

Childhood hearing surveillance activity in Italy: preliminary recommendations

L'attività di sorveglianza audiologica pediatrica in Italia: raccomandazioni preliminari

E. ORZAN¹, F. RUTA², P. BOLZONELLO³, R. MARCHI¹, F. CESCHIN⁴, E. CICIRIELLO¹

¹ Audiology and Otolaryngology Unit, Institute for Maternal and Child Health, IRCCS “Burlo Garofolo”, Trieste, Italy; ² Family Paediatrician, Azienda per l'Assistenza Sanitaria n. 5 “Friuli Occidentale”, Italy; ³ Service of Audiology, Azienda per l'Assistenza Sanitaria n. 5 “Friuli Occidentale”, Hospital of Pordenone, Italy; ⁴ Family Paediatrician Azienda per l'Assistenza Sanitaria n. 5 “Friuli Occidentale”, Italy

SUMMARY

Following the positive outcomes of the newborn hearing screening programmes already underway in several Italian regions, it is now necessary to address the identification of childhood hearing impairments that missed the neonatal screening programme or have delayed onset. Within the framework of the Ministry of Health project CCM 2013 “Preventing Communication Disorders: a Regional Program for early Identification, Intervention and Care of Hearing Impaired Children”, a group of professionals identified three main recommendations that can be useful to improve hearing surveillance activity within the regional and state Italian Health System. The family paediatrician is recognised as having a key role in ongoing monitoring of hearing capacity and development of the growing child.

KEY WORDS: Health surveillance • Hearing loss children • Paediatrician • Paediatric primary care • Regular health visits • SWOT analysis

RIASSUNTO

Sulla base dei risultati dei programmi di screening uditivo neonatale attivi in diverse regioni italiane, si è resa necessaria una riflessione riguardo all'identificazione delle ipoacusie sfuggite al programma di screening neonatale, delle ipoacusie progressive o insorte dopo la nascita. Nell'ambito del progetto del Ministero della Salute CCM 2013 “Programma regionale di identificazione, intervento e presa in carico precoci per la prevenzione dei disturbi comunicativi nei bambini con deficit uditivo” un gruppo di esperti ha identificato tre principali raccomandazioni utili per ottimizzare l'attività di sorveglianza audiologica pediatrica nell'ambito del Servizio Sanitari Nazionale. Il pediatra di famiglia è riconosciuto come la figura chiave per il monitoraggio delle capacità uditive e dello sviluppo del bambino.

PAROLE CHIAVE: Sorveglianza sanitaria • Ipoacusia infantile • Pediatra di famiglia • Pediatria di base • Bilanci di salute • Analisi SWOT

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Introduction

In many countries and in most Italian regions hearing screening tests are routinely carried out in all newborns to detect and treat permanent hearing impairments (PHI) as early as possible. Research has shown that congenital hearing problems are usually diagnosed much sooner if newborns are routinely screened^{1,2}. Moreover, children whose PHI was diagnosed through universal newborn hearing screening (UNHS) receive earlier treatment and have better early language development than in children whose PHI was diagnosed and treated later^{3,4}. Yet the prevalence of confirmed childhood PHI increases with age and concerns have been expressed about hearing impaired children not picked up by the newborn screen⁵. There are many factors that may contribute to the increase

in cumulative prevalence of PHI with age. Some are related to the limitations of the UNHS system, i.e. cases that missed the neonatal screening test, false negative cases and cases loss to follow-up⁶. Other factors are related to the unpredictable timing of onset and the different aetiologies responsible for late-onset or progressive cases. This is, for example, the case of some genetic forms of PHI or of other PHI in “at risk” children such as, for example, after meningitis, chemotherapy or CMV infection⁷. Unfortunately, many young children may not have another hearing screening test after the newborn period, and previous investigations have shown that a large proportion of children with PHI have undiagnosed delayed-onset hearing loss⁸. The frequent timeliness of service delivery (in diagnostic audiologic evaluations, hearing aid fitting

or early intervention services) is an additional important issue for late-identified children with PHI. Walker et al. advised that children referred from the UNHS should receive earlier intervention than those identified later⁹.

In summary, literature data convey important implications for paediatric primary audiology service providers. Many critical issues on the efficacy of current childhood PHI detection programmes have been identified^{10,11}, while few healthcare organisation system proposals provide for effective management of the problem⁹. To our knowledge, there are no studies that have explored the efficacy of the childhood hearing surveillance (CHS) activity in Italy. This study aims to highlight the strengths and weaknesses of current assets in order to achieve preliminary recommendations that can minimise the loss to follow-up cases and identify as early as possible delayed-onset or progressive PHI in preschool children in Italy.

Materials and methods

A group of professionals working in tertiary care referral centres for childhood PHI was asked to complete a survey. To examine issues related to CHS activity in Italy, the survey asked participants to report at least 2 strengths, weaknesses, opportunities and threats for use in strategic planning. This phase has been conducted with the principles of a SWOT analysis. The acronym SWOT stands for Strength (S), Weaknesses (W), Opportunities (O) and Threats (T), and corresponds to what the comments of the participants have pointed out. The responses obtained were reviewed by the specialists responsible for this work's area. To generate recommendations from the SWOT analysis, a TOWS matrix was used to match the external threats and opportunities with internal weaknesses and strengths of the organisation or programme¹². The detailed description of the SWOT and TOWS matrix analysis procedure can be found elsewhere in this issue. The study and the survey was focused on this specific aim: minimise the *loss to follow-up* cases and obtain early identification of *late onset* or progressive PHI in early childhood.

Results

The study group included 24 professionals with specific roles in the prevention, diagnosis and treatment of paediatric hearing loss (especially in tertiary care referral centres where there is an active UNHS programme) (Table I). All participants completed the SWOT questionnaire. The 219 open-ended answers (S = 56, W = 58, O = 48, T = 57) were reviewed by the authors and grouped in main key points (Table II).

Strength key points analysis

Effectiveness of Family Paediatrician (FP) activity and FP regional network activity

Table I. Duties of the rehabilitation professionals (n = 24) involved in the survey.

| Professional duty | n |
|---|----|
| Otolaryngologist/Physician in Audiology | 10 |
| Audiologist/Hearing Acoustician | 4 |
| Physician in Neonatology | 1 |
| Speech and language therapist | 2 |
| Psychologist | 1 |
| Cochlear implant technical specialist | 1 |
| Parent/Association | 2 |
| Primary Care Paediatrician | 3 |

In this category answers regarding a complete, uniform and shared education and activity of FP (n = 14) were included: guarantee that the UNHS is carried out and correctly reported (n = 7), correct administration of parental questionnaires aimed to support the observation of child's auditory capacity (n = 3), organisation of a FP network covering the entire territory to ensure homogeneity of the service offered (n = 5). In some areas, FP education

Table II. Main key points extrapolated from the questionnaires.

Table IIa. *Strengths.*

| Strength key points | N (%) |
|---|----------|
| Effectiveness of FP activity and FP regional network activity | 29 (52%) |
| Strong organisation and procedures | 22 (39%) |
| Public awareness | 5 (9%) |

Table IIb. *Weaknesses.*

| Weakness key points | N (%) |
|---------------------------------|----------|
| Inefficient system organisation | 36 (62%) |
| Inefficiency of FP CHS activity | 13 (22%) |
| Inefficient communication | 9 (16%) |

Table IIc. *Opportunities.*

| Opportunity key points | N (%) |
|---|----------|
| Development of innovative and efficient healthcare organisation | 18 (38%) |
| CHS awareness and knowledge | 15 (31%) |
| Up-to-date technology | 14 (29%) |
| Other | 1 (2%) |

Table IId. *Threats.*

| Threats key points | N (%) |
|---|-----------|
| Cultural, ethnical and territorial differences | 25 (59.5) |
| Scarce equity of service and non-homogeneous training | 30 (53%) |
| Multiculturalism | 9 (16%) |
| Conflict of interest | 10 (17%) |
| Lack of funds | 7 (12%) |
| Other | 1 (2%) |

is proposed at least every two years. FPs regularly visit infants and children and become the key figure that can share parental concerns related to child's hearing and language development. They ascertain UNHS results and subsequent delivery steps, and monitor and follow-up auditory, language and behavioural development.

Strong organisation and procedures

This category includes all the themes concerning the organisation and procedures for screening and surveillance governance: effective and systematic collaboration among I, II and III level centres (i.e. integration between hospitals and FP, facilitation of procedures, easy access to useful contacts and to professionals) (n = 7); correct and efficient organisation of data processing within the regional program that also includes retrieving missed appointments and communication of results to the family (n = 7); the organisation of dedicated administrative reference points for easy access to II and III level centres (n = 8).

Public awareness

This category includes the themes regarding the presence of family associations that are aware of and support CHS activity. By acting at the territorial level, associations can further support the tertiary referral centre and the FP in providing the correct information to families (n = 5).

Weakness key points analysis

Inefficient system organisation

In this category, we entered all the answers that reported various issues related to an ineffective organisation of CHS activity. In particular, the network activity can be compromised by scarce communication between the tertiary referral centre and the FP: i.e. inadequate sharing of audiologic evaluations outcomes and scarce awareness regarding CHS within paediatrician's community (n = 11). Difficulties in recalling patients who do not show up at to FP visits or who missed UNHS and second level appointments (n = 8). Other issues relate to difficulties in aligning CHS activity. This is the case when a regional FP network is missing, when there are no guidelines regarding the CHS role of the FP or when there is no FP coverage in the territory (n = 6). Finally, this category includes lack of dedicated administrative offices or excessive workload of professionals (n = 6).

Inefficiency of FP CHS activity

This theme includes the answers related to the lack of professional education or professional training in paediatric audiology addressed to the FP (n = 6) and to the presence of insufficient and inefficient evaluation tools for CHS (n = 7).

Inefficient communication

This theme includes the answers concerning the insufficient information given to families about the importance

of CHS in childhood (n = 9), and in particular information on late onset or progressive hearing loss and information on the meaning of a positive UNHS result.

Opportunities key points analysis

Development of innovative and efficient healthcare organisation

This category includes all the remarks that can help to ameliorate the organisational set up: interdisciplinary work-up for a complete evaluation of the child (n = 4); shared education and training for professionals and parents (n = 4); stronger involvement of FP in CHS programmes (n = 4); use UNHS models as a source for CHS programme planning (n = 2); obtain a FP assignment for the newborn before the hospital discharge or link screening results to vaccination data (n = 2).

CHS awareness and knowledge

This category includes the themes regarding CHS awareness within families, institutions and associations (n = 15). There is, for example, the possibility to publicise information by means of posters or brochures displayed in FP waiting rooms, schools, vaccinations centres, etc. Other opportunities can emerge from successful experiences of the UNHS.

Up-to-date technology

This theme includes the answers regarding utilising technological advances that can ameliorate databases and software in order to ease collection/reaching/sharing of data within professionals and clinics (n = 8). This theme also includes the opportunity to develop and design new audiologic tools to FP CHS activity (n = 6).

Other answers in this field of the key points analysis are unspecified (n = 1).

Threats key points analysis

Scarce equity of service and non-homogeneous training

This category includes answers concerning the lack of shared education and training of FP, with inadequate knowledge about epidemiology of non-congenital PHI, about auditory deprivation effects and the role of the FP in CHS activities (n = 11). The lack of shared education can predispose to communication difficulties between stakeholders (n = 6).

Some answers also pointed out the lack of national uniformity (legislations, medical report models...) (n = 7) and the lack of general awareness on the problem (n = 2).

Multiculturalism

In this category, participants raised concerns regarding the interaction with families belonging to different cultures: cultural and linguistic barriers can sometimes be a serious obstacle for acceptance of CHS activities (n = 9).

Conflict of interest

Since the FP receives economic reimbursement when performing the Boel Test during regular visits, in Italy some concerns have raised the risk that this observational tool will continue to be used even if ascertained as having scarce sensibility and specificity (n = 8). Other conflicts of interest have been identified in the possible overlap in diagnostic activity between II and III level services, or between different III level centres, especially in regions with a higher number of III level centres (n = 2).

Lack of funds

This category includes the answers concerning the lack of funds and projects aimed at paediatric audiology and CHS research, with subsequent reduced staff and scarcity of funds for research projects (n = 7). Other answers in this field of the key-points analysis are unspecified (n = 2).

Discussion

The JCIH outlined risk factors registries that can be used in association with parental and professional concerns to identify infants and small children with HI missed by UNHS (“infants who pass the neonatal screening, but have a risk factor should have at least 1 diagnostic audiology assessment by 24-30 months of age”) ⁷. In practice, several critical matters emerged regarding the efficacy of these registries and considerable debate exists over the most effective method for PHI detection post-UNHS ^{13 14}. The following issues were preliminarily considered by the working group:

- within 7-9 years of age, the prevalence of PHI is about twice that expected from the results of the UNHS ¹⁰;
- some children who develop PHI have no risk factors and have no discoverable cause ¹⁵;
- there is a high lost contact rate in CHS and children with only a single risk factor are more likely to not attend a CHS appointment ¹⁴;

- sometimes it is difficult to establish a good alliance with the family. This situation can lead to late recognition of risk factors and poor adherence to CHS appointments;
- delayed onset of PHI is linked to a limited ability to achieve early diagnosis and early cochlear implantation of a significant number of children ¹⁶;
- a single additional hearing screening at primary school entrance (5-7 years of age) could not detect new cases early enough, possibly leading to a lack of timeliness of service delivery ⁸;
- there is no consensus about the most suitable methodology to screen auditory capacity in the preschool age ¹⁴;
- the FP represents a key figure for childhood health CHS in the Italian National Health System. The FP visits regularly carried out at 1, 3, 6, 8, 12, 18, 24 and 36 months of the growing child, place the FP in an optimal position to ascertain the UNHS results and subsequent service delivery steps, to monitor and follow-up auditory, language and behavioral development and to screen the child’s hearing capacity. In some areas and regions, FP education is already established and regularly proposed ¹⁷.

Following the SWOT analysis, a TOWS matrix was created, which compares Strengths-Opportunities, Weaknesses-Threats, Weaknesses-Opportunities. Analysing and discussing the data obtained from this research, 10 recommendations were obtained (Table III). These recommendations can represent the starting point for focusing the attention on population needs, on optimising resources and on generating positive changes. With this point of view, the strategic analysis identified three main themes or recommendations that may contribute to an efficient CHS system in Italy. These are:

1. Reassessment of the role of the FP though continuing

Table III. TOWS matrix (see text for explanation).

| | | Internal | |
|----------|-------------------|--|---|
| | | Strength (S) | Weakness (W) |
| External | Opportunities (O) | <p>SO strategy</p> <ol style="list-style-type: none"> 1. Develop an innovative organisation that exploits the good coverage of the FP on the territory, its training, a common database and the use of an objective screening tool. 2. Take advantage of the good organization of the UNHS to increase awareness of families, FP, institutions and associations on CHS. | <p>WO strategy</p> <ol style="list-style-type: none"> 1. Leverage on the current cultural awareness to organise effective public CHS activity. 2. Use technological developments to improve tools, methods and systems for effective CHS and cooperation between FP and III level centres. |
| | Threats (T) | <p>ST strategy</p> <ol style="list-style-type: none"> 1. Capitalise on well trained FPs to become regional referents for CHS activity on the territory and favour paediatric networks. 2. Endorse FP and III level centres as having an effective role and awareness to promote tools and protocols that can overcome conflicts of interest. 3. Take advantage of the most efficient existing programs to convey financial supports and national uniformity. 4. Overcome multicultural issues raising cultural awareness. | <p>WT strategy</p> <ol style="list-style-type: none"> 1. Identify ineffective FP activity in order to minimize risk of pitfalls and non-uniform service. 2. Set up a system for prompt identification of service course weaknesses and their revision. |

- education within the hearing screening national system/programme;
2. UNHS and CHS community awareness and attendance;
 3. Organisational set-up and procedural uniformity of childhood CHS activity within the Italian National Health System.

The first theme is considered essential for a successful CHS regional or national programme: permanent and effective education and training of the FP needs to be consistent in the entire territory, formally included within the UNHS programmes and operationally linked with I, II and III level activities. The recognition and establishment of the role of the FP for the CHS within early identification and intervention programmes will “naturally” replace ineffective CHS activity^{8 10 11}.

There have been some initial network experiences for pediatric audiology in Italy, promoted by Italian Paediatrician Federation (FIMP). This initiative also includes training and educational courses taught by audiologists and otolaryngologists. New cost-effective tools and procedures of audiological screening to be used in primary care are currently tested in an ongoing study at IRCCS Burlo Garofolo in Trieste¹⁹.

The second main theme can partially integrate the first. The FP is indeed in the role of educating caregivers to be alert and sensible observers of their child’s development. Moreover, the FP can increase family cooperation to attend appointments offered by the CHS programme. Several methods that can maximise the FP’s role were identified: posters that highlight and advise essential information can be displayed in clinics and FP waiting rooms; a cooperation with the National Health Service and FP/family associations can popularise leaflets and short videos that inform the community about the critical issues on auditory development and deficit; interdisciplinary events and meetings can raise awareness and publicise the opportunities and benefits of early intervention. It is the belief of the working group that all the projects aimed to strengthen CHS actions within the FP activity should be agreed between the parties, i.e. National Health System, FP organisations, family associations, and UNHS programmes.

The third main recommendation relates to the national CHS set-up, in order to make its activity more practical. This theme is also linked to the first regarding the FP’s role, and specifically the CHS audiologic screening tools and methods to be used by the FP. Current FP referral procedures comprise parental and professional concerns, questionnaires, high-risk lists and subjective observational tools, i.e. the Boel Test which, similar to the Distraction Test, observes the gaze orientation after a sound stimulus. The first three did not achieve national uniformity and the Boel test yields only a very inaccurate global estimate of

the child’s hearing capacity in the best case, with many false positive and false negative results⁵.

Few scientific studies have reported on alternative methods and tools that would be effective at the level of the FP CHS^{18 19}.

Finally, this working group endorses a public health CHS structure, in terms of an ongoing systematic collection, analysis and interpretation of data, closely integrated with UNHS programmes and with the timely dissemination of these data to professionals responsible for preventing and treating the disease^{8 10 11 20 21}.

Conclusions

Postneonatal routes of identification are today considered essential. While being aware of their current ineffectiveness, they need to be maintained and improvements investigated in order to obtain an operative policy for detection and an early management for children not identified by the UNHS program. The Italian National Health System recognises the FP as a key figure for childhood health and CHS. The FP ascertains the UNHS results and the following delivery steps. The regular visits to the FP place the FP in an optimal position to monitor auditory capacity of the growing child. New audiologic tools for FP CHS activities should be developed and designed. The FP should be included in interdisciplinary education and training. It is indeed necessary to implement a public health system in parallel to the UNHS that can more effectively empower early referral and identification of hearing impaired children not identified by newborn screening.

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