen in other countries [36,40]. Such a database could facilitate tracking the program's development and provide valuable information regarding the effects that changes in the program might have on participation rate. Specifically, a comprehensive database could also identify children that, for unknown reasons, have been lost to the UNHS program. This would particularly relevant for the children at risk of congenital hearing loss, as this group was found to have a lower participation rate.

4.7 Unanswered questions and future research

This study has identified groups of children at risk of non-participation. We, however, do not know why these children were not screened, and can only speculate as to the cause beyond possible factors investigated here. Future research should focus on identifying and understanding the reasons behind non-participation, for instance, by means of in-depth interviews.

5. Conclusion

The Danish UNHS program has not yet reached the international standard participation rate of 95%. Even though this study found an increased participation rate, as compared to previous findings from the Danish Health Authority, it is not satisfactory that the hearing status of 7% of Danish newborns

is unknown. In order to ensure that no children with congenital hearing loss are lost to the program, it is important that healthcare professionals are aware of which groups of children are at a higher risk of not being screened. As we have shown, however, many different variables influence the risk of non-participation, an optimization of the program cannot be expected from one single initiative. It will instead require continuous monitoring of the hearing screening program, a strong interdisciplinary collaboration between relevant health care professionals and further insight into the reasons behind non-participation in a Danish context.

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Table 1: Socio-economic and demographic characteristics of the study population

	Screened (n = 234,365)		Unscreened (n = 16,716)		Total (N = 251,081)	
	n	**(%)**	**n**	**(%)**	**n**	**(%)**
Sex of infant						
Male	119,980	(93.24)	8,694	(6.76)	128,674	(51.25)
Female	114,385	(93.45)	8,022	(6.55)	122,407	(48.75)
Ethnicity of infant						
Danish	212,300	(94.02)	13,492	(5.98)	225,792	(89.93)
Non-Danish	21,031	(88.37)	2,767	(11.63)	23,798	(9.48)
Unknown	1,034	(69.35)	457	(30.65)	1,491	(0.59)
Ethnicity of mother						
Danish	198,863	(94.15)	12,367	(5.85)	211,230	(84.13)
Non-Danish/Western country	9,879	(92.50)	801	(7.50)	10,680	(4.25)
Non-Danish/ Non-western country	24,536	(90.02)	2,721	(9.98)	27,257	(10.86)
Unknown	1,087	(56.79)	827	(43.21)	1,914	(0.76)
Ethnicity of father						
Danish	197,652	(94.13)	12,320	(5.87)	209,972	(83.63)
Non-Danish/Western country	8,502	(92.23)	716	(7.77)	9,218	(3.67)
Non-Danish/ Non-western country	21,859	(88.57)	2,821	(11.43)	24,680	(9.83)
Unknown	6,352	(88.09)	859	(11.91)	7,211	(2.87)
Parental cohabitation						
No	19,938	(86.39)	3,142	(13.61)	23,080	(9.19)
Yes	214,427	(94.05)	13,574	(5.95)	228,001	(90.81)

Parity						
1	104,503	(94.65)	5,904	(5.35)	110,407	(43.97)
2	85,413	(94,07)	5,386	(5.93)	90,799	(36.16)
3	30,732	(91.88)	2,715	(8.12)	33,447	(13.32)
4	7,107	(88.67)	908	(11.33)	8,015	(3.19)
>4	2,965	(83.12)	602	(16.88)	3,567	(1.42)
Unknown	3,645	(75.22)	1,201	(24.78)	4,846	(1.93)
Work affiliation of mother						
Employed	189,803	(94.57)	10,901	(5.43)	200,704	(79.94)
Non-employed	20,755	(89.10)	2,538	(10.90)	23,293	(9.28)
Student	11,263	(92.91)	859	(7.09)	12,122	(4.83)
Child (under 15 years old)	20	(100.00)	0	(0.00)	20	(0.01)
Unknown	12,524	(83.82)	2,418	(16.18)	14,942	(5.95)
Work affiliation of father						
Employed	205,151	(93.95)	13,210	(6.05)	218,361	(86.97)
Non-employed	12,287	(88.86)	1,541	(11.14)	13,828	(5.51)
Student	4,865	(93.18)	356	(6.82)	5,221	(2.08)
Child (under 15 years old)	9	(90.00)	1	(10.00)	10	(0.00)
Unknown	12,053	(88.23)	1,608	(11.77)	13,661	(5.44)
Education of mother*[1]						
7-12 years	52,749	(91.14)	5,128	(8.86)	57,877	(23.05)
10-12 years	62,598	(94.34)	3,754	(5.66)	66,352	(26.43)
≥ 13 years	108,309	(94.95)	5,755	(5.05)	114,064	(45.43)
Unknown	10,709	(83.74)	2,079	(16.26)	12,788	(5.09)
Education of father*[1]						
7-12 years	52,844	(91.44)	4,945	(8.56)	57,789	(23.02)
10-12 years	85,912	(94.08)	5,404	(5.92)	91,316	(36.37)

≥ 13 yeras	81,599	(94.65)	4,611	(5.35)	86,210	(34.34)
Unknown	14,010	(88.86)	1,756	(11.14)	15,766	(6.28)
Family income*[2]						
Group 1	39,286	(90.07)	4,329	(9.93)	43,615	(17.37)
Group 2	40,652	(93.20)	2,964	(6.80)	43,616	(17.37)
Group 3	41,336	(94.77)	2,281	(5.23)	43,617	(17.37)
Group 4	41,661	(95.52)	1,956	(4.48)	43,617	(17.37)
Group 5	41,691	(95.58)	1,926	(4.42)	43,617	(17.37)
Unknown	29,739	(90.12)	3,260	(9.88)	32,999	(13.14)

	Mean (SD)	**Mean (SD)**	**Mean (SD)**
Age of mother	30.9 (4.94)	30.7 (5.50)	30.9 (4.98)
Age of father	33.5 (5.81)	33.9 (6.68)	33.5 (5.87)

*[1] **7-10 years**: Basic school/high school; **10-12 years**: Vocational training; **≥13 years**: higher education.

*[2] Family income was stratified into five groups each covering 20 percentiles. **Group 1** covered the families with the 20 % lowest income and G**roup 5** the families with the 20 % highest income. For a detailed description and definition of the studied variables see Supplementary Table 1.

Table 2: Miscellaneous peripartum factors in the study population

	Screened (n = 234,365)		Unscreened (n = 16,716)		Total (N = 251,081)	
	n	**(%)**	**n**	**(%)**	**n**	**(%)**
Changed residence within 1 month of birth						
No	230,060	(93.40)	16,252	(6.60)	246,312	(98.10)
Yes	4,305	(90.27)	464	(9.73)	4,769	(1.90)
Moved out of Denmark within 1 month of birth						
No	234,296	(93.34)	16,705	(6.66)	251,001	(99.97)
Yes	69	(86.25)	11	(13.75)	80	(0.03)
Place of birth						
Hospital	232,322	(93.89)	15,120	(6.11)	247,442	(98.55)
Home	1,800	(72.55)	681	(27.45)	2,481	(0.99)
Unknown	243	(20.98)	915	(79.02)	1,158	(0.46)
Type of birth						
Vaginal	182,731	(93.40)	12,911	(6.60)	195,642	(77.92)
Planned cesarean	21,806	(93.09)	1,619	(6.91)	23,425	(9.33)
Acute cesarean	29,828	(93.17)	2,186	(6.83)	32,014	(12.75)
Multiparous birth						
No	224,678	(93.34)	16,036	(6.66)	240,714	(95.87)
Yes	9,687	(93.44)	680	(6.56)	10,367	(4.13)
	Median (range)		**Median (range)**		**Median (range)**	
Number of days infant admitted at the hospital in relation to birth	3 (7.74)		3 (12.87)		3 (8.15)	
Number of readmissions for the infant the first month of life	1 (0.60)		1 (0.72)		1 (0.61)	

Table 3: Medical risk factors for congenital hearing loss in the study population

	Screened (n = 234,365)		Unscreened (n = 16,716)		Total (N = 251,081)	
	n	**(%)**	**n**	**(%)**	**n**	**(%)**
Birth weight						
< 1500g	1,305	(88.96)	162	(11.04)	11,230	(0.58)
≥ 1500g	229,728	(94.07)	14,700	(6.01)	244,428	(97.35)
Unknown	3,332	(64.25)	1,854	(35.75)	5,186	(2.07)
Assisted ventilation						
No	233,616	(93.38)	16,567	(6.62)	250,183	(99.64)
Yes	749	(83.41)	149	(16.59)	898	(0.36)
Admitted to NICU >48hours						
No	220,190	(93.48)	15,359	(6.52)	235,549	(93.81)
Yes	14,175	(91.26)	1,357	(8.74)	15,532	(6.19)
APGAR score						
< 7	1,538	(90.21)	167	(9.79)	1,705	(0.68)
≥ 7	231,232	(93.94)	14,905	(6.06)	246,137	(98.03)
Unknown	1,595	(49.24)	1,644	(50.76)	3,239	(1.29)
Congenital malformations						
Non	214,629	(93.35)	15,292	(6.65)	229,921	(91.57)
Cranial malformations	2,686	(93.33)	192	(6.67)	2,878	(1.15)
Other malformations	17,050	(93.26)	1,232	(6.74)	18,282	(7.28)

Table 4: Logistic regression model to predict non-participation in the Danish UNHS program

	OR (95% CI) Crude (N = 251.081)	*P*	OR (95% CI) Adjusted (N = 247.014)	*P*
Sex of infant				
Male	1		1	
Female	1.03 (1.00 to 1.07)	**0.041**	1.02(0.98 to 1.05)	0.33
Ethnicity of infant				
Danish	1		1	
Non-Danish	0.48 (0.46 to 0.50)	< **0.0001**	0.96 (0.91 to 1.02)	0.20
Unknown	0.14 (0.13 to 0.16)	< **0.0001**	0.58 (0.48 to 70)	<**0.0001**
Parental cohabitation				
Yes	1		1	
No	0.40 (0.39 to 0.42)	< **0.0001**	0.84 (0.80 to 0.89)	< **0.0001**
Parity				
1	1		1	
2	0.90 (0.86 to 0.93)	< **0.0001**	0.85 (0.82 to 0.89)	< **0.0001**
3	0.64 (0.61 to 0.67)	< **0.0001**	0.67 (0.63 to 0.70)	< **0.0001**
4	0.44 (0.41 to 0.48)	< **0.0001**	0.56 (0.52 to 0.61)	< **0.0001**
>4	0.28 (0.25 to 0.30)	< **0.0001**	0.43 (0.38 to 0.47)	< **0.0001**
Unknown	0.17 (0.16 to 0.18)	< **0.0001**	1.02 (0.88 to 1.17)	0.82
Work affiliation of mother				
Employed	1		1	
Unemployed	0.47 (0.45 to 0.49)	< **0.0001**	0.78 (0.73 to 0.82)	< **0.0001**
Student	0.75 (0.70 to 0.81)	< **0.0001**	0.93 (0.86 to 1.01)	0.09
Child (under 15 years old)	empty		empty	
Unknown	0.30 (0.28 to 0.31)	< **0.0001**	0.76 (0.70 to 0.81)	< **0.0001**
Work affiliation of father				
Employed	1		1	
Unemployed	0.51 (0.49 to 0.54)	< **0.0001**	0.89 (0.84 to 0.95)	**0.001**
Student	0.88 (0.79 to 0.98)	**0.022**	1.08 (0.96 to 1.22)	0.20
Child (under 15 years old)	0.58 (0.73 to 4.57)	0.61	0.60 (0.07 to 5.02)	0.64
Unknown	0.48 (0.46 to 0.51)	< **0.0001**	0.83 (0.78 to 0.89)	< **0.0001**
Education of mother				
7-12	1		1	
10-12	1.62 (1.55 to 1.69)	< **0.0001**	1.28 (1.22 to 1.34)	< **0.0001**
≥ 13	1.83 (1.76 to 1.90)	< **0.0001**	1.33 (1.26 to 1.40)	< **0.0001**
Unknown	0.50 (0.47 to 0.53)	< **0.0001**	1.05 (0.97 to 1.15)	0.23
Family income				
Group 1	1		1	
Group 2	1.51 (1.44 to 1.59)	< **0.0001**	1.16 (1.09 to 1.23)	< **0.0001**
Group 3	2.00 (1.89 to 2.10)	< **0.0001**	1.34 (1.25 to 1.42)	< **0.0001**
Group 4	2.35 (2.22 to 2.48)	< **0.0001**	1.46 (1.37 to 1.56)	< **0.0001**
Group 5	2.39 (2.26 to 2.52)	< **0.0001**	1.45 (1.35 to 1.55)	< **0.0001**
Unknown	1.01 (0.96 to 1.05)	0.83	0.99 (0.94 to 1.05)	0.84
Changed residence within 1 month of birth				

No	1		1	
Yes	0.66 (0.59 to 0.72)	< **0.0001**	0.82 (0.74 to 0.91)	< **0.0001**
Moved out of Denmark within 1 month of birth				
No	1		1	
Yes	0.45 (0.24 to 0.85)	**0.013**	1.69 (0.79 to 3.60)	0.18
Place of birth				
Hospital	1		1	
Home	0.17 (0.16 to 0.19)	< **0.0001**	0.58 (0.42 to 0.80)	**0.001**
Unknown	0.02 (0.01 to 0.02)	< **0.0001**	empty	
Type of birth				
Vaginal	1		1	
Planned cesarean	0.95 (0.90 to 1.00)	0.07	0.87 (0.82 to 0.92)	< **0.0001**
Acute cesarean	0.960.92 to 1.01)	0.13	0.90 (0.85 to 0.95)	< **0.0001**
Multiparous birth				
No	1		1	
Yes	1.02 (0.94 to 1.10)	0.68	1.10 (1.01 to 1.20)	**0.028**
Age of mother	1.01(1.003 to 1.009)	< **0.0001**	0.997 (0.99 to 1.00)	0.10
Age of father*	0.98 (0.985 to 0.99)	< **0.0001**		
Number of days infant admitted at the hospital in relation to birth**	0.99 (0.988 to 0.991)	< **0.0001**	0.996 (0.994 to 0.999)	**0.001**
Number of readmissions for the infant the first month of life***	0.78 (0.76 to 0.80)	< **0.0001**	0.84 (0.82 to 0.86)	< **0.0001**
Birth weight				
≥ 1500g	1		1	
< 1500g	0.52 (0.44 to 0.61)	<**0.0001**	0.99 (0.80 to 1.23)	0.96
Unknown	0.12 (0.11 to 0.12)	<**0.0001**	0.69 (0.61 to 0.78)	< **0.0001**
Assisted ventilation				
No	1		1	
Yes	0.36 (0.30 to 0.43)	< **0.0001**	0.65 (0.52 to 0.80)	< **0.0001**
Admitted to NICU >48hours				
No	1		1	
Yes	0.73 (0.69 to 0.77)	< **0.0001**	0.92 (0.85 to 0.99)	**0.036**
APGAR score				
≥ 7	1		1	
< 7	0.59 (0.51 to 0.70)	< **0.0001**	0.91 (0.76 to 1.08)	0.26
Unknown	0.63 (0.06 to 0.07)	< **0.0001**	0.68 (0.56 to 0.82)	< **0.0001**
Congenital malformations				
Non	1		1	
Cranial malformations	0.997 (0.86 to 1.15)	0.97	0.98 (0.84 to 1.14)	0.81
Other malformations	0.986 (0.93 to 1.05)	0.65	1.02 (0.96 to 1.09)	0.55

OR: Odds ratios; CI: Confidence Intervals
*n=246,698 **n=247,829 ***n=249,059.

Table 5: Participation rate for children with a medical risk factor for congenital hearing loss and for healthy newborns.

	Screened (%)	Unscreened (%)	Total (%)
Risk	17,323 (91.52)	1,605 (8.48)	18,928 (7.54)
No Risk	217,042 (93.49)	15,111 (6.51)	232,153 (92.46)
Total	234,365 (93.34)	16,716 (6.66)	251,081 (100)

Supplementary Table 1: Definition and categorization of variables

Variables	Definition
Ethnicity Danish Non-Danish/Western Country Non-Danish/ Non-Western country	***Danish***: Where at least one of the parents is both a Danish citizen and born in Denmark. ***Non-Danish***: None of the parents are both a Danish citizen and born in Denmark. ***Western countries***: The 28 member states of the European Union, Andorra, Australia, Canada, Iceland, Liechtenstein, Monaco, New Zealand, Norway, San Marino, Switzerland, Vatican State and the USA. ***Non-western countries***: all other countries.
Parental cohabitation	The mother and father are registered on the same address on the date of the birth.
Work affiliation Employed Non-employed Student Child	Work affiliation is based on the SOCIO_02 classification from Statistic Denmark. The parents' affiliation is based on the primary source of income. ***Employed***: In a job for more than half of the year. ***Non-employed****:* Without a job for at least half of the year. This category also counts for persons on maternity leave, sick leave and pension. ***Student****:* An individual in school who turns at least 15 years of age before the end of the year. ***Child****:* Under the age of 15 years.
Education 7-12 years 10-12 years ≥13 years	The highest attained education extracted on the 1st of October the year of the birth. ***7-10****:* Basic school/high school ***10-12****:* Vocational training ***≥13****:* higher education.
Family income Group 1 Group 2 Group 3 Group 4 Group 5	Family income after taxation and interest per person, adjusted for number of persons in the household. All negative income was set to an income of 0 DKK. Income was further stratified into five groups each covering 20 percentiles. Group 1 covered the families with the 20 % lowest income and group 5 the families with the 20 % highest income. The income was extracted for the year prior to the birth of the child, so that the decrease in income because of maternity leave would not affect

	the outcome.
Parity 1 2 3 4 >4	The number of times the mother has given birth, including the actual birth. Still born babies are also included in the count of parity.
Change of residence within 1 month of birth	1 month defined as 30 days after the birth of the child.
Multiparous birth No Yes	**No**: birth of a single child **Yes**: birth of twins, triplets etc.
Apgar score ≥7 <7	Apgar score as assessed 5 min. after birth. The categorization of above and below 7 is based on the Joint Committee on infant hearing's identification of < 7 as a risk factor for congenital hearing loss.
Congenital malformations None Cranial malformations Other malformations	In the Danish Medical Birth Registry up to 20 different diagnoses of malformation can be registered for each child. For a list of ICD-10 codes used for identifying cranial malformations see Supplementary Table 2. As the registry covers all congenital malformations, it was decided to also include malformations not related to hearing loss as an extra category.

Supplementary Table 2: Distribution of ICD-10 diagnoses for the 2,878 children with cranial malformations related to hearing loss.

ICD-10 codes	Description	ICD-10 codes not regarded as relevant for congenital hearing loss and therefore not included	Number of children with each diagnosis in this study
Q00	Anencephaly and similar Malformations		0
Q01	Frontal encephalocele		13
Q02.9	Microcephalus		73
Q16	Congenital malformations of ear causing impairment of hearing		49
Q17	Other malformations of ear	Q17.5 Prominent ear (bat ear)	125
Q18	Other congenital malformations of face and neck	Q18.6 macrocheilia (hypertrophy of lip)	181
Q35	Cleft palate		254
Q36	Cleft lip		166
Q37	Cleft palate with cleft lip		393
Q67*	Congenital musculoskeletal deformities of head, face, spine and chest	Q67.5: congenital deformity of spine Q67.6: Pectus excavatum (funnel chest) Q67.7 Pectus carinatum (pigeon chest) Q67.8: Other congenital deformities of chest	1274
Q75	Other congenital malformations of skull and face bones		1216
Q87.0	Congenital malformation syndromes predominantly affecting facial appearance		78
Total			3822**

*263 children were found to be registered with the diagnosis code Q67. As this is a general diagnosis and does not explain whether the malformations are centered to the head, face, spine or chest, all 263 observations were excluded from the analysis.

** Some children had more than one diagnosis; therefore the number of diagnoses was higher than the number of children in the group of children with risk factors.