Vital Surveillances

County-Level Hotspot Identification and Spatial Regression Analysis of Health Loss from Kashin–Beck Disease — China, 2019 and 2023

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ABSTRACT

Introduction: We analyzed the spatial distribution of years lived with disability (YLDs) among patients with Kashin–Beck disease (KBD) at the county level across the country, identified hotspot regions and the primary areas of disease burden. This provides a foundation for the prevention and control of KBD and the rational allocation of healthcare resources to regions with high disease burden.

Methods: The data were obtained from the National KBD Surveillance System. Spatial autocorrelation analysis was conducted to assess spatial clustering and to identify hotspots of YLDs in patients with KBD. Geographically weighted regression (GWR) models were used to identify counties with limited economic and healthcare resources and a high burden of health losses.

Results: Spatial aggregation of YLDs among patients with KBD was observed nationwide, with hotspots concentrated in diseased counties in western China, including Shaanxi, Gansu, and Sichuan, and in the northern regions of Heilongjiang and Inner Mongolia. Among the variables, the number of health technicians was negatively correlated with the YLD rate of patients with KBD across 2 years ($P<0.05$). Significant geographical differences were found in the spatial distribution of YLDs, with key disease burden areas in 85 northern counties, including Heilongjiang, Jilin, and Inner Mongolia, and 145 western counties, including Shaanxi, Shanxi, and other provincial-level administrative divisions.

Conclusions: YLDs among patients with KBD at the county level in China demonstrated spatial clustering, with hotspots primarily in the western regions. Strengthening the recruitment and training of health professionals in high-burden, underserved areas may help improve the quality of life of patients.

Kashin–Beck disease (KBD) is an endemic chronic osteoarticular disorder primarily affecting children (*1*). It is characterized by joint pain, limb deformities, shortened extremities, and growth retardation, often causing lifelong disability and reduced quality of life (*2*). KBD has historically been endemic to 379 counties across 13 provincial-level administrative divisions (PLADs) in China, largely along the northeast-to-southwest belt. Comprehensive interventions, including water and grain substitution, socioeconomic development, and improved healthcare, have caused its elimination and zero incidence (*3*–*4*). Nevertheless, a substantial number of individuals live with KBD and experience a significantly lower quality of life (*5*). However, targeted health interventions for these patients are not sufficiently supported by scientific evidence.

Currently, spatial epidemiological research on KBD is largely limited to individual PLADs or counties and lacks high-resolution county-level data at the national scale. This gap limits the effectiveness of the prevention and control strategies. Therefore, we conducted a spatial analysis of years lived with disability (YLDs) among patients with KBD across China's counties to assess spatial distribution patterns, identify high-burden areas, provide a theoretical basis for improving patient outcomes, optimizing healthcare resource allocation, and informing targeted prevention and control policies.

METHODS

The data for this study were sourced from the National KBD Surveillance System and the National Case Surveys conducted in 2019 and 2023 (*6*). The system conducts epidemiological surveys in KBD-endemic regions using active case-finding methods, including household visits and physical examinations. It identifies individuals based on KBD diagnostic criteria (WS/T 207-2010), which include clinically diagnosed and imaging-confirmed cases. The dataset included the demographic information of the respondents, such as sex, age, occupation, place of residence, and disease severity. The per capita gross domestic product (GDP) was extracted from the China County Statistical Yearbook, and per capita income was derived from the same surveillance system. Data on the number of hospital beds and healthcare technicians were obtained from the National Bureau of Statistics of China. Missing values in county-level economic and healthcare indicators were addressed through multiple imputations by chained equations (MICE) (*7*). Specifically, this method preserves the inherent variability of the original data and, consequently, improves the accuracy and statistical efficiency of regression estimates.

We used YLDs to assess the loss of healthy life expectancy in patients with KBD due to the non-lethal nature of KBD. This measure effectively reflects the burden of non-fatal disability caused by different severities of KBD at both the individual and societal levels (*8*).

The YLD was calculated as follows:

$$\text{YLD} = N \times DW,$$

where N represents the number of patients with KBD and DW denotes the disability weight. Because previous studies have not reported the disability weights for KBD and its clinical manifestations resemble those of rheumatoid arthritis (*9*), disability weights for rheumatoid arthritis from the Global Burden of Disease (GBD) 2019 study were used as proxies for KBD. These values were 0.117, 0.317, and 0.581 for degrees I, II, and III, respectively (*10*).

The ArcGIS software (version 10.8; ESRI, California, USA) was used to assess the overall spatial aggregation of YLDs among patients with KBD using global spatial autocorrelation, whereas the Getis-Ord Gi^* statistic was employed to identify local hotspots (*11*). The ordinary least squares (OLS) model was used to initially examine the relationships between per capita gross domestic product (GDP), per capita income, number of hospital beds, number of health technicians, and the YLD rate among patients with KBD. Multicollinearity was assessed using the variance inflation factor (VIF), with VIF values of <10 indicating acceptable collinearity. Subsequently, a geographically weighted regression (GWR) model was used to conduct localized estimations, revealing spatial non-stationarity in the relationships between each variable and the YLD rate across endemic areas and counties. These patterns were visualized using local regression coefficients. Key areas for disease management and priority interventions in adult patients with KBD have been identified (*12*–*13*).

RESULTS

General KBD Characteristics

KBD is endemic to 13 Chinese PLADs. Data during the first national surveillance in 2019 were collected from 325 endemic counties, reporting 164,914 prevalent cases, including 105,142 degree I, 46,702 degree II, and 13,070 degree III cases. By 2023, data collected from 379 endemic counties indicated 165,348 prevalent cases, including 103,969 degree I, 49,796 degree II, and 11,583 degree III cases.

Spatial Distribution of YLDs in Patients with KBD

Significant regional differences were observed in the YLD rates and the YLDs of KBD in 2019 and 2023. The regions with severe loss of healthy life expectancy in 2019 were the disease area counties of Yantang County (497.87 YLD/10,000, 2,074.54 YLDs) and Aba County (289.53 YLD/10,000, 1,604.27 YLDs), both are in Sichuan Province. By 2023, the regions with severe losses were Heshui County (86.02 YLD/10,000, 1,621.58 YLDs) and Ning County (44.68 YLD/10,000, 1,367.49) in Gansu Province.

Moran's I indices for KBD-related YLDs and YLD rates were 0.11 and 0.07, respectively, in 2019, and 0.17 and 0.08, respectively, in 2023, with P<0.05 in both years. These results indicate significant spatial clustering of KBD-related health losses at the county level nationwide (Supplementary Figure S1, available at https://weekly.chinacdc.cn/).

The Getis-Ord-Gi^* statistic was applied to further identify hotspots. In 2019, the hotspot areas of YLD among patients with KBD were primarily concentrated in the central and western regions of China, covering 155 endemic counties across PLADs, including Henan, Shaanxi, Qinghai, Gansu, Sichuan, and Shanxi (Table 1). The hotspot areas had shifted by 2023, with concentrations in both the western and northern regions of China spanning PLADs such as Shaanxi, Qinghai, Gansu, Sichuan, Inner Mongolia, and Heilongjiang and comprising 90 endemic counties

TABLE 1. Hotspot areas of KBD YLDs rates at the county level in 2019 identified by local Getis-Ord Gi* analysis.

Hotspot type	PLADs	Counties
Hotspot: 99% confidence	Gansu	Qinzhou District, Maiji District, Qingshui County, Qin'an County, Wushan County, Zhangjiachuan Hui Autonomous County, Kongtong District, Jingchuan County, Lingtai County, Chongxin County, Zhuanglang County, Huating City, Xifeng District, Qingcheng County, Huan County, Huachi County, Heshui County, Zhengning County, Ning County, Zhenyuan County, Longxi County, Weiyuan County, Zhang County, Min County, Wudu District, Cheng County, Wen County, Dangchang County, Kang County, Xihe County, Li County, Hui County, Liangdang County, Kangle County, Hezheng County, Zhuoni County, Luqu County
	Henan	Shanzhou District, Lushi County, Lingbao City
	Qinghai	Guide County, Xinghai County, Banma County
	Shaanxi	Baqiao District, Lintong District, Lantian County, Zhouzhi County, Wangyi District, Yintai District, Yaozhou District, Yijun County, Weibin District, Jintai District, Chencang District, Fengxiang County, Qishan County, Fufeng County, Mei County, Long County, Qianyang County, Linyou County, Feng County, Taibai County, Sanyuan County, Jingyang County, Qian County, Liquan County, Yongshou County, Changwu County, Xunyi County, Chunhua County, Binzhou City, Linwei District, Huazhou District, Tongguan County, Heyang County, Chengcheng County, Pucheng County, Baishui County, Fuping County, Hancheng City, Baota District, Ansai District, Zhidan County, Ganquan County, Fu County, Luochuan County, Yichuan County, Huanglong County, Huangling County, Nanzheng District, Xixiang County, Mian County, Ningqiang County, Lueyang County, Hanbin District, Shiquan County, Ningshan County, Shangzhou District, Luonan County, Zhen'an County, Zhashui County
	Shanxi	JiShan County, Pinglu County, Ruicheng County
	Sichuan	Beichuan Qiang Autonomous County, Pingwu County, Jiangyou City, Wangcang County, Qingchuan County, Dazhu County, Yucheng District, Hanyuan County, Shimian County, Tianquan County, Tongjiang County, Nanjiang County, Maerkang City, Wenchuan County, Mao County, Songpan County, Jiuzhaigou County, Jinchuan County, Xiaojin County, Heishui County, Rangtang County, Aba County, Ruo'ergai County, Hongyuan County, Luding County, Danba County, Daofu County, Ganzi County, Xinlong County, Dege County, Seda County, Mianning County
Hotspot: 95% confidence	Henan	Luoning County, Mianchi County
	Inner Mongolia	Uxin Banner
	Shanxi	Wanrong County, Wenxi County, Jiang County, Yuanqu County, Xia County, Xiangfen County, Ji County, Xiangning County, Daning County, Yonghe County
	Shaanxi	Yanchang County
Hotspot: 90% confidence	Shanxi	Xi County, Pu County, Shilou County
	Shaanxi	Yuyang District

Abbreviation: KBD=Kashin–Beck disease; YLDs=years lived with disability; PLAD=provincial-level administrative division.

(Table 2).

Spatial Regression Analysis of YLDs in Patients with KBD

The data were standardized to eliminate the effects of different units and magnitudes among the variables. An OLS model was used to assess the association between the YLD rate of patients with KBD and the different socioeconomic factors globally. All independent variables had VIF values less than 10, indicating acceptable levels of multicollinearity. In 2019, the YLD rates of patients with KBD were positively associated with per capita GDP, per capita income, and the number of hospital beds, although none of these associations were statistically significant. In 2023, the YLD rates were negatively associated with per capita GDP, the number of hospital beds, and positively associated with per capita income, with none of these associations reaching statistical significance. Furthermore, a significant negative association (P<0.05) was observed between the number of health technicians and YLD rates in both 2019 and 2023 (Table 3).

We used corrected Akaike information criterion (AICc) as the model selection metric to apply the GWR model that yielded AICc values of 769.14 and 845.17 in 2019 and 2023, respectively, whereas the OLS model produced values of 859.77 and 863.15, respectively. Thus, the GWR model provided a better fit for both years.

The GWR parameter estimation results revealed varying coefficients across different counties, further confirming the spatial heterogeneity in the determinants of YLD rates for KBD (Table 3). The OLS model results demonstrated that the number of

TABLE 2. Hotspot areas of KBD YLDs rates at the county level in 2023 identified by local Getis-Ord Gi* analysis.

Hotspot type	PLADs	Counties
Hotspot: 99% confidence	Gansu	Qinzhou District, Qin'an County, Wushan County, Longxi County, Weiyuan County, Zhang County, Min County, Wudu District, Cheng County, Wen County, Dangchang County, Kang County, Xihe County, Li County, Hui County, Kangle County, Hezheng County, Zhuoni County, Luqu County
	Inner Mongolia	Morin Dawa Daur Autonomous Banner
	Qinghai	Guide County, Xinghai County, Banma County
	Shaanxi	Nanzheng District, Ningqiang County, Lueyang County
	Sichuan	Beichuan Qiang Autonomous County, Pingwu County, Jiangyou City, Wangcang County, Qingchuan County, Yucheng District, Hanyuan County, Shimian County, Tianquan County, Maerkang City, Wenchuan County, Lixian County, Mao County, Songpan County, Jiuzhaigou County, Jinchuan County, Xiaojin County, Heishui County, Rangtang County, Aba County, Ruo'ergai County, Hongyuan County, Luding County, Danba County, Daofu County, Ganzi County, Xinlong County, Dege County, Seda County, Mianning County
	Xizang	Jiangda County, Gongjue County
Hotspot: 95% confidence	Gansu	Maiji District, Qingshui County, Zhangjiachuan Hui Autonomous County, Zhuanglang County, Liangdang County
	Heilongjiang	Nenjiang City, Mohe City, Huma County, Tahe County
	Inner Mongolia	Arun Banner, Eerguna City, Genhe City
	Shaanxi	Feng County, Xixiang County, Mian County
	Sichuan	Tongjiang County, Nanjiang County
	Xizang	Chaya County, Mangkang County
Hotspot: 90% confidence	Gansu	Kongtong District, Chongxin County, Huating City
	Heilongjiang	Aihui District
	Shaanxi	Weibin District, Jintai District, Chencang District, Fengxiang County, Long County, Qianyang County, Taibai County, Shiquan County
	Xizang	Zuogong County

Abbreviation: KBD=Kashin–Beck disease; YLDs=years lived with disability; PLAD=provincial-level administrative division.

health technicians was always negatively associated with YLD rates in both years. This negative correlation was observed in 230 counties across 10 PLADs, including Shanxi, Inner Mongolia, Jilin, Heilongjiang, Gansu, Qinghai, Sichuan, Henan, Shaanxi, and Xizang (Supplementary Table S1, available at https://weekly.chinacdc.cn/). Thus, there is a need to increase human resource allocation in primary healthcare to strengthen disease prevention and control systems.

DISCUSSION

In this study, we selected YLDs over single measures, such as prevalence, to more comprehensively reflect the population-level and societal burden of nonfatal disability associated with KBD. In addition, the use of YLDs provides a more in-depth evaluation of the disease burden among existing patients with KBD across the country.

The YLDs among patients with KBD exhibited significant spatial clustering at the county level. The extent of YLD hotspots declined in 2023 compared to that in 2019; however, persistent hotspots remained, primarily located in 81 endemic counties in Shaanxi, Gansu, and Sichuan in the western region and 9 counties in Heilongjiang and Inner Mongolia in the northern region. The formation of these hotspots may be associated with long-standing geographic and environmental characteristics at the district and county levels. Historically, regional geographic environments have shaped unique living and dietary patterns among local populations, influencing the incidence of KBD to varying extents over time. Consequently, substantial regional disparities in KBD prevalence have emerged, contributing to the current spatial heterogeneity in the YLD rates among patients with KBD (*14*).

Previous studies have reported that KBD occurs in areas with lagging economic development (*15*). However, we found no statistically significant correlation between YLD rates and local economic levels in both 2019 and 2023. The implementation of policies such as the National Twelfth Five-Year Plan for the Prevention and Control of Endemic Diseases

TABLE 3. Descriptive statistics of OLS analysis results and GWR model parameter estimates.

Year	Variable	OLS model		GWR model			
		Coefficient	*P*	Max	Min	Mean	SD
2019	Per capita GDP	0.060	0.299	14.950	−0.995	0.686	2.124
	Per capita income	0.018	0.757	0.627	−0.303	0.638	0.219
	Number of hospital beds	0.003	0.971	0.303	−0.641	0.020	0.133
	Number of health technicians	−0.184	0.007*	1.818	−1.037	−0.150	0.244
2023	Per capita GDP	−0.008	0.887	0.251	−0.335	−0.028	0.114
	Per capita income	0.064	0.269	0.482	−0.204	0.012	0.140
	Number of hospital beds	−0.053	0.359	0.168	−0.385	−0.082	0.153
	Number of health technicians	−0.135	0.019*	0.295	−0.577	−0.149	0.173

Abbreviation: SD=standard deviation; OLS=ordinary least squares; GWR=geographically weighted regression; GDP=gross domestic product.

* P<0.05, denoting statistical significance.

and the Three-Year Special Action Plan for Tackling Endemic Diseases (2018–2020), which include relocation, targeted care, health poverty relief, and increased funding for disease control, may have significantly improved the economic conditions in KBD-endemic areas. Narrowing regional economic disparities, and increased investment in medical resources (e.g., hospital construction, medical equipment, and drug supply) in these regions have effectively alleviating uneven distribution of healthcare resources.

Despite this progress, our study identified a significant negative correlation between the number of health technicians and YLD rates for both 2019 and 2023. Thus, a shortage of specialized health personnel may limit patients' access to adequate treatment and rehabilitation services, thereby negatively affecting their quality of life.

Therefore, targeted interventions should be introduced to identify areas with high YLD rates and insufficient healthcare personnel. These include the recruitment of professional health technicians, tailored training programs, and optimized medical resource allocation to enhance KBD prevention and treatment in underserved areas. In addition, efforts should be made to increase the supply of medications and medical equipment to primary healthcare institutions, ensure the availability of symptomatic treatments, and ultimately improve the quality of life of patients with KBD.

Future studies could include additional metrics such as functional impairment scores (e.g., WOMAC and HAQ) and quality of life measures (e.g., SF-36) among patients with KBD to enhance the framework for evaluating the health burden of KBD and provide a more comprehensive and systematic understanding of its impact.

This study had two main limitations. Firstly, no established studies have defined the disease-specific disability weights for patients with KBD. Therefore, disability weights for rheumatoid arthritis from the GBD 2019 study were used as proxies. Secondly, a small proportion (<5%) of cases were missing due to population movement, mortality, and urbanization and were therefore excluded from the spatial analysis. Although this proportion was minor, it was unlikely to affect the overall representativeness of the dataset substantially.

Conflicts of interest: No conflicts of interest.

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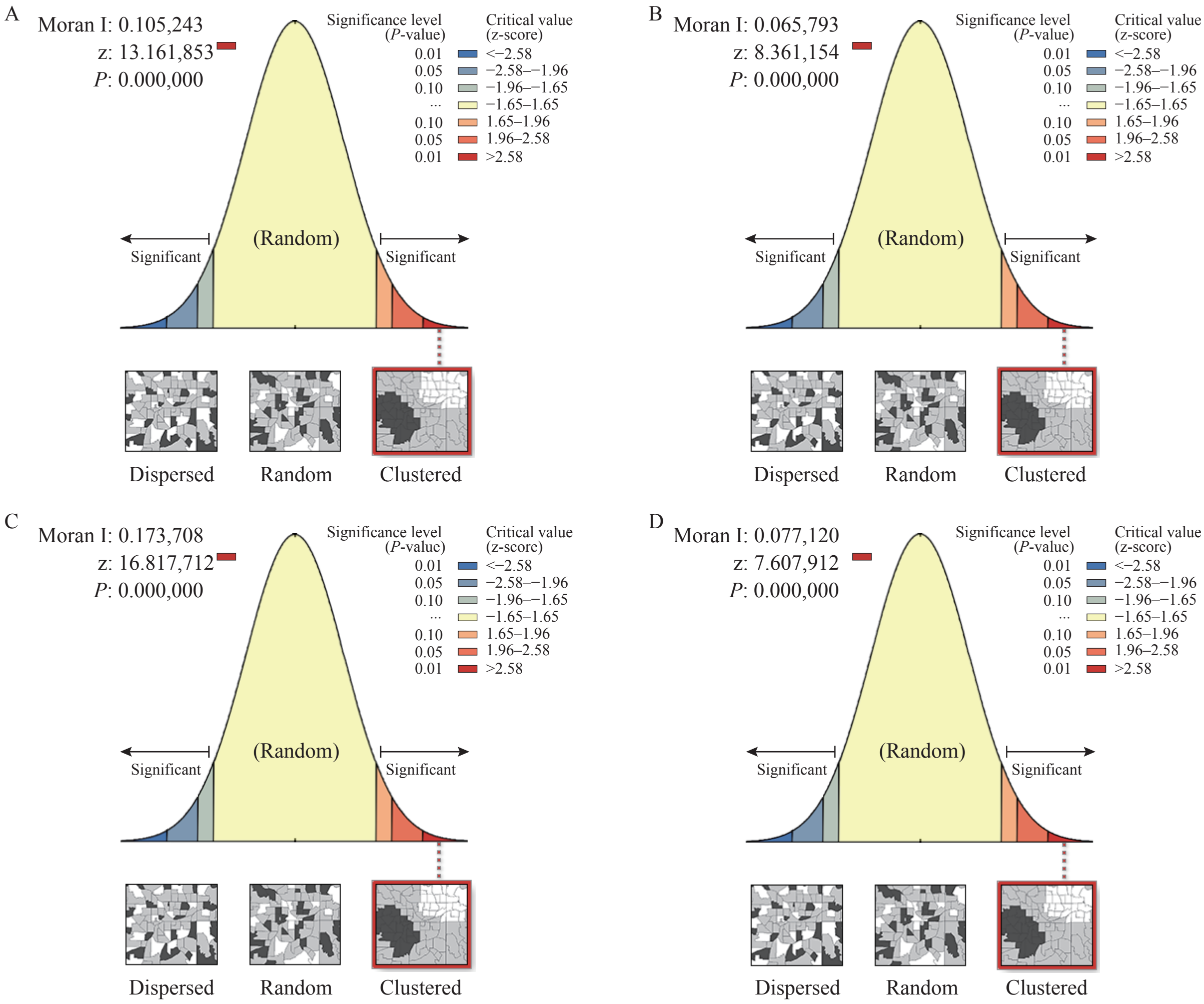
SUPPLEMENTARY MATERIAL



SUPPLEMENTARY FIGURE S1. Statistical plots of global spatial autocorrelation test results for YLDs and YLD rates among KBD patients. (A) YLDs among KBD patients in 2019; (B) YLD rates among KBD patients in 2019; (C) YLDs among KBD patients in 2023; (D) YLD rates among KBD patients in 2023.
Abbreviation: KBD=Kashin–Beck disease; YLDs=years lived with disability.

SUPPLEMENTARY TABLE S1. Counties with an inverse relationship between YLDs rate and number of health technicians for patients with KBD in 2019 and 2023.

PLADs	Number of counties	Counties
Heilongjiang	57	Acheng District, Yilan County, Fangzheng County, Bayan County, Mulan County, Tonghe County, Yanshou County, Shangzhi City, Wuchang City, Meilisi Daur District, Longjiang County, Yi'an County, Gannan County, Fuyu County, Keshan County, Kedong County, Baiquan County, Jiguan District, Hengshan District, Didao District, Lishu District, Chengzihe District, Mashan District, Jidong County, Hulin City, Mishan City, Luobei County, Lingdong District, Sifangtai District, Jixian County, Raohe County, Longfeng District, Ranghulu District, Datong District, Lindian County, Jiayin County, Nancha County, Tieli City, Suburban District, Huachuan County, Tongjiang City, Fujin City, Boli County, Xi’an District, Linkou County, Suifenhe City, Hailin City, Ning’an City, Muling City, Dongning City, Xunke County, Bei’an City, Wudalianchi City, Mingshui County, Suileng County, Zhaodong City, Hailun City
Shaanxi	55	Baqiao District, Lintong District, Lantian County, Zhouzhi County, Wangyi District, Yintai District, Yaozhou District, Yijun County, Weibin District, Jintai District, Chencang District, Fengxiang County, Qishan County, Fufeng County, Mei County, Long County, Qianyang County, Linyou County, Taibai County, Sanyuan County, Jingyang County, Qian County, Liquan County, Yongshou County, Changwu County, Xunyi County, Chunhua County, Binzhou City, Linwei District, Huazhou District, Tongguan County, Heyang County, Chengcheng County, Pucheng County, Baishui County, Fuping County, Hancheng City, Baota District, Ansai District, Yanchang County, Zhidan County, Ganquan County, Fu County, Luochuan County, Yichuan County, Huanglong County, Huangling County, Yuyang District, Shenmu City, Hanbin District, Ningshan County, Shangzhou District, Luonan County, Zhen’an County, Zhashui County
Shanxi	33	Guangling County, Tunliu District, Changzi County, Wuxiang County, Qin County, Qinyuan County, Qinshui County, Yushe County, Zuoquan County, Heshun County, Wenxi County, Jishan County, Jiang County, Yuanqu County, Xia County, Pinglu County, Ruicheng County, Xiangfen County, Gu County, Anze County, Fushan County, Ji County, Xiangning County, Daning County, Xi County, Yonghe County, Pu County, Fenxi County, Huozhou City, Lishi District, Shilou County, Fangshan County, Jiaokou County
Sichuan	26	Beichuan Qiang Autonomous County, Pingwu County, Yucheng District, Hanyuan County, Shimian County, Tianquan County, Maerkang City, Wenchuan County, Mao County, Songpan County, Jiuzhaigou County, Jinchuan County, Xiaojin County, Heishui County, Rangtang County, Aba County, Ruo’ergai County, Hongyuan County, Luding County, Danba County, Daofu County, Ganzi County, Xinlong County, Dege County, Seda County, Mianning County
Jilin	25	Shuangyang District, Yushu City, Longtan District, Chuanying District, Fengman District, Yongji County, Jiaohe City, Huadian City, Shulan City, Panshi City, Yitong Manchu Autonomous County, Xi’an District, Fusong County, Jingyu County, Qian Gorlos Mongol Autonomous County, Changling County, Qianguo County, Yanji City, Tumen City, Dunhua City, Hunchun City, Longjing City, Helong City, Wangqing County, Antu County
Gansu	22	Qinzhou District, Maiji District, Qingshui County, Qin’an County, Wushan County, Zhangjiachuan Hui Autonomous County, Kongtong District, Zhuanglang County, Huating City, Huan County, Huachi County, Heshui County, Zhengning County, Weiyuan County, Zhang County, Min County, Dangchang County, Li County, Kangle County, Hezheng County, Zhuoni County, Luqu County
Henan	5	Luoning County, Shanzhou District, Mianchi County, Lushi County, Lingbao City
Inner Mongolia	3	Uxin Banner, Zhalaite Banner, Tuquan County
Qinghai	3	Guide County, Xinghai County, Banma County
Xizang	1	Xietongmen County

Abbreviation: KBD=Kashin–Beck disease; YLDs=years lived with disability; PLAD=provincial-level administrative division.