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ABSTRACT

Objective. The aim of this study was to determine whether a failure of neonatal hearing screening affected the anxiety level of parents of high-risk infants.

Methods. 288 parents of infants included in the neonatal hearing screening protocol of our Institution were tested with the Spielberger State-Trait Anxiety Inventory and with an open-question questionnaire investigating parents attitude to hearing problems in their child, done at the time of audiological follow-up. 105 were parents of high-risk infants who had been discharged from neonatal intensive care unit and 183 of low-risk infants discharged from well-baby nursery.

Results. No differences in anxiety levels were seen between parents of high-risk infants passing and failing neonatal hearing screening using homogeneous case-control pairs. Additionally, no differences in level of anxiety were found between parents of high- and low-risk infants failing neonatal auditory screening.

Conclusions. Failure of neonatal auditory screening does not affect the anxiety levels of parents of high-risk infants at post discharge from neonatal intensive care unit. This finding is a key factor to be considered when evaluating the costs and benefits of tests for universal neonatal hearing screening.

TITLE PAGE

Failure of hearing screening in high-risk neonates does not increase parental anxiety

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INTRODUCTION

Neonatal hearing screening programmes have been implemented worldwide following recommendations of the American Academy of Paediatrics [1] and the Joint Committee of Infant Hearing [2]. Detection of hearing impairment within 3-6 months of life and early habilitation are of great importance for language acquisition [1,2].

Screening for hearing impairment is generally achieved by testing all neonates at birth by evoked otoacoustic emissions [3] and/or automatic auditory brainstem responses (a-ABR) [4,5]. During the neonatal period, several audiological or neural factors may have a transient interference with these tests giving rise to false-positive results, particularly in those admitted to neonatal intensive care units (NICU) [4].

Research has shown that a false-positive result in the neonatal screening for other disorders such as congenital hypothyroidism [6], phenylketonuria [7], Down syndrome [8] or cystic fibrosis [9] may cause unnecessary long-lasting parental concern. Parental anxiety may adversely affect the parent-child relationship because of parents continuing to worry about their child's health, despite the previously positive result having been found to be false [10,11]. The so-called "vulnerable child" syndrome may thus develop [12].

The issue of long-lasting parental anxiety in relation to the outcome of neonatal auditory screenings has been of concern especially since the majority of infants failing auditory screening are subsequently found to have normal hearing [13-18].

In the population of low-risk neonates admitted to the well-baby nursery, the false-positive outcome of neonatal hearing screening was not shown to produce long-lasting parental anxiety [15,16], but earlier studies did not focus on parents of high-risk infants discharged from NICU. In this population, which is at higher risk of hearing loss, a higher anxiety could be expected; indeed, NICU admission for more than 48 hours represents the single best audiological risk factor [19]. Moreover, NICU admitted infants are at risk for major health problems which can influence per se the emotional state of the parents [20].

The STAI-Y questionnaire allows evaluation of the emotional response of the subject to a stressful event and of the general feeling of anxiety; it has been shown to be sensitive enough for identification of raised levels of parental anxiety in cases of serious illness in their children [15] particularly in parents of infants discharged from NICU [20].

The aim of this study was to determine the parental anxiety related to possible hearing problems in the population of parents of high-risk NICU admitted infants who failed the neonatal hearing screening.

METHODS

Setting

The neonatal hearing screening program of the Paediatric University Hospital of Padua is based, in the third level NICU, on transient evoked otoacoustic emissions and a-ABR done before discharge and repeated at follow-up between 3 and 5 months. By contrast in the well-baby nursery infants are screened with transient otoacoustic emissions before discharge, and retest is done only in case of failure. The outcome of the screening is defined "pass" when the screen is normal in both ears, "fail" when it is not normal in one or both ears and retest is needed. Infants failing retest have a complete audiological work-up.

Subjects

The study included parents of all neonates discharged from our third level NICU (high-risk group) and well-baby nursery (low-risk group) who participated in the auditory follow-up program in the period between February 2010 and December 2010. Parents gave written informed consent to the study. In order to maximise the reliability and validity of the study only Italian-speaking parents were selected. The questionnaire was administered by an interviewer to each participant privately.

A total of 288 parents (200 mothers and 88 fathers) of 233 children (143 infants from well-baby nursery all "fail", 90 from NICU - 43 (47.8%) "fail" and 47 (52.2%) "pass") completed the STAI-Y questionnaire at the time of post discharge auditory follow-up before they received results of retesting.

A subgroups of parents (54 pairs including mothers and fathers) of the same high-risk neonate performed also an open-question questionnaire investigating parents attitude to hearing problems in their child and qualitative level of the emotional feeling.

Clinical data of the NICU admitted infants, retrieved from the medical letter given to parents at discharge, are summarized in Table I.

TABLE I about here

The Ethical Committee of the Hospital approved the study.

Measures

Anxiety level of the parents was evaluated using the 40-items version of the Spielberger State-Trait Anxiety Inventory questionnaire Y-form (STAI-Y) [24]. The STAI-Y is the leading measure of personal anxiety worldwide and has been used and validated by health professionals in a variety of

different settings; the Italian edition was adapted by Pedrabissi and Santinello in 1989 [25]. Twenty of the forty STAI-Y statements describe the anxiety state – that is the emotional answer to an event (STAI-Y1), the other twenty statements describe the anxiety trait, that is the general feeling of the subject (STAI-Y2). Each item counts four possible answers. For each parent, scores were combined to form a sum-STAI-Y. Scores range from 20 (no anxiety) to 80 (high anxiety). Scores higher than two standard deviations above the mean were considered clinically relevant. We performed both STAI-Y1 and STAI-Y2 scores in order to exclude possible confounding effects due to differences in the baseline anxiety level of groups. The effect of specific risk factors for parental anxiety in NICU admitted infants (resuscitation at birth, mechanical ventilation, apnoeas)[26] was also investigated.

To measure the hearing-specific anxiety, parents had to replay with four possible answers ("It can be", "It would be better", "Perhaps", "Indifferent") to a questionnaire with three open-questions investigating parents attitude to hearing problems in their child and qualitative level of their emotional feeling.

The a-ABR and transient evoked otoacoustic emissions recordings were performed using Accu-Screen PRO-GN Otometrics/Madsen Electronics/Copenhagen Denmark (www.gnotometrics.com). A binomial statistical test gives automatically a response score ('Pass' or 'Refer'). For a-ABR the waveform obtained following stimulation at 35 dB nHL at a rate of approximately 55 Hz is compared to a template, derived from a composed waveform obtained from a number of normal neonates. The transient evoked otoacoustic emissions were elicited following nonlinear click sequence at 73 dB SPL (corresponding to 35 dB nHL), generated by a small probe positioned in the neonate external canal, the sounds emitted by active mechanical processes in the outer hair cells are recorded by a microphone included in the probe; the statistical probability that an emission has been recorded at a succession of points ranging from 6 to 12 ms after the end of the stimulus determines the result.

No sedation was given and whenever possible neonates were tested in sleeping/quiet state, after feeding and/or bathing [4].

Statistical analyses

The group of parents of high-risk infants discharged from NICU and failing neonatal hearing screening was compared with two different groups: 1) parents of infants discharged from NICU and passing the auditory screening and 2) parents of infants discharged from well-baby nursery and failing the auditory screening. Student's t test for independent groups was used to compare the mean of the state (STAI-Y1) and trait (STAI-Y2) scores in different groups. Homogeneous case-control pairs ("pass-fail") were matched for same gestational age ± 1 and same modality of partum caesarean or vaginal. Mann-Whitney U test was used to investigate the effect on STAI-Y scores of risk factors for parental anxiety other than failure of neonatal screening.

P values <0.05 were considered significant.

RESULTS

The STAI-Y1 and the STAI-Y2 scores of mothers and fathers of neonates failing and passing neonatal hearing screening in NICU and well-baby nursery are summarised in Table II.

TABLE II about here

We did not found differences in the STAI-Y2 scores between groups; a clinically relevant elevation in the trait-anxiety scores was seen in only two mothers of high-risk infants failing hearing screening (T scores of 60 and 66).

The STAI-Y1 scores of mothers and fathers of high-risk infants passing and failing neonatal auditory screening were not significantly different.

The STAI-Y1 scores of mothers and fathers of infants failing the auditory screening discharged from NICU and from well-baby nursery were not significantly different.

There were no significant differences of anxiety level between mothers and fathers of the same infants.

By contrast, considering the influence of medical risk factors other than failure of neonatal auditory screening in infants discharged from NICU, we found that the state anxiety scores were significantly higher in parents of infants who were resuscitated at birth (U=335,5; p=.030), while mechanical ventilation and apnoeas did not appear to influence parental anxiety levels. No significant interaction was found between resuscitation and outcome of neonatal hearing screening.

The responses to the open-question questionnaire are in agreement with results obtained using STAI-Y questionnaire. At the first question only 14,81% of parents seemed to be worried about outcome of hearing screening against 53,7% which were not worried. At the second question, 57,4% denied to have adopted special care concerning hearing impairment. At the third question, only 22,22% affirmed that it could have been better to anticipate rescreen. No differences were found between responses given by the parents of children from NICU and those from well-baby nursery.

DISCUSSION

The results of the present study show that parents of infants who had health problems requiring admission to NICU at birth and who have a "normal" background anxiety, have levels of anxiety similar whether their child passed or did not pass the neonatal auditory screening. Furthermore the level of anxiety of parents of high- and low-risk infants was similar.

Parents were tested before the auditory follow-up test, thus they were not influenced by the outcome of their auditory follow-up.

Previous studies in parents of low-risk infants discharged from well-baby nursery showed no differences in parental anxiety evaluated with the Spielberger State-Trait Anxiety Inventory questionnaire (STAI-Y), whether or not their child passed auditory hearing screening. However, earlier studies evaluating the possibility of long-lasting parental anxiety with non-standardized protocols in mixed population of low- and high-risk infants gave conflicting results [13-18].

Parents of high-risk NICU-admitted infants constitute a unique population since the risk of hearing loss is higher in their children because of major exposure to ototoxic drugs, neonatal asphyxia and infective events. The 2007 JCIH guidelines, stated that admission to NICU for more than 48 hours, represents the highest risk factor for hearing impairment [19] and population based studies indicate a prevalence of hearing loss of 2–4% in graduates from NICU in contrast with about 1/1000 live births in the general population [19]. Furthermore, NICU admitted infants are at risk for major health problems which can influence the emotional state of the parents. The majority of NICU admitted infants are born preterm and this condition has been reported to produce high levels of state and trait anxiety, persisting even if the child growth is without further problems [20]. We took into account prematurity since we used case-control pairs homogeneous for gestational age, but didn't find differences between NICU admitted infants failing and passing the neonatal

hearing screening. However, possible subtle anxiety differences related to hearing outcome could have been masked, in high-risk infants, by worries for major health pathologies. In fact we found that the neonatal factor influencing the state anxiety scores of parents of high-risk infants was resuscitation at birth.

A limitation of our study was the impossibility of having data on STAI-Y in parents of low-risk infants passing the neonatal hearing screening, because our protocol for neonatal auditory screening do not include follow-up in that population.

To know that failure of the hearing screening in high-risk infants do not represent a significant problem for the parents and their relationship with their child is important because the high rate of false-positive results remains an open issue, particularly in NICU-admitted infants[4]. In this population the different screening tools, otoacustic emissions, a-ABR and brainstem auditory evoked potentials, have shown different rates of false positive results [4].

Otoacustic emissions are acoustic responses produced in the inner ear by physiologic activity of the outer hair cells, measured with a sensitive small microphone positioned in the external auditory canal; they reflect mechanical processes which do not contribute to hearing but provide an indication of the integrity of the cochlea [3]. The a-ABR is based upon automated detection of the neural response generated at brainstem level following acoustic stimuli; the obtained waveform is tested statistically eliminating the need for specialist interpretation [5]. Otoacustic emissions and a-ABR provide non-invasive, fast, easy to perform and painless recordings and do not require specialist interpretation but, particularly in NICU admitted, ventilated infants, low post-natal clearance of middle ear fluid and reduced tympanic membrane mobility as well as transient dysfunctions of the central auditory pathways such as those occurring in perinatal asphyxia [27] may interfere with these tests and give rise to false-positive outcome of hearing screening [28]. By contrast, BAEPs are less subjected to false positive results [4] but have the disadvantage of long recording time and of requiring a specialist interpretation.

The demonstration of lack of parental anxiety after a failure of neonatal hearing screening, is a key factor to be considered when evaluating the costs and benefits of tests for universal neonatal hearing screening.

DECLARATION OF INTEREST

The authors report no declarations of interest.

REFERENCES

[1] American Academy of Pediatrics task force on newborn and infant hearing. Newborn and infant hearing loss: detection and intervention. Pediatrics 1999;133(2):527-529.

[2] Joint Committee on Infant Hearing. Year 2000 position statement: principles and guidelines for hearing detection and intervention programs. Am J Audiol 2000;9:9–29.

[3] Kemp DT. Stimulated acoustic emissions from within the human auditory system. J Acoust Soc Am 1978;64:1386–1391.

[4] Suppiej A, Rizzardi E, Zanardo V, et al. Reliability of hearing screening in high-risk neonates: comparative study of otoacoustic emission, automated and conventional auditory brainstem response. Clin Neurophysiol 2007;118:869-876.

[5] Iwsaki S, Hayashi Y, Seki A, et al. A model of two-stage newborn hearing screening with automated auditory brainstem response. Int J Pediatr Otorhinolaryngol 2003;67:99–104.

[6] Tymstra T. False positive results in screening tests: experiences of parents of children screened for congenital hypothyroidism. Fam Pract 1986;3(2):92-96.

[7] Rothenberg MD, Sills EM. Introgenesis: the PKU anxiety syndrome. Pediatr Pulmonol 1989;6:28-42.

[8] Marteau TM, Kidd J, Michie S, et al. Anxiety, knowledge and satisfaction in women receiving false positive results on routine prenatal screening: a randomized controlled trial. J Psychosom Obstet Gynaecol 1993;14:185-196.

[9] Tluczek A, Mischler EH, Bowers B, et al. Psychological impact of false-positive results when screening for cystic fibrosis. Pediatr Pulmonol Suppl 1991;7:29-37.

[10] McCormick MC, Shapiro S, Starfield B. Factors associated with maternal opinion of infant development: clues to the vulnerable child? Pediatrics 1982;69:537-543.

[11] Tluczek A, Mischler EH, Farrell PM, et al. Parents' knowledge of neonatal screening and response to false positive cystic fibrosis testing. J Dev Behav Pediatr 1992;13:181-186.

[12] Green M, Solnit AJ. Reactions to the threatened loss of a child: a vulnerable child syndrome. Pediatrics 1964;34(1):58-66.

[13] deUzcategiu CA, Yoshinga-Itano C. Parents' reactions to newborn hearing screening. Audiology Today 1997;24-27.

[14] Weichbold V, Welzl-Mueller K. Universelles Neugeborenen-Hörscreening – Einstellung und Ängste der Mütter. Phoniatrie und Padaudiologie 2000;48:606-612.

[15] Van der Ploeg CPB, Lanting CI, Kauffman-de Boer MA, et al. Examination of long-lasting parental concern after false-positive results of neonatal hearing screening. Arch Dis Child 2008;93:508-511.

[16] Watkin PM, Baldwin M, Dixon R, et al. Maternal anxiety and attitudes to universal neonatal hearing screening. Br J Audiol 1998;32(1):27–37.

[17] Clemens, CJ, Davis SA, Baileyet AR. The false-positive in universal newborn hearing screening. Pediatrics 2000;106(1):E7.

[18] Weichbold V, Welzl-Mueller K. Maternal concern about positive test results in universal newborn hearing screening. Pediatrics 2001;108(5):1111-1116.

[19] Joint Committee on Infant Hearing. Position Statement 2007: Principles and Guidelines for Early Hearing Detection and Intervention Programs Pediatrics 2007;120:898-921.

[20] Zanardo V, Freato F, Cereda C. Level of anxiety in parents of high-risk premature twins. Acta Genet Med Gemellol (Roma) 1998,47(1):13-18.

[21] de Vries LS, Eken P, Dubowitz LMS. The spectrum of leukomalacia using cranial ultrasound. Behav Brain Res 1992;49:1-6.

[22] Fielder A, Quinn GW, Taylor D, Hoyt CS, editors. Pediatric ophthalmology and strabismus. New York: Saunders; 2004.

[23] American College of Obstetricians and Gynecologists ACOG. Neonatal encephalopathy and cerebral palsy: defining the pathogenesis and pathophysiology. 1st ed. Washington, DC 2003.

[24] Spielberger CD. Manual for the Stait-Trait Anxiety Inventory (Form Y). Palo Alto, CA: Consulting Psychologists Press; 1983.

[25] Pedrabissi L, Santinello M. Verifica della validità dello STAI forma Y di Spielberger. Bollettino di Psicologia applicata 1989;191:11-14.

[26] Zanardo V and Freato F. Home oxygen therapy in infants with bronchopulmonary dysplasia: assessment of parental anxiety. Early Human Development 2001;65:39– 46.

[27] Jiang ZD. Maturation of peripheral and brainstem auditory function in the first year following perinatal asphyxia: a longitudinal study. J Speech Hear Res 1998;41(1):83–93.

[28] Valkama AM, Tolonen EU, Kerttula Lia, et al. Brainstem size and function at term age in relation to later neurosensory disability in high-risk, preterm infants. Acta Paediatr 2001;90:909–915.

Table I Clinical data of NICU infants included in the study

Clinical data	Cases (%)
Prematurity	27/31(87)
Hyperbilirubinemia	15/31(48)
PVL	2/31 (6)
BPD	10/31(32)
Anaemia	6/31 (19)
Apnoeas	4/31 (13)
Infections	5/31 (16)
Hypoglycaemia	6/31 (19)
ROP	3/31 (9)
Birth asphyxia	3/31 (9)
PDA	8/31 (26)

Legend: Prematurity (birth at gestational age < 38 weeks), PVL: periventricular leucomalacia (de Vries et al.,[21]) BPD: broncopulmonary dysplasia (oxygen dependence at 36 weeks corrected gestational age), ROP: retinopathy of prematurity (Fielder et al.,[22]), birth asphyxia[23], PDA: patent ductus arteriosus (needing medical or surgical treatment).

Table II Results of STAI-Y 1 and 2 T scores (Mean ± Standard Deviation) in the three study groups: NICU "Fail", Well-baby Nursery "Fail", NICU "Pass".

	NICU "Fail"	Nursery "Fail"	NICU "Pass"
Mother's STAI-Y 1	45.5±7.0	46.3±8.2	43.0±6.2
Mother's STAI-Y 2	45.0±8.1	43.6±8.2	44.1±9.6

Father's STAI -Y 1	46.0±6.8	45.7±7.0	47.1±7.3
Father's STAI -Y 2	45.5±5.3	44.9±7.1	45.7±7.3