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Prevalence of Childhood Hearing Impairment of Different Severities in Urban and Rural Areas: A Nationwide Population-Based Study in Taiwan

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Prevalence of childhood hearing impairment of different severities in urban and rural areas: a nationwide population-based study in Taiwan

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Short title: Childhood hearing impairment of different severities in urban and rural areas

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Abstracts

Objective

Childhood hearing impairment (CHI) is a major developmental disability, but data at the national level are limited, especially those on different severities. We conducted a study to fill this data gap.

Design

A nationwide longitudinal study.

Setting

To provide services to disabled citizens, the Taiwanese government maintains a registry of certified cases. Using data from this registry, we estimated prevalence rates of CHI of different severities from 2004 to 2010, and made comparisons between urban (with > 50% of the population living in metropolitan regions) and rural areas.

Participants

Taiwanese citizens ≤ 17 years old.

Primary outcomes measure

To qualify for CHI disability benefits, a child must have an unaided pure-tone better ear hearing level (BEHL) at 0.5, 1, and 2 kHz with an average ≥ 55 decibels (dB), confirmed by an otolaryngologist. The severity was classified by BEHL as mild (55-69 dB), moderate (70-89 dB), and severe (≥ 90 dB).

Results

The registered cases under 17 years old decreased annually from 4075 in 2004 to 3533 in 2010, but changes in the prevalence rate were small, ranging from 7.62/10000 in 2004 to 7.91/10000 in 2006. The prevalence rates of mild CHI increased in all areas over time, but not those of moderate or severe CHI. Rural areas had higher overall prevalence rates than urban areas in all years, with rate ratios (RRs) between

1.01 and 1.09. By severity, rural areas had higher prevalence rates of mild (RRs between 1.08 and 1.25) and moderate (RRs between 1.06 and 1.21) CHI, but had lower prevalence rates of severe CHI (RRs between 0.92 and 0.99).

Conclusion

While rural areas had higher overall prevalence rates of CHI than urban areas, the RRs decreased with CHI severity. Further studies that identify factors affecting the rural-urban difference might help the prevention of CHI.

Keywords: child; disability; hearing impairment; hearing loss; prevalence; severity; Taiwan; urbanization

Strengths and limitations of this study

- Data on the same population were collected over a seven-year period, which allows the assessment of time trends.
- The study number of cases was large, over 3533 cases in 2010 alone.
- We have information on severity, which is rarely reported by large-scale studies.
- This study used administrative data, which do not cover cases who are not detected or who have never received services from the administration.
- Data on individual cases were not provided by the registry, which hindered more detailed analyses.

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INTRODUCTION

Hearing impairment (HI) is a global problem, and the World Health Organization estimated that 360 million people (including 32 million children) have disabling HI ¹. Most patients live in low- and middle-income countries, and 25% are born with or acquire HI during childhood. Compared with infants born in resource-rich countries, infants born in resource-poor countries have a nearly two-fold risk ²⁻⁴. The costs of the education support to children with better ear hearing level (BEHL) > 50 dB was estimated as \$3.9 billion ⁵. Compared to normal children, patients with childhood HI (CHI) have difficulties in language development, speech production, and cognition, which in turn affect their academic performance, vocational attainment, and socioemotional competence ^{2,6,7}.

The World Health Assembly affirmed the importance of interventions in control preventable HI ⁸ and recommended population-based epidemiological studies to determine the prevalence rate and causes of hearing impairment in all nations for targeting of preventive efforts ². The reported prevalence rates of CHI varied widely around the world ⁹⁻¹³. Most studies focus on either rural or urban populations, even though comparing the difference between the two is important. The urban–rural differences might be attributable to differences in cultural perceptions regarding the impact of HI, diagnosis, and treatment ¹⁴, but efforts are needed to investigate the differences further.

In 1980, the Taiwanese government constructed a system to certify disabled residents and to provide them with various services. The central government keeps a registry of certified cases ¹⁵, presenting a rare opportunity for studying CHI at the national level. The objectives of this study were to estimate the prevalence rates of CHI of different severities and to evaluate the differences between urban and rural areas.

METHODS

The disability registry system in Taiwan

In Taiwan, the Disabled Welfare Act was promulgated in 1980¹⁶. Accordingly, the local governments began to certify seven types of patients with disabilities, including “hearing impairment or balance disability.” When the Act was revised to become the People with Disabilities Rights Protection Act in 1997¹⁷, the “hearing impairment or balance disability” category was divided into two: HI and “balance disability”.

Individuals can make applications for certification through their local government in the residential area^{18 19}. These local governments report certified cases to the central government. The registry of cases was first maintained by the Ministry of the Interior and then by the Ministry of Health and Welfare after the re-organization of the government in 2013²⁰.

Case definition of childhood hearing impairment

When a child is suspected of having HI, parents or guardians can apply for certification. To qualify for disability benefits, a child must have an unaided pure-tone BEHL at 0.5, 1 and 2 kHz (pure-tone average, PTA) with an average ≥ 55 decibels (dB), and confirmation by an otolaryngologist accredited by the government^{18 19}. In cases with suspected malingering or difficulties in testing, auditory brainstem response is applied.

According to the Taiwan government¹⁹, the severity of HI is defined as “mild” with $PTA \geq 55$ dB BEHL and < 70 dB BEHL, “moderate” with $PTA \geq 70$ dB BEHL and < 90 dB BEHL, and “severe” with $PTA \geq 90$ dB BEHL.

Data collection

Using the nationwide registry data of HI, we conducted a cohort study which included all children (≤ 17 years old) with citizenship in Taiwan. Each year, the government publishes a Statistical Yearbook¹⁵. We obtained the data from the central government, but they are available since 2004 only. Furthermore, with the re-organization of administrative regions in 2011, one of the rural regions was merged into an urban region. While the impact of the reorganization on the classification was small, it made the population subdivisions incomparable before and after the reorganization. Therefore, we only analyzed the data until 2010. To calculate the prevalence rates, we obtained the total number of individuals in each age group from the Monthly Bulletin of Interior Statistics²¹.

According to the Directorate-General of Budget, Accounting and Statistics, we defined an “urban area” as a city or county with $> 50\%$ of the population living in metropolitan regions²². Error! Reference source not found. In Taiwan, there are 7 cities and 18 counties, of which 7 cities and 5 counties were categorized as urban areas, and the remaining 13 counties were categorized as rural areas.

Statistical analysis

We estimated the prevalence rate of CHI in a rural or urban area by dividing the number of cases by the number of individuals each year and evaluated the trend over time. According to the yearbooks¹⁵, we categorized the age into five groups (< 3 years, 3-5 years, 6-11 years, 12-14 years, and 15-17 years). We calculated the overall prevalence rates, as well as the prevalence rates by severity, and evaluated the trends over time.

To evaluate the differences between urban and rural areas, we estimated the prevalence rate ratio (RR) by dividing the prevalence rate of rural areas by that of urban areas. A 95% confidence interval (CI) was calculated for each RR to evaluate

its statistical significance.

We used the Chi-square test for trend to evaluate trends of changes in the prevalence rates over time and across age groups. To evaluate trends of changes in prevalence RRs over time and across age groups, we used linear regressions. In addition, we used ANOVA for repeated measures to evaluate trends of changes in the prevalence RRs among three different severity groups.

We conducted the analyses using SAS 9.1 (SAS Institute Inc., Cary, North Carolina, USA) and performed all statistical tests at the significance level of 0.05. The study protocol was reviewed and approved by the Institution Review Board of the Ditmanson Medical Foundation Chia-Yi Christian Hospital.

RESULTS

The trend of the overall prevalence rate by area

From 2004 to 2010, the registered cases under 17 years old decreased from 4075 to 3533, with a decreasing time trend. However, the changes in prevalence rates were small, ranging from 7.62/10000 in 2004 to 7.91/10000 in 2006, without a remarkable time trend, mainly because of the decreasing number of newborns each year. The prevalence rates in rural areas fluctuated between 7.70/10,000 and 8.18/10,000, without remarkable time trends (Table 1). The prevalence rates in urban areas also fluctuated, between 7.50/10,000 and 7.85/10,000, without remarkable time trends (Table 2). Rural areas had higher overall prevalence rates in all years, and the rural-to-urban prevalence RRs ranged from 1.01 to 1.09 (with $p < 0.05$ in 2008 and 2009), without remarkable time trends (Table 3).

The trends of prevalence rates by age

In rural areas, the prevalence rates in age groups < 3 years, 3-5 years, 6-11 years,

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12-14 years, and 15-17 years was 2.31-4.90/10000, 5.91-7.75/10000, 7.61-8.39/10000, 8.97-9.73/10000, and 9.71-12.00/10000, respectively (Table 1). In each year, prevalence rates increased with age ($p < 0.01$ for all Chi-square tests for trend). Over time, the prevalence rates increased in age groups < 3 years ($p < 0.01$, increased by 71.0% from 2004 to 2010.) and 3-5 years ($p < 0.05$, increased by 23.6%), but decreased in the age group 15-17 years ($p < 0.01$, decreased by 19.1%).

In urban areas, the prevalence rates in age groups < 3 years, 3-5 years, 6-11 years, 12-14 years, and 15-17 years was 2.24-4.01/10000, 5.90-6.82/10000, 7.37-7.84/10000, 8.18-9.25/10000, and 9.21-11.17/10000, respectively (Table 2). In each year, prevalence rates increased with age ($p < 0.01$ in all years). The prevalence rates increased in the age group < 3 years over time ($p < 0.01$, increased by 79.0%), but decreased in age groups 12-14years and 15-17 years ($p < 0.05$ for both, decreased by 8.3% and 17.6%, respectively).

Table 1. The prevalence rate (per 10,000 children) of hearing impairment in rural areas by age in Taiwan.

Year	< 3year		3-5 year		6-11 year		12-14 year		15-17 year		0-17 year	
	N	Prev. [†]	N	Prev.	N	Prev.	N	Prev.	N	Prev.	N	Prev.
2004	50	2.31	159	6.27	438	8.05	256	9.13	343	12.00	1246	7.88
2005	49	2.43	150	6.16	403	7.61	270	9.73	315	10.87	1187	7.70
2006	70	3.68	154	6.99	420	8.04	259	9.31	301	10.68	1204	8.06
2007	89	4.90	122	5.91	422	8.39	251	9.05	277	9.77	1161	8.00
2008	68	3.90	143	7.43	389	8.10	248	8.99	284	10.10	1132	8.06
2009	68	4.06	132	7.21	378	8.34	255	9.23	282	9.98	1115	8.18
2010	61	3.95	135	7.75	353	8.07	234	8.97	274	9.71	1057	8.07

[†]Prev.: the prevalence, estimated by dividing the number of cases by the population in each age group in each year.

Table 2. The prevalence rate (per 10,000 children) of hearing impairment in urban areas by age in Taiwan.

Year	< 3year		3-5 year		6-11 year		12-14 year		15-17 year		0-17 year	
	N	Prev. [†]	N	Prev.	N	Prev.	N	Prev.	N	Prev.	N	Prev.
2004	104	2.24	364	6.14	989	7.37	621	8.96	751	11.17	2829	7.51
2005	111	2.52	334	5.90	1013	7.71	610	8.88	767	11.05	2835	7.66
2006	140	3.26	326	6.38	998	7.65	639	9.25	734	10.80	2837	7.85
2007	138	3.24	325	6.69	957	7.62	629	9.05	709	10.30	2758	7.77
2008	132	3.10	315	6.82	907	7.54	600	8.66	643	9.43	2597	7.50
2009	147	3.50	283	6.28	889	7.84	567	8.18	652	9.52	2538	7.50
2010	159	4.01	293	6.55	853	7.74	536	8.22	635	9.21	2476	7.53

[†]Prev.: the prevalence, estimated by dividing the number of cases by the population in each age group in each year.

Table 3. The rural-to-urban prevalence rate ratio of hearing impairment by age in Taiwan.

	< 3 year		3-5 year		6-11 year		12-14 year		15-17 year		0-17 year	
Year	Rate ratio	(95% CI) [†]	Rate ratio	(95% CI)	Rate ratio	(95% CI)	Rate ratio	(95% CI)	Rate ratio	(95% CI)	Rate ratio	(95% CI)
2004	1.03	(0.74,1.45)	1.02	(0.85,1.23)	1.09	(0.98,1.22)	1.02	(0.88,1.18)	1.07	(0.95,1.22)	1.05	(0.98,1.12)
2005	0.96	(0.69,1.35)	1.05	(0.86,1.27)	0.99	(0.88,1.11)	1.10	(0.95,1.26)	0.98	(0.86,1.12)	1.01	(0.94,1.08)
2006	1.13	(0.85,1.50)	1.10	(0.90,1.33)	1.05	(0.94,1.18)	1.01	(0.87,1.16)	0.99	(0.87,1.13)	1.03	(0.96,1.10)
2007	1.51	(1.16,1.97)*	0.88	(0.72,1.09)	1.10	(0.98,1.23)	1.00	(0.86,1.16)	0.95	(0.83,1.09)	1.03	(0.96,1.10)
2008	1.26	(0.94,1.69)	1.09	(0.89,1.33)	1.07	(0.95,1.21)	1.04	(0.89,1.20)	1.07	(0.93,1.23)	1.08	(1.00,1.15*)
2009	1.16	(0.87,1.55)	1.15	(0.93,1.41)	1.06	(0.94,1.20)	1.13	(0.97,1.31)	1.05	(0.91,1.21)	1.09	(1.02,1.17)*
2010	0.98	(0.73,1.32)	1.18	(0.96,1.45)	1.04	(0.92,1.18)	1.09	(0.94,1.27)	1.05	(0.92,1.21)	1.07	(1.00,1.15)

[†]CI: confidence interval; **p* < 0.05.

9 The trends of prevalence rates by severity and area

10 For mild CHI, the overall prevalence rates increased over time in both rural and urban
11 areas ($p < 0.05$ for both) (Table 4). For moderate CHI, overall prevalence rates decreased
12 over time in urban areas ($p < 0.01$), but no remarkable trends were observed in rural areas.
13 For severe CHI, the changes in overall prevalence rates were small in both rural and urban
14 areas and without any remarkable time trends.

15 Rural areas had higher overall prevalence rates of mild CHI in all years, and the
16 differences reached statistical significance in all years except 2005 and 2007. Rural areas
17 also had higher prevalence rates of moderate CHI in all years, but the difference reached
18 statistical significance in 2008 only. For severe CHI, prevalence rates in urban areas were
19 slightly higher in all years, but none of the differences reached statistical significance (Table
20 4). The changes in rural-to-urban RR were small in all severity groups and without any
21 remarkable time trends. Nevertheless, the mean of rural-to-urban RR in mild, moderate, and
22 severe CHI was 1.15, 1.10, and 0.96, respectively, indicating a decreasing trend ($p < 0.01$).
23 The rural-to-urban RR decreased with severity in all years except for 2007 and 2008.

Table 4. The overall rural-to-urban prevalence rate ratio of hearing impairment by severity in Taiwan.

Year	Mild				Moderate				Severe			
	Prev. [†]		Rate ratio	(95% CI [‡])	Prev.		Rate ratio	(95% CI)	Prev.		Rate ratio	(95% CI)
	Rural	Urban			Rural	Urban			Rural	Urban		
2004	2.44	2.13	1.15	(1.01,1.29)*	2.03	1.92	1.06	(0.93,1.21)	3.42	3.47	0.99	(0.89,1.09)
2005	2.49	2.28	1.09	(0.97,1.23)	1.95	1.85	1.06	(0.92,1.21)	3.26	3.53	0.92	(0.83,1.02)
2006	2.69	2.38	1.13	(1.00,1.27)*	1.97	1.84	1.07	(0.93,1.23)	3.41	3.62	0.94	(0.85,1.04)
2007	2.59	2.41	1.08	(0.95,1.22)	2.09	1.86	1.12	(0.98,1.29)	3.31	3.50	0.95	(0.85,1.05)
2008	2.64	2.32	1.14	(1.01,1.29) *	2.12	1.76	1.21	(1.05,1.39) *	3.30	3.42	0.96	(0.87,1.07)
2009	2.93	2.35	1.25	(1.10,1.40) *	1.98	1.74	1.14	(0.99,1.31)	3.27	3.41	0.96	(0.86,1.07)
2010	2.95	2.45	1.21	(1.07,1.36) *	1.76	1.64	1.07	(0.92,1.25)	3.37	3.45	0.98	(0.87,1.09)

[†]Prev.: the prevalence, estimated by dividing the number of cases by the population in each age group in each year; [‡]CI: confidence interval; **p* < 0.05.

1 DISCUSSION

2 Prevalence data on CHI between urban and rural areas from large-scale studies are
3 limited, and the reported prevalence rates have a wide range. Conducting a search with the
4 combined keywords of “hearing impairment,” “child,” “dB HL,” “rural,” “urban,” and
5 “prevalence” in the PubMed database, we identified 16 studies on the prevalence rate of
6 low-frequency CHI which defined HI by dB hearing level (HL) values and included rural
7 and/or urban participants (Table 5)^{2 9-11 13 20 23-32}. The variation in reported prevalence rates
8 may mainly be attributable to differences in case definition, age range, and case-finding
9 methods³³. Factors such as genetic makeup, health-care accessibility, and socioeconomic
10 status, may also have contributions¹¹⁻¹³. The differences make comparisons among studies
11 difficult. For example, the case definition of severe for CHI in our study was ≥ 90 dB BEHL,
12 and the 3.4/10000 prevalence rate in the rural areas in 2010 was lower than those reported by
13 a study in Saudi Arabia (3.9/10000)⁹ and a study in India (35.2/10000)¹⁰ adopting similar
14 criteria. However, the age ranges used were different, making the comparison difficult. The
15 above limitations highlight the need for standardization to enhance the quality and
16 comparability of study results. Standardization can allow direct future comparisons of studies
17 as well as establish normative baseline data to illuminate potential intervention strategies¹².

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18 **Table 5. The prevalence rate (per 10000 children) of low-frequency hearing impairment (≥ 30 dB hearing level [HL] in the better ear)**
19 **defined by dB values in different studies.**

Study (year)	Country	Case-finding method	Case number (area)	Age (year)	Case definition	Prevalence
Seely et al. (1995) ²³	West Africa	Two-stage screening	2015 (rural)	5-15	Average of 0.5, 1, and 2 kHz	
					> 40 dB HL in the better ear	297.8
					> 60 dB HL in the better ear	129.0
					> 80 dB HL in the better ear	99.3
Minja & Machemba (1996) ²⁴	Tanzania	Two-stage screening	127 (rural)	5-20	Average of 0.5, 1, and 2 kHz	
					> 40 dB HL in the better ear	0.0
			675 (urban)	5-19	Average of 0.5, 1, and 2 kHz	
					> 40 dB HL in the better ear	163.0
Morioka et al. (1996) ²⁵	China	Population registry survey	282 (rural)	7-17	Average of 0.5, 1, and 2 kHz	
					≥ 35 dB HL in the better ear	496.5
Jacob et al. (1997) ¹⁰	India	Population registry survey	284 (rural)	6-10	Average of 0.5, 1, 2, 4kHz	
					> 40 dB HL in the better ear	1091.5
					> 65 dB HL in the better ear	70.4
Kaewboonchoo et al. (1998) ¹¹	China	Population registry survey	442 (urban)	6-19	> 90 dB HL in the better ear	35.2
					Average of 0.5, 1, and 2 kHz	
					≥ 35 dB HL in the better ear	113.1
Olusanya et al. (2000) ²⁷	Nigeria	Two-stage screening	359(urban)	4.5-10.9	Average of 0.5, 1, 2, 4kHz	
					> 40 dB HL in the better ear	55.7
Czechowicz et al. (2010) ²	Peru	Population registry survey	335 (rural)	6-19	Average of 0.5, 1, 2, 4kHz	
					> 40 dB HL in the better ear	238.8
					> 55 dB HL in the better ear	119.4

						> 70 dB HL in the better ear	29.9
						Average of 0.5, 1, 2, 4kHz	
						≥ 30 dB HL in the better ear	151.3
	Schmitz et al. ^{Error! Reference source not found.} (2010) ²⁹	Nepal	Population registry survey	3646 (rural)	15-23	> 40 dB HL in the better ear	71.5
						> 60 dB HL in the better ear	38.5
						> 80 dB HL in the better ear	33.0
						Average of 0.5, 1, 2, 4kHz	
	Bagshaw et al. ^{Error! Reference source not found.} (2011) ²⁰	Nepal	Population registry survey (with a diagnosis of otitis media with effusion)	70 (rural)	4-13	> 40 dB HL in the better ear	1000.0
				51 (urban)	4-13	Average of 0.5, 1, 2, 4kHz	
						> 40 dB HL in the better ear	0.0
	Gondim et al. (2012) ³⁰	Brazil	Population registry survey	90 (urban)	4-19	Average of 1, 2, 4kHz	
						> 30 dB HL in the better ear	111.1
						Average of 1, 2, 4kHz	
	Al-Rowaily et al. (2012) ⁹	Saudi Arabia	Two-stage screening	2574(urban)	4-8	> 40 dB HL in the better ear	73.8
						> 90 dB HL in the better ear	3.9
						Averaged of 0.5, 1, 2 kHz	
						≥ 55 dB HL in the better ear	8.1
						≥ 70 dB HL in the better ear	5.1
	Our study	Taiwan	National registry (reporting)	1309068 (rural)	0-17	≥ 90 dB HL in the better ear	3.4
				3286699 (urban)		≥ 55 dB HL in the better ear	7.5
						≥ 70 dB HL in the better ear	5.1
						≥ 90 dB HL in the better ear	3.5

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21 Our major finding of a higher prevalence rate of CHI in rural areas was consistent with
22 the results of previous studies in other countries^{20 24}. In 2009 the rural-urban prevalence RR
23 was 0.96 (not statistically significant) in severe cases and 1.14 (not statistically significant)
24 in moderate cases, but it was 1.25 (statistically significant) in mild cases, making the overall
25 RR (1.09) statistically significant. In a study in Tanzania, in which 802 primary school
26 children were examined using pure tone audiometry and HI was defined as a low-frequency
27 PTA threshold of > 5 dB HL in the frequencies of 0.5, 1, and 2 kHz²⁴, the prevalence rate of
28 CHI was 1102.4/10000 among rural children, while it was only 755.6/10000 among the
29 urban children ($p < 0.05$). Similarly, in a survey in Nepal, school children with a diagnosis of
30 otitis media with effusion (aged from 4 to 13 years) underwent audiometric assessment, and
31 the prevalence rate of HI, defined as a middle-frequency PTA threshold of > 25 dB HL in
32 the frequencies of 0.5, 1, 2, and 4 kHz, was higher among rural children (2700.0/10000 vs.
33 400.0/10000, $p < 0.05$)²⁰.

34 Some studies comparing CHI between urban and rural areas reported findings that are
35 different from our observations. A study in China examined 6626 residents with an age
36 range from 1 month to 90 years using the WHO definitions of HI and found no differences
37 between urban and rural areas (19.7% vs. 15.7% reduction in dB HL, $p > 0.05$)³⁴. However,
38 the report did not have separate data on CHI specifically, and therefore it is difficult to draw
39 a conclusion on the difference in CHI. A study in Tanzania examined 854 schoolchildren
40 from one urban district and one rural district by screening audiometry (air conduction) and
41 found that the prevalence of bilateral HI was higher in the urban district (10.5% vs. 4.7%)³⁵.
42 However, they did not include sensory HI, and therefore it is difficult to compare their data
43 with our findings directly.

44 Some studies have investigated the possible etiological factors of the high prevalence
45 rate of CHI in rural populations. A study on 335 school children between 6 and 19 years of
46 age in an impoverished area of Peru identified the following risk factors for CHI: neonatal

jaundice, seizure, hospitalization, recurrent otitis media, past otorrhea, family history of HI at < 35 years, tympanic membrane abnormality, cerumen impaction, and eustachian tube dysfunction². This study proposed that untreated middle ear disease in the context of limited access to pediatric care may be a major risk factor for rural CHI. In a rural primary school in south India, hearing assessments were performed on 284 students (from 6 to 10 years old), and middle ear disease was found to be the predominant cause of CHI¹⁰. An investigation of HI in 75 Yemeni children (0.6-15 years) with chronic suppurative otitis media found that middle ear disease predominantly caused a HI of 26 to 60 dB HL³⁶. According to these findings, middle ear disease appears to be major cause of CHI in rural areas, mainly leading to HI in the range of 26-60 dB HL. In our study, we found that mild CHI (55-69 dB BEHL) was more prevalent in the rural areas in all years, with most of the rural-to-urban RRs reaching statistical significance, while the prevalence rates of CHI in the other two higher severity categories (≥ 70 dB HL) were similar between rural and urban areas. Therefore, we speculate that a higher prevalence of untreated middle ear disease in rural areas contributed, at least in part, to the rural-urban differences observed in our study.

In each year, the prevalence rates of CHI in both rural and urban areas increased with age. This finding was also noted in the Metropolitan Atlanta Developmental Disabilities Surveillance Program in the United States³⁷, which found that the prevalence rate of CHI > 40 dB HL increased steadily from 6.7/10000 among 3-year-old children to 13.8/10000 for 10-year-olds. Likewise, a study in the United Kingdom found that the prevalence rate of CHI > 40 dB HL rose from 9.1/10000 among 3 year-old children to 16.5/10000 among children 9 to 16 years old³⁸. Because HI was rarely fatal and a substantial proportion of serious cases were not curable³³, it is reasonable that age appears to be a main determinant of the prevalence rate of CHI. In addition, both newly acquired HI and the progress of impairment severity might also contribute to the increasing trend in the prevalence of CHI associated with age.

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73 We found that the prevalence rates of CHI in the age group < 3 years significantly
74 increased over the years. In urban areas, the rates increased significantly by 79 % from 2004
75 to 2010, and they increased significantly by 71% in rural areas. We speculated that one of the
76 main causes of this was the implementation of the newborn hearing screening (NHS)
77 program in Taiwan. As early diagnosis and early intervention of congenital HI has
78 demonstrated effectiveness in reducing its negative impacts on a child’s development, the
79 Health Promotion Administration of Taiwan began the promotion of NHS using otoacoustic
80 emission and automated auditory brainstem response in 2003 ³⁹. We believe that through
81 increasing awareness of parents and professionals and promoting easier access to NHS, the
82 registration of CHI cases have increased. According to Taiwan’s official reports, the
83 participation rates of newborn hearing screenings have increased from 4.0% in 2002 to
84 71.1% in 2010, and 97.8% of the baby-delivering institutions offered NHS services in 2013⁴⁰.
85 Another possible cause is that Taiwan Health Promotion Administration has also
86 implemented the Hearing Screening Plan for Pre-School Age Children in communities and
87 kindergartens. In 2013, for example, 138197 children were thus screened, yielding a
88 screening rate of 81.6%, compared to 30.3% in 2002 ⁴⁰.
89 In contrast with previous studies, our study has some unique features. While most
90 previous studies were cross-sectional surveys, we have data on the same population over
91 time. In most previous large-scale studies, data collection was just a one-time effort, but our
92 study included seven years-worth of data, which allows for the assessment of time trends. In
93 addition, our study has a very large number of cases, over 3533 cases in 2010 alone, and
94 therefore we can generate reliable statistical estimates. We also have specific information on
95 severity, which is rarely reported by large-scale studies.
96 However, our study has some limitations. We used “administrative prevalence” data,
97 which did not cover cases that were not detected or never received services from the
98 administration. Also, data on individual cases provided by the registry were limited, which

99 hindered the study of the etiology of the differences between rural and urban areas.
100 Investigations to clarify the etiology of the difference should be performed, which would
101 help prevention and health education to reduce the risk of CHI. Furthermore, we used
102 city/county as the unit for observation, but there may be both urban and rural townships
103 within a county. Therefore, using township as the unit of study may lead to more precise
104 classification. Unfortunately, such data were unavailable from the Taiwan government.
105 Nonetheless, this limitation tends to under estimate the difference in CHI prevalence
106 between rural and urban areas, instead of overestimating it, and since we observed a
107 statistically significant difference, its effect is unlikely to change our conclusions.

108 In conclusion, we found that the prevalence of CHI had remained similar from 2004 to
109 2010 in Taiwan. During this period, rural areas generally had higher prevalence rates than
110 urban areas. This difference was attributable to the higher prevalence rates of mild CHI
111 (55-69 dB BEHL). The rural-to-urban prevalence RRs generally decreased with severity. In
112 addition, we found that the prevalence rate in the age group < 3 years had increased
113 remarkably in both rural and urban areas, which might be attributable to the implementation
114 of the NHS program. We hope these findings can cast some light on the prevention and
115 control of CHI.

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124 **Contributor statement**

125 All authors participated in the design or implementation of the study or the analysis and
126 interpretation of findings. D-C Lai, Y-C Tseng, C-Y Lin, and H-R Guo were involved in all
127 phases of the study. D-C Lai and H-R Guo contributed to the study design. H-R Guo was the
128 principal investigator. D-C Lai, C-Y Lin, and H-R Guo contributed to the implementation of
129 the study, statistical analysis, and interpretation of the efficacy results. D-C Lai, Y-C Tseng,
130 C-Y Lin, and H-R Guo had access to the study data and contributed to data interpretation.
131 The report was drafted by C-Y Lin, and all authors reviewed and revised drafts of the
132 manuscript and approved the final version.

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138 **Competing interests**

139 C-Y Lin, Y-C Tseng, D-C Lai and H-R Guo declare that they have no conflicts of interest.

141 **Ethics approval**

142 The Institution Review Board of the Ditmanson Medical Foundation Chia-Yi Christian
143 Hospital.

145 **Provenance and peer review**

146 Not commissioned; externally peer reviewed.

147 **Data sharing statement**

148 No additional data available.

For peer review only

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STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cohort studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(a) Indicate the study’s design with a commonly used term in the title or the abstract	1-2
		(b) Provide in the abstract an informative and balanced summary of what was done and what was found	2-3
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	5
Objectives	3	State specific objectives, including any prespecified hypotheses	5
Methods			
Study design	4	Present key elements of study design early in the paper	7
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	6-7
Participants	6	(a) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up	6-7
		(b) For matched studies, give matching criteria and number of exposed and unexposed	Not applicable
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	7-8
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	6-7
Bias	9	Describe any efforts to address potential sources of bias	7
Study size	10	Explain how the study size was arrived at	Not applicable
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	6-7
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding	7-8
		(b) Describe any methods used to examine subgroups and interactions	7-8
		(c) Explain how missing data were addressed	Not applicable
		(d) If applicable, explain how loss to follow-up was addressed	Not applicable
		(e) Describe any sensitivity analyses	Not applicable
Results			

Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	8
		(b) Give reasons for non-participation at each stage	Not applicable
		(c) Consider use of a flow diagram	Not applicable
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	8-9
		(b) Indicate number of participants with missing data for each variable of interest	Not applicable
		(c) Summarise follow-up time (eg, average and total amount)	8
Outcome data	15*	Report numbers of outcome events or summary measures over time	10-11
Main results	16	(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	8-13
		(b) Report category boundaries when continuous variables were categorized	8-9
		(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	8-9
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	12-13
Discussion			
Key results	18	Summarise key results with reference to study objectives	20
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	20
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	17-19
Generalisability	21	Discuss the generalisability (external validity) of the study results	19
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	21

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at <http://www.plosmedicine.org/>, Annals of Internal Medicine at <http://www.annals.org/>, and Epidemiology at <http://www.epidem.com/>). Information on the STROBE Initiative is available at www.strobe-statement.org.

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Prevalence of Childhood Hearing Impairment of Different Severities in Urban and Rural Areas: A Nationwide Population-Based Study in Taiwan

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Prevalence of childhood hearing impairment of different severities in urban and rural areas: a nationwide population-based study in Taiwan

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[†]These authors contributed equally to the work.

Short title: Childhood hearing impairment of different severities in urban and rural areas

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Abstracts

Objective

Childhood hearing impairment (CHI) is a major developmental disability, but data at the national level are limited, especially those on different severities. We conducted a study to fill this data gap.

Design

A nationwide study on the basis of a reporting system.

Setting

To provide services to disabled citizens, the Taiwanese government maintains a registry of certified cases. Using data from this registry, we estimated prevalence rates of CHI of different severities from 2004 to 2010, and made comparisons between urban and rural areas.

Participants

Taiwanese citizens ≤ 17 years old.

Primary outcomes measure

To qualify for CHI disability benefits, a child must have an unaided pure-tone better ear hearing level at 0.5, 1, and 2 kHz with an average ≥ 55 decibels (dB), confirmed by an otolaryngologist. The severity was classified by pure-tone better ear hearing level as mild (55-69 dB), moderate (70-89 dB), and severe (≥ 90 dB).

Results

The registered cases under 17 years old decreased annually from 4075 in 2004 to 3533 in 2010, but changes in the prevalence rate were small, ranging from 7.62/10000 in 2004 to 7.91/10000 in 2006. The prevalence rates of mild CHI increased in all areas over time, but not those of moderate or severe CHI. Rural areas had higher overall prevalence rates than urban areas in all years, with rate ratios (RRs) between

1.01 and 1.09. By severity, rural areas had higher prevalence rates of mild (RRs between 1.08 and 1.25) and moderate (RRs between 1.06 and 1.21) CHI, but had lower prevalence rates of severe CHI (RRs between 0.92 and 0.99).

Conclusion

While rural areas had higher overall prevalence rates of CHI than urban areas, the RRs decreased with CHI severity. Further studies that identify factors affecting the rural-urban difference might help the prevention of CHI.

Keywords: child; disability; hearing impairment; hearing loss; prevalence; severity; Taiwan; urbanization

Strengths and limitations of this study

- Data on the same population were collected over a seven-year period, which allows the assessment of time trends.
- The study number of cases was large, over 3533 cases in 2010 alone.
- We have information on severity, which is rarely reported by large-scale studies.
- This study used administrative data, which do not cover cases who are not detected or who have never received services from the administration.
- Data on individual cases were not provided by the registry, which hindered more detailed analyses.

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INTRODUCTION

Hearing impairment (HI) is a global problem, and the World Health Organization estimated that 360 million people (including 32 million children) have disabling HI ¹. Most patients live in low- and middle-income countries, and 25% are born with or acquire HI during childhood. Compared with infants born in resource-rich countries, infants born in resource-poor countries have a nearly two-fold risk ²⁻⁴. The costs of the education support to children with better ear hearing level (BEHL) > 50 dB was estimated as \$3.9 billion ⁵. Compared to normal children, patients with childhood HI (CHI) have difficulties in language development, speech production, and cognition, which in turn affect their academic performance, vocational attainment, and socioemotional competence ^{2,6,7}.

The World Health Assembly affirmed the importance of interventions in control preventable HI ⁸ and recommended population-based epidemiological studies to determine the prevalence rate and causes of hearing impairment in all nations for targeting of preventive efforts ². The reported prevalence rates of CHI varied widely around the world ⁹⁻¹³. Most studies focus on either rural or urban populations, even though comparing the difference between the two is important. The urban–rural differences might be attributable to differences in cultural perceptions regarding the impact of HI, diagnosis, and treatment ¹⁴, but efforts are needed to investigate the differences further.

In 1980, the Taiwanese government constructed a system to certify disabled residents and to provide them with various services. The central government keeps a registry of certified cases ¹⁵, presenting a rare opportunity for studying CHI at the national level. The objectives of this study were to estimate the prevalence rates of CHI of different severities and to evaluate the differences between urban and rural areas.

METHODS

The disability registry system in Taiwan

In Taiwan, the Disabled Welfare Act was promulgated in 1980¹⁶. Accordingly, the local governments began to certify seven types of patients with disabilities, including “hearing impairment or balance disability.” When the Act was revised to become the People with Disabilities Rights Protection Act in 1997¹⁷, the “hearing impairment or balance disability” category was divided into two: HI and “balance disability”.

Individuals can make applications for certification through their local government in the residential area¹⁸⁻²⁰. These local governments report certified cases to the central government. The registry of cases was first maintained by the Ministry of the Interior and then by the Ministry of Health and Welfare after the re-organization of the government in 2013¹⁹. Because the registry identifies cases by the unique National Identification Numbers, each case is identified as one entity only.

Case definition of childhood hearing impairment

When a child is suspected of having HI, parents or guardians can apply for certification. To qualify for disability benefits, a child must have an unaided pure-tone BEHL at 0.5, 1 and 2 kHz (pure-tone average, PTA) with an average ≥ 55 decibels (dB), and confirmation by an otolaryngologist accredited by the government^{18 19}.

Different hearing tests are used to check for hearing disability in children less than 5 years of age. Neonatal hearing impairment is identified by the otoacoustic emissions screening with referral for diagnostic auditory brainstem response assessment. Visual reinforcement audiometry and play audiometry are used to test hearing impairment in older babies and young children. In cases with suspected malingering or difficulties in

testing, an auditory brainstem response is applied.

According to the Taiwanese government ¹⁹, the severity of HI is defined as “mild” with PTA \geq 55 dB BEHL and $<$ 70 dB BEHL, “moderate” with PTA \geq 70 dB BEHL and $<$ 90 dB BEHL, and “severe” with PTA \geq 90 dB BEHL. In order to continue to receive the disability benefits, a registered case needs to be re-evaluated every three years by an otolaryngologist accredited by the government.

Data collection

Using the nationwide registry data of HI, we conducted a study which included all children (\leq 17 years old) with Taiwanese citizenship. Each year, the government publishes a Statistical Yearbook ¹⁵. We obtained the data from the central government, but they are available since 2004 only. Furthermore, with the re-organization of administrative regions in 2011, one of the rural regions was merged into an urban region. While the impact of the reorganization on the classification was small, it made the population subdivisions incomparable before and after the reorganization. Therefore, we only analyzed the data until 2010. To calculate the prevalence rates, we obtained the total number of individuals in each age group from the Monthly Bulletin of Interior Statistics ²¹. The number were used as the denominators in estimating prevalence rates because the case ascertainment of the registry is through reporting by caregivers, and all eligible children are under continuous watch of the caregivers and will be reported when they become cases.

According to the Directorate-General of Budget, Accounting and Statistics, we defined an “urban area” as a city or county with $>$ 50% of the population living in metropolitan regions ²² **Error! Reference source not found.** In Taiwan, there are 7 cities and 18 counties, of which 7 cities and 5 counties were categorized as urban areas, and the remaining 13 counties were categorized as rural areas.

Statistical analysis

We estimated the prevalence rate of CHI in a rural or urban area by dividing the number of cases by the number of individuals each year and evaluated the trend over time. According to the yearbooks¹⁵, we categorized the age into five groups (< 3 years, 3-5 years, 6-11 years, 12-14 years, and 15-17 years). We calculated the overall prevalence rates, as well as the prevalence rates by severity, and evaluated the trends over time.

To evaluate the differences between urban and rural areas, we estimated the prevalence rate ratio (RR) by dividing the prevalence rate of rural areas by that of urban areas. A 95% confidence interval (CI) was calculated for each RR to evaluate its statistical significance.

We used the Chi-square test for trend to evaluate trends of changes in the prevalence rates over time and across age groups. To evaluate trends of changes in prevalence RRs over time and across age groups, we used linear regressions. In addition, we used ANOVA for repeated measures to evaluate trends of changes in the prevalence RRs among three different severity groups.

We conducted the analyses using SAS 9.1 (SAS Institute Inc., Cary, North Carolina, USA) and performed all statistical tests at the significance level of 0.05. The study protocol was reviewed and approved by the Institution Review Board of the Ditmanson Medical Foundation Chia-Yi Christian Hospital.

RESULTS

The trend of the overall prevalence rate by area

From 2004 to 2010, the registered cases under 17 years old decreased from 4075 to 3533, with a decreasing time trend. However, the changes in prevalence rates were

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small, ranging from 7.62/10000 in 2004 to 7.91/10000 in 2006, without a remarkable time trend, mainly because of the decreasing number of newborns each year. The prevalence rates in rural areas fluctuated between 7.70/10,000 and 8.18/10,000, without remarkable time trends (Table 1). The prevalence rates in urban areas also fluctuated, between 7.50/10,000 and 7.85/10,000, without remarkable time trends (Table 2). Rural areas had higher overall prevalence rates in all years, and the rural-to-urban prevalence RRs ranged from 1.01 to 1.09 (with $p < 0.05$ in 2008 and 2009), without remarkable time trends (Table 3).

The trends of prevalence rates by age

In rural areas, the prevalence rates in age groups < 3 years, 3-5 years, 6-11 years, 12-14 years, and 15-17 years was 2.31-4.90/10000, 5.91-7.75/10000, 7.61-8.39/10000, 8.97-9.73/10000, and 9.71-12.00/10000, respectively (Table 1). In each year, prevalence rates increased with age ($p < 0.01$ for all Chi-square tests for trend). Over time, the prevalence rates increased in age groups < 3 years ($p < 0.01$, increased by 71.0% from 2004 to 2010.) and 3-5 years ($p < 0.05$, increased by 23.6%), but decreased in the age group 15-17 years ($p < 0.01$, decreased by 19.1%).

In urban areas, the prevalence rates in age groups < 3 years, 3-5 years, 6-11 years, 12-14 years, and 15-17 years was 2.24-4.01/10000, 5.90-6.82/10000, 7.37-7.84/10000, 8.18-9.25/10000, and 9.21-11.17/10000, respectively (Table 2). In each year, prevalence rates increased with age ($p < 0.01$ in all years). The prevalence rates increased in the age group < 3 years over time ($p < 0.01$, increased by 79.0%), but decreased in age groups 12-14years and 15-17 years ($p < 0.05$ for both, decreased by 8.3% and 17.6%, respectively).

Table 1. The prevalence rate (per 10,000 children) of hearing impairment in rural areas by age in Taiwan.

Year	< 3year			3-5 year			6-11 year			12-14 year			15-17 year			0-17 year		
	N	Prev. [†]	(95% CI [‡])	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)
2004	50	2.31	(1.75,3.05)	159	6.27	(5.37,7.32)	438	8.05	(7.33,8.84)	256	9.13	(8.08,10.32)	343	12.00	(10.80,13.34)	1246	7.88	(7.46,8.33)
2005	49	2.43	(1.84,3.22)	150	6.16	(5.25,7.23)	403	7.61	(6.90,8.39)	270	9.73	(8.63,10.96)	315	10.87	(9.74,12.14)	1187	7.70	(7.28,8.15)
2006	70	3.68	(2.91,4.65)	154	6.99	(5.97,8.19)	420	8.04	(7.30,8.84)	259	9.31	(8.25,10.52)	301	10.68	(9.54,11.96)	1204	8.06	(7.62,8.53)
2007	89	4.90	(3.99,6.03)	122	5.91	(4.95,7.05)	422	8.39	(7.63,9.23)	251	9.05	(8.00,10.24)	277	9.77	(8.69,10.99)	1161	8.00	(7.55,8.47)
2008	68	3.90	(3.08,4.95)	143	7.43	(6.31,8.75)	389	8.10	(7.33,8.94)	248	8.99	(7.94,10.18)	284	10.10	(8.99,11.35)	1132	8.06	(7.61,8.54)
2009	68	4.06	(3.20,5.15)	132	7.21	(6.08,8.55)	378	8.34	(7.54,9.22)	255	9.23	(8.16,10.43)	282	9.98	(8.89,11.22)	1115	8.18	(7.72,8.68)
2010	61	3.95	(3.07,5.07)	135	7.75	(6.55,9.17)	353	8.07	(7.27,8.96)	234	8.97	(7.90,10.20)	274	9.71	(8.63,10.93)	1057	8.07	(7.60,8.58)

[†]Prev.: the prevalence, estimated by dividing the number of cases by the population in each age group in each year; [‡]CI: confidence interval.

Table 2. The prevalence rate (per 10,000 children) of hearing impairment in urban areas by age in Taiwan.

Year	< 3year			3-5 year			6-11 year			12-14 year			15-17 year			0-17 year		
	N	Prev. [†]	(95% CI [‡])	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)
2004	104	2.24	(1.85,2.71)	364	6.14	(5.54,6.81)	989	7.37	(6.92,7.84)	621	8.96	(8.29,9.70)	751	11.17	(10.40,12.00)	2829	7.51	(7.24,7.80)
2005	111	2.52	(2.10,3.04)	334	5.90	(5.30,6.56)	1013	7.71	(7.25,8.20)	610	8.88	(8.20,9.61)	767	11.05	(10.29,11.86)	2835	7.66	(7.38,7.95)
2006	140	3.26	(2.76,3.85)	326	6.38	(5.73,7.12)	998	7.65	(7.19,8.14)	639	9.25	(8.56,10.00)	734	10.80	(10.04,11.61)	2837	7.85	(7.57,8.14)
2007	138	3.24	(2.75,3.83)	325	6.69	(6.00,7.46)	957	7.62	(7.15,8.12)	629	9.05	(8.37,9.78)	709	10.30	(9.57,11.09)	2758	7.77	(7.48,8.06)
2008	132	3.10	(2.62,3.68)	315	6.82	(6.11,7.62)	907	7.54	(7.07,8.05)	600	8.66	(8.00,9.38)	643	9.43	(8.73,10.19)	2597	7.50	(7.21,7.79)
2009	147	3.50	(2.98,4.12)	283	6.28	(5.59,7.05)	889	7.84	(7.34,8.37)	567	8.18	(7.53,8.88)	652	9.52	(8.82,10.28)	2538	7.50	(7.22,7.80)
2010	159	4.01	(3.43,4.68)	293	6.55	(5.84,7.35)	853	7.74	(7.24,8.28)	536	8.22	(7.55,8.95)	635	9.21	(8.52,9.96)	2476	7.53	(7.24,7.84)

[†]Prev.: the prevalence, estimated by dividing the number of cases by the population in each age group in each year; [‡]CI: confidence interval.

Table 3. The rural-to-urban prevalence rate ratio of hearing impairment by age in Taiwan.

	< 3 year		3-5 year		6-11 year		12-14 year		15-17 year		0-17 year	
Year	Rate ratio	(95% CI) [†]	Rate ratio	(95% CI)	Rate ratio	(95% CI)	Rate ratio	(95% CI)	Rate ratio	(95% CI)	Rate ratio	(95% CI)
2004	1.03	(0.74,1.45)	1.02	(0.85,1.23)	1.09	(0.98,1.22)	1.02	(0.88,1.18)	1.07	(0.95,1.22)	1.05	(0.98,1.12)
2005	0.96	(0.69,1.35)	1.05	(0.86,1.27)	0.99	(0.88,1.11)	1.10	(0.95,1.26)	0.98	(0.86,1.12)	1.01	(0.94,1.08)
2006	1.13	(0.85,1.50)	1.10	(0.90,1.33)	1.05	(0.94,1.18)	1.01	(0.87,1.16)	0.99	(0.87,1.13)	1.03	(0.96,1.10)
2007	1.51	(1.16,1.97)*	0.88	(0.72,1.09)	1.10	(0.98,1.23)	1.00	(0.86,1.16)	0.95	(0.83,1.09)	1.03	(0.96,1.10)
2008	1.26	(0.94,1.69)	1.09	(0.89,1.33)	1.07	(0.95,1.21)	1.04	(0.89,1.20)	1.07	(0.93,1.23)	1.08	(1.00,1.15)*
2009	1.16	(0.87,1.55)	1.15	(0.93,1.41)	1.06	(0.94,1.20)	1.13	(0.97,1.31)	1.05	(0.91,1.21)	1.09	(1.02,1.17)*
2010	0.98	(0.73,1.32)	1.18	(0.96,1.45)	1.04	(0.92,1.18)	1.09	(0.94,1.27)	1.05	(0.92,1.21)	1.07	(1.00,1.15)

[†]CI: confidence interval; **p* < 0.05.

9 The trends of prevalence rates by severity and area

10 For mild CHI, the overall prevalence rates increased over time in both rural and urban
11 areas ($p < 0.05$ for both) (Table 4). For moderate CHI, overall prevalence rates decreased
12 over time in urban areas ($p < 0.01$), but no remarkable trends were observed in rural areas.
13 For severe CHI, the changes in overall prevalence rates were small in both rural and urban
14 areas and without any remarkable time trends.

15 Rural areas had higher overall prevalence rates of mild CHI in all years, and the
16 differences reached statistical significance in all years except 2005 and 2007. Rural areas
17 also had higher prevalence rates of moderate CHI in all years, but the difference reached
18 statistical significance in 2008 only. For severe CHI, prevalence rates in urban areas were
19 slightly higher in all years, but none of the differences reached statistical significance (Table
20 4). The changes in rural-to-urban RR were small in all severity groups and without any
21 remarkable time trends. Nevertheless, the mean of rural-to-urban RR in mild, moderate, and
22 severe CHI was 1.15, 1.10, and 0.96, respectively, indicating a decreasing trend ($p < 0.01$).
23 The rural-to-urban RR decreased with severity in all years except for 2007 and 2008.

Table 4. The overall rural-to-urban prevalence rate ratio of hearing impairment by severity in Taiwan.

Year	Mild				Moderate				Severe			
	Prev. [†]		Rate ratio	(95% CI [‡])	Prev.		Rate ratio	(95% CI)	Prev.		Rate ratio	(95% CI)
	Rural	Urban			Rural	Urban			Rural	Urban		
2004	2.44	2.13	1.15	(1.01,1.29)*	2.03	1.92	1.06	(0.93,1.21)	3.42	3.47	0.99	(0.89,1.09)
2005	2.49	2.28	1.09	(0.97,1.23)	1.95	1.85	1.06	(0.92,1.21)	3.26	3.53	0.92	(0.83,1.02)
2006	2.69	2.38	1.13	(1.00,1.27)*	1.97	1.84	1.07	(0.93,1.23)	3.41	3.62	0.94	(0.85,1.04)
2007	2.59	2.41	1.08	(0.95,1.22)	2.09	1.86	1.12	(0.98,1.29)	3.31	3.50	0.95	(0.85,1.05)
2008	2.64	2.32	1.14	(1.01,1.29)*	2.12	1.76	1.21	(1.05,1.39)*	3.30	3.42	0.96	(0.87,1.07)
2009	2.93	2.35	1.25	(1.10,1.40)*	1.98	1.74	1.14	(0.99,1.31)	3.27	3.41	0.96	(0.86,1.07)
2010	2.95	2.45	1.21	(1.07,1.36)*	1.76	1.64	1.07	(0.92,1.25)	3.37	3.45	0.98	(0.87,1.09)

[†]Prev.: the prevalence, estimated by dividing the number of cases by the population in each age group in each year; [‡]CI: confidence interval; **p* < 0.05.

DISCUSSION

Prevalence data on CHI between urban and rural areas from large-scale studies are limited, and the reported prevalence rates range widely. Conducting a search with the combined keywords of “hearing impairment,” “child,” “dB HL,” “rural,” “urban,” and “prevalence” in the PubMed database, we identified 16 studies on the prevalence rate of low-frequency CHI which defined HI by dB hearing level (HL) values and included rural and/or urban participants, and 11 of them used cutoffs ≥ 30 dB (Table 5)^{2 9-11 13 20 23-32}. The variation in reported prevalence rates may mainly be attributable to differences in case definition, age range, and case-finding methods³³. Factors such as genetic makeup, health-care accessibility, and socioeconomic status, may also have contributions¹¹⁻¹³. The differences make comparisons among studies difficult. For example, the case definition of severe for CHI in our study was ≥ 90 dB BEHL, and the 3.4/10000 prevalence rate in the rural areas in 2010 was lower than those reported by a study in Saudi Arabia (3.9/10000)⁹ and a study in India (35.2/10000)¹⁰ adopting similar criteria. However, the age ranges used were different, making the comparison difficult. The above limitations highlight the need for standardization to enhance the quality and comparability of study results. For example, the World Health Organization (WHO) recommends disabling hearing impairment in children be defined as a permanent unaided BEHL > 30 dB taken as the average BEHL for frequencies 0.5, 1, 2, and 4 kHz, while we were unable to adopt the standards because the lack of data on individual cases, of the 11 previous study identified from the systematic literature review, none adopted the WHO standards, even though most of them had data on individual cases. Standardization can allow direct future comparisons of studies as well as establish normative baseline data to illuminate potential intervention strategies¹².

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5 24 **Table 5. The prevalence rate (per 10000 children) of low-frequency hearing impairment (≥ 30 dB hearing level [HL] in the better ear)**
6 25 **defined by dB values in different studies.**

Study (year)	Country	Case-finding method	Case number (area)	Age (year)	Case definition	Prevalence
Seely et al. (1995) ²³	West Africa	Two-stage screening	2015 (rural)	5-15	Average of 0.5, 1, and 2 kHz	
					> 40 dB HL in the better ear	297.8
					> 60 dB HL in the better ear	129.0
					> 80 dB HL in the better ear	99.3
Minja & Machemba (1996) ²⁴	Tanzania	Two-stage screening	127 (rural)	5-20	Average of 0.5, 1, and 2 kHz	
					> 40 dB HL in the better ear	0.0
			675 (urban)	5-19	Average of 0.5, 1, and 2 kHz	
					> 40 dB HL in the better ear	163.0
Morioka et al. (1996) ²⁵	China	Population registry survey	282 (rural)	7-17	Average of 0.5, 1, and 2 kHz	
					≥ 35 dB HL in the better ear	496.5
Jacob et al. (1997) ¹⁰	India	Population registry survey	284 (rural)	6-10	Average of 0.5, 1, 2, 4kHz	
					> 40 dB HL in the better ear	1091.5
					> 65 dB HL in the better ear	70.4
Kaewboonchoo et al. (1998) ¹¹	China	Population registry survey	442 (urban)	6-19	> 90 dB HL in the better ear	35.2
					Average of 0.5, 1, and 2 kHz	
					≥ 35 dB HL in the better ear	113.1
Olusanya et al. (2000) ²⁷	Nigeria	Two-stage screening	359(urban)	4.5-10.9	Average of 0.5, 1, 2, 4kHz	
					> 40 dB HL in the better ear	55.7
Czechowicz et al. (2010) ²	Peru	Population registry survey	335 (rural)	6-19	Average of 0.5, 1, 2, 4kHz	
					> 40 dB HL in the better ear	238.8
					> 55 dB HL in the better ear	119.4

						> 70 dB HL in the better ear	29.9
						Average of 0.5, 1, 2, 4kHz	
						≥ 30 dB HL in the better ear	151.3
Schmitz et al. Error! Reference source not found. (2010) ²⁹	Nepal	Population registry survey	3646 (rural)	15-23		> 40 dB HL in the better ear	71.5
						> 60 dB HL in the better ear	38.5
						> 80 dB HL in the better ear	33.0
						Average of 0.5, 1, 2, 4kHz	
Bagshaw et al. Error! Reference source not found. (2011) ²⁰	Nepal	Population registry survey (with a diagnosis of otitis media with effusion)	70 (rural)	4-13		> 40 dB HL in the better ear	1000.0
			51 (urban)	4-13		Average of 0.5, 1, 2, 4kHz	
						> 40 dB HL in the better ear	0.0
						Average of 1, 2, 4kHz	
Gondim et al. (2012) ³⁰	Brazil	Population registry survey	90 (urban)	4-19		> 30 dB HL in the better ear	111.1
						Average of 1, 2, 4kHz	
Al-Rowaily et al. (2012) ⁹	Saudi Arabia	Two-stage screening	2574(urban)	4-8		> 40 dB HL in the better ear	73.8
						> 90 dB HL in the better ear	3.9
						Averaged of 0.5, 1, 2 kHz	
						≥ 55 dB HL in the better ear	8.1
						≥ 70 dB HL in the better ear	5.1
Our study	Taiwan	National registry (reporting)	1309068 (rural)	0-17		≥ 90 dB HL in the better ear	3.4
			3286699 (urban)			≥ 55 dB HL in the better ear	7.5
						≥ 70 dB HL in the better ear	5.1
						≥ 90 dB HL in the better ear	3.5

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Our major finding of a higher prevalence rate of CHI in rural areas was consistent with the results of previous studies in other countries^{20 24}. In 2009 the rural-urban prevalence RR was 0.96 (not statistically significant) in severe cases and 1.14 (not statistically significant) in moderate cases, but it was 1.25 (statistically significant) in mild cases, making the overall RR (1.09) statistically significant. In a study in Tanzania, in which 802 primary school children were examined using pure tone audiometry and HI was defined as a low-frequency PTA threshold of > 5 dB HL in the frequencies of 0.5, 1, and 2 kHz²⁴, the prevalence rate of CHI was 1102.4/10000 among rural children, while it was only 755.6/10000 among the urban children ($p < 0.05$). Similarly, in a survey in Nepal, school children with a diagnosis of otitis media with effusion (aged from 4 to 13 years) underwent audiometric assessment, and the prevalence rate of HI, defined as a middle-frequency PTA threshold of > 25 dB HL in the frequencies of 0.5, 1, 2, and 4 kHz, was higher among rural children (2700.0/10000 vs. 400.0/10000, $p < 0.05$)²⁰.

Some studies comparing CHI between urban and rural areas reported findings that are different from our observations. A study in China examined 6626 residents with an age range from 1 month to 90 years using the WHO definitions of HI and found no differences between urban and rural areas (19.7% vs. 15.7% reduction in dB HL, $p > 0.05$)³⁴. However, the report did not have separate data on CHI specifically, and therefore it is difficult to draw a conclusion on the difference in CHI. A study in Tanzania examined 854 schoolchildren from one urban district and one rural district by screening audiometry (air conduction) and found that the prevalence of bilateral HI was higher in the urban district (10.5% vs. 4.7%)³⁵. However, they did not include sensory HI, and therefore it is difficult to compare their data with our findings directly.

Some studies have investigated the possible etiological factors of the high prevalence rate of CHI in rural populations. A study on 335 school children between 6 and 19 years of age in an impoverished area of Peru identified the following risk factors for CHI: neonatal

jaundice, seizure, hospitalization, recurrent otitis media, past otorrhea, family history of HI at < 35 years, tympanic membrane abnormality, cerumen impaction, and eustachian tube dysfunction². This study proposed that untreated middle ear disease in the context of limited access to pediatric care may be a major risk factor for rural CHI. In a rural primary school in south India, hearing assessments were performed on 284 students (from 6 to 10 years old), and middle ear disease was found to be the predominant cause of CHI¹⁰. An investigation of HI in 75 Yemeni children (0.6-15 years) with chronic suppurative otitis media found that middle ear disease predominantly caused a HI of 26 to 60 dB HL³⁶. According to these findings, middle ear disease appears to be major cause of CHI in rural areas, mainly leading to HI in the range of 26-60 dB HL. In our study, we found that mild CHI (55-69 dB BEHL) was more prevalent in the rural areas in all years, with most of the rural-to-urban RRs reaching statistical significance, while the prevalence rates of CHI in the other two higher severity categories (≥ 70 dB HL) were similar between rural and urban areas. Therefore, we speculate that a higher prevalence of untreated middle ear disease in rural areas contributed, at least in part, to the rural-urban differences observed in our study.

In each year, the prevalence rates of CHI in both rural and urban areas increased with age. This finding was also noted in the Metropolitan Atlanta Developmental Disabilities Surveillance Program in the United States³⁷, which found that the prevalence rate of CHI > 40 dB HL increased steadily from 6.7/10000 among 3-year-old children to 13.8/10000 for 10-year-olds. Likewise, a study in the United Kingdom found that the prevalence rate of CHI > 40 dB HL rose from 9.1/10000 among 3 year-old children to 16.5/10000 among children 9 to 16 years old³⁸. Because HI was rarely fatal and a substantial proportion of serious cases were not curable³³, it is reasonable that age appears to be a main determinant of the prevalence rate of CHI. In addition, both newly acquired HI and the progress of impairment severity might also contribute to the increasing trend in the prevalence of CHI associated with age.

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We found that the prevalence rates of CHI in the age group < 3 years significantly increased over the years. In urban areas, the rates increased significantly by 79 % from 2004 to 2010, and they increased significantly by 71% in rural areas. We speculated that one of the main causes of this was the implementation of the newborn hearing screening (NHS) program in Taiwan. As early diagnosis and early intervention of congenital HI has demonstrated effectiveness in reducing its negative impacts on a child’s development, the Health Promotion Administration of Taiwan began the promotion of NHS using otoacoustic emission and automated auditory brainstem response in 2003 ³⁹. We believe that through increasing awareness of parents and professionals and promoting easier access to NHS, the registration of CHI cases have increased. According to Taiwan’s official reports, the participation rates of newborn hearing screenings have increased from 4.0% in 2002 to 71.1% in 2010, and 97.8% of the baby-delivering institutions offered NHS services in 2013⁴⁰. Another possible cause is that Taiwan Health Promotion Administration has also implemented the Hearing Screening Plan for Pre-School Age Children in communities and kindergartens. In 2013, for example, 138197 children were thus screened, yielding a screening rate of 81.6%, compared to 30.3% in 2002 ⁴⁰.

In contrast with previous studies, our study has some unique features. While most previous studies were cross-sectional surveys, we have data on the same population over time. In most previous large-scale studies, data collection was just a one-time effort, but our study included seven years-worth of data, which allows for the assessment of time trends. In addition, our study has a very large number of cases, over 3533 cases in 2010 alone, and therefore we can generate reliable statistical estimates. We also have specific information on severity, which is rarely reported by large-scale studies.

However, our study also has some limitations. We used “administrative prevalence” data, which did not cover cases that were not detected or never received services from the administration. Also, data on individual cases provided by the registry were limited, which

105 hindered the study of the etiology of the differences between rural and urban areas.
106 Investigations to clarify the etiology of the difference should be performed, which would
107 help prevention and health education to reduce the risk of CHI. Furthermore, we used
108 city/county as the unit for observation, but there may be both urban and rural townships
109 within a county. Therefore, using township as the unit of study may lead to more precise
110 classification. Unfortunately, such data were unavailable from the Taiwan government.
111 Nonetheless, this limitation tends to under estimate the difference in CHI prevalence
112 between rural and urban areas, instead of overestimating it, and since we observed a
113 statistically significant difference, its effect is unlikely to change our conclusions.

114 In conclusion, we found that the prevalence of CHI had remained similar from 2004 to
115 2010 in Taiwan. During this period, rural areas generally had higher prevalence rates than
116 urban areas. This difference was attributable to the higher prevalence rates of mild CHI
117 (55-69 dB BEHL). The rural-to-urban prevalence RRs generally decreased with severity. In
118 addition, we found that the prevalence rate in the age group < 3 years had increased
119 remarkably in both rural and urban areas, which might be attributable to the implementation
120 of the NHS program. We hope these findings can cast some light on the prevention and
121 control of CHI.

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128 analysis.

130 **Contributor statement**

131 All authors participated in the design or implementation of the study or the analysis and
132 interpretation of findings. D-C Lai, Y-C Tseng, C-Y Lin, and H-R Guo were involved in all
133 phases of the study. D-C Lai and H-R Guo contributed to the study design. H-R Guo was the
134 principal investigator. D-C Lai, C-Y Lin, and H-R Guo contributed to the implementation of
135 the study, statistical analysis, and interpretation of the efficacy results. D-C Lai, Y-C Tseng,
136 C-Y Lin, and H-R Guo had access to the study data and contributed to data interpretation.
137 The report was drafted by C-Y Lin, and all authors reviewed and revised drafts of the
138 manuscript and approved the final version.

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144 **Competing interests**

145 C-Y Lin, Y-C Tseng, D-C Lai and H-R Guo declare that they have no conflicts of interest.

147 **Ethics approval**

148 The Institution Review Board of the Ditmanson Medical Foundation Chia-Yi Christian
149 Hospital.

151 **Provenance and peer review**

152 Not commissioned; externally peer reviewed.

153 **Data sharing statement**

154 No additional data available.

For peer review only

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STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of *cohort studies*

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(a) Indicate the study’s design with a commonly used term in the title or the abstract	1-2
		(b) Provide in the abstract an informative and balanced summary of what was done and what was found	2-3
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	5
Objectives	3	State specific objectives, including any prespecified hypotheses	5
Methods			
Study design	4	Present key elements of study design early in the paper	7
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	6-7
Participants	6	(a) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up	6-7
		(b) For matched studies, give matching criteria and number of exposed and unexposed	Not applicable
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	7-8
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	6-7
Bias	9	Describe any efforts to address potential sources of bias	7
Study size	10	Explain how the study size was arrived at	Not applicable
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	6-7
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding	7-8
		(b) Describe any methods used to examine subgroups and interactions	7-8
		(c) Explain how missing data were addressed	Not applicable
		(d) If applicable, explain how loss to follow-up was addressed	Not applicable
		(e) Describe any sensitivity analyses	Not applicable
Results			

Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	8
		(b) Give reasons for non-participation at each stage	Not applicable
		(c) Consider use of a flow diagram	Not applicable
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	8-9
		(b) Indicate number of participants with missing data for each variable of interest	Not applicable
		(c) Summarise follow-up time (eg, average and total amount)	8
Outcome data	15*	Report numbers of outcome events or summary measures over time	10-11
Main results	16	(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	8-13
		(b) Report category boundaries when continuous variables were categorized	8-9
		(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	8-9
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	12-13
Discussion			
Key results	18	Summarise key results with reference to study objectives	20
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	20
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	17-19
Generalisability	21	Discuss the generalisability (external validity) of the study results	19
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	21

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at <http://www.plosmedicine.org/>, Annals of Internal Medicine at <http://www.annals.org/>, and Epidemiology at <http://www.epidem.com/>). Information on the STROBE Initiative is available at www.strobe-statement.org.

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Prevalence of Childhood Hearing Impairment of Different Severities in Urban and Rural Areas: A Nationwide Population-Based Study in Taiwan

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Prevalence of childhood hearing impairment of different severities in urban and rural areas: a nationwide population-based study in Taiwan

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[†]These authors contributed equally to the work.

Short title: Childhood hearing impairment of different severities in urban and rural areas

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Word count: 2976

Abstracts

Objective

Childhood hearing impairment (CHI) is a major developmental disability, but data at the national level are limited, especially those on different severities. We conducted a study to fill this data gap.

Design

A nationwide study on the basis of a reporting system.

Setting

To provide services to disabled citizens, the Taiwanese government maintains a registry of certified cases. Using data from this registry, we estimated prevalence rates of CHI of different severities from 2004 to 2010, and made comparisons between urban and rural areas.

Participants

Taiwanese citizens ≤ 17 years old.

Primary outcomes measure

To qualify for CHI disability benefits, a child must have an unaided pure-tone better ear hearing level at 0.5, 1, and 2 kHz with an average ≥ 55 decibels (dB), confirmed by an otolaryngologist. The severity was classified by pure-tone better ear hearing level as mild (55-69 dB), moderate (70-89 dB), and severe (≥ 90 dB).

Results

The registered cases under 17 years old decreased annually from 4075 in 2004 to 3533 in 2010, but changes in the prevalence rate were small, ranging from 7.62/10000 in 2004 to 7.91/10000 in 2006. The prevalence rates of mild CHI increased in all areas over time, but not those of moderate or severe CHI. Rural areas had higher overall prevalence rates than urban areas in all years, with rate ratios (RRs) between

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1.01 and 1.09. By severity, rural areas had higher prevalence rates of mild (RRs between 1.08 and 1.25) and moderate (RRs between 1.06 and 1.21) CHI, but had lower prevalence rates of severe CHI (RRs between 0.92 and 0.99).

Conclusion

While rural areas had higher overall prevalence rates of CHI than urban areas, the RRs decreased with CHI severity. Further studies that identify factors affecting the rural-urban difference might help the prevention of CHI.

Keywords: child; disability; hearing impairment; hearing loss; prevalence; severity; Taiwan; urbanization

Strengths and limitations of this study

- Data on the same population were collected over a seven-year period, which allows the assessment of time trends.
- The study number of cases was large, over 3533 cases in 2010 alone.
- We have information on severity, which is rarely reported by large-scale studies.
- This study used administrative data, which do not cover cases who are not detected or who have never received services from the administration.
- Data on individual cases were not provided by the registry, which hindered more detailed analyses.

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INTRODUCTION

Hearing impairment (HI) is a global problem, and the World Health Organization estimated that 360 million people (including 32 million children) have disabling HI ¹. Most patients live in low- and middle-income countries, and 25% are born with or acquire HI during childhood. Compared with infants born in resource-rich countries, infants born in resource-poor countries have a nearly two-fold risk ²⁻⁴. The costs of the education support to children with better ear hearing level (BEHL) > 50 dB was estimated as \$3.9 billion ⁵. Compared to normal children, patients with childhood HI (CHI) have difficulties in language development, speech production, and cognition, which in turn affect their academic performance, vocational attainment, and socioemotional competence ^{2,6,7}.

The World Health Assembly affirmed the importance of interventions in control preventable HI ⁸ and recommended population-based epidemiological studies to determine the prevalence rate and causes of hearing impairment in all nations for targeting of preventive efforts ². The reported prevalence rates of CHI varied widely around the world ⁹⁻¹³. Most studies focus on either rural or urban populations, even though comparing the difference between the two is important. The urban–rural differences might be attributable to differences in cultural perceptions regarding the impact of HI, diagnosis, and treatment ¹⁴, but efforts are needed to investigate the differences further.

In 1980, the Taiwanese government constructed a system to certify disabled residents and to provide them with various services. The central government keeps a registry of certified cases ¹⁵, presenting a rare opportunity for studying CHI at the national level. The objectives of this study were to estimate the prevalence rates of CHI of different severities and to evaluate the differences between urban and rural areas.

METHODS

The disability registry system in Taiwan

In Taiwan, the Disabled Welfare Act was promulgated in 1980¹⁶. Accordingly, the local governments began to certify seven types of patients with disabilities, including “hearing impairment or balance disability.” When the Act was revised to become the People with Disabilities Rights Protection Act in 1997¹⁷, the “hearing impairment or balance disability” category was divided into two: HI and “balance disability”.

Individuals can make applications for certification through their local government in the residential area¹⁸⁻²⁰. These local governments report certified cases to the central government. The registry of cases was first maintained by the Ministry of the Interior and then by the Ministry of Health and Welfare after the re-organization of the government in 2013¹⁹. Because the registry identifies cases by the unique National Identification Numbers, each case is identified as one entity only.

Case definition of childhood hearing impairment

When a child is suspected of having HI, parents or guardians can apply for certification. To qualify for disability benefits, a child must have an unaided pure-tone BEHL at 0.5, 1 and 2 kHz (pure-tone average, PTA) with an average ≥ 55 decibels (dB), and confirmation by an otolaryngologist accredited by the government^{18 19}.

Different hearing tests are used to check for hearing disability in children less than 5 years of age. Neonatal hearing impairment is identified by the otoacoustic emissions screening with referral for diagnostic auditory brainstem response assessment. Visual reinforcement audiometry and play audiometry are used to test hearing impairment in older babies and young children. In cases with suspected malingering or difficulties in

testing, an auditory brainstem response is applied.

According to the Taiwanese government ¹⁹, the severity of HI is defined as “mild” with PTA ≥ 55 dB BEHL and < 70 dB BEHL, “moderate” with PTA ≥ 70 dB BEHL and < 90 dB BEHL, and “severe” with PTA ≥ 90 dB BEHL. In order to continue to receive the disability benefits, a registered case needs to be re-evaluated every three years by an otolaryngologist accredited by the government.

Data collection

Using the nationwide registry data of HI, we conducted a study which included all children (≤ 17 years old) with Taiwanese citizenship. Each year, the government publishes a Statistical Yearbook ¹⁵. We obtained the data from the central government, but they are available since 2004 only. Furthermore, with the re-organization of administrative regions in 2011, one of the rural regions was merged into an urban region. While the impact of the reorganization on the classification was small, it made the population subdivisions incomparable before and after the reorganization. Therefore, we only analyzed the data until 2010. To calculate the prevalence rates, we obtained the total number of individuals in each age group from the Monthly Bulletin of Interior Statistics ²¹. The number were used as the denominators in estimating prevalence rates because the case ascertainment of the registry is through reporting by caregivers, and all eligible children are under continuous watch of the caregivers and will be reported when they become cases.

According to the Directorate-General of Budget, Accounting and Statistics, we defined an “urban area” as a city or county with > 50% of the population living in metropolitan regions ²² Error! Reference source not found. In Taiwan, there are 7 cities and 18 counties, of which 7 cities and 5 counties were categorized as urban areas, and the remaining 13 counties were categorized as rural areas.

Statistical analysis

We estimated the prevalence rate of CHI in a rural or urban area by dividing the number of cases by the number of individuals each year and evaluated the trend over time. According to the yearbooks¹⁵, we categorized the age into five groups (< 3 years, 3-5 years, 6-11 years, 12-14 years, and 15-17 years). We calculated the overall prevalence rates, as well as the prevalence rates by severity, and evaluated the trends over time.

To evaluate the differences between urban and rural areas, we estimated the prevalence rate ratio (RR) by dividing the prevalence rate of rural areas by that of urban areas. A 95% confidence interval (CI) was calculated for each RR to evaluate its statistical significance.

We used the Chi-square test for trend to evaluate trends of changes in the prevalence rates over time and across age groups. To evaluate trends of changes in prevalence RRs over time and across age groups, we used linear regressions. In addition, we used ANOVA for repeated measures to evaluate trends of changes in the prevalence RRs among three different severity groups.

We conducted the analyses using SAS 9.1 (SAS Institute Inc., Cary, North Carolina, USA) and performed all statistical tests at the significance level of 0.05. The study protocol was reviewed and approved by the Institution Review Board of the Ditmanson Medical Foundation Chia-Yi Christian Hospital.

Patient and Public Involvement

This study was based on secondary data analysis, and there was no patient or public involvement.

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RESULTS

The trend of the overall prevalence rate by area

From 2004 to 2010, the registered cases under 17 years old decreased from 4075 to 3533, with a decreasing time trend. However, the changes in prevalence rates were small, ranging from 7.62/10000 in 2004 to 7.91/10000 in 2006, without a remarkable time trend, mainly because of the decreasing number of newborns each year. The prevalence rates in rural areas fluctuated between 7.70/10,000 and 8.18/10,000, without remarkable time trends (Table 1). The prevalence rates in urban areas also fluctuated, between 7.50/10,000 and 7.85/10,000, without remarkable time trends (Table 2). Rural areas had higher overall prevalence rates in all years, and the rural-to-urban prevalence RRs ranged from 1.01 to 1.09 (with $p < 0.05$ in 2008 and 2009), without remarkable time trends (Table 3).

The trends of prevalence rates by age

In rural areas, the prevalence rates in age groups < 3 years, 3-5 years, 6-11 years, 12-14 years, and 15-17 years was 2.31-4.90/10000, 5.91-7.75/10000, 7.61-8.39/10000, 8.97-9.73/10000, and 9.71-12.00/10000, respectively (Table 1). In each year, prevalence rates increased with age ($p < 0.01$ for all Chi-square tests for trend). Over time, the prevalence rates increased in age groups < 3 years ($p < 0.01$, increased by 71.0% from 2004 to 2010.) and 3-5 years ($p < 0.05$, increased by 23.6%), but decreased in the age group 15-17 years ($p < 0.01$, decreased by 19.1%).

In urban areas, the prevalence rates in age groups < 3 years, 3-5 years, 6-11 years, 12-14 years, and 15-17 years was 2.24-4.01/10000, 5.90-6.82/10000, 7.37-7.84/10000, 8.18-9.25/10000, and 9.21-11.17/10000, respectively (Table 2). In each year, prevalence rates increased with age ($p < 0.01$ in all years). The prevalence rates increased in the age group < 3 years over time ($p < 0.01$, increased by 79.0%), but

decreased in age groups 12-14years and 15-17 years ($p < 0.05$ for both, decreased by 8.3% and 17.6%, respectively).

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1 **Table 1. The prevalence rate (per 10,000 children) of hearing impairment in rural areas by age in Taiwan.**

Year	< 3year			3-5 year			6-11 year			12-14 year			15-17 year			0-17 year		
	N	Prev. [†]	(95% CI [‡])	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)
2004	50	2.31	(1.75,3.05)	159	6.27	(5.37,7.32)	438	8.05	(7.33,8.84)	256	9.13	(8.08,10.32)	343	12.00	(10.80,13.34)	1246	7.88	(7.46,8.33)
2005	49	2.43	(1.84,3.22)	150	6.16	(5.25,7.23)	403	7.61	(6.90,8.39)	270	9.73	(8.63,10.96)	315	10.87	(9.74,12.14)	1187	7.70	(7.28,8.15)
2006	70	3.68	(2.91,4.65)	154	6.99	(5.97,8.19)	420	8.04	(7.30,8.84)	259	9.31	(8.25,10.52)	301	10.68	(9.54,11.96)	1204	8.06	(7.62,8.53)
2007	89	4.90	(3.99,6.03)	122	5.91	(4.95,7.05)	422	8.39	(7.63,9.23)	251	9.05	(8.00,10.24)	277	9.77	(8.69,10.99)	1161	8.00	(7.55,8.47)
2008	68	3.90	(3.08,4.95)	143	7.43	(6.31,8.75)	389	8.10	(7.33,8.94)	248	8.99	(7.94,10.18)	284	10.10	(8.99,11.35)	1132	8.06	(7.61,8.54)
2009	68	4.06	(3.20,5.15)	132	7.21	(6.08,8.55)	378	8.34	(7.54,9.22)	255	9.23	(8.16,10.43)	282	9.98	(8.89,11.22)	1115	8.18	(7.72,8.68)
2010	61	3.95	(3.07,5.07)	135	7.75	(6.55,9.17)	353	8.07	(7.27,8.96)	234	8.97	(7.90,10.20)	274	9.71	(8.63,10.93)	1057	8.07	(7.60,8.58)

2 [†]Prev.: the prevalence, estimated by dividing the number of cases by the population in each age group in each year; [‡]CI: confidence interval.

4 **Table 2. The prevalence rate (per 10,000 children) of hearing impairment in urban areas by age in Taiwan.**

Year	< 3year			3-5 year			6-11 year			12-14 year			15-17 year			0-17 year		
	N	Prev. [†]	(95% CI [‡])	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)	N	Prev.	(95% CI)
2004	104	2.24	(1.85,2.71)	364	6.14	(5.54,6.81)	989	7.37	(6.92,7.84)	621	8.96	(8.29,9.70)	751	11.17	(10.40,12.00)	2829	7.51	(7.24,7.80)
2005	111	2.52	(2.10,3.04)	334	5.90	(5.30,6.56)	1013	7.71	(7.25,8.20)	610	8.88	(8.20,9.61)	767	11.05	(10.29,11.86)	2835	7.66	(7.38,7.95)
2006	140	3.26	(2.76,3.85)	326	6.38	(5.73,7.12)	998	7.65	(7.19,8.14)	639	9.25	(8.56,10.00)	734	10.80	(10.04,11.61)	2837	7.85	(7.57,8.14)
2007	138	3.24	(2.75,3.83)	325	6.69	(6.00,7.46)	957	7.62	(7.15,8.12)	629	9.05	(8.37,9.78)	709	10.30	(9.57,11.09)	2758	7.77	(7.48,8.06)
2008	132	3.10	(2.62,3.68)	315	6.82	(6.11,7.62)	907	7.54	(7.07,8.05)	600	8.66	(8.00,9.38)	643	9.43	(8.73,10.19)	2597	7.50	(7.21,7.79)
2009	147	3.50	(2.98,4.12)	283	6.28	(5.59,7.05)	889	7.84	(7.34,8.37)	567	8.18	(7.53,8.88)	652	9.52	(8.82,10.28)	2538	7.50	(7.22,7.80)
2010	159	4.01	(3.43,4.68)	293	6.55	(5.84,7.35)	853	7.74	(7.24,8.28)	536	8.22	(7.55,8.95)	635	9.21	(8.52,9.96)	2476	7.53	(7.24,7.84)

5 [†]Prev.: the prevalence, estimated by dividing the number of cases by the population in each age group in each year; [‡]CI: confidence interval.

Table 3. The rural-to-urban prevalence rate ratio of hearing impairment by age in Taiwan.

	< 3 year		3-5 year		6-11 year		12-14 year		15-17 year		0-17 year	
Year	Rate ratio	(95% CI) [†]	Rate ratio	(95% CI)	Rate ratio	(95% CI)	Rate ratio	(95% CI)	Rate ratio	(95% CI)	Rate ratio	(95% CI)
2004	1.03	(0.74,1.45)	1.02	(0.85,1.23)	1.09	(0.98,1.22)	1.02	(0.88,1.18)	1.07	(0.95,1.22)	1.05	(0.98,1.12)
2005	0.96	(0.69,1.35)	1.05	(0.86,1.27)	0.99	(0.88,1.11)	1.10	(0.95,1.26)	0.98	(0.86,1.12)	1.01	(0.94,1.08)
2006	1.13	(0.85,1.50)	1.10	(0.90,1.33)	1.05	(0.94,1.18)	1.01	(0.87,1.16)	0.99	(0.87,1.13)	1.03	(0.96,1.10)
2007	1.51	(1.16,1.97)*	0.88	(0.72,1.09)	1.10	(0.98,1.23)	1.00	(0.86,1.16)	0.95	(0.83,1.09)	1.03	(0.96,1.10)
2008	1.26	(0.94,1.69)	1.09	(0.89,1.33)	1.07	(0.95,1.21)	1.04	(0.89,1.20)	1.07	(0.93,1.23)	1.08	(1.00,1.15)*
2009	1.16	(0.87,1.55)	1.15	(0.93,1.41)	1.06	(0.94,1.20)	1.13	(0.97,1.31)	1.05	(0.91,1.21)	1.09	(1.02,1.17)*
2010	0.98	(0.73,1.32)	1.18	(0.96,1.45)	1.04	(0.92,1.18)	1.09	(0.94,1.27)	1.05	(0.92,1.21)	1.07	(1.00,1.15)

[†]CI: confidence interval; * $p < 0.05$.

9 **The trends of prevalence rates by severity and area**

10 For mild CHI, the overall prevalence rates increased over time in both rural and urban
11 areas ($p < 0.05$ for both) (Table 4). For moderate CHI, overall prevalence rates decreased
12 over time in urban areas ($p < 0.01$), but no remarkable trends were observed in rural areas.
13 For severe CHI, the changes in overall prevalence rates were small in both rural and urban
14 areas and without any remarkable time trends.

15 Rural areas had higher overall prevalence rates of mild CHI in all years, and the
16 differences reached statistical significance in all years except 2005 and 2007. Rural areas
17 also had higher prevalence rates of moderate CHI in all years, but the difference reached
18 statistical significance in 2008 only. For severe CHI, prevalence rates in urban areas were
19 slightly higher in all years, but none of the differences reached statistical significance (Table
20 4). The changes in rural-to-urban RR were small in all severity groups and without any
21 remarkable time trends. Nevertheless, the mean of rural-to-urban RR in mild, moderate, and
22 severe CHI was 1.15, 1.10, and 0.96, respectively, indicating a decreasing trend ($p < 0.01$).
23 The rural-to-urban RR decreased with severity in all years except for 2007 and 2008.

Table 4. The overall rural-to-urban prevalence rate ratio of hearing impairment by severity in Taiwan.

Year	Mild				Moderate				Severe			
	Prev. [†]		Rate ratio	(95% CI [‡])	Prev.		Rate ratio	(95% CI)	Prev.		Rate ratio	(95% CI)
	Rural	Urban			Rural	Urban			Rural	Urban		
2004	2.44	2.13	1.15	(1.01,1.29)*	2.03	1.92	1.06	(0.93,1.21)	3.42	3.47	0.99	(0.89,1.09)
2005	2.49	2.28	1.09	(0.97,1.23)	1.95	1.85	1.06	(0.92,1.21)	3.26	3.53	0.92	(0.83,1.02)
2006	2.69	2.38	1.13	(1.00,1.27)*	1.97	1.84	1.07	(0.93,1.23)	3.41	3.62	0.94	(0.85,1.04)
2007	2.59	2.41	1.08	(0.95,1.22)	2.09	1.86	1.12	(0.98,1.29)	3.31	3.50	0.95	(0.85,1.05)
2008	2.64	2.32	1.14	(1.01,1.29)*	2.12	1.76	1.21	(1.05,1.39)*	3.30	3.42	0.96	(0.87,1.07)
2009	2.93	2.35	1.25	(1.10,1.40)*	1.98	1.74	1.14	(0.99,1.31)	3.27	3.41	0.96	(0.86,1.07)
2010	2.95	2.45	1.21	(1.07,1.36)*	1.76	1.64	1.07	(0.92,1.25)	3.37	3.45	0.98	(0.87,1.09)

[†]Prev.: the prevalence, estimated by dividing the number of cases by the population in each age group in each year; [‡]CI: confidence interval;

* $p < 0.05$.

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DISCUSSION

Prevalence data on CHI between urban and rural areas from large-scale studies are limited, and the reported prevalence rates range widely. We conducted a search of literature in the PubMed database and identified 16 studies on the prevalence rate of low-frequency CHI which defined HI by dB hearing level (HL) values and included rural and/or urban participants, and 11 of them used cutoffs ≥ 30 dB (Table 5)^{2 9-11 13 20 23-32}. The variation in reported prevalence rates may mainly be attributable to differences in case definition, age range, and case-finding methods³³. Factors such as genetic makeup, health-care accessibility, and socioeconomic status, may also have contributions¹¹⁻¹³. The differences make comparisons among studies difficult. For example, the case definition of severe for CHI in our study was ≥ 90 dB BEHL, and the 3.4/10000 prevalence rate in the rural areas in 2010 was lower than those reported by a study in Saudi Arabia (3.9/10000)⁹ and a study in India (35.2/10000)¹⁰ adopting similar criteria. However, the age ranges used were different, making the comparison difficult. The above limitations highlight the need for standardization to enhance the quality and comparability of study results. For example, the World Health Organization (WHO) recommends disabling hearing impairment in children be defined as a permanent unaided BEHL > 30 dB taken as the average BEHL for frequencies 0.5, 1, 2, and 4 kHz, while we were unable to adopt the standards because the lack of data on individual cases, of the 11 previous study identified from the systematic literature review, none adopted the WHO standards, even though most of them had data on individual cases. Standardization can allow direct future comparisons of studies as well as establish normative baseline data to illuminate potential intervention strategies¹².

Table 5. The prevalence rate (per 10000 children) of low-frequency hearing impairment (≥ 30 dB hearing level [HL] in the better ear) defined by dB values in different studies.

Study (year)	Country	Case-finding method	Case number (area)	Age (year)	Case definition	Prevalence
Seely et al. (1995) ²³	West Africa	Two-stage screening	2015 (rural)	5-15	Average of 0.5, 1, and 2 kHz	
					> 40 dB HL in the better ear	297.8
					> 60 dB HL in the better ear	129.0
					> 80 dB HL in the better ear	99.3
Minja & Machemba (1996) ²⁴	Tanzania	Two-stage screening	127 (rural)	5-20	Average of 0.5, 1, and 2 kHz	
					> 40 dB HL in the better ear	0.0
			675 (urban)	5-19	Average of 0.5, 1, and 2 kHz	
					> 40 dB HL in the better ear	163.0
Morioka et al. (1996) ²⁵	China	Population registry survey	282 (rural)	7-17	Average of 0.5, 1, and 2 kHz	
					≥ 35 dB HL in the better ear	496.5
Jacob et al. (1997) ¹⁰	India	Population registry survey	284 (rural)	6-10	Average of 0.5, 1, 2, 4kHz	
					> 40 dB HL in the better ear	1091.5
					> 65 dB HL in the better ear	70.4
					> 90 dB HL in the better ear	35.2
Kaewboonchoo et al. (1998) ¹¹	China	Population registry survey	442 (urban)	6-19	Average of 0.5, 1, and 2 kHz	
					≥ 35 dB HL in the better ear	113.1
Olusanya et al. (2000) ²⁷	Nigeria	Two-stage screening	359(urban)	4.5-10.9	Average of 0.5, 1, 2, 4kHz	
					> 40 dB HL in the better ear	55.7
Czechowicz et al. (2010) ²	Peru	Population registry survey	335 (rural)	6-19	Average of 0.5, 1, 2, 4kHz	
					> 40 dB HL in the better ear	238.8
					> 55 dB HL in the better ear	119.4

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						> 70 dB HL in the better ear	29.9
						Average of 0.5, 1, 2, 4kHz	
						≥ 30 dB HL in the better ear	151.3
Schmitz et al. source not found.	Error! Reference (2010) ²⁹	Nepal	Population registry survey	3646 (rural)	15-23	> 40 dB HL in the better ear	71.5
						> 60 dB HL in the better ear	38.5
						> 80 dB HL in the better ear	33.0
						Average of 0.5, 1, 2, 4kHz	
Bagshaw et al. source not found.	Error! Reference (2011) ²⁰	Nepal	Population registry survey (with a diagnosis of otitis media with effusion)	70 (rural)	4-13	> 40 dB HL in the better ear	1000.0
				51 (urban)	4-13	Average of 0.5, 1, 2, 4kHz	
						> 40 dB HL in the better ear	0.0
Gondim et al. (2012) ³⁰		Brazil	Population registry survey	90 (urban)	4-19	Average of 1, 2, 4kHz	
						> 30 dB HL in the better ear	111.1
						Average of 1, 2, 4kHz	
Al-Rowaily et al. (2012) ⁹		Saudi Arabia	Two-stage screening	2574(urban)	4-8	> 40 dB HL in the better ear	73.8
						> 90 dB HL in the better ear	3.9
						Averaged of 0.5, 1, 2 kHz	
						≥ 55 dB HL in the better ear	8.1
						≥ 70 dB HL in the better ear	5.1
Our study		Taiwan	National registry (reporting)	1309068 (rural) 3286699 (urban)	0-17	≥ 90 dB HL in the better ear	3.4
						≥ 55 dB HL in the better ear	7.5
						≥ 70 dB HL in the better ear	5.1
						≥ 90 dB HL in the better ear	3.5

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Our major finding of a higher prevalence rate of CHI in rural areas was consistent with the results of previous studies in other countries^{20 24}. In 2009 the rural-urban prevalence RR was 0.96 (not statistically significant) in severe cases and 1.14 (not statistically significant) in moderate cases, but it was 1.25 (statistically significant) in mild cases, making the overall RR (1.09) statistically significant. In a study in Tanzania, in which 802 primary school children were examined using pure tone audiometry and HI was defined as a low-frequency PTA threshold of > 5 dB HL in the frequencies of 0.5, 1, and 2 kHz²⁴, the prevalence rate of CHI was 1102.4/10000 among rural children, while it was only 755.6/10000 among the urban children ($p < 0.05$). Similarly, in a survey in Nepal, school children with a diagnosis of otitis media with effusion (aged from 4 to 13 years) underwent audiometric assessment, and the prevalence rate of HI, defined as a middle-frequency PTA threshold of > 25 dB HL in the frequencies of 0.5, 1, 2, and 4 kHz, was higher among rural children (2700.0/10000 vs. 400.0/10000, $p < 0.05$)²⁰.

Some studies comparing CHI between urban and rural areas reported findings that are different from our observations. A study in China examined 6626 residents with an age range from 1 month to 90 years using the WHO definitions of HI and found no differences between urban and rural areas (19.7% vs. 15.7% reduction in dB HL, $p > 0.05$)³⁴. However, the report did not have separate data on CHI specifically, and therefore it is difficult to draw a conclusion on the difference in CHI. A study in Tanzania examined 854 schoolchildren from one urban district and one rural district by screening audiometry (air conduction) and found that the prevalence of bilateral HI was higher in the urban district (10.5% vs. 4.7%)³⁵. However, they did not include sensory HI, and therefore it is difficult to compare their data with our findings directly.

Some studies have investigated the possible etiological factors of the high prevalence rate of CHI in rural populations. A study on 335 school children between 6 and 19 years of age in an impoverished area of Peru identified the following risk factors for CHI: neonatal

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4 52 jaundice, seizure, hospitalization, recurrent otitis media, past otorrhea, family history of HI
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6 53 at < 35 years, tympanic membrane abnormality, cerumen impaction, and eustachian tube
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8 54 dysfunction². This study proposed that untreated middle ear disease in the context of limited
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10 55 access to pediatric care may be a major risk factor for rural CHI. In a rural primary school in
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12 56 south India, hearing assessments were performed on 284 students (from 6 to 10 years old),
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14 57 and middle ear disease was found to be the predominant cause of CHI¹⁰. An investigation of
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16 58 HI in 75 Yemeni children (0.6-15 years) with chronic suppurative otitis media found that
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18 59 middle ear disease predominantly caused a HI of 26 to 60 dB HL³⁶. According to these
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20 60 findings, middle ear disease appears to be major cause of CHI in rural areas, mainly leading
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22 61 to HI in the range of 26-60 dB HL. In our study, we found that mild CHI (55-69 dB BEHL)
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24 62 was more prevalent in the rural areas in all years, with most of the rural-to-urban RRs
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26 63 reaching statistical significance, while the prevalence rates of CHI in the other two higher
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28 64 severity categories (≥ 70 dB HL) were similar between rural and urban areas. Therefore, we
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30 65 speculate that a higher prevalence of untreated middle ear disease in rural areas contributed,
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32 66 at least in part, to the rural-urban differences observed in our study.
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35 67 In each year, the prevalence rates of CHI in both rural and urban areas increased with
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37 68 age. This finding was also noted in the Metropolitan Atlanta Developmental Disabilities
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39 69 Surveillance Program in the United States³⁷, which found that the prevalence rate of CHI >
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41 70 40 dB HL increased steadily from 6.7/10000 among 3-year-old children to 13.8/10000 for
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43 71 10-year-olds. Likewise, a study in the United Kingdom found that the prevalence rate of CHI
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45 72 > 40 dB HL rose from 9.1/10000 among 3 year-old children to 16.5/10000 among children 9
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47 73 to 16 years old³⁸. Because HI was rarely fatal and a substantial proportion of serious cases
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49 74 were not curable³³, it is reasonable that age appears to be a main determinant of the
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51 75 prevalence rate of CHI. In addition, both newly acquired HI and the progress of impairment
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53 76 severity might also contribute to the increasing trend in the prevalence of CHI associated
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55 77 with age.
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We found that the prevalence rates of CHI in the age group < 3 years significantly increased over the years. In urban areas, the rates increased significantly by 79 % from 2004 to 2010, and they increased significantly by 71% in rural areas. We speculated that one of the main causes of this was the implementation of the newborn hearing screening (NHS) program in Taiwan. As early diagnosis and early intervention of congenital HI has demonstrated effectiveness in reducing its negative impacts on a child's development, the Health Promotion Administration of Taiwan began the promotion of NHS using otoacoustic emission and automated auditory brainstem response in 2003³⁹. We believe that through increasing awareness of parents and professionals and promoting easier access to NHS, the registration of CHI cases have increased. According to Taiwan's official reports, the participation rates of newborn hearing screenings have increased from 4.0% in 2002 to 71.1% in 2010, and 97.8% of the baby-delivering institutions offered NHS services in 2013⁴⁰. Another possible cause is that Taiwan Health Promotion Administration has also implemented the Hearing Screening Plan for Pre-School Age Children in communities and kindergartens. In 2013, for example, 138197 children were thus screened, yielding a screening rate of 81.6%, compared to 30.3% in 2002⁴⁰.

In contrast with previous studies, our study has some unique features. While most previous studies were cross-sectional surveys, we have data on the same population over time. In most previous large-scale studies, data collection was just a one-time effort, but our study included seven years-worth of data, which allows for the assessment of time trends. In addition, our study has a very large number of cases, over 3533 cases in 2010 alone, and therefore we can generate reliable statistical estimates. We also have specific information on severity, which is rarely reported by large-scale studies.

However, our study also has some limitations. We used "administrative prevalence" data, which did not cover cases that were not detected or never received services from the administration. Also, data on individual cases provided by the registry were limited, which

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104 hindered the study of the etiology of the differences between rural and urban areas.
105 Investigations to clarify the etiology of the difference should be performed, which would
106 help prevention and health education to reduce the risk of CHI. Furthermore, we used
107 city/county as the unit for observation, but there may be both urban and rural townships
108 within a county. Therefore, using township as the unit of study may lead to more precise
109 classification. Unfortunately, such data were unavailable from the Taiwan government.
110 Nonetheless, this limitation tends to under estimate the difference in CHI prevalence
111 between rural and urban areas, instead of overestimating it, and since we observed a
112 statistically significant difference, its effect is unlikely to change our conclusions.

113 In conclusion, we found that the prevalence of CHI had remained similar from 2004 to
114 2010 in Taiwan. During this period, rural areas generally had higher prevalence rates than
115 urban areas. This difference was attributable to the higher prevalence rates of mild CHI
116 (55-69 dB BEHL). The rural-to-urban prevalence RRs generally decreased with severity. In
117 addition, we found that the prevalence rate in the age group < 3 years had increased
118 remarkably in both rural and urban areas, which might be attributable to the implementation
119 of the NHS program. We hope these findings can cast some light on the prevention and
120 control of CHI.

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127 analysis.

129 Contributor statement

130 All authors participated in the design or implementation of the study or the analysis and
131 interpretation of findings. D-C Lai, Y-C Tseng, C-Y Lin, and H-R Guo were involved in all
132 phases of the study. D-C Lai and H-R Guo contributed to the study design. H-R Guo was the
133 principal investigator. D-C Lai, C-Y Lin, and H-R Guo contributed to the implementation of
134 the study, statistical analysis, and interpretation of the efficacy results. D-C Lai, Y-C Tseng,
135 C-Y Lin, and H-R Guo had access to the study data and contributed to data interpretation.
136 The report was drafted by C-Y Lin, and all authors reviewed and revised drafts of the
137 manuscript and approved the final version.

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143 Competing interests

144 C-Y Lin, Y-C Tseng, D-C Lai and H-R Guo declare that they have no conflicts of interest.

146 Ethics approval

147 The Institution Review Board of the Ditmanson Medical Foundation Chia-Yi Christian
148 Hospital.

150 Provenance and peer review

151 Not commissioned; externally peer reviewed.

152 Data sharing statement

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153 No additional data available.

For peer review only

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STROBE 2007 (v4) Statement—Checklist of items that should be included in reports of cohort studies

Section/Topic	Item #	Recommendation	Reported on page #
Title and abstract	1	(a) Indicate the study’s design with a commonly used term in the title or the abstract	1-2
		(b) Provide in the abstract an informative and balanced summary of what was done and what was found	2-3
Introduction			
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported	5
Objectives	3	State specific objectives, including any prespecified hypotheses	5
Methods			
Study design	4	Present key elements of study design early in the paper	7
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection	6-7
Participants	6	(a) Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up	6-7
		(b) For matched studies, give matching criteria and number of exposed and unexposed	Not applicable
Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable	7-8
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group	6-7
Bias	9	Describe any efforts to address potential sources of bias	7
Study size	10	Explain how the study size was arrived at	Not applicable
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why	6-7
Statistical methods	12	(a) Describe all statistical methods, including those used to control for confounding	7-8
		(b) Describe any methods used to examine subgroups and interactions	7-8
		(c) Explain how missing data were addressed	Not applicable
		(d) If applicable, explain how loss to follow-up was addressed	Not applicable
		(e) Describe any sensitivity analyses	Not applicable
Results			

Participants	13*	(a) Report numbers of individuals at each stage of study—eg numbers potentially eligible, examined for eligibility, confirmed eligible, included in the study, completing follow-up, and analysed	8
		(b) Give reasons for non-participation at each stage	Not applicable
		(c) Consider use of a flow diagram	Not applicable
Descriptive data	14*	(a) Give characteristics of study participants (eg demographic, clinical, social) and information on exposures and potential confounders	8-9
		(b) Indicate number of participants with missing data for each variable of interest	Not applicable
		(c) Summarise follow-up time (eg, average and total amount)	8
Outcome data	15*	Report numbers of outcome events or summary measures over time	10-11
Main results	16	(a) Give unadjusted estimates and, if applicable, confounder-adjusted estimates and their precision (eg, 95% confidence interval). Make clear which confounders were adjusted for and why they were included	8-13
		(b) Report category boundaries when continuous variables were categorized	8-9
		(c) If relevant, consider translating estimates of relative risk into absolute risk for a meaningful time period	8-9
Other analyses	17	Report other analyses done—eg analyses of subgroups and interactions, and sensitivity analyses	12-13
Discussion			
Key results	18	Summarise key results with reference to study objectives	20
Limitations	19	Discuss limitations of the study, taking into account sources of potential bias or imprecision. Discuss both direction and magnitude of any potential bias	20
Interpretation	20	Give a cautious overall interpretation of results considering objectives, limitations, multiplicity of analyses, results from similar studies, and other relevant evidence	17-19
Generalisability	21	Discuss the generalisability (external validity) of the study results	19
Other information			
Funding	22	Give the source of funding and the role of the funders for the present study and, if applicable, for the original study on which the present article is based	21

*Give information separately for cases and controls in case-control studies and, if applicable, for exposed and unexposed groups in cohort and cross-sectional studies.

Note: An Explanation and Elaboration article discusses each checklist item and gives methodological background and published examples of transparent reporting. The STROBE checklist is best used in conjunction with this article (freely available on the Web sites of PLoS Medicine at <http://www.plosmedicine.org/>, Annals of Internal Medicine at <http://www.annals.org/>, and Epidemiology at <http://www.epidem.com/>). Information on the STROBE Initiative is available at www.strobe-statement.org.