Development of an effective public health screening program to assess hearing disabilities among newborns in Shanghai: a prospective cohort study

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Running head: Public health screening of newborn hearing

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Summary

Background An effective, systematic program of screening, diagnosis, and intervention against hearing loss in infants could help them avoid developmental impediments and could help society stem preventable health-care burdens. We therefore assessed the feasibility and outcomes of a government administered and financed, public health universal newborn hearing screening program (UNHSP) for neonates born in Shanghai.

Methods From March 2002 to June 2007, we conducted a two-stage hearing loss screening program for neonates born at all 105 delivery hospitals in Shanghai. Institutional participants in the program followed standardized testing criteria and procedures. The first-stage screening occurred in hospital third day post birth; positive infants underwent a second-stage outpatient screening on day 42. Positive infants were examined at clinical diagnosis centers and interventions were conducted at rehabilitation centers. In 2003, a random sample of parents were interviewed on the program and their level of stress over learning their child screened positive.

Findings Overall, our program screened 72·98% of eligible infants and provided effective interventions within 6 months for 86·31% to those with hearing loss. During the five-year study, first-stage screenings assessed 90·86% of 616 780 eligible infants and found 12·16% positive. Day 42 screenings had a 65·68% participation rate and a positive rate of 14·75%. Of these, 0·146%, were deemed permanently deaf within 3

months of birth. Parental satisfaction measures exceeded 90%.

Interpretation A government-sponsored public health program to screen, diagnose, treat, and intervene all permanently deaf newborns can be effectively implemented and can achieve outcomes that surpass comparable clinical initiatives.

Introduction

Worldwide, significant bilateral hearing loss is found in between 1 and 3 of every 1 000 newborns in well-baby populations and in 2% to 4% of infants in neonate intensive care units.[1, 2, 3] Failure to detect hearing loss in a timely manner, and to intervene as possible, can lead to a disabilities in linguistic and language functioning and cognitive development and to difficulties in social functioning. Research indicates that diagnosing hearing loss in newborns within 3 months of birth and implementing appropriate treatment and interventions within 6 months of birth normalizes cognitive and language development, therefore allowing hearing-loss infants to develop at a pace similar to that of infants without hearing loss. Such studies also show that the long-term quality of life for intervened infants surpasses what would have been possible had treatment been delayed or absent.[4, 5]

Research also shows more traditional detection efforts are inadequate: Only about 50% of neonates with congenital hearing loss are detected using the high-risk registry that screens for, among other items, a family history of deafness. In addition, physician examinations and parental observations only occasionally succeed in identifying congenital hearing loss in infants under the age of 1. To improve the early detection of infant hearing loss, in the past 15 years many countries and cities have instituted universal newborn hearing screening programs (UNHSPs) or have set up a series of screening measures.[1, 4, 6, 7, 8]

In the United States and elsewhere,[9] UNHSPs have spread gradually,[10, 11, 12] spurred by the endorsements of a 1993 National Institutes of Health Consensus Development Conference and, in 1994, by the American Academy of Pediatrics (AAP). The AAP's findings set a goal of universal detection of hearing loss in infants before 3 months of age coupled with appropriate intervention no later than 6 months of age. Both groups advocated hearing loss screening for all newborns before hospital discharge. [1, 4, 13, 14]

In 2003, an analysis of UNHSPs in the United States showed 58% employed a two-stage process—initial inpatient screening in birth hospital followed by outpatient screening before 3 months of age—and achieved a first-stage screening rate of 70% among eligible newborns. After a second-stage screening, 56% of infants were referred for diagnosis by 3 months of age and 53% of the infants diagnosed with hearing loss received an appropriate intervention by 6 months of age.[10]

Most clinical UNHSPs in developed countries use Otoacoustic Emission (OAE) screening protocols, which include Transient Evoked Otoacoustic Emission (TEOAE) and Distortion Products Otoacoustic Emission (DPOAE), and Automated Auditory Brainstem Response (AABR). [15, 16, 17] Approximately 53% of U.S. screening programs use TEOAE and 67% use AABR, with some employing both methods.[10] In general, these programs start with an initial screening test 24 to 48 hours following birth and either undertake a second inpatient or outpatient screening for newborns who test

positive in the first stage or refer newborns with positive results directly to a diagnosis center. [18, 19, 20, 21]

According to an epidemiological survey undertaken in Shanghai, between 600 and 700 of the 78 762 infants born in 1999 had some congenital defect. Furthermore, the rate of disability for children under 7 years of age was 0·968% with 16% of that group suffering from hearing loss.[22]

In 2000 Shanghai's government initiated a strategy for developing effective monitoring and intervention techniques that might limit or prevent disabilities in newborns. The strategy concentrates on three areas of public health importance: genetic testing and counseling before pregnancy, prenatal screening for congenital disabilities (such as Down's syndrome), and newborn screening for hearing loss. To address the strategy's hearing loss screening concentration, we developed a public health universal hearing loss screening procedure that was systematically designed to ensure cross-program consistency in all key aspects: instrumentation, diagnosis and screening procedures, interventions, administration and organization, personnel training, and quality control of methods and outcomes. Overall, we sought to ensure our program would meet the needs of Shanghai's population and meet—or exceed—world standards.

Methods

Network infrastructure

All 105 of Shanghai's delivery hospitals, both obstetrics and gynecology hospitals and obstetrics and gynecology departments in general hospitals, participated in the study. Diagnostic centers were set up in three institutions—the Shanghai Children's Medicine Center and Xinhua Hospital, both affiliated with Jiaotong University; and Fudan Pediatric Hospital, affiliated with Fudan University—and rehabilitation centers were organized at Xinhua Hospital, the EENT Hospital at Fudan University, and the Shanghai Municipal Federation of Disabled People, a rehabilitation center that assists those with disabilities. The 19 district-level women and children healthcare networks assisted in managing this stage of the process. Administration was centralized at the Shanghai Institute for Women and Children Healthcare, an institute organized and supported by the Shanghai Municipal Health Bureau.

In order to standardize screening results, in 2001 we purchased 130 units of the GSI 60 TEOAE system (Grasen-Stadler GSI 60, Grasen-Stadler, Inc, A Welch Allyn Co., Milford, NJ, USA). This instrument was selected after an analysis of its technical performance and cost, a review that used reports on its ease of use and clinical performance from experts in hearing screening systems. We adopted the World Health Organization's criteria for hearing screening (Table 1). Regular training sessions were arranged for the hospital personnel who would be conducting the screenings; participants were granted work permits upon passing a proficiency examination.

Nominal fees of 20RMB (2·50 US$) for one ear and 40RMB (5·00 US$) for both ears were set. Standardized processes were established for information collection and quality control.

Pilot studies

We carried out a one-year pilot study (September 2000 to August 2001) to determine participation rates for screenings on an outpatient basis alone (day 42), and for inpatient screenings conducted on either the first, second, or third day following birth. In addition, we conducted an internal comparison of the inpatient results, including images of the outer surfaces of the infants' tympanums and results from tests using high frequency acoustic impedance technology, to determine the status of eardrum effusion in neonates assessed on each of the three days. A total of 5 000 infants were screened in the pilot study: 2 000 during outpatient checkups at birth hospitals on day 42 and 1 000 on each of the first, second, and third days following birth prior to hospital discharge.

Based on the results of the pilot studies, we produced two documents: *"The Strategy of Launching Universal Newborn Hearing Screening Program in Shanghai"* and *"The Plan for Universal Newborn Hearing Screening, Diagnosis and Intervention in Shanghai."* These documents detailed the objectives, methods, and requirements necessary for an effective screening program, the screening process (Figure 1) and the diagnosis and intervention processes (Figure 2) such a program should follow, as well

as the quality controls, administrative responsibilities, and costs—to both families and program. The documents and their recommendations were adopted by the Shanghai Municipal Health Bureau in early 2002 and used as the blueprint for implementing our city-wide feasibility study of a UNHSP that would address the public health needs of Shanghai infants who are born with hearing loss. To emphasize its powerful support for our large feasibility study of the hearing loss screening program, the Shanghai Municipal Health Bureau officially launched the program on March 3, 2002, in conjunction with World Ears Day.

Screening feasibility study

From March 2002 through June 2007, 560 412 infants born in any of the study's 105 delivery hospitals were screened for hearing loss. Infants receiving positive reports received a second screening on day 42 following birth, conducted on an outpatient basis at an infant's birth hospital. All screenings were performed by hospital personnel trained in the use of the equipment. Infants receiving a second positive report were referred to one of the three diagnostic centers in the study where they were given an otoscopic exam, checked for the presence of tympanic effusion, and given DPOAE, AABR, and hearing behavior assessments (Figure 2).

Interventions included the use of audiphones within 6 months of diagnosis for infants with slight to moderate hearing loss as well as for some with severe hearing loss.

Interventions for infants with severe and extremely severe hearing loss consisted of the surgical implantation of artificial cochlear devices within 1 year of birth. Implantations were performed at two hospitals; all infants who needed implants received them. In addition, all intervened infants received aural and language training at one of the three rehabilitation centers participating in the study.

Sociological measures

In 2003 at 6 of the participating hospitals, 400 parents, either the father or the mother, of infants receiving positive results in the first-stage screening were randomly selected for face-to-face interviews. Interview questions were designed to assess parental satisfaction with the screening process. In addition, 110 of the 400 parents responded to questions that would help assess the level of anxiety or stress they felt upon learning their infants had initially screened positive for hearing loss.

Results

A comparison of data from the pilot study indicated first-stage inpatient screenings would be most effective, that is, less likely to result in false positive reports, if conducted on the 3rd day following birth. Imaging of the outer surface of the tympanum and acoustic checks using high frequency impedance detection technology showed tympanic effusion rates decreased dramatically in the three-day period following birth.

According to our data, an effusion rate of up to 70% was found in the 1 000 infants screened on the first day post birth. This rate dropped to 30% in infants screened the second day and approached zero in the 1 000 infants screened on the third day post birth. Since the presence of amniotic fluid interfered with acoustic transmissions, we determined that TEOAE screenings would yield the fewest false positive results if conducted on the third day following birth.

Our preliminary assessment also supported a two-stage screening process begun on the third day following birth and repeated if needed on day 42. We found that initial inpatient screening achieved high participation, but also produced a high number of false positives. Families receiving a positive diagnosis were motivated to attend the second screening: The majority of Chinese families have one child, so learning that the child may be hearing impaired caused parents great anxiety. The second screening, with its promise of further defining, perhaps even nullifying, the initial diagnosis, spurred compliance. In addition, by ensuring diagnosis by 3 months of age and intervention by 6 months, families were encouraged to pursue the timely potential benefits of participation in the program.

By lessening the number of false positives, the two-stage screening also greatly reduced the number of infants referred for diagnosis. If the 13·14% of infants who screened positive in the initial stage had all been referred for diagnosis, we estimate we would have needed 12 diagnosis centers to handle the referral load. With the two-stage

testing, referrals totaled fewer than 2 000 per year, a number that could be handled by three centers efficiently and well.

During the study period, 616 780 infants were born in delivery hospitals in Shanghai. Of that group of eligible infants, 90·86% (560 412) received an initial hearing screening test on the 3rd day after birth, with 12·16% (68 152) testing positive for hearing loss. A second-stage screening, conducted on an outpatient basis 42 days following birth, had a participation rate of 65·68% (44 763 infants). Positive results were reported in 14·75% (6 601) of the infants. Overall, the two-stage screening process achieved a 72·98% screening rate among eligible infants (Figure 3, Table 4).

An accounting of the 34·32% (23 389) of neonates who failed to participate in the second screening despite receiving positive results in the first screening revealed 7·30% (4 972) were directly taken to diagnosis centers after their first screenings, 8·08% (5 506) were lost to contact, and 18·94% (12 911) were infants of temporary workers who had moved back their original residential provinces following their child's birth. (Figure 3)

To help understand the success of our high rate of participation in both stages of screening, it is useful to know that annually 51% of the 100 000 infants born in Shanghai are children of parents who have moved to the city to take temporary work. These parents usually return to their home province after childbirth. If they then return

to Shanghai after recuperating from delivery, their newborns are generally left with the grandparents in the home province. Ninety percent of the infants born to these temporary workers, known as the “floating population,” are not taken to their birth hospital for a return visit on day 42.

Among Shanghai residents, failure to return for a second screening on day 42 (15% of the remaining 49% of infants and families who did not follow-up), was attributed to economic concern, a lack of faith in the process, afraid of getting in trouble because of a negative diagnosis, and the like.

Of the infants referred to diagnostic centers, 1·46 per 1 000 of those from the first-stage screening (a total of 818) received a diagnosis of permanent deafness within 3 months of birth. The majority of diagnoses showed slight to moderate hearing loss in referred infants (33·98%, or 278 infants, and 55·37%, or 453 infants, respectively) with only a 3·42% (28 infants) found to have extremely severe hearing loss. Bilateral deafness was diagnosed in 82·40% of the referred newborns (674) and single-ear deafness in 17·60% (144) of the infants (Table 1).

Among the permanently deaf infants receiving interventions, 86·31% (706) had effective interventions within 6 months. (Table 4). All bilateral audiphones, internal ear moulds, and batteries were provided free of charge by Shanghai’s Municipal Federation of Disabled People. Cochlear implants were provided free of charge by two

Shanghai-based nongovernmental organizations: the Inpatient Health Insurance Foundation for Children and the Charity Foundation.

Interviews with 400 randomly selected parents revealed 90% were satisfied with the screening process and their knowledge of it (Table 2). At the same time, the cohort of 110 parents interviewed for their level of anxiety indicated they had suffered a high degree of mental stress upon learning their child had tested positive for hearing loss (Table 3).

Discussion

Our study showed a government-sponsored public health universal newborn hearing screening program can be effectively coordinated and carried out in a large, complex city such as Shanghai. To our knowledge, our effort has developed the first successful model of a public health UNHSP.

The integrated design of our study included an initial inpatient screening, a second screening on an outpatient basis, and coordinated diagnosis and intervention services (including free audiphones, surgical implantation of cochlear devices, and aural and language training). It resulted in high rates of participation for both the initial and the second screenings; reduced false positive rates at second screenings; widespread access to participating centers, and prompt and effective interventions that placed no economic burden on the parents of the infants.

In addition, we believe our study shows that implementing a well-organized program that adheres to uniform screening, training, and intervention standards can achieve high compliance rates and high rates of satisfaction from families involved in the program. Our outcomes—such as percentage participation at first and second stage screenings and percentage of infants who receive diagnoses and interventions in timely manner as suggested by international guidelines—exceed those of clinical programs conducted in many developed countries. Furthermore, the success of our program spurred the Central Government of China in 2005 to add the program to its list of legislated screening programs. Our UNHSP model has since been launched in 27 provinces, municipalities, and autonomous regions (of a total of 31 in mainland China).

The study's outcomes are not without shortfalls and limitations, however. Why did 9% of eligible infants fail to take part in the inpatient screening? Although we had a high rate of participation in the 3rd day screening, we had no mechanism in place to follow-up on groups that failed to show for the second screening, such as the 26·24% who were the children of "floating population" workers who temporarily move to Shanghai from other provinces. Subsequent efforts will need to investigate whether these infants received diagnoses and, when necessary, appropriate interventions at their local hospitals. We also need to reconcile the reasons and outcomes of the 8·08% lost to contact following their initial positive screening, and we need to determine ways to improve the intervention phase so that all infants who receive diagnoses of permanent

deafness are treated within 6 months. Our study intervened in 706 of the 818 cases diagnosed; 112 who had been diagnosed with permanent deafness were left without proper treatment within the appropriate time period. In addition to addressing these questions, we hope to further analyze data from this study to determine the overall cost effectiveness of the program.

Contributors: X Sun was the main author, principal investigator, project manager, and developer of the project's systematic design. X Shen designed the study methodology and assisted in managing the project. J Lv oversaw the sociological assessment and the statistical analysis of the study. Z Xu managed the diagnostic centers and training program; and H Wu managed operations in the clinical and rehabilitation centers.

Conflict of interest statement: The authors report no conflict of interest.

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References

1. American Academy of Pediatrics. Newborn and infant hearing loss: detection and intervention. *Pediatrics* 1999; **103:** 527–30.
2. Green GE, Scott DA, McDonald JM, Woodworth GG, Sheffield VC, Smith RJH. Carrier rates in the midwestern United States for *GJB2* mutations causing inherited deafness. *JAMA* 1999; **281:** 2211–16.
3. Zhao P. Neonatal Hearing Screening. *Foreign Medical Sciences [Section of Maternal and Child Health]* 2001; **12:** 113–16.
4. American Academy of Pediatrics, Joint Committee on Infant Hearing. 1994 Position Statement, *Pediatrics* 1995; **95:** 152–56.
5. Shen X. The meaning of developing neonatal hearing screening in China. *Journal of Clinical Pediatrics* 2001; **19:** 67.
6. Kanne TJ, Schaefer L, Perkins JA. Potential pitfalls of initiating a newborn hearing screening program. *Arch Otolaryngol Head Neck Surg* 1999; **125:** 28–32.
7. Wessex Universal Neonatal Hearing Screening Trial Group. Controlled trial of universal neonatal screening for early identification of permanent childhood Hearing Impairment. *Lancet* 1998; **352:** 1957–64.
8. Grandori F. The European consensus development conference on neonatal hearing screening (Milan, May 15-16, 1998). *Arch Otolarynglo Head Neck Surg* 1999; **125:** 118.
9. White KR, Behrens TR. The Rhode Island Hearing Assessment Project:

implications for universal newborn hearing screening. *Semin Hear* 1993; **14:** 1–122.

10. White KR. The current status of EHDI programs in the United States. *Ment Retard Dev Disabil Res Rev* 2003; **9:** 79–88.
11. Morton CC, Nance WE. Newborn hearing screening—A silent revolution. *N Engl J Med* 2006; **354:** 2151–64.
12. Wu S. Current research status of children's hearing impairment. *Foreign Medical Sciences [Section of Pediatrics]* 2000; **27:** 254–57.
13. NIH consensus statement: early identification of hearing impairment in infants and young children. National Institute of Health, Rockville, MD. 1993; **11:** m1–24.
14. Centers for Disease Control and Prevention. Infants tested for hearing loss—United States, 1999–2001 *Morb Mortal Wkly Rep* 2003; **52:** 981–84.
15. Xu Z, Li J, Hu T, Sun J, Shen X. Sensitivity of distortion product otoacoustic emissions and auditory brain-stem response in neonatal hearing screening, a comparative study. *Chinese Medical Journal*, 2003; **83:** 278–80.
16. Li J, Xu Z, Tao Z. Otoacoustic emissions and auditory brainstem response in high risk neonatal hearing screening. *J Audiology and Speech Path* 2004; **12:** 5–7.
17. Shen X. Neonatal hearing screening. *Chinese Journal of Pediatrics* 2002; **40:** 56–58.
18. U.S. Preventive Services Task Force (USPSTF). Newborn Hearing Screening:

Recommendations and Rationale. *Am J Nurs* 2002; **102:** 83–89.

19. U.S. Preventive Services Tast Force (USPSTF). Newborn Hearing Screening: Recommendations and Rationale. *Am Fam Physician* 2001; **64:** 1995–99.
20. Uus K, Bamford J. Effectiveness of population-based newborn hearing screening in England: ages of interventions and profile of cases. *Pediatrics* 2006; **117:** 887–93.
21. Lin HC, Shu MT, Chang K, Bruna SM. A Universal Newborn Hearing Screening Program in Taiwan. *Int J Pediatr Otorhinolaryngol* 2002; **63:** 209–18.
22. Zhao JR, Ge HL, Zhen M. An epidemiological survey on birth deficient and the disabled morbidities of children under 7 years old in Shanghai. *Personal communication* 1999.

Tables

Table 1: Degree of hearing loss among newborns in Shanghai, 2002–2007

	Criteria	Congenital Deaf	Proportion□%□
Degree	voice frequency□500, 1000, 2000Hz□ pure-tone threshold average□PAT□		
Slight	26-40dbHL	278	33.98
Medium	41-60dbHL	453	55.37
Severe	61-90dbHL	59	7.21
Extremely severe	□90dbHL	28	3.42
Total		818	100
Single or bilateral			
Single		144	17.60
Bilateral		674	82.40
Total		818	100

Table 2: Knowledge and attitudes of parents toward hearing loss screening program

	Yes	No
1□Have you heard of the newborn hearing loss screening program?	70.0	30.0
2□Did you get information on the program from the hospital?	62.7	37.3
3□Are you willing to have your child screened?	98.2	1.8
4□Did the doctor inform you of the process and possible results of the screening?	55.5	44.5
5□Have you given permission to have your child screened?	90.0	10.0
6□Did your child receive a positive result from the first screening?	10.9	89.1
7□Do you trust this screening result?	95.5	4.5
8□Do you think your child should be further screened for hearing loss?	93.6	6.4
9□Do you think all newborns should be screened for hearing loss?	93.6	6.4

Table 3: Psychological responses of parents of child found to have hearing loss

Psychological Response	Proportion□□□	Range
Anxiety	53.30	1
Concern for child's future	40.00	2
Poor Appetite	16.70	3
Self-abased and depressed	10.00	4
Sleepless	6.60	5
Fail to concentrate when working	6.60	6
Denying child's hearing problem	6.60	7
Economic concern for treatment	6.60	8

Table 4: Number of newborns screened in program, 2002—2007

		March 3-December 2002	2003	2004	2005	2006	January-June 2007	**Total**
Live birth		67418	83817	114072	125130	144994	81449	616780
3rd day Screening	number	56191	78595	105873	117371	129738	72664	560412
	positive	7040	8771	12147	13812	16836	9546	68152
3rd day screening rate %		83.35	93.88	92.81	93.80	89.48	89.19	90.86
3rd screening positive rate %		12.53	11.16	11.47	11.77	12.98	13.14	12.16
42nd Screening	number	4500	5534	8264	9545	10651	6269	44763
	positive	468	751	1359	1444	1638	941	6601
42nd screening rate %		63.92	63.09	68.03	69.11	63.26	65.67	65.68
42nd screening positive rate %		10.40	13.57	16.44	15.13	15.38	15.01	14.75
Diagnosis in 3M		94	118	153	157	189	107	818
Diagnosis rate ‰ in 3M		1.67	1.50	1.45	1.34	1.46	1.47	1.46
Intervention in 6M		80	89	141	139	158	99	706
Intervention rate % in 6M		85.11	75.42	92.16	88.54	83.60	92.52	86.31

FIGURES

Figure 1: Administrative process for universal newborn hearing screening program in Shanghai

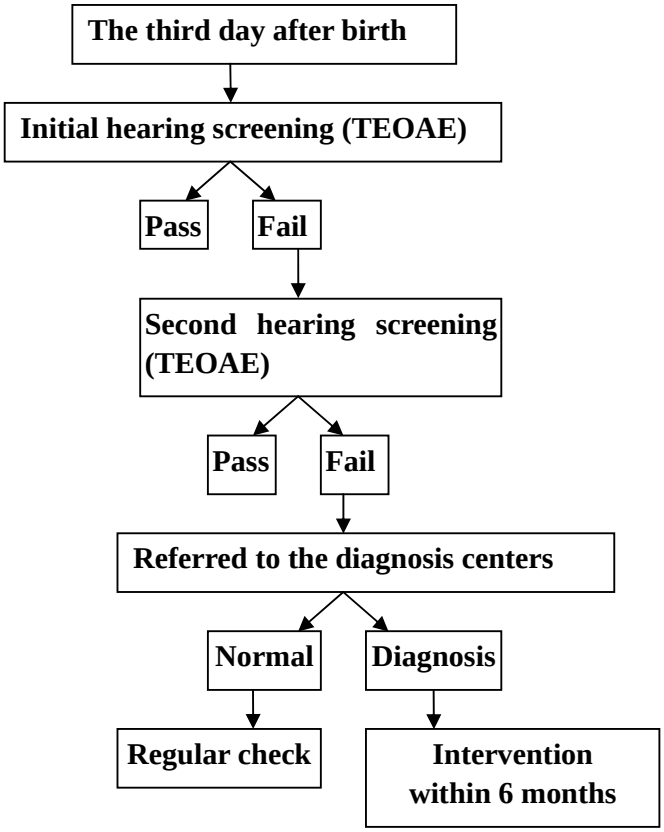


Figure 2: Process for diagnosing hearing loss in newborns

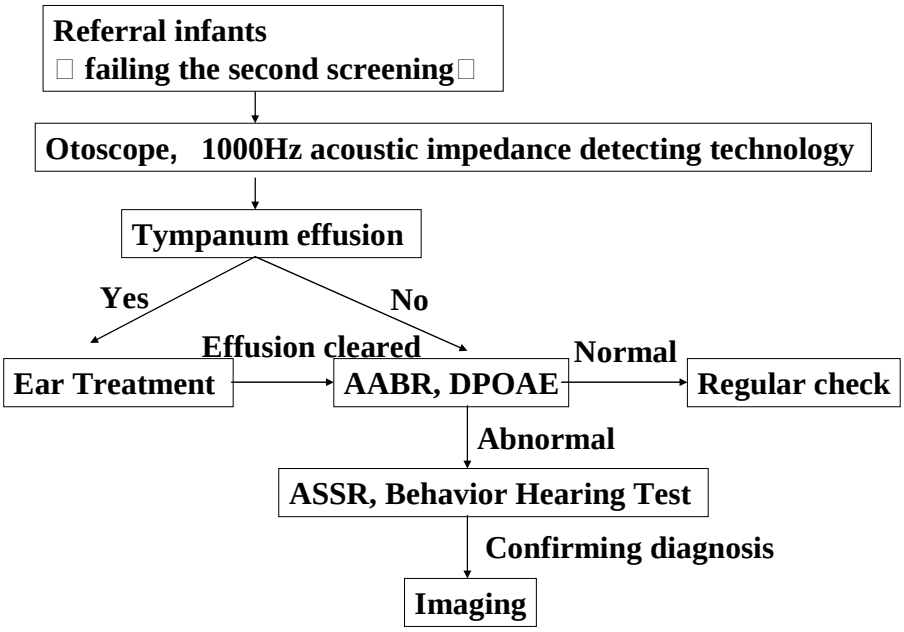
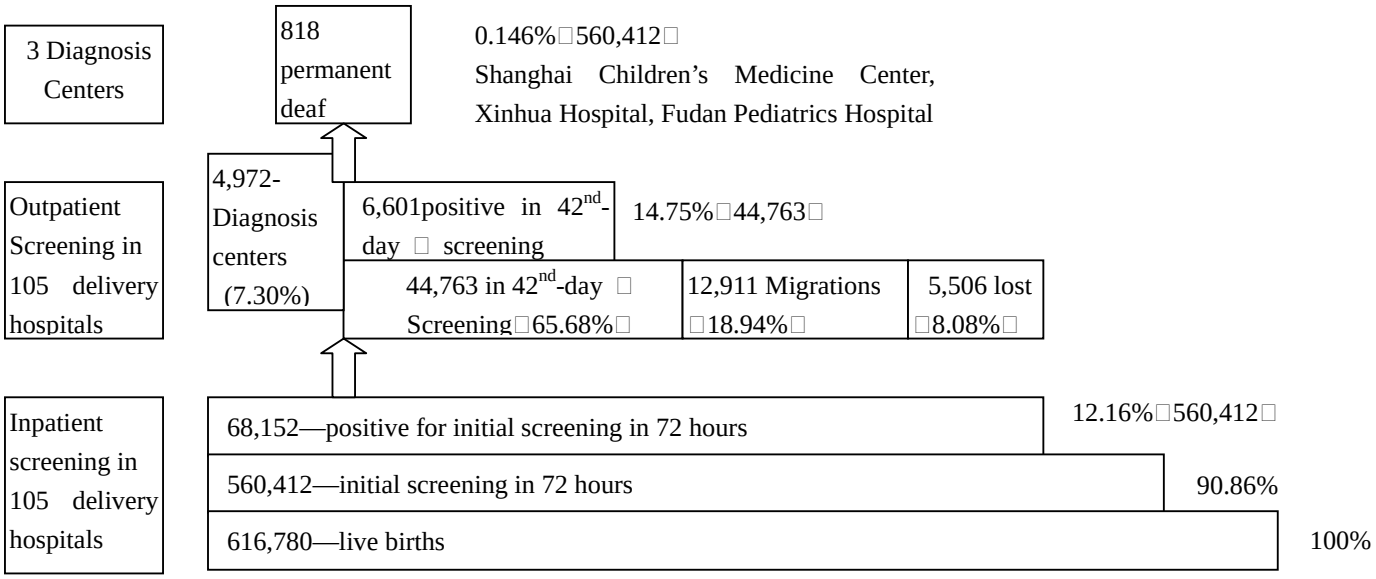


Figure 3: Statistics of universal newborn hearing screening program in Shanghai,



2002–2007