

Preplanned Studies

Congenital Clubfoot Prevalence and Folic Acid Supplementation — Five Counties, Shanxi Province, China, 2003–2024

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Summary

What is already known about this topic?

Congenital clubfoot is a common musculoskeletal deformity and its prevalence varies across regions. In recent years, the overall prevalence of congenital clubfoot among perinatal infants has shown an upward trend in China.

What is added by this report?

This study demonstrated a significant increasing trend in isolated congenital clubfoot after 2012, whereas non-isolated cases continued to decline in the well-recognized high-prevalence region of birth defects in northern China, highlighting the distinct temporal patterns between congenital clubfoot subtypes.

What are the implications for public health practice?

Continued surveillance is needed to monitor changing patterns of congenital clubfoot subtypes and clarify factors underlying the increase in isolated congenital clubfoot.

live births, including 0.26 per 1,000 for isolated cases and 0.20 per 1,000 for non-isolated cases. Joinpoint analysis identified a 2012 inflection for isolated congenital clubfoot (annual percentage change [APC]=−11.85% pre-2012 and 11.89% post-2012). The ITS showed no immediate effect of the folic acid policy; however, isolated congenital clubfoot increased after 2012 (trend prevalence rate ratio [PRR]=1.230, $P<0.05$).

Conclusion: Isolated and non-isolated congenital clubfoot showed distinct temporal patterns. Continued surveillance is needed to clarify the increase in isolated cases after 2012.

ABSTRACT

Introduction: Congenital clubfoot is a common musculoskeletal defect that can occur as a secondary manifestation of neural tube defects (NTDs). This study estimated the prevalence and trends of congenital clubfoot in Shanxi Province, an area with a high incidence of NTDs.

Methods: Data from a population-based birth defect surveillance system covering five counties in Shanxi Province during 2003–2024 were analyzed. The cases were classified as isolated (congenital clubfoot only) or non-isolated (congenital clubfoot with additional congenital anomalies). The prevalence of congenital clubfoot was calculated and trends in prevalence were analyzed using joinpoint regression and interrupted time series (ITS) analysis.

Results: Among 296,064 births, the overall prevalence of congenital clubfoot was 0.46 per 1,000

Congenital clubfoot is one of the most common musculoskeletal deformities resulting from structural abnormalities of the foot and lower leg that cause abnormal positioning of the foot and ankle joints (1). Congenital clubfoot is generally classified as isolated or complex (non-isolated), with the complex type accounting for approximately 20%–25% of patients and occurring with other congenital anomalies (2). Genetic susceptibility and environmental exposure can contribute to the development of congenital clubfoot. Folic acid (FA) supplementation, a well-established preventive measure for neural tube defects (NTDs), is associated with reduced risk of congenital clubfoot (3). Congenital clubfoot frequently occurs as a secondary manifestation in infants with spina bifida, suggesting a potential developmental association between NTDs and congenital clubfoot (4).

Shanxi Province in northern China has historically been recognized as a region with a high incidence of NTDs. A previous study in this region showed a substantial decline in overall birth defects, particularly NTDs and orofacial clefts, whereas the incidence of congenital heart defects increased. However, the prevalence and temporal trends of congenital clubfoot in this population have not been systematically

examined. Given the close association between congenital clubfoot and NTDs, and the potential long-term disability caused by congenital clubfoot, understanding its epidemiological patterns is important for public health. Therefore, this study aimed to estimate the birth prevalence of isolated and non-isolated congenital clubfoot in Shanxi Province from 2003 to 2024, with a particular focus on cases of spina bifida.

Data from a population-based birth defect surveillance system covering five counties in Shanxi Province from 2003 to 2024, an area with a relatively high prevalence of NTDs, were analyzed. A population-based birth defect surveillance system was introduced to the study area in October 2002 (5–6). All live births (≥ 28 weeks), all stillbirths (≥ 20 weeks), and pregnancy terminations at any gestational age following the prenatal diagnosis of birth defects were recorded. Live-born infants with birth defects were included in the surveillance system if they had a gestational age of at least 20 weeks, and a birth defect was diagnosed by 6 weeks of age. For stillbirths and pregnancy terminations, the diagnoses were established at the time of delivery or termination. Congenital clubfoot cases were coded as Q66 according to the International Statistical Classification of Diseases and Related Health Problems, 10th revision (ICD-10), initially diagnosed by local maternal-fetal specialists and confirmed by pediatricians at Peking University. All congenital clubfoot cases were classified as isolated (congenital clubfoot without any other anomalies) or non-isolated (congenital clubfoot accompanied by additional congenital anomalies) using the categorization derived from the original reported data.

The prevalence per 1,000 live births was calculated for total, isolated, and non-isolated congenital clubfoot; the denominator was the total number of all live births, while the numerator was all cases observed in the surveillance system. Trends were assessed using the Cochran-Armitage trend tests. To reduce random year-to-year fluctuations due to small case counts, three-year centered rolling averages were calculated, smoothing short-term variability while preserving overall trends. Joinpoint regression analysis was performed to identify significant changes in the trend slopes and estimate the annual percentage change (APC) and average annual percentage change (AAPC). The maximum number of joint points was set to three. Model selection was based on the Monte Carlo permutation test with a significance level of 0.05. Based on the joinpoint results and relevant policy

interventions, an interrupted time-series (ITS) analysis was conducted using generalized linear models with a quasi-Poisson distribution, including the logarithm of total live births as an offset to account for overdispersion. The models incorporated terms for baseline time trend (underlying temporal pattern), level changes at each intervention or joint point (immediate changes in prevalence), and post-intervention trend changes (changes in slope over time). Three ITS models were evaluated: 1) the 2009 FA policy, 2) the 2012 trend inflection, and 3) a combination of these. In ITS, prevalence rate ratios (PRRs) quantify the relative changes in case counts. Categorical variables are summarized as frequencies and percentages, and group differences were assessed using the chi-square test. Statistical significance was defined as a two-tailed $P < 0.05$. Analyses were performed using SPSS (version 25.0, SPSS Inc., Chicago, IL, USA), R (version 4.5.1, R Foundation for Statistical Computing, Vienna, Austria; packages DescTools, dplyr, tidyr, zoo, and ggplot2), and JoinPoint (version 5.0.2, Statistical Research and Applications Branch, National Cancer Institute, Bethesda, MD, USA; Information Management Services, Inc., Calverton, MD, USA).

From 2003 to 2024, the Shanxi surveillance system recorded 296,064 births and 137 cases of congenital clubfoot (Supplementary Table S1, available at <https://weekly.chinacdc.cn/>), with 56.2% (77/137) isolated congenital clubfoot and 43.8% (60/137) non-isolated congenital clubfoot. No significant differences were observed between the two groups in terms of maternal residence, infant sex, or clubfoot laterality, whereas the gestational age differed.

The overall prevalence of congenital clubfoot was 0.46 per 1,000 live births [95% confidence interval (CI): 0.39, 0.55], including 0.26 per 1,000 for isolated cases (95% CI: 0.21, 0.32) and 0.20 per 1,000 for non-isolated cases (95% CI: 0.16, 0.26). From 2003 to 2024, the prevalence of total and non-isolated congenital clubfoot gradually declined, with values falling from 0.93/1,000 and 0.37/1,000 in 2003 to 0.55/1,000 (2024, total cases) and 0.07/1,000 (2022–2024, non-isolated cases), respectively, whereas isolated congenital clubfoot showed no significant downward trend. Trend analysis confirmed a statistically significant decrease in non-isolated congenital clubfoot ($P < 0.05$) (Figure 1A–C). The incidence of non-isolated congenital clubfoot with spina bifida was 0.14/1,000 overall. It decreased from 0.31/1,000 in 2003 to 0.07/1,000 in 2022–2024 ($P < 0.05$) with a consistent downward trend (Figure 1D).

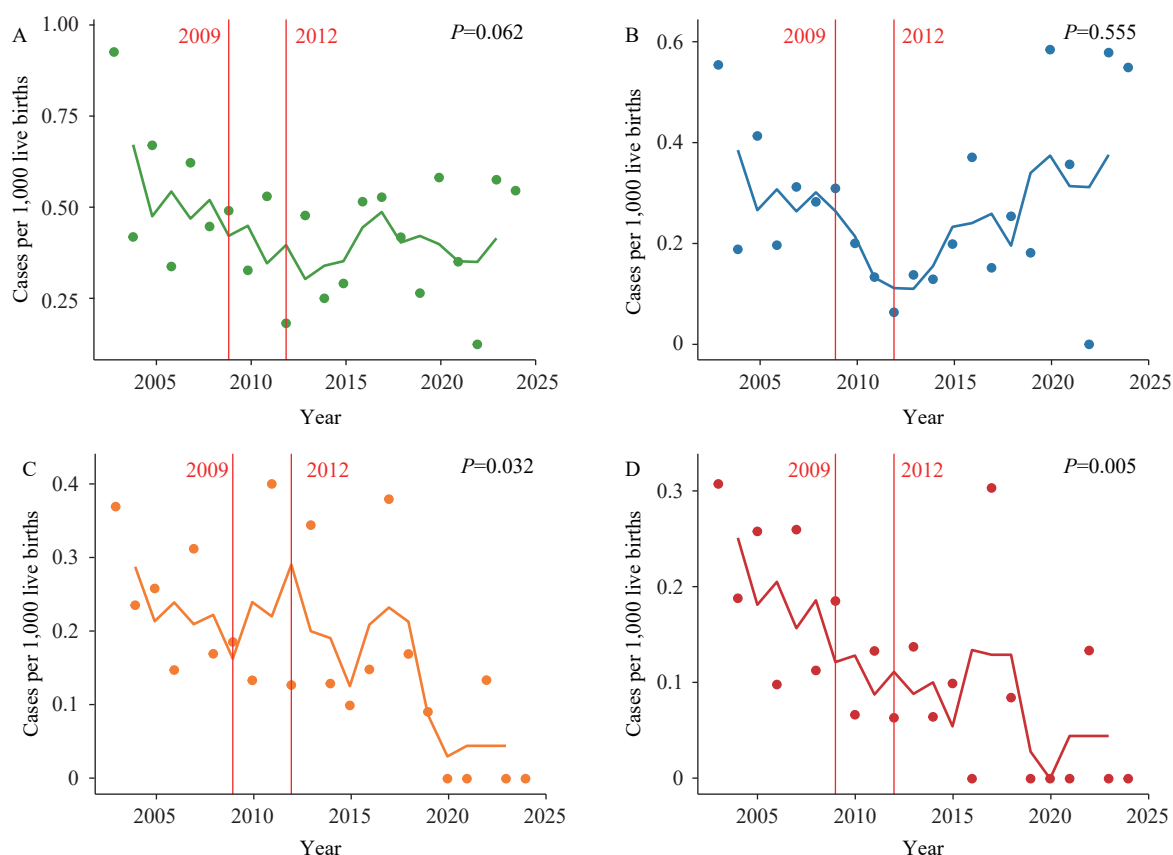


FIGURE 1. Time trends in the prevalence of congenital clubfoot in five counties, Shanxi Province, 2003–2024. (A) Total congenital clubfoot. (B) Isolated congenital clubfoot. (C) Non-isolated congenital clubfoot. (D) Non-isolated congenital clubfoot with spina bifida.

Note: Data for 2003 were available for only four counties: Zezhou, Pingding, Taigu, and Shouyang. For 2023 and 2024, data were available for Xiyang, Pingding, Taigu, and Shouyang. The data for all five counties are presented for all other years.

Abbreviation: NTDs=neural tube defects.

* P for Cochran-Armitage trend test.

Joinpoint regression analysis further examined temporal changes in prevalence (Table 1). No significant joinpoints were identified for total congenital clubfoot, non-isolated congenital clubfoot, or non-isolated congenital clubfoot with spina bifida, so trends were summarized using the AAPC. Total congenital clubfoot showed a modest but significant decline (AAPC=−2.62%, $P=0.048$), while non-isolated congenital clubfoot showed no significant overall trend (AAPC=−2.84%, $P=0.129$). Non-isolated congenital clubfoot with spina bifida declined significantly (AAPC=−4.21%, $P=0.035$). In contrast, isolated congenital clubfoot exhibited a significant joinpoint in 2012, with a decreasing trend before 2012 (APC=−11.85%, $P=0.016$), followed by an increasing trend thereafter (APC=11.89%, $P=0.023$).

Interrupted time series (ITS) models were used to assess the association between the 2009 FA supplementation policy and the 2012 statistical

inflection point (Table 2). For patients with isolated congenital clubfoot, no significant immediate changes were observed after either intervention. A significant increasing trend was detected after 2012 (trend PRR=1.230, 95% CI: 1.074, 1.408, $P<0.05$). No significant trend change was observed following the FA policy (trend PRR=1.163, 95% CI: 0.948, 1.426). For total congenital clubfoot, non-isolated congenital clubfoot, and non-isolated congenital clubfoot with spina bifida, neither the level nor trend changes associated with the FA policy or the 2012 inflection point were statistically significant. The combined ITS models yielded similar results (Supplementary Table S2 and Supplementary Figure S1, available at <https://weekly.chinacdc.cn/>).

DISCUSSION

In this population-based study covering over

TABLE 1. APC and AAPC in the prevalence of congenital clubfoot in five counties, Shanxi Province, 2003–2024.

Types of cases	Period	APC (95% CI)	P
Total congenital clubfoot*			
Trend	2003–2024	–2.62 (–5.15, –0.02)	0.048
Isolated congenital clubfoot			
Trend1	2003–2012	–11.85 (–20.21, –2.60)	0.016
Trend2	2012–2024	11.89 (1.74, 23.07)	0.023
AAPC	2003–2024	1.02 (–5.26, 7.72)	0.756
Non-isolated congenital clubfoot*			
Trend	2003–2024	–2.84 (–6.46, 0.92)	0.129
Non-isolated congenital clubfoot with spina bifida*			
Trend	2003–2024	–4.21 (–7.93, –0.32)	0.035

Abbreviation: APC=annual percentage change; AAPC=average annual percentage change; CI=confidence interval.

* Trends were summarized using the AAPC.

TABLE 2. Changes in congenital clubfoot prevalence following folic acid supplementation and the joinpoint identified in 2012 in five counties, Shanxi Province, 2003–2024: an interrupted time series analysis.

Types of cases	Model	PRR (95% CI)
Total congenital clubfoot	Time	0.909 (0.799, 1.034)
	FA policy level change	1.007 (0.536, 1.893)
	FA policy trend change	1.099 (0.958, 1.261)
	Time	0.941 (0.874, 1.013)
	2012 joinpoint level change	0.871 (0.473, 1.602)
	2012 joinpoint trend change	1.091 (0.987, 1.206)
Isolated congenital clubfoot	Time	0.909 (0.751, 1.101)
	FA policy level change	0.697 (0.264, 1.838)
	FA policy trend change	1.163 (0.948, 1.426)
	Time	0.909 (0.820, 1.008)
	2012 joinpoint level change	0.732 (0.301, 1.780)
	2012 joinpoint trend change	1.230 (1.074, 1.408)
Non-isolated congenital clubfoot	Time	0.909 (0.736, 1.123)
	FA policy level change	1.543 (0.564, 4.222)
	FA policy trend change	1.015 (0.809, 1.275)
	Time	0.979 (0.870, 1.103)
	2012 joinpoint level change	1.116 (0.442, 2.821)
	2012 joinpoint trend change	0.912 (0.764, 1.088)
Non-isolated congenital clubfoot with spina bifida	Time	0.884 (0.693, 1.128)
	FA policy level change	1.095 (0.313, 3.834)
	FA policy trend change	1.048 (0.798, 1.375)
	Time	0.897 (0.774, 1.039)
	2012 joinpoint level change	1.113 (0.304, 4.082)
	2012 joinpoint trend change	1.036 (0.822, 1.306)

Note: For each intervention, two PRRs are presented: one for level change (immediate effect) and one for trend change (change in the slope over time). All the models were adjusted for the underlying time trend and included an offset term for the logarithm of total births. Only separate policy models are presented in this table; the results from the model that includes both policies simultaneously are provided in [Supplementary Table S2](https://weekly.chinacdc.cn/) (available at <https://weekly.chinacdc.cn/>).

Abbreviation: PRR=prevalence rate ratio; FA=folic acid; CI=confidence interval.

290,000 births, the overall prevalence of congenital clubfoot was estimated to be 0.46 per 1,000 live births, with isolated congenital clubfoot accounting for 0.26 per 1,000 births and non-isolated congenital clubfoot for 0.20 per 1,000 births. An increasing trend in isolated congenital clubfoot prevalence was observed after the joinpoint identified in 2012, whereas no statistically significant changes were identified for the FA policy.

Compared with the global data, the prevalence of congenital clubfoot in Shanxi Province was relatively low. The reported rates range from 1.18 per 1,000 globally to 1.29 per 1,000 people in the United States (7). In China, the prevalence varies across regions. A cohort in Beijing from 2013 to 2018 showed a prevalence of 0.69 per 1,000 singleton infants (3), while lower rates were observed in Hunan and Guangxi provinces. Overall, the prevalence in Shanxi Province was slightly higher than the national average, but remained lower than that in some regions. Both isolated and non-isolated congenital clubfoot are relatively rare in Shanxi, comparable to Sweden (2016–2019), where isolated cases occurred at 1.24 per 1,000 live births and non-isolated cases comprised 8% of all cases, resulting in an overall prevalence of 1.35 per 1,000 (8).

Our study demonstrated a marked decline in the incidence of non-isolated congenital clubfoot accompanied by spina bifida over the past two decades. Among the NTD subtypes, spina bifida is particularly prone to coexisting musculoskeletal abnormalities, with congenital clubfoot being one of the most common. Neurological deficits caused by NTDs may impair fetal motor function, thereby reducing intrauterine movement and contributing to congenital clubfoot development (9). Previous evidence has reported that approximately 30% of NTD fetuses show congenital clubfoot on ultrasound before 24 weeks of gestation, with additional cases developing later in gestation, suggesting a progressive risk as pregnancy advances (10). Therefore, dynamic monitoring of foot morphology is crucial for fetuses with NTDs.

For isolated congenital clubfoot, the observed significant increase in prevalence after 2012 suggests a distinct temporal pattern compared to non-isolated congenital clubfoot. A population-based study of perinatal infants in China (2011–2020) also reported an increasing trend, consistent with the findings of this study on isolated congenital clubfoot (11). Several factors may have contributed to this change. Improved standardization of classification and reporting may

have reduced under-reporting and enhanced surveillance completeness. Meanwhile, advances in prenatal screening and imaging, including better ultrasound technology and increased diagnostic expertise, may have led to a greater detection of fetal limb anomalies, particularly in milder cases. Additionally, environmental risk factors and adverse prenatal exposure may have become more prominent, potentially affecting the genetic susceptibility. Notably, the joinpoint identified in 2012 represents a data-driven temporal inflection rather than a defined intervention, and its underlying causes remain uncertain. Therefore, these interpretations should be considered as exploratory.

From a broader public health perspective, congenital clubfoot represents not only a structural deformity but also a long-term neurological impairment, repeated surgical interventions, and even amputation, leading to functional disability and imposing significant psychosocial and economic burdens on affected families and the healthcare system. Although a declining trend was observed, the persistence of isolated and non-isolated cases indicated that congenital limb deformities remain an important challenge. Future efforts should integrate strengthened prenatal surveillance, long-term monitoring of congenital anomalies and their subtypes, and continued investigations of genetic and environmental mechanisms to reduce the burden of congenital clubfoot and related congenital anomalies.

Our study has several strengths. First, it is based on a population-based surveillance system with an extended study period and involves both perinatal and pre-perinatal cases, thereby providing a more comprehensive assessment of the occurrence of congenital anomalies. Second, this study conducted a detailed analysis of the prevalence and trends of isolated and non-isolated clubfoot, which has rarely been reported in China.

The findings in this report are subject to at least two limitations. First, the surveillance data were restricted to five counties, which may have limited generalizability. Second, the absence of maternal sociodemographic and lifestyle information precluded exploration of specific risk factors.

Conflicts of interest: No conflicts of interest.

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SUPPLEMENTARY MATERIALS

SUPPLEMENTARY TABLE S1. Characteristics of infants with congenital clubfoot in five counties, Shanxi Province, 2003–2024.

Characteristics	Isolated congenital clubfoot (n=77)	Non-isolated congenital clubfoot (n=60)	χ^2	P
Residence				
Rural	25 (32.5)	13 (21.7)	1.963	0.161
Urban	52 (67.5)	47 (78.3)		
Infant sex				
Male	43 (55.8)	37 (61.7)	0.866	0.352
Female	34 (44.2)	21 (35.0)		
Unknown	0 (0.0)	2 (3.3)		
Gestational weeks				
<28	8 (10.4)	19 (31.7)	9.648	0.002
≥28	69 (89.6)	41 (68.3)		
Laterality				
Bilateral	16 (20.8)	11 (18.3)	2.512	0.113
Unilateral	28 (26.3)	8 (13.4)		
Missing	33 (42.9)	41 (68.3)		
Delivery time				
Before 2009	36 (46.8)	28 (46.7)	<0.001	0.992
After 2009	41 (53.2)	32 (53.3)		

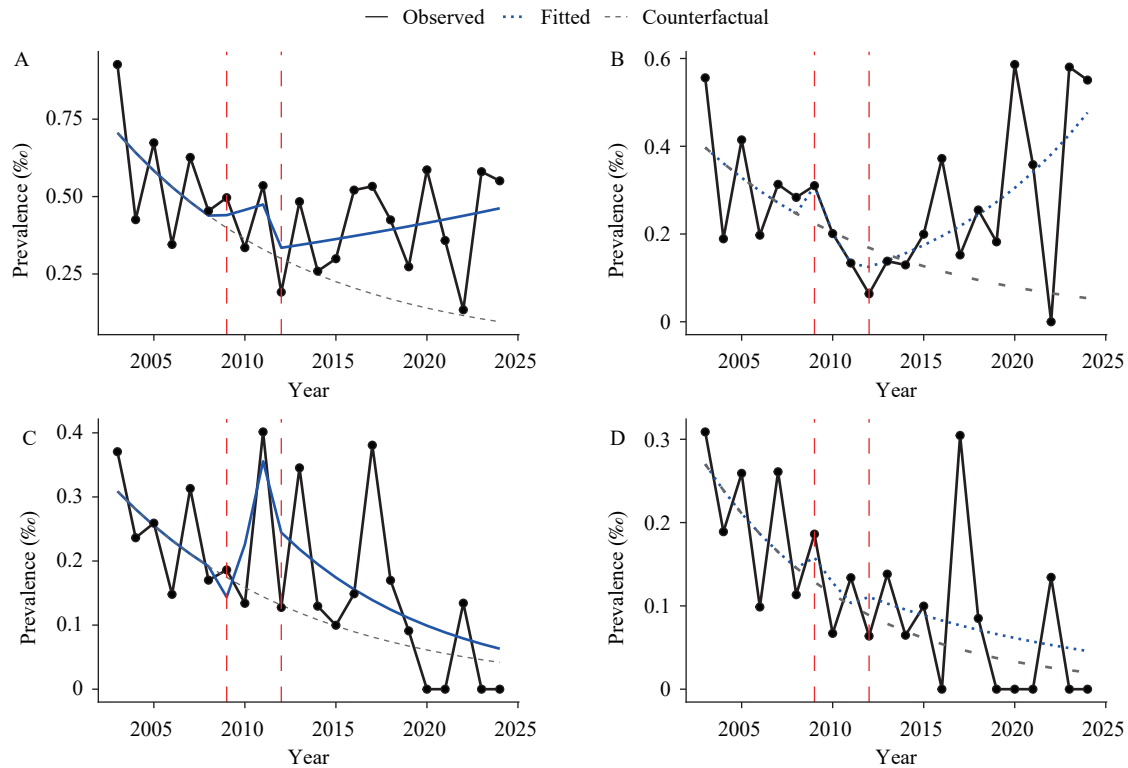
Note: Data for 2003 were available for only four counties: Zezhou, Pingding, Taigu, and Shouyang. For 2023 and 2024, data were available for Xiyang, Pingding, Taigu, and Shouyang. Data for all five counties were presented for all other years.

SUPPLEMENTARY TABLE S2. Results of interrupted time series models including both the folic acid supplementation policy and the joinpoint identified in 2012.

Types of cases	Model	PRR (95% CI)
Total congenital clubfoot	Time	0.909 (0.796, 1.038)
	FA policy level change	1.104 (0.493, 2.473)
	FA policy trend change	1.142 (0.706, 1.846)
	2012 joinpoint level change	0.679 (0.229, 2.012)
	2012 joinpoint trend change	0.989 (0.620, 1.578)
Isolated congenital clubfoot	Time	0.909 (0.761, 1.086)
	FA policy level change	1.380 (0.488, 3.901)
	FA policy trend change	0.721 (0.347, 1.498)
	2012 joinpoint level change	1.434 (0.229, 8.983)
	2012 joinpoint trend change	1.706 (0.834, 3.488)
Non-isolated congenital clubfoot	Time	0.909 (0.739, 1.119)
	FA policy level change	0.823 (0.216, 3.134)
	FA policy trend change	1.731 (0.842, 3.559)
	2012 joinpoint level change	0.437 (0.103, 1.845)
	2012 joinpoint trend change	0.568 (0.281, 1.146)
Non-isolated congenital clubfoot with spina bifida	Time	0.884 (0.683, 1.143)
	FA policy level change	1.228 (0.250, 6.040)
	FA policy trend change	0.916 (0.321, 2.613)
	2012 joinpoint level change	1.322 (0.104, 16.759)
	2012 joinpoint trend change	1.148 (0.408, 3.231)

Note: The full model includes both policies to evaluate their independent effects after mutual adjustments.

Abbreviation: PRR=prevalence rate ratio; FA=folic acid; CI=confidence interval.



SUPPLEMENTARY FIGURE S1. Fitted trends and counterfactual estimates of congenital clubfoot prevalence from interrupted time series analysis, 2003–2024. (A) Total congenital clubfoot; (B) isolated congenital clubfoot; (C) non-isolated congenital clubfoot; (D) non-isolated congenital clubfoot with spina bifida.

Note: The black solid line represents the observed prevalence of clubfoot (per 1,000 live births). The blue solid line shows the fitted values from the quasi-Poisson ITS model incorporating two temporal change points: the 2009 folic acid policy and a statistical jointpoint in 2012 identified by joinpoint regression. The grey dotted line indicates a counterfactual trend, representing the expected prevalence in the absence of these changes. The red dashed vertical lines mark the timing of the 2009 folic acid policy and the jointpoint identified in 2012.

Abbreviation: ITS=Interrupted time series.