Socio-demographic determinants of hearing impairment studied in 103 835 term babies

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Background: Serious hearing problems appear in approximately one in 1000 newborns. In 2000, the Joint Committee on Infant Hearing defined a list of risk factors for neonatal hearing impairment relating to health, physical characteristics and family history. The aim of this study is to determine which personal, environmental and social factors are associated with the prevalence of congenital hearing impairment (CHI). Methods: The entire population of 103 835 term newborns in Flanders, Belgium, was tested by a universal neonatal hearing screening (UNHS) programme using automated auditory brainstem responses (AABR). In the case of a positive result, a CHI diagnosis was verified in specialized referral centres. Socio-demographic risk factors were investigated across the entire population to study any relationship with CHI. Results: The prevalence of bilateral CHI of 35 dB nHL (normal hearing level) or more was 0.87/1000 newborns. The sensitivity and specificity of the screening test were 94.02 and 99.96%, respectively. The socio-demographic factors of gender, birth order, birth length, feeding type, level of education and origin of the mother were found to be independent predictors of CHI. Conclusions: The socio-demographic risk factors found to be associated with CHI extend the list of classic risk factors as defined by the American Academy of Pediatrics (AAP). Assessment of these additional factors may alert the treating physician to the increased risk of newborn hearing impairment and urge the need for accurate follow-up. Moreover, this extended assessment may improve decision making in medical practice and screening policy.
Introduction

Bilateral congenital hearing impairment (CHI) affects about 1.3/1000 newborns. In babies admitted to a Neonatal Intensive Care Unit (NICU), the prevalence of bilateral hearing loss rises to 1.9%. The American Academy of Pediatrics (AAP) has defined risk factors for CHI including pre- and perinatal factors, genetic factors and craniofacial abnormalities. Universal neonatal hearing screening (UNHS) is advocated by the AAP, as only 50–60% of infants with CHI have any of these risk factors. Since 1998, a UNHS programme has been successfully implemented by the Flemish governmental organization Kind & Gezin (K&G, Child & Family), in Flanders, the northern Dutch speaking part of Belgium. The Flanders region constitutes ~60% of the entire population of Belgium. The full programme comprised screening, referral, diagnosis, treatment and home-based guidance as well as registration of all findings in a central database, in accordance with the Guidelines of the Joint Committee of Infant Hearing and the European consensus statement.

This study presents the outcomes of the UNHS programme in Flanders during 2003 and 2004, and addresses the personal, environmental and social factors associated with the prevalence of CHI, providing an extension of the list of classic risk factors defined by the AAP.

Methods

The UNHS programme was free of charge to all newborns in Flanders. The methodology of the screening programme, based on an automated auditory brainstem response (AABR) device (the Algo Portable Screener by Natus Medical Inc.) has been described in detail elsewhere. Screening sound intensity was 35 dB nHL (normal hearing level) and the results were reported as either 'pass' (negative) or 'refer' (positive). 'Pass' indicated that there was a 99.98% chance of normal hearing, while 'refer' indicated that the instrument could not achieve this degree of confidence. It is based on automated comparison of the auditory brainstem responses of the tested baby with a 'built-in' normal hearing pattern response. When the machine was unable to provide a 'pass' or 'refer' reading within 20 min, the test was stopped and scored as 'aborted test'. Babies who fell into the 'refer' or 'aborted test' categories, underwent a repeat assessment within 48 h. When this second test was scored as 'refer', the baby was referred to a certified centre of expertise to ascertain CHI. These centres follow a strict protocol implying diagnostic examinations, multidisciplinary surveys, integrated approach and standardized reporting to K&G. Babies with a repeated 'aborted test' outcome were not referred, because the 'aborted test' was most often attributable to obvious reasons such as continuous crying of the baby.

The degree of sensorineural hearing impairment, determined by the specialized referral centres, served as the criterion for the diagnosis. If the hearing impairment was 35 dB nHL or more, the baby was coded as congenitally unilaterally or bilaterally hearing impaired (CHI), or otherwise as normal.

When CHI was determined, a standardized protocol of additional comprehensive tests was carried out to determine the aetiology including the use of imaging studies, and paediatric, ophthalmological and genetic consultation. Following each hearing test, the results were uploaded automatically to a central database containing the data of all newborn babies along with selected demographic and socio-economic data, including the familial situation. In a relatively small proportion of babies AAP risk factors were known, but were insufficient for an analysis of the entire group. The UNHS protocol also ensured that all the investigation results and diagnoses from referred babies were reported to the central database.

Hearing impaired children who were missed for screening or children with a false-negative screening result or a progressive or late onset hearing loss, are generally only identified as hearing impaired after a few years, but they always ended in one of the specialized referral centres in Flanders. As part of the protocol for collaboration, the centres report automatically these children to Child and Family. Consequently, these data are integrated into the central K&G database, which so truly covers the total population. Sensitivity and specificity were only calculated 3 years after the screening, to be sure that babies with later discovered CHI were included in the analysis.

During the period of testing (2003–04), 125 595 newborns in Flanders (excluding the Brussels region) were eligible for testing. Babies who died within the first 3 weeks after birth (N = 832) were excluded. Seven thousand babies were not tested, due to reasons like moving, refusal to participate, disinterest of parents or other causes of drop out. Babies tested by private health workers from outside the K&G organization were also excluded (N = 4599) since the methodology of testing was sometimes different, and quality control of the data could not be performed.

In order to have a homogeneous data set of newborns, the cohort of NICU in-patients was excluded because it was commonly known that a significant relationship existed between CHI and NICU admission.

Based on the gestational age and birthweight, babies were classified as either premature or not in accordance with the World Health Organisation (WHO) criteria (weight ≤2.5 kg or gestational age ≤37 weeks). After exclusion of these babies (N = 9329), the final analysis for this study was performed on 103 835 newborns. The final diagnosis after investigation can be ‘confirmed unilateral or bilateral CHI’, ‘normal hearing’ or ‘finally normal, but temporary hearing impairment (HI)’.

Babies diagnosed with temporary hearing loss, caused by, for example, middle ear effusion [otitis media with effusion (OME)], were not considered as having CHI. Despite the correct conclusion of the Algo test, these babies regained normal hearing after treatment. For the calculation of the prevalence of CHI, this group was therefore considered to be ‘normal’ but from the point of view of sensitivity and specificity of the Algo screening test, they were considered as true ‘positive’. For some referred babies, the diagnosis was not conclusive, due to lack of information, chronic effusion of the middle ear, fluctuating hearing impairment or multiple disabilities.

Socio-demographic data were available for all babies, enabling links to be drawn to the results of the hearing screening.

Statistical analysis was performed with SPSS v18 (Statistical Package for Social Sciences). Level of significance for all test statistics was chosen at 5%. Chi-square test was used in general except for birth order, where linear-by-linear association chi-square test was used since the categories in the cross table had an increasing order. Continuous data were compared using analysis of variance (ANOVA) or t-test, given the large number of samples. In order to determine which factors were independent, a backward logistic regression analysis was performed, with prior prevalence of the hearing impairment set to 0.001503, as provided by the data. Twelve variables were investigated for this logistic regression: gender, birth order, birthweight, birth length, head circumference, gestational age in weeks, delivery (caesarean or vaginal), feeding type (breast or bottle fed), age of the mother in years, dwelling (urban or rural), origin of the mother (Western Europe, Eastern Europe, Northern Africa, Turkey, Black Africa or other) and educational level of the mother. Based on the available registered data, educational level of the mother has been defined as ‘Low’ equal to no or primary education only, ‘Medium’ equal to vocational education and ‘High’ equal to secondary and tertiary education.
Results

The average test age of the newborn babies was 25.44 days [standard deviation (SD) = 12.5 days]. Over all tests performed, 86.7% were performed between the age of 7 and 35 days.

From the 103,835 screened babies, the Algo test yielded a ‘pass’ result for 103,139 (99.330%), a ‘refer’ result for 273 (0.263%) and a ‘aborted test’ outcome for 423 (0.407%) babies.

All 273 babies with a ‘refer’ result were referred for further investigation in a specialized referral centre, 95% percent of them before the age of 17 weeks.

Additionally, referral centres reported that 25 babies with an ‘aborted test’ outcome and 14 babies with a ‘pass’ result were later on investigated in specialized referral centres, because there was a matter of concern from health workers or the parents.

For 21 investigated babies, no further conclusions were able to be drawn. Thirty-five referred babies were ‘lost to follow-up’ often caused by moving outside Flanders.

Table 1 summarizes the screening outcome as well as the final diagnosis as obtained by the specialized referral centres. Based on these numbers, test sensitivity and specificity are calculated.

A final diagnosis regarding hearing status was obtained for 256 of the referred babies. Out of this group, 62 babies were diagnosed as normal.

CHI of 35 dB nHL or more (the screening level) was confirmed in 156 babies (a rate of 1.502/1000).

Ninety babies had confirmed bilateral hearing loss of 35 dB nHL or more (a rate of 0.867/1000).

A group of 38 referred babies was diagnosed with ‘temporary’ hearing loss and were not considered as having CHI.

From the 14 babies with an initial ‘pass’ result who were investigated and reported by the reference centres, 10 were confirmed with CHI. These constituted the false negatives in this study, that is 10/103,835 or 0.0096%.

From the 25 investigated children with an ‘aborted test’ outcome, 4 were confirmed with HI, being 4/423 or 0.95%.

When looking at the 103,357 babies with ‘pass’ or ‘refer’ and a known diagnosis, 173 babies had a true-positive screening test result (0.167%) and 11 babies a false-negative result (0.011%). On the other hand, 103,128 babies had a true negative (99.778%) and 45 babies a false-positive test result (0.435%).

In the population of term babies, the sensitivity of the Algo screening test (true-positive rate) yielded 94.02% and the specificity (true-negative rate) yielded 99.96%.

The positive predictive value yielded 79.36% and the negative predictive value yielded 99.99%.

In these calculations, the 423 ‘aborted test’ cases were not included, because they were not referred for investigation due to the fail result. The calculations also did not include the 56 cases that were lost to follow-up or cases where the diagnosis was not conclusive.

This study also investigated the association of socio-demographic factors with the prevalence of CHI.

The relevant characteristics of the study population with respect to CHI prevalence are presented in table 2.

Boys were significantly more affected by CHI than girls.

The risk of CHI increased significantly with birth order. When adjustment was made for the age of the mother (looked at by decade), the link remained for the group of mothers in their thirties.

A significant association between CHI prevalence and origin of the mother was observed in mothers from Eastern Europe. Apart from mothers of Eastern European origin, there was no difference in prevalence of CHI between the different regions of origin of the mother.

A significant association was demonstrated regarding the educational level of the mother, ranked according to the highest achieved level, on the prevalence of CHI. This association remained significant even when only first born babies of Belgian mothers were considered.

Additionally, there was a significant association between breast feeding and the prevalence of CHI. This effect remained after adjustment for the origin of the mother.

The birthweight of the babies with CHI was significantly smaller than their normal hearing counterparts. Gender also had an influence on birthweight, but ANOVA analysis indicated that there was no significant interaction effect, that is, affected babies had lower weights, regardless of gender.

In addition, the length of the babies with CHI was significantly smaller than those babies without CHI, even when taking into account the involvement of the gender (P < 0.001). Thus there was no significant interaction effect.

Our study found that the factors of head circumference, delivery (caesarean or vaginal), dwelling (urban or rural), age of the mother at birth and gestational age of the term babies were not associated with CHI prevalence.

The outcome of the multivariate logistic regression analysis yielded a classification table (table 3) with an overall correct classification of 63% and an equation with the following variables: gender, birth length, birth order, type of feeding, origin of the mother and educational level of the mother.

As presented in table 4 below, the odds ratio [with 95% (confidence interval CI)] of CHI for male over female is 1.7 (1.2–2.4). This means that for a boy the odds for having CHI is 1.7 times higher than for a girl, when all other factors are kept constant.

The odds ratio of CHI when a baby is bottle or mixed fed is 1.75 (1.2–2.5), that is, the odds for having CHI when bottle or mixed fed is 1.75 times higher than when breast fed.

The odds ratio of CHI with mothers of low level education vs those with high level education is 2.46 (1.3–5.8) and 1.31 for medium level of education over a high level of education (0.9–1.9).

The odds ratio for CHI with mothers of Eastern Europe origin is 2.72 (1.3–5.8) compared with mothers from other parts of Europe.

The odds ratio for birth length is 0.86 (0.85–0.87) per increasing length in centimetres (cm), indicating that with each cm increase in birth length the odds ratio of CHI decreases.

The odds ratio for birth order is 1.14 (0.96–1.36). This means that for each successive baby, the odds for having CHI is 1.14 times higher.

Discussion

This study presents the outcome of a UNHS programme in Flanders in 2003 and 2004 with an AABR device, on 103,835 term babies.

The study demonstrated confirmed unilateral and bilateral hearing impairment of 35 dB nHL or more in 1.502/1000 term
newborns. Confirmed bilateral hearing impairment of 35 dB nHL or more was found in 0.867/1000 term newborns.

In this study, 0.95% of the babies with an aborted test were in fact hearing impaired. Therefore, it is important that babies with a hearing test that has not led to a ‘pass’ or ‘refer’ within 20 min would be further investigated for hearing impairment.

This large data set allowed investigation into the question of which socio-demographic determinants are associated with the prevalence of CHI.

Although the effect of these socio-demographic factors on health and more specifically on newborn hearing impairment is still poorly understood, in general, several factors seem to be linked and contribute to this correlation, such as poverty, smoking, working conditions, poorer hygiene, fetal alcohol syndrome, cytomegalovirus (CMV) or other infections, inadequate prenatal care, single parent, consanguinity, unemployment and quality of housing. Factors such as origin of the mother, birth order or education level may be linked to poverty, but all appear to have strong independent contributions to CHI, as shown by the logistic regression.

The study found that gender contributed to CHI as well as decreased birth length. It is known that birth length correlates with smoking, but smoking data were not available in the database. Additionally, underprivileged circumstances or suboptimal feeding in pregnancy may contribute to lower birth length.

Increasing birth order also results in a higher risk of CHI. In literature, increasing number of pregnancies has also been shown to be linked to congenital CMV infection as well as to poverty.

### Table 2 Determinant characteristics of the study population (103 835 term babies)

<table>
<thead>
<tr>
<th>Term baby population</th>
<th>% (N)</th>
<th>CHI prevalence %</th>
<th>Statistical test and P-value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Boys</td>
<td>51.5 (53 437)</td>
<td>1.78</td>
<td>0.018</td>
</tr>
<tr>
<td>Girls</td>
<td>48.5 (50 398)</td>
<td>1.21</td>
<td></td>
</tr>
<tr>
<td>Birth order</td>
<td></td>
<td></td>
<td>Linear-by-linear ass. (0.017)</td>
</tr>
<tr>
<td>First child</td>
<td>47.1 (48 840)</td>
<td>1.31</td>
<td></td>
</tr>
<tr>
<td>Second child</td>
<td>34.8 (36 073)</td>
<td>1.47</td>
<td></td>
</tr>
<tr>
<td>Third child</td>
<td>12.4 (12 920)</td>
<td>2.01</td>
<td></td>
</tr>
<tr>
<td>Fourth child</td>
<td>3.6 (3775)</td>
<td>2.38</td>
<td></td>
</tr>
<tr>
<td>Fifth or more child</td>
<td>2.1 (2161)</td>
<td>2.54</td>
<td></td>
</tr>
<tr>
<td>Origin of the mother</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Western Europe</td>
<td>85.6 (88 797)</td>
<td>1.41</td>
<td></td>
</tr>
<tr>
<td>North Africa (Maghreb)</td>
<td>4.6 (4798)</td>
<td>2.29</td>
<td></td>
</tr>
<tr>
<td>Eastern Europe</td>
<td>2.8 (2878)</td>
<td>3.82</td>
<td>0.011</td>
</tr>
<tr>
<td>Turkey</td>
<td>3.1 (3222)</td>
<td>1.24</td>
<td></td>
</tr>
<tr>
<td>Black Africa</td>
<td>1.2 (1273)</td>
<td>2.36</td>
<td></td>
</tr>
<tr>
<td>Rest of the world</td>
<td>2.7 (2811)</td>
<td>0.71</td>
<td></td>
</tr>
<tr>
<td>Education level of the mother</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Low</td>
<td>3.3 (3381)</td>
<td>0.355</td>
<td>0.001</td>
</tr>
<tr>
<td>Medium</td>
<td>28.0 (29007)</td>
<td>0.193</td>
<td></td>
</tr>
<tr>
<td>High</td>
<td>59.0 (61303)</td>
<td>0.121</td>
<td></td>
</tr>
<tr>
<td>Unknown</td>
<td>9.7 (10088)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Feeding type</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Breastfeeding</td>
<td>64.4 (66 831)</td>
<td>1.21</td>
<td>0.002</td>
</tr>
<tr>
<td>Bottle or mixed feeding</td>
<td>35.6 (36 938)</td>
<td>2.00</td>
<td></td>
</tr>
<tr>
<td>Biometric parameters</td>
<td>Normal hearing</td>
<td>CHI</td>
<td></td>
</tr>
<tr>
<td>Length (cm)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>50.6 (2.0)</td>
<td>50.1 (1.9)</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Female</td>
<td>49.8 (1.9)</td>
<td>49.1 (2.1)</td>
<td></td>
</tr>
<tr>
<td>Weight (kg)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>3.48 (0.43) (53 013)</td>
<td>3.37 (0.41) (94)</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Female</td>
<td>3.34 (0.41) (50 028)</td>
<td>3.24 (0.41) (61)</td>
<td></td>
</tr>
<tr>
<td>Head circumference (cm)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>34.8 (1.4)</td>
<td>34.7 (1.4)</td>
<td>0.095</td>
</tr>
<tr>
<td>Female</td>
<td>34.2 (1.3)</td>
<td>33.9 (1.4)</td>
<td></td>
</tr>
<tr>
<td>% (N)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Way of giving birth</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Vaginal delivery</td>
<td>83.9 (87 119)</td>
<td>1.50</td>
<td>NS</td>
</tr>
<tr>
<td>Caesarean section</td>
<td>16.1 (16 696)</td>
<td>1.50</td>
<td></td>
</tr>
<tr>
<td>Area of living of the mother</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Urban</td>
<td>71.8 (74 510)</td>
<td>1.53</td>
<td>NS</td>
</tr>
<tr>
<td>Rural</td>
<td>28.2 (29 325)</td>
<td>1.43</td>
<td></td>
</tr>
<tr>
<td>Mother's age</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Average years (minimum 13 years–maximum 50 years)</td>
<td>28.98 (4.71)</td>
<td>NS</td>
<td></td>
</tr>
<tr>
<td>Gestational age</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Average weeks (minimum 37 weeks–maximum 48 weeks)</td>
<td>39.29 (1.12 weeks)</td>
<td>NS</td>
<td></td>
</tr>
</tbody>
</table>
If one looks at the rate of antibodies to CMV in developed countries, 50–60% of the pregnant, middle to upper class women are positive, compared with 70–85% of those in lower socio-economic groups. 16,17

When the mother originates from an Eastern European country, the odds for CHI increases with a factor of 2.72, while no other significant effect from origin appears to determine CHI. A possible explanation might be the high proportion of Roma people among the immigrants from Eastern Europe. Neurological diseases in newborns are more prevalent among the Roma population and this has been linked to consanguinity. 18,19

Our study also showed that a lower educational level of the mother increases the risk of CHI. Similar findings were obtained by Lee et al. 20 in Hispanic schoolchildren. As shown in several studies, the level of education is an important factor in health disparities. 21 More highly educated mothers are more likely to have a healthy lifestyle. 22

A similar intricate effect emerges from the impact of breastfeeding on the prevalence of CHI. Breastfed newborns were less likely to have CHI than their bottle fed counterparts.

Although feeding type is linked to education level, origin of the mother, environmental factors, but also to poverty and smoking habits, our logistic regression analysis has shown that feeding type appears as an independent variable, which contributes to the prevalence of CHI. Despite the fact that breastfeeding is a postnatal factor, there is an intricate and subtle relationship with the health status of the baby and the mother. This study remains inconclusive on the exact mechanism of the complex relationship of feeding type with CHI. Since poor people are less likely to breastfeed, we hypothesize that breastfeeding, through the path of poverty is linked to CHI. 23,24

The table of the logistic regression states ‘sensitivity’ and ‘specificity’ values for the model fit of 63.2 and 64.2%, respectively (table 3), which are usually considered as rather low, but when an event occurs only in 0.15% of the cases, a combination of factors that identifies a disease in 64% of the cases, is quite powerful.

The logistic regression analysis yielded gender, birth order, birth length, feeding type, level of education and origin of the mother as independent socio-demographic factors to predict CHI.

When this combination of factors is found in an individual baby, care needs to be taken to make sure that results are accurate. Aborted tests should be investigated more than ever and tests suggesting referral should also be followed up aggressively and with great attention. 20,25 When in a population, the organization of UNHS for the total target group is not feasible, then policy makers must give priority to the underprivileged people in the organization of screening. They are a key risk group that can be screened with the highest cost-effectiveness.

The strength of this study is undoubtedly the huge sample size of the investigated group, covering the total population of term babies in a geographic region (Flanders). Additionally, the 3-year time span between screening and analysis ensured that late discovered CHI cases were finally also included in the analysis. Furthermore, all data were obtained using the exact same protocol and procedures converging in a central database system. Next, we adopted multivariate statistics to uncover combinations of factors that are associated with CHI, rather than reporting differences on each separate variable. As such, intricate mechanisms emerge, and this study, therefore, is complementary to other studies where individual factors are investigated. This rather unique approach, therefore, allowed this study to uncover that hearing impairment in term babies could be linked to a cluster of socio-demographic factors.

On the other hand, unfortunately, the lack of knowledge on AAP risk factors in this population did not permit an AAP risk assessment. Neither was any data available regarding the smoking habits and alcohol consumption during pregnancy, partially caused by restrictive privacy policy and regulations. This is obviously a limitation. Future studies should therefore include all these aforementioned factors to uncover more aetiological mechanisms of CHI. Additionally, the current revealed non-trivial factors urge certainly the need for further studies in the field of socio-demographic risk factors for CHI.

The observation by the AAP that 40–50% of babies with CHI do not display risk factors belonging to the classical list may partially be due to the fact that socio-demographic risk factors are not included. We hypothesize that inclusion of the here reported risk factors might reduce the aforementioned percentages.

We can conclude that the classic risk factors for CHI can be extended with an additional socio-demographic cluster of factors, including: gender, birth order, birth length, feeding type, education level and origin of the mother. The data indicates that, additional to the increased risk of other dysfunctions or pathology, newborns in underprivileged families also have a higher risk of hearing impairment. Thus, when a physician in the context of neonatal hearing impairment is confronted with a newborn boy of small length, bottle fed, is the second or subsequent baby of a lower educated European mother, he/she has to be aware that this baby is at higher risk of CHI and should certainly be closely investigated.

Consideration of these socio-demographic factors improves decision making in individual medical practice and in public health policy on screening priorities.

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Conflicts of interest: None declared.

Key points

- The total population of term babies in Flanders was studied.
- Babies with a hearing test that has not led to a 'pass' or 'refer' within 20 min should be further investigated for hearing impairment.
- Socio-demographic factors were identified as independent determinants for CHI.
- Presence of the herein specified cluster of risk factors should urge the involved clinicians to pursue abnormal or aborted hearing screening results.
- Underprivileged people are a key risk group for CHI.

References