Six month impact of false positives in an Australian infant hearing screening programme

Z Poulakis, M Barker, M Wake

Aims: To assess short and longer term parent reported impacts of false positive referrals in the Victorian Infant Hearing Screening Program (VIHSP).

Methods: Mailed retrospective case-control survey of infants consecutively referred to VIHSP between December 1998 and April 1999 for whom audiology did not confirm permanent hearing loss, comprising 137 infants screened with a neonatal risk factor questionnaire and 148 older infants screened with two consecutive behavioural (distraction) tests. The two control groups comprised non-referred screened infants matched by domicile, age, and gender. Main outcome measures were parent reported emotions experienced before and after child’s audiology test, parent estimated impact of hearing loss, the Child Vulnerability Scale, audiology assessment satisfaction questionnaire, and questions relating to their child’s hearing and language development.

Results: Final sample: at risk cases (AR) 108 (79% response), at risk controls 64 (51%); distraction test cases (DT) 103 (70%); distraction test controls 53 (41%). Parents across all groups believed that hearing loss would have major effects on a child’s language (91–96%), schooling (81–91%), and employment opportunities (67–75%). Before audiology, 71% (AR) and 72% (DT) of case parents were anxious/worried, falling to 4% and 15% afterwards. After the test 82% (AR) and 79% (DT) reported relief, but 19% and 18% continued to feel worried. Ongoing concerns about hearing, language, development, and general health were comparable for AR cases compared to controls, and for DT cases compared to controls.

Conclusions: Hearing screening tests are generally well received. Parents are realistic about the impact of childhood hearing loss and report a range of negative emotions when a false positive hearing screen requires referral. Although most are reassured by a normal test, a substantial number report continuing concern.

Infant hearing screening, in both targeted and universal programmes, is now a widely accepted method for the early detection of hearing loss. As with any population based programme, benefits of screening should outweigh potential harms. The prevalence of the target condition (significant bilateral hearing loss very early in life) is low, and thus the positive predictive value of hearing screens is generally also low. While some have reported positive predictive values as high as 17% or more, recent US Preventive Task Force systematic review reported values ranging from just 2.2% to 11.7%. In other words, the great majority of babies with “failed” screens have normal hearing.

This high false positive rate has led a number of authors to raise the issue of potential negative impacts of hearing screening.

Studies of programmes screening for other medical disorders indicate that false positive test results cause substantial parental concern and anxiety which may last for months or years. Studies that have directly assessed the negative effects of hearing screening programmes are to date inconsistent.

It appears that positive (“refer”) screen do engender at least transient negative emotions in a proportion of parents. Most studies, however, indicate that the immediate concern and distress engendered in parents by false positive tests is minor, and acceptance of hearing screening is high. Other studies have reported equivalent levels of stress or anxiety in mothers whose infants receive a refer result when compared to those who receive a pass result, or to controls not receiving neonatal hearing testing at all.

There is little information about longer term impacts of false positive screens. Clemens, Davis, and Bailey (2000) surveyed 49 mothers whose infants had “failed” an initial automated ABR screen in a two stage UNHS programme. Twelve per cent of those mothers surveyed a number of months after UNHS (mean 4.9, range 2–13) reported lasting “mild anxiety” even after their child passed second stage screening. The Wessex study’s universal hearing screening programme did not lead to increased maternal state or trait anxiety or more negative attitudes towards the baby 2–10 months after a screen “refer” result, compared to mothers whose babies passed the screen. However, it did not report on ongoing parent concern about hearing, language, or development, or more specific residual anxieties than can be measured with the Spielberger State-Trait Anxiety Inventory.

In this case-control study, we report on psychological and social impacts of false positive hearing screens approximately six months after the diagnostic audiology test in otherwise healthy infants. We hypothesised that parent reported anxiety and other negative emotions prior to diagnostic audiology would largely resolve following a diagnostic audiology test result that did not confirm permanent hearing impairment. We also hypothesised that parents would not view their child as more vulnerable to hearing, language, or general health problems several months later, and that most parents would be aware of the likely impacts of significant hearing loss for a child.

Abbreviations: AR, at risk; DT, distraction test; MCH, maternal and child health; UNHS, universal newborn hearing screening; VIHSP, Victorian Infant Hearing Screening Program.
**METHODS**

**Setting**
The birth rate in the state of Victoria, Australia, is approximately 60 000 per annum, with approximately 110 children diagnosed with permanent hearing loss managed with hearing aids from each annual birth cohort.24 A universal two tier infant screening programme, the Victorian Infant Hearing Screening Program (VIHSP), has been operating since late 1992. All newborn infants are screened by their maternal and child health (MCH) nurse using a written questionnaire to screen for the presence of one or more of eight risk factors for hearing loss (table 1). If any risk factor is present, infants are referred for diagnostic audiological evaluation as soon as possible after birth. Five per cent of each annual birth cohort (approximately 3000 infants) are referred for assessment via this pathway; at 7–9 months, all infants who have not already had formal audiological testing are again screened by their MCH nurse with a behavioural screening test based on the Ewing distraction test; a further 5% of the birth cohort is referred for diagnostic audiological evaluation following two failed behavioural screens. The great majority of referred infants do not have hearing loss.25

**Design**
This project was a retrospective community based case-control survey. It was approved by the Ethics in Human Research Committee at the Royal Children's Hospital, Victoria, Australia.

**Cases**
Subjects were selected from all infants referred to VIHSP between December 1998 and April 1999 whose subsequent audiological assessment indicated that permanent hearing loss was not present. Of the 691 referral forms with complete data received in that period, 368 (53%) were referrals for at risk infants and 251 (47%) were for infants failing a distraction test screen twice. Sample sizes of approximately 150 were randomly selected to be able to detect a 5% mean difference in scores between groups with alpha set at 0.05 and power of 0.90.

We wished to focus on the impact of a false positive hearing test in otherwise healthy children. Therefore, infants with a risk factor that considerably increased the likelihood of ongoing health or developmental problems were excluded (see table 1). A total of 137 (37%) of the at risk referrals met inclusion criteria and comprised the at risk case group.

Of the 251 referrals for children who failed the distraction test screen twice, we randomly selected 148, including equal numbers of children with conductive loss and normal hearing, to comprise the distraction test case group.

**Controls**
Every child in Victoria is allocated to a single MCH nurse serving a small geographical area (approximately 100–150 births per year per nurse) based on street address. Each case child's MCH nurse was asked to select a control infant by matching the child from her centre closest to the case group infant on age and sex. The additional criterion for inclusion into the at risk control group was that the infant did not have a risk factor for hearing loss. The additional criterion for inclusion into the distraction test control group was that the infant had not failed two distraction test screens.

Questionnaire packages were mailed by the researchers to the parents of all children in two case groups in August 1999, with reminder letters at six and 12 weeks. Parents of control children were sent the study package (and reminders where appropriate) by the MCH nurse who notified the study group when questionnaires were sent. The identity of the controls was concealed to the study group.

**Measures**
All questionnaires contained sections eliciting information on the infant's hearing history/results, family demographics, parents' thoughts about their child's health and development, and estimated impact of childhood hearing loss. The questionnaires sent to the case groups contained additional sections assessing parents' experience of the time before and after their child's audiological assessment and their evaluation of the hearing screening and/or audiological assessment process.

Emotions felt by parents at the time leading up to and immediately after their child's audiology test were assessed using 10 questions based on the 1997 study of de Uzcategui and Yoshinaga-Itano.26 Each item was scored on a three point scale (“not at all”, “a little”, “a lot”).

Parents' perceptions about their child's general susceptibility to ill health were assessed using the validated Child Vulnerability Scale.27 Higher total scores reflect increased perceived vulnerability to general ill health, with a total cut off score of 10 used to indicate high vulnerability. We added four questions about parents' perceptions of their child's current hearing and language development (“I have to call my child several times before he/she responds”, “My child's hearing doesn't seem as good as other children”, “I worry about my child's overall development compared to other children”, and “I worry about my child's language compared to other children”). All items were scored on a four point scale (“strongly agree”, “agree”, “disagree”, “strongly disagree”). Parents were also asked “How worried are you now about your child's hearing?” (“not at all”, “a little”, or “a lot”).

Parents' thoughts regarding the likely impact of childhood hearing loss on a number of child and family outcomes were assessed using a 13 item questionnaire developed for the study. Child variables were language, schooling, intelligence, ability to make friends, and employment opportunities. Family related variables were family activities or outings, seeing friends or relatives, confidence in being a parent, relationships, health (including stress), parent employment opportunities, parent income and earnings, and time spent with the deaf child. Parents estimated the impact hearing loss would have on each area by rating each item on a four point scale (“no effect”, “small effect”, “medium effect”, “big effect”).

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**Table 1** Risk factors for which referral for ABR testing recommended by VIHSP*

<table>
<thead>
<tr>
<th>Risk factors included in this study:</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. Family history of congenital hearing impairment</td>
</tr>
<tr>
<td>2. Exchange transfusion or serum bilirubin level greater than 350 µmol/l</td>
</tr>
<tr>
<td>3. Minor congenital abnormalities of the head and neck</td>
</tr>
<tr>
<td>4. Parental concern</td>
</tr>
<tr>
<td>5. Rubella, cytomegalovirus, or toxoplasmosis during pregnancy or other perinatal infections</td>
</tr>
<tr>
<td>6. Birth asphyxia as defined by an Apgar score of less than 4 at five minutes of age</td>
</tr>
<tr>
<td>7. Birth weight below 1500 g</td>
</tr>
<tr>
<td>8. Major congenital abnormalities of the head and neck</td>
</tr>
<tr>
<td>9. Other risk factors, e.g. bacterial meningitis, developmental delay</td>
</tr>
</tbody>
</table>

*Based on Joint Committee on Infant Hearing 1994 position statement.
Parents evaluated the hearing screening and diagnostic assessment process using the MOS-9, a widely used nine item satisfaction scale.

Data were stored and analysed using Microsoft Access 97 and SPSS.26

**RESULTS**

The sample

Questionnaires were returned for 108 (79%) of the children in the at risk case group, and 103 (70%) in the distraction test case group. On average, 5.4 (SE 0.14) months had elapsed between definitive test results and questionnaire completion for at risk cases, and 6.0 (SE 0.17) months for distraction test cases. The study group received notification from MCH nurses that 126 of a possible 137 (92%) questionnaires had actually been sent to at risk control children, and that 129 of a possible 148 (87%) questionnaires had been sent to distraction test control children. We received 64 (51% of the target) questionnaires back from the at risk control group and 53 (41%) from distraction test case group. We received 129 of a possible 148 (87%) questionnaires had been sent to at risk control children, and that 6.0 (SE 0.17) months for definitive audiology testing.

Table 2 shows parent reported outcomes of audiology assessments for the 212 case children. Of the entire group of children in the study, 22 (7%) were referred to an ear, nose, and throat surgeon, and of these, 13 had tympanotomy tubes.

Emotions before and after testing

Table 3 shows the retrospectively reported emotions experienced by parents during the period after their child's identification of risk/failed screen, but before their attendance for definitive audiology compared to the emotions reported after definitive audiology testing.

<table>
<thead>
<tr>
<th>Emotion</th>
<th>% at risk (n=108)</th>
<th>% distraction test (n=104)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Before</td>
<td>After</td>
</tr>
<tr>
<td>Angry</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Sad/depressed</td>
<td>13</td>
<td>0</td>
</tr>
<tr>
<td>Powerless/helpless</td>
<td>24</td>
<td>0</td>
</tr>
<tr>
<td>Impatient</td>
<td>25</td>
<td>2</td>
</tr>
<tr>
<td>Shocked</td>
<td>9</td>
<td>n/a</td>
</tr>
<tr>
<td>Relieved</td>
<td>n/a</td>
<td>82</td>
</tr>
<tr>
<td>Upset</td>
<td>9</td>
<td>0</td>
</tr>
<tr>
<td>Anxious/worried</td>
<td>71</td>
<td>4</td>
</tr>
<tr>
<td>Stressed</td>
<td>29</td>
<td>1</td>
</tr>
<tr>
<td>Guilty</td>
<td>7</td>
<td>0</td>
</tr>
<tr>
<td>Confused</td>
<td>7</td>
<td>0</td>
</tr>
</tbody>
</table>

n/a, not asked

*McNemar test; †Fisher’s exact test used when one cell expected value less than 5.

Valid percentage figures are reported throughout. All variables dichotomised into “not at all” versus “a little” and “a lot” combined.

In both case groups, most negative emotions resolved and most parents reported feeling relieved after their child’s definitive hearing test. More parents in the distraction test case group reported negative emotions after their child’s definitive hearing test than parents of children in the at risk case group (table 3).

**Concerns six months after testing**

Nineteen percent of case parents were still “a little” worried about their child’s hearing at the time of completing the questionnaire, which was on average 5.6 months (SE 0.21) after receiving a normal hearing result. There were no differences in the proportions of parents of at risk or distraction test cases who were still worried about their child’s hearing (19% versus 18%, $\chi^2 = 0.9, p = 0.45$). However, of those parents in the distraction test case group who were still worried about their child’s hearing, 78% had children with demonstrated conductive losses (mainly mild).

Parent perceptions of their child’s hearing, language, and general development did not differ between cases and controls in either the at risk or distraction test groups. Mean Child Vulnerability Scale scores were similar across the four groups ($F = 0.5, p = 0.70$), as were the proportions of children whose Child Vulnerability Scale score was above the cut off of 10 ($\chi^2 = 4.3, p = 0.23$).

Parent perceived impact of hearing loss

Many parents thought hearing loss, if present, would have “medium” or “big” impacts for a child and his/her family; these beliefs were almost identical across the four groups and were therefore combined. Overall, 93% of parents thought that a child’s language would be affected, followed by schooling (88%), employment opportunities (72%), ability to make friends (59%), and intelligence (34%). Perceived medium to large family impacts were also frequent; 55% thought a deaf child would require more of their time, 49% believed a child’s hearing loss would increase parent stress, 43% believed it would alter their own employment opportunities, 42% income and earnings, and 40% their confidence in being a parent.

Perceptions of testing

Parents of children in the at risk case group were more satisfied overall with the audiology testing process than parents of children in the distraction test case group (mean = 1.8 (SD 0.6) versus 2.0 (SD 0.8); $t = 4.9, p < 0.01$). Only 6% of at risk parents and 6% of distraction test parents rated the audiological assessment as “somewhat difficult/pleasing”. However, 28% of distraction test case parents rated the distraction test
process as either “fair” or “poor” compared to just 10% of controls ($\chi^2 = 6.7, p = 0.01$).

**DISCUSSION**

This retrospective case-control study investigated parent reported emotions after false positive results on two types of hearing screen (newborn risk factor questionnaire and 7–9 month distraction test) approximately six months after diagnostic audiology testing. As predicted, these parents of children without permanent hearing loss were very aware of the impact such a loss might have on a range of child and family functioning domains, regardless of their case or control status.

Consistent with other research, anxiety and worry were frequently reported before definitive audiology testing, suggesting the immediate impacts of positive screens are negative emotions. However, as hypothesised, most parents in both groups reported feeling relieved after diagnostic audiology, and negative emotions largely resolved.21

The screening and audiology assessments were generally well received by all groups, with parents of the distraction test case group being slightly less satisfied. This may be related to how audiology practices prioritise waiting lists, with younger infants (that is, newborns with a risk factor for hearing impairment) tending to be seen sooner, while older infants may wait longer for diagnostic assessment. Paradoxically, parents of the distraction test case group may feel their child has already “failed” two hearing tests (the distraction screens), and therefore perceive the urgency and importance of the diagnostic assessment to be paramount, especially when a significant number of these babies do have conductive hearing loss. Moreover, parents of distraction test cases tended to report negative emotions after definitive audiology testing more frequently than did parents of at risk cases.

A substantial proportion of parents across both case groups (19%) were still “a little” worried about their child’s hearing approximately six months after receiving a normal result from definitive audiology testing. In the distraction test group, most of these cases were children with ongoing conductive hearing problems. However, this was not the case in the at risk group. We were not able to determine whether this continuing concern reflected development of conductive losses or heightened concern because of awareness of a risk factor even in the presence of a normal audiology result. However, parents reported concerns about language and general development and perceived vulnerability to ill health did not differ among the four groups in this study.

To our knowledge, this is only the second controlled study to investigate longer term concerns about hearing in parents of children who have been referred for diagnostic audiology via a screening process, and where diagnostic results indicate a normal result. It adds to the findings from the Wessex study22 by focusing on potential lasting concerns relating to children’s hearing, including language, development, and health, rather than on the more general constructs of overall maternal anxiety and positive/negative attitudes towards the child. The inclusion of two control groups matched on age and sex with which to compare the two case groups strengthens this study’s findings.

Limitations include the rather low response rate for children in the two control groups. However, this did not detract from those comparisons (the majority) which focused on the case groups only. Retrospective collection of data, resulting in several months having elapsed between the screening and diagnostic process data collection, could have resulted in biased recall and an understimation of the negative emotions experienced at the time. However, this did not apply to the longer term outcomes of particular interest to this study.

Finally, the condition of interest in this study (permanent hearing loss) has a low prevalence in the population (0.2%),23 and therefore the positive predictive value of screening tests is also low.24 It is possible that parents that would be reporting greater levels of concern for conditions where positive results on screening tests are more predictive of the presence of the target condition. Low levels of concern, however, were clearly not related to lack of knowledge about the condition’s likely impact.

Hearing screening programmes are generally well accepted and result in few negative emotional reactions. Some parent anxiety is unavoidable; a certain level of anxiety may even motivate parents to attend diagnostic testing. Staff who are involved with hearing screening programmes should be aware of the emotions likely to be experienced by parents whose children do not pass the hearing screen, and that these emotions are not all immediately and completely alleviated by a normal result at diagnostic testing. However, we can also be reassured that these negative emotions do not appear to translate to long term concerns about hearing, language, development, and general health, even when parents are aware of the likely impacts of deafness on a young child.

**ACKNOWLEDGEMENTS**

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**REFERENCES**

ARCHIVIST

Choosing deafness

Dialectes Have you heard, Archivistes, that a couple in America wanted, and arranged to have, a deaf child?

Archivistes Why and how did they do that?

Dialectes Well, they are a lesbian couple who are both deaf and they wanted children through donor insemination. As sperm donor they chose a deaf friend who could trace deafness back through five generations of his family. Their first child, a girl born 5 years ago, is deaf and now they have a boy who also is deaf.

Archivistes As so often, Dialectes, you have explained how but not why.

Dialectes They wanted their children to be deaf like themselves.

Archivistes It must surely be wrong to impose a handicap on an unborn child.

Dialectes They argue that the harm that results from being deaf is socially imposed. The deaf are a minority group who are discriminated against as are girls and black people. Nobody thinks it wrong to want a girl or a black baby.

Archivistes But the deaf can't hear.

Dialectes Quite so, Archivistes.

Archivistes How can it be argued that being unable to hear is desirable?

Dialectes Deaf people have to suffer the unthinking prejudices of hearing people that deny them opportunities that could be opened to them. They may find comfort in their own deaf community and some, it seems, believe that membership of this community is so rewarding that their children should also be members even though the condition of membership is lack of hearing.

Archivistes Are all the disadvantages of deafness socially imposed?

Dialectes Many are, but not all. Being unable to hear music, birdsong, a baby crying, a car horn, or a fire alarm are not social impositions. Of course we should do as much as possible to minimise the disadvantages but it seems unlikely that they can be eliminated.

Archivistes When does an impairment become a disadvantage?

Dialectes When it prevents the impaired person doing what she or he wants to do.

Archivistes Would it be justified to select for impairment if parents and others believed that the impairment would not be a disadvantage?

Dialectes You have not listened carefully, Archivistes. I am the only one who can decide what I want to do. To select for impairment is to deprive the child of choice; it is an infringement of liberty. It is unlikely that a child would choose to have no choice.

Archivistes Many people with handicaps lead rewarding, successful, and apparently happy lives, Dialectes.

Dialectes That is true, but acknowledging it and admiring such people is a far cry from wanting a child to be born impaired. That is something we should not wish for.

References

1 Spriggs M. Lesbian couple create a child who is deaf like them. J Med Ethics 2002;28:283.

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