SHORT COMMUNICATION

Dependence of Lyme disease incidence in children on environmental factors in Wielkopolska Province (West-Central Poland)

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A – Research concept and design, B – Collection and/or assembly of data, C – Data analysis and interpretation,
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▌Abstract

Introduction and Objective. Lyme borreliosis (LB) causes hundreds of thousands of new human infections worldwide annually. This is the first study connecting the LB risk to children with environmental factors.

Materials and Method. The potential impacts were assessed of environmental factors (deer density in forests, coverage of potential broadleaved forest plant communities, urbanization index) on the number of LB cases in children. Analysis covered the medical records of 196 children diagnosed with LB (ICD- A69.2) from 1 January 2012 – 30 October 2021 in Wielkopolska Province (Poland).

Results. All examined factors were positively correlated with LB cases. The highest correlation with the number of patients diagnosed with LB was presented by the degree of urbanization (percentage of the population living in cities in the total inhabitants of the study region). The number of cases was much higher in the second research period (2017–2021).

Conclusions. The number of LB cases in children is increasing as the coverage of potential broadleaved forest plant communities increases. The number of cases among males is positively correlated with the coverage. Deer density is positively correlated with the number of LB cases among children – the higher the deer density, the greater the risk of LB infection. LB cases in children are positively correlated with the urbanization index – the more people that live in cities, the greater the risk to children of LB infection.

▌Key words
humans, child, urbanization, deer, forests, Lyme disease

INTRODUCTION

Lyme borreliosis (LB) causes hundreds of thousands of new human infections worldwide annually, with numerous cases contracted in Central Europe among adults as well as children [1, 2]. The number of cases correlates with the number of infected ticks (*Ixodes ricinus*), which are vectors of *Borrelia burgdorferi* sensu lato, the causative agent of LB and other tick-borne diseases. A considerable amount of work has been undertaken to identify the most important environmental factors having an impact on the number of infected ticks, and consequently, on the number of LB cases. For example, studies from North America and Europe have examined the relationship between ticks and deer, but the results are not consistent; some results support the dilution effect of deer [3–6] due to deer not being a reservoir host for *B. burgdorferi* s.l. Whereas others support the amplification effect [7–11] as deer are the reproductive host for ticks and thus increase the numbers of ticks in the environment. This inconsistent relationship is due to the complex ecology of borrelia. Its reservoir host such as rodents and birds, must share the ecosystem with the reproductive hosts of the vector, in order to produce high risk levels of LB in the environment.

This explains that the second factor which contributes to the number of LB cases is forest cover in the research area [12, 13].

In Poland, a correlation between mixed and broadleaved forest sites and tick numbers was established due to the environment this vegetation creates for ticks host species (rodents and deer) [14]. Another factor is living in urban or rural areas; in urban parks, the number of ticks is lower [15, 16]. Regarding the prevalence and risk of LB infection, the data are contradictory; in some areas, the risk is lower [17], whereas in others, it is similar or higher [18, 19] than that in the surrounding forests. Studies connecting the LB risk in children with the environment are scarce [20], which makes the results of this study highly valuable.

OBJECTIVE

Th aim of the study was to assess the environmental factors that influence the number of LB cases in children. It was hypothesised that (1) the number of LB cases is positively correlated with the coverage of potential broadleaved forest plant communities, (2) the number of LB cases is positively correlated with the number of deer in forests, and (3) the number of LB cases is lower in urban than in rural areas in counties located in the Wielkopolska Province of West-Central Poland.

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MATERIALS AND METHOD

The analysis covered the medical records of 206 children (<18 years of age) diagnosed with LB (ICD- A69.2), hospitalised at the Department of Infectious Diseases and Paediatric Neurology of the Karol Jonscher Hospital University of Medical Sciences in Poznań, or consulted at the Outpatient Clinic of Infectious Diseases of the Karol Jonscher Hospital in Poznań, Poland, from 1 January 2012 – 30 October 2021. The study was approved by the Institutional and Licensing Committee and performed in accordance with ethical standards in the 1964 Declaration of Helsinki, and its later amendments. The local Ethics Committee of the Poznań University of Medical Science approved this study (Protocol Code: 65/19, dated 10 January 2019). All methods were performed in accordance with the above guidelines and regulations. Informed consent was obtained from all parents or legal guardians of the subjects.

The Polish Society of Epidemiologists and Doctors of Infectious Diseases criteria were adopted as the basis for the diagnosis of LB [21]. To meet the requirements of an epidemiological analysis, the study population was limited only to patients from Wielkopolska Province or, according to the administration name, the Greater Poland Voivodeship (west-central Poland). The basis for qualification for inclusion in the study was the child's place of residence, declared by the parents or guardians, and the area where the tick bite had occurred, based on the zip code. Children from outside the district and children living in Wielkopolska Province who reported having been bitten by ticks outside the province (n = 10) were not subjected to further analysis. On this basis, 196 children were qualified for further research. To analyse the aimed correlations, the study group (n = 196) was divided according to gender (male, female, total) and the period in which the disease occurred (2012–2016 and 2017–2021). Analysis of these two periods confirmed a significant increase in the paediatric number of LB cases, whereas no statistical differences were found in the distribution of the patients' gender and age [22].

The LB cases among children (Tab. S1) were calculated per 100,000 children (age 0–18) and per 100 km² (total research area: 29,827 km²) for two periods (2012–2016 and 2017–2021) and for the entire research period (10 years), divided into males, females and total. Demographics were obtained from *Statistics Poland* for 2015, 2017 and 2019 [23–25]. Coverage of potential broadleaved and coniferous forest plant communities values for each county were calculated using the official State Forests data portal [26] (Tab. S2). The density (ind. per km²) of cervids (fallow, deer, red deer, roe deer and moose) per county was calculated for the same time periods as LB cases, based on data obtained from the Polish Hunting Association in Czempiń, Kościan county, Poznań Province (Tab. S3). As urbanisation index, the percentage of inhabitants of a region living in towns and cities was assumed (Sup. Table S4).

Statistical analysis. Data normality was checked using the Shapiro-Wilk test. Due to lack of normality, dependence between LB cases per 100,000 inh. and 100 km² and environmental factors (coverage of potential broadleaved forest plant communities, deer density, urbanization index) was analysed using the rho Spearman correlation. Additionally, Kruskal-Wallis test was performed in order to confirm significant correlations. A level of significance of p (< 0.05) was assumed in all statistical tests, and all statistical analyses were conducted using Statistica 13 software [27].

For maps, the ArcMap 9.3 software (ESRI Inc.) was employed.

RESULTS

From the total of 196 cases, 100 were male and 96 female. The first study period consisted of 52 cases and the second 144 cases. The highest number of cases in a single county was 99 and the lowest – 0. Out of 31 counties, seven recorded no LB cases during the study period (Tab. 1).

Number of LB cases per 100 km². The highest numbers of LB cases per 100 km² in children in the first research period (2012–2016) were observed in the following counties: Poznań (1.5), Nowy Tomyśl (0.4), Chodzież (0.3) and Środa Wielkopolska (0.3). In the second research period (2017–2021), the highest numbers were observed in Poznań (3.1), Kościan (1.3) and Śrem (1.2). The highest numbers of LB cases in children per 100 km² in the entire research period (2012–2021) were observed in the counties of Poznań (4.6), Śrem (1.4), Kościan (1.3) and Nowy Tomyśl (1.0) (Fig. 1a).

Number of LB cases per 100,000 children. The highest numbers of LB cases per 100,000 children in the first research period (2012–2016) was observed in the counties of Nowy Tomyśl (29.0), Chodzież (23.3), Poznań (22.4), Środa Wielkopolska (19.8) and Piła (16.6), and in the second research period (2017–2021), the highest numbers were found in Kościan (67.7), Śrem (63.6), Szamotuły (61.0), Pleszew (43.9) and Leszno (43.5). The highest numbers of LB cases per 100,000 children throughout the research period (2012–2021) were observed in Śrem (72.7), Nowy Tomyśl (72.5), Kościan (67.7), Poznań (66.4) and Szamotuły (61.0) (Fig.1b).

Comparison of the first (2012–2016) and second (2017–2021) study periods showed a considerable increase (277%) in the number of LB cases in children throughout the entire study region.

Coverage of potential broadleaved forest plant communities, deer density and urbanization index. The highest coverage of potential broadleaved forest plant communities was found for Złotów (16.3%), followed by Poznań (15.8%) and Międzychód (15.6%) (Tab. S2). The counties with the highest deer density (ind. per km²) were Międzychód (5.8), Gostyń (5.8) and Śrem (5.4) (Tab. S3). The highest urbanization index was found for Poznań (72.4%), followed by Piła (64.3%) and Gniezno (61.5%) (Tab. S4).

Correlation between coverage of potential broadleaved forest plant communities and LB cases. Overall, coverage of potential broadleaved forest plant communities of a county was positively correlated with LB cases (cases per 100,000 children) irrespective of the gender for the second study period (2017–21) (r =.359; p=.047) and in the entire research period (2012–2021) (r =.369; p=.041). The number of LB cases (cases per 100 km²) for both genders was also positively correlated with the coverage for the years from 2017–2021 (r =.356, p=.049) and the whole research period (2012–21) (r =.372, p=.039). The *post-hoc* test confirmed these correlations (Tab. 1, 2; Fig. 1).

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Table 1. Spearman's rank correlation coefficients (r) and results of Kruskal-Wallis tests (H) between number of LB cases among children (<18 years old) per 100,000 children in Wielkopolska Province, between counties and environmental factors.

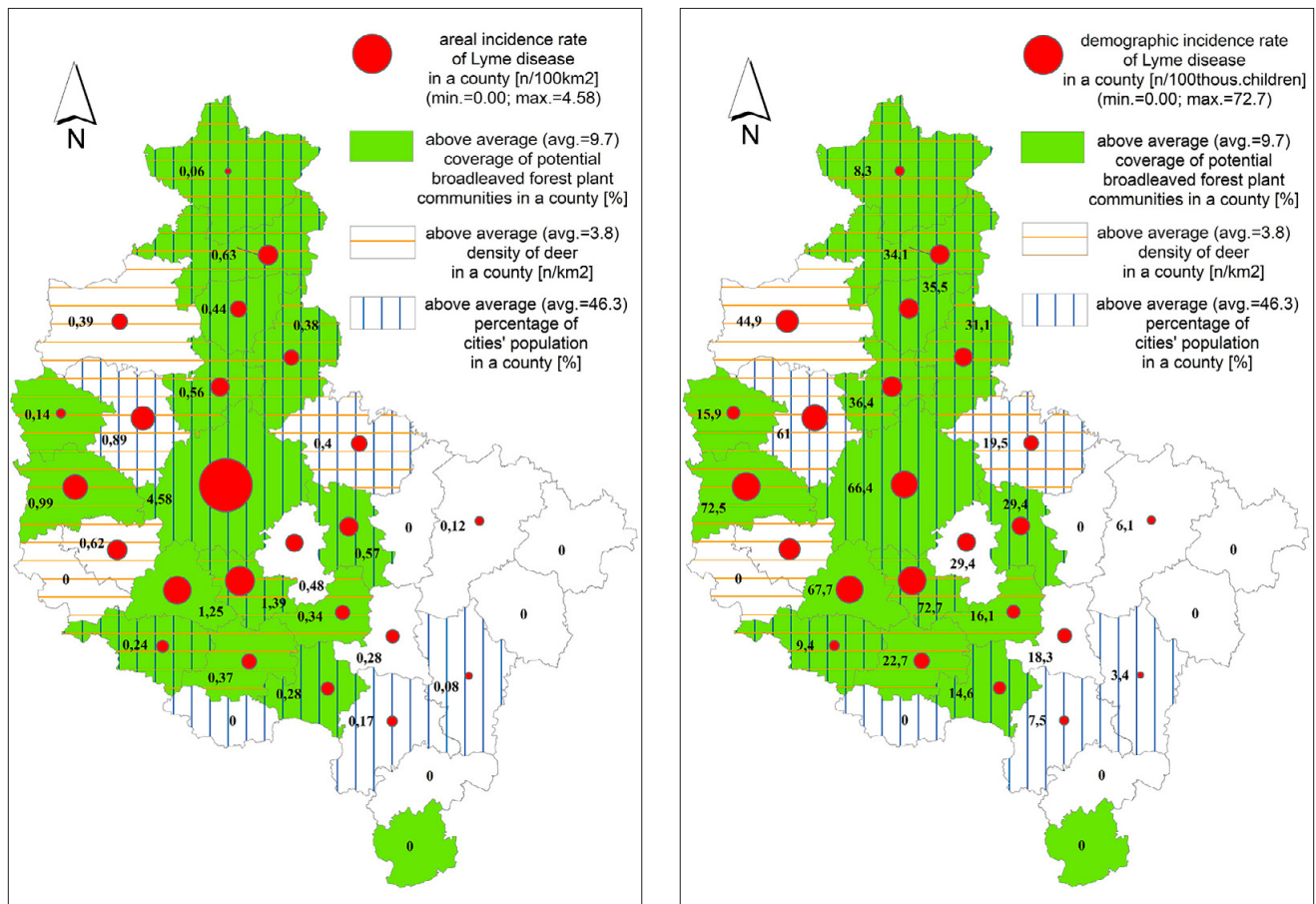| | No. of LB cases among children (<18 years old) per 100,000 children | | | | | | | | |
	2012–2016			2017–2021			2012–2021		
	Female	Male	Total	Female	Male	Total	Female	Male	Total
N	31	21	52	65	79	144	96	100	196
Mean density of deer									
Spearman's r	0.123	0.274	0.257	0.188	0.596***	0.489**	0.157	0.492**	0.415*
p-value	0.51	0.14	0.16	0.31	<0.001	0.01	0.40	0.01	0.02
Kruskal-Wallis H					7.33	5.88		6.08	5.13
p-value					0.01	0.02		0.01	0.02
Coverage of potential broadleaved forest plant communities									
Spearman's r	0.215	0.229	0.286	0.208	0.297	0.359*	0.222	0.345	0.369*
p-value	0.25	0.22	0.12	0.26	0.11	0.04	0.23	0.06	0.04
Kruskal-Wallis H						4.61			5.32
p-value						0.03			0.02
Urbanization index									
Spearman's r	0.077	0.417*	0.272	0.418*	0.300	0.430*	0.316	0.382*	0.432*
p-value	0.68	0.02	0.14	0.02	0.10	0.02	0.08	0.03	0.02
Kruskal-Wallis H		3.36		3.42		1.83		1.37	1.42
p-value		0.07		0.06		0.18		0.24	0.23

Significance levels: ***<0.001; **<0.01; *<0.05

Table 2. Spearman's rank correlation coefficients (r) and results of Kruskal-Wallis tests (H) between number of LB cases among children (<18 years old) per 100 km^2 in Wielkopolska Province, between counties and environmental factors.

	No. of LB cases among children (<18 years old) per 100 km^2								
	2012–2016			2017–2021			2012–2021		
	Female	Male	Total	Female	Male	Total	Female	Male	Total
N	31	21	52	65	79	144	96	100	196
Mean density of deer									
Spearman's r	0.111	0.266	0.250	0.144	0.537**	0.410*	0.115	0.427*	0.328
p-value	0.55	0.15	0.17	0.44	<0.01	0.02	0.54	0.02	0.07
Kruskal-Wallis H					5.29	3.79		4.42	
p-value					0.02	0.05		0.04	
Coverage of potential broadleaved forest plant communities									
Spearman's r	0.232	0.253	0.319	0.213	0.325	0.356*	0.241	0.346	0.372*
p-value	0.21	0.17	0.08	0.25	0.08	0.04	0.19	0.06	0.04
Kruskal-Wallis H						4.61			5.13
p-value						0.03			0.02
Urbanization index									
Spearman's r	0.102	0.435*	0.301	0.444*	0.361*	0.502**	0.356*	0.419*	0.505**
p-value	0.58	0.01	0.10	0.01	0.04	<0.01	0.04	0.02	<0.01
Kruskal-Wallis H		3.36		3.72	1.54	3.06	1.12	2.36	2.53
p-value		0.07		0.05	0.214	0.08	0.28	0.12	0.11

Significance levels: ***<0.001; **<0.01; *<0.05

Correlation between deer density and LB cases. Deer density was positively correlated with the number of LB cases among males in both calculation variants (per 100,000 children. and per 100 km^2) for the second part of the research period (2017–2021) (r =.596; p<.001; r =.537; p=.002) and for the entire research period (2012–2021) (r =.492; p=.005; r =.427; p=.017) The *post-hoc* test confirmed above correlations (Tab. 1, 2; Fig. 1).

Correlation between urbanization index and LB cases. The number of LB cases was positively correlated with the urbanization index, especially in the second research period (2017–2021). When calculating LB cases per 100,000 children, there was a correlation for males for the first study period (r =.417; p=.019) and for the whole research period (r =.382; p=.034). The number of cases among females per 100,000 children correlated with the urbanization index in the second period (r =.418; p=.019). The highest positive correlation

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Figure 1. Areal (A) and demographic (B) incidence rate of LB in a county on the background of: above-average coverage of potential broadleaved forest plant communities, above-average density of deer, and above-average percentage of cities' population in a county.

was observed for the entire research period, irrespective of gender (r =.432; p=.015). The same pattern of correlation was observed when calculating the number of cases per 100 km², even obtaining a stronger correlation (Tab. 1, 2).

DISCUSSION

During the research period, the number of LB cases in children in the study area increased, which is in line with regional, national [28] and European trends [1]. This might be due to the climate warming, as frost in this region is currently rare; ticks were collected from roe deer even in December and January [29]. Higher numbers of ticks and tick borne diseases are considered a result of climate change in Europe [30], and findings obtained confirm this statement.

Correlation between environmental factors was much more common for males, eight times in contrast to twice for females, most likely because of the way children spend their free time. According to a Global Matrix 4.0 survey, boys in Poland spend more time outdoors than girls [31], which increases the chances of being bitten by ticks and contracting LB. This difference in the time spent outdoors between genders has been established for Europe, the United States and Japan [32].

The number LB cases among children was correlated with environmental factors with which the number of ticks is also correlated. In Poland, the correlation between forest cover and tick number was reported by Zając et al. [33]. For

Wielkopolska Province, Wierzbicka et al. [14] reported a positive correlation between broadleaved forest sites and number of ticks.

The positive correlation was found between deer density and number of LB cases in Wielkopolska region points to cervids in this area (mainly red and Roe) being an amplification host for LB cases in the region [7–11, 34].

The urbanization index showed the highest correlation with LB cases in children in the study area. This can be explained by the higher spirochete infection rate of *I. ricinus* in city parks than in forests across Poland [16, 35, 36] and neighbouring countries [18]. This could potentially be driven by blackbirds (*Turdus merula*) (a reservoir host) [37] and deer (a reproductive host) living in urban parks which has shown a correlation with LB cases in the literature [15].

The results of this study and the growing trend in the number of LB cases closely correspond to the data published annually by the National Institute of Public Health – National Institute of Hygiene on the incidence of LB for the entire country [38]. As noted by L. Szenborn, in recent years, there has been a rapid increase in the incidence rate of LB, which can be explained by an increase in accurate diagnosis. This is a result of increasing physician awareness and knowledge of LB, as well as an increase in public awareness [39]. Since 1996, physicians have been obliged to report all cases of LB to State sanitary and epidemiological stations which, in accordance with the Regulation of the Minister of Health in 2019, requires physicians to report not only cases of diagnosed LB, but also suspected cases of Lyme disease,

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and death due to LB [40]. These obligatory measures have resulted in a better monitoring of the number of cases and improved preventive measures.

CONCLUSIONS

Over the period of 10 years, clinical data on LB cases in children were collected and compared with environmental data, resulting in the following conclusions:

1) The number of LB cases in children is growing as the coverage of potential broadleaved forest plant communities increases. The number of cases among males is positively correlated with the coverage.
2) Deer density is positively correlated with the number of LB cases among children in Wielkopolska Province – the higher the deer density, the greater the risk of LB infection.
3) LB cases in children are positively correlated with the urbanization index – the more people that live in cities, the greater the risk of LB infection in children.

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Supplemented Material

Table S1. Number of LB cases among children (<18 years old) in Wielkopolska Province according to county between 2012–2021

County	Research period split by gender					
	2012–2016		2017–2021		2012–2021	
	females	males	females	males	females	males
Chodzież	1	1	1	0	2	1
Czarnków	1	1	2	3	3	4
Gniezno	0	0	4	1	4	1
Gostyń	2	0	0	1	2	1
Grodzisk Wlkp.	1	0	1	2	2	2
Jarocin	0	0	2	0	2	0
Kalisz	0	0	1	0	1	0
Kępno	0	0	0	0	0	0
Koło	0	0	0	0	0	0
Konin	0	0	0	2	0	2
Kościan	0	0	8	1	8	1
Krotoszyn	1	0	1	0	2	0
Leszno	0	0	0	2	0	2
Międzychód	0	0	0	1	0	1
Nowy Tomyśl	2	2	1	5	3	7
Oborniki	0	0	1	3	1	3
Ostrów Wlkp.	0	0	1	1	1	1
Ostrzeszów	0	0	0	0	0	0
Piła	0	4	0	4	0	8
Pleszew	0	0	1	1	1	1
Poznań	20	12	27	40	47	52
Rawicz	0	0	0	0	0	0
Słupca	0	0	0	0	0	0
Szamotuły	0	0	6	4	6	4
Środa Wlkp.	2	0	0	1	2	1
Śrem	0	1	4	3	4	4
Turek	0	0	0	0	0	0
Wągrowiec	1	0	1	2	2	2
Wolsztyn	0	0	0	0	0	0
Września	0	0	2	2	2	2
Złotów	0	0	1	0	1	0
Total	31	21	65	79	96	100
	52		144		196	

Table S2. Coverage of potential forest plant communities in Wielkopolska Province according to county (data obtained from the State Forests official website www.bdl.lasy.gov.pl)

County	Coverage of potential forest plant communities (%)		
	Coniferous	Broadleaved	Total
Chodzież	25.7	10.5	36.2
Czarnków	45.6	6.8	52.4
Gniezno	6.0	8.9	14.9
Gostyń	3.0	11.3	14.3
Grodzisk Wlkp.	6.8	7.5	24.3
Jarocin	8.8	9.9	18.7
Kalisz	16.1	3.8	19.9
Kępno	7.7	12.5	20.2
Koło	6.0	6.0	12.0
Konin	9.7	6.4	16.1
Kościan	3.3	10.5	13.9
Krotoszyn	5.1	13.9	19.0
Leszno	10.8	13.8	24.6
Międzychód	30.3	15.6	45.9
Nowy Tomyśl	28.9	10.2	39.1
Oborniki Wlkp.	21.8	10.3	32.1
Ostrów Wlkp.	19.7	9.3	29.0
Ostrzeszów	30.0	5.3	35.3
Piła	17.6	11.8	29.4
Pleszew	14.3	5.3	19.6
Poznań	6.4	15.8	22.2
Rawicz	8.2	7.0	15.2
Słupca	10.4	5.6	16.0
Szamotuły	22.6	9.2	31.8
Środa Wlkp.	9.3	7.3	16.7
Śrem	9.2	10.8	20.0
Turek	18.6	6.2	24.8
Wągrowiec	6.4	13.1	19.5
Wolsztyn	23.0	8.1	31.0
Września	8.7	10.6	19.3
Złotów	31.6	16.3	48.0

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Table S3. Mean density of deer species (fallow deer, red deer, roe deer and moose) in Wielkopolska Province according to county between 2012–21 (data obtained from Polish Hunting Association in Czempin)

County	Mean deer density [ind. per km²]		
	2012–2016	2017–2021	2012–2021
Chodzież	2.1	2.3	2.2
Czarnków	5.0	5.5	5.3
Gniezno	3.9	4.5	4.2
Gostyń	5.7	5.8	5.8
Grodzisk Wlkp.	4.1	4.7	4.4
Jarocin	3.5	4.5	4.0
Kalisz	1.7	2.0	1.8
Kępno	3.6	3.9	3.7
Koło	2.3	2.5	2.4
Konin	2.0	2.5	2.2
Kościan	3.6	3.4	3.5
Krotoszyn	3.1	3.1	3.1
Leszno	4.5	5.3	4.9
Międzychód	5.9	5.7	5.8
Nowy Tomyśl	4.8	4.6	4.7
Oborniki Wlkp.	4.2	5.1	4.6
Ostrów Wlkp.	3.5	4.0	3.7
Ostrzeszów	2.8	3.1	2.9
Piła	4.7	5.1	4.9
Pleszew	3.0	3.1	3.0
Poznań	3.3	3.9	3.6
Rawicz	3.6	3.7	3.6
Słupca	2.7	3.3	3.0
Szamotuły	3.8	4.1	3.9
Środa Wlkp.	2.5	3.1	2.8
Śrem	4.9	5.9	5.4
Turek	2.1	2.3	2.2
Wągrowiec	4.2	5.3	4.8
Wolsztyn	4.2	4.1	4.2
Września	3.0	3.8	3.4
Złotów	4.6	4.9	4.8

Table S4. Percentage of human population living in towns and cities of Wielkopolska Province according to county (data from Statistics Poland – Statistical Office in Poznan, http://poznan.stat.gov.pl)

County	Inhabitants of towns and cities (%)
Chodzież	54.9
Czarnków	45.8
Gniezno	61.5
Gostyń	42.1
Grodzisk Wlkp.	38.5
Jarocin	41.6
Kalisz	57.7
Kępno	25.0
Koło	36.9
Konin	45.0
Kościan	46.2
Krotoszyn	59.6
Leszno	57.4
Międzychód	45.1
Nowy Tomyśl	45.7
Oborniki Wlkp.	49.0
Ostrów Wlkp.	52.0
Ostrzeszów	32.4
Piła	64.3
Pleszew	35.3
Poznań	72.4
Rawicz	47.1
Słupca	28.3
Szamotuły	46.4
Środa Wlkp.	39.9
Śrem	55.4
Turek	37.8
Wągrowiec	47.4
Wolsztyn	23.0
Września	52.8
Złotów	49.7

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