







## Оригінальні дослідження / Original Researches

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# Geographic information system in monitoring the orphan and socially significant diseases in children

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**Abstract.** Background. The purpose was to enhance the monitoring of orphan and socially significant diseases (ODs and SSDs) in children by utilizing a geographic information system (GIS) and examining the relationship between the prevalence of growth hormone deficiency (GHD), cystic fibrosis (CF), acute lymphoblastic leukemia (ALL), type 1 diabetes mellitus (T1DM) and eco-geophysical factors in the surrounding area. Materials and methods. Monitoring the cases of GHD, CF, ALL and T1DM in children in the Odesa region was carried out from 2016 to 2020. We recorded the findings of 862 children, among whom 92 had GHD, 54 had CF, 88 had ALL, and 628 had T1DM. The study used clinical and epidemiological techniques, as well as a local GIS. To analyze the data, we used a local GIS medico-social layer and overlaid it with the eco-geophysical layer of the same GIS. The study utilized information, which was obtained through the Black Sea geophysical expedition conducted beforehand in the Odesa region. The prevalence of diseases was analyzed by  $\chi^2$  test. A p-value < 0.05 was considered statistically significant. **Results.** When testing the "null hypothesis" regarding the distribution of GHD, CF, ALL, and T1DM cases in children across three physical-geographical zones in the Odesa region, the study found significant differences in disease prevalence among the different areas. The forest-steppe zone, particularly the Ananiv district, had the highest prevalence of GHD. Meanwhile, the highest occurrence of T1DM was registered in the Mykolaivka district of the steppe zone. In the Transnistrian zone, the Biliaivka district had the highest rates of T1DM in children, while the Ovidiopol district had the highest rates of CF, and Odesa had the highest rates of ALL. By examining the prevalence of GHD, CF, ALL, and T1DM in children alongside the mapping of geophysical, and environmental anomalies in the Odesa region, it was discovered that eco-geophysical factors play a major role. Conclusions. The use of the GIS method in the epidemiological study of GHD, CF, ALL, and T1DM in children contributes to the improvement of monitoring the orphan and socially significant diseases.

**Keywords:** geographic information system; orphan and socially significant diseases; children

## Introduction

Monitoring and tracking of orphan diseases (ODs) in specific populations is essential for planning and implementing effective preventive measures, evaluating intervention strategies, and creating epidemiological forecasts. The socially significant diseases (SSDs) are illnesses, which rank highest in the morbidity and death rate in a country. The ODs are congenital or acquired diseases that occur with a frequency of no more than 1:2,000 [1]. It is important to control the prevalence of SSDs and ODs because they create a significant social and economic burden on society. This is due to the psychological problems that children face, which can lead to disability and a reduced quality of life.

Additionally, there are challenges with diagnosis, treatment, rehabilitation, upbringing, and education [2].

Medical geographic information system (GIS) is a system that acquire, store, analyze, and display geographically linked data. By bridging the fields of biomedical and social sciences, this approach can effectively improve disease monitoring [3, 4]. Mapping health information alongside environmental and socioeconomic data can reveal the relationship between the two. By comparing eco-geophysical data and medico-social parameters, including health indicators, a map can highlight the correlations between environment and health [5, 6]. A recent study found that an orphan disease such as growth hormone deficiency (GHD)

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occurs unevenly among children in different areas of the Odesa region [7]. The findings were interesting to compare with the regional prevalence of other ODs and SSDs: cystic fibrosis (CF), acute lymphoblastic leukemia (ALL), and type 1 diabetes mellitus (T1DM).

The **purpose** was to enhance the monitoring of ODs and SSDs in children utilizing a GIS and examining the relationship between the prevalence of GHD, CF, ALL, T1DM and eco-geophysical factors in the surrounding area.

## Materials and methods

A study was conducted at the Odesa Regional Children's Clinical Hospital (ORCCH), following the principles of the Declaration of Helsinki. It involved population studies and epidemiological monitoring of children with GHD, CF, ALL, and T1 DM across 26 administrative districts and three natural physical-geographical zones in the Odesa region, including forest-steppe, steppe, and Transnistrian. The data was collected using GIS technology. The results of the clinical and epidemiological study were entered into the medico-social layer of the local GIS and mapped by overlaying the data of the eco-geophysical layer [5-7]. The latter was represented by territorial maps of geophysical, hydrogeological and ecological anomalies in the Odesa region based on the results of the earlier Black Sea geophysical expedition [8]. The medico-social layer included data on the regional prevalence of childhood diseases at the time of the expedition. The categorical variables were expressed as frequency and analyzed by  $\chi^2$  test. A p-value < 0.05 was considered statistically significant.

#### Results

From 2016 to 2020, the outpatient department of the ORCCH kept track of children with ODs and SSDs in the Odesa region. During this time, they registered 92 children with GHD, 54 with CF, 88 with ALL, and 628 with T1DM. In 2020, the prevalence of these conditions in the Odesa region was as follows: GHD -1 in 5,096 children, CF -1 in 8,682 children, ALL -1 in 5,328 children, and T1DM -1 in 747 children. We proposed a "null hypothesis" that assumes an even distribution of GHD, CF, ALL, and T1DM in children across the Odesa region, considering their spontaneous and sporadic occurrence. Using the Pearson criterion to test this hypothesis, we obtained the results shown in Table 1, which indicate the prevalence of these diseases throughout the region.

Studies have shown that the distribution of OD and SSD cases in the districts of Odesa region is not uniform, which contradicts the original "null hypothesis". There are notable

variations in the prevalence of these diseases across different areas. Some conditions are more common in some areas and less common in others, and the generalized data for the Odesa region do not reflect the specifics of the territorial distribution of cases of OD and SSD in children.

The data presented in Table 2 shows the prevalence of ODs and SSDs in the forest-steppe zone of the Odesa region. The occurrence of GHD is higher in the forest-steppe zone, particularly in the Ananiv district, compared to the average regional values (p < 0.01). The frequency of other diseases (CF, ALL, T1DM) is distributed in this zone according to the "null hypothesis". The Ananiv district has gravitational, magnetic, geological, and geophysical anomalies, a break in the earth's crust, and a higher level of uranium content, as reported by the Black Sea geophysical expedition [8].

In Table 3, you can find information about the occurrence of ODs and SSDs in the steppe zone of the Odesa region. Between 2016 and 2020, a significantly higher rate of T1DM was found in children in the steppe zone, particularly in the Mykolaivka district (p < 0.001). The other health conditions studied were distributed more evenly. Previous geophysical studies in the Mykolaivka district have identified several anomalies, including gravitational, magnetic, geological, and geophysical ones, as well as an increased amount of uranium [8].

The data presented in Table 4, shows the occurrence of ODs and SSDs in the Transnistrian zone of the Odesa region. The findings from a four-year monitoring period indicate that T1DM rates have been steadily increasing in children from the Biliaivka district, while CF rates have been on the rise in the Ovidiopol district, and ALL rates in the city of Odesa. Furthermore, the Black Sea geophysical expedition data showed that the Transnistrian zone has several eco-geophysical anomalies, including insufficient zinc, molybdenum, and cobalt levels [8].

## **Discussion**

We analyzed the prevalence of ODs and SSDs in children across different administrative districts and geographical zones in the Odesa region, using local GIS data. The study supported the experts' view on the usefulness of GIS as a tool for collecting, storing, analyzing, and visualizing geographic data and related medical information [9–11]. The results showed that the "null hypothesis" of a uniform distribution of GHD, CF, ALL and T1DM cases in children throughout the Odesa region was not confirmed. Specifically, we observed a higher prevalence of GHD in children in the Ananiv district located in the forest-steppe zone of the Odesa region. Additionally, within the steppe zone, we detected a higher oc-

Table 1. Differences in the prevalence of orphan and socially significant diseases among children in districts of the Odesa region

ODs/SSDs	n*	χ²	р						
CF	54	42.25	< 0.05						
ALL	88	48.67	< 0.01						
GHD	92	41.11	< 0.05						
T1DM	628	96.01	< 0.001						

Note: \* — number of cases (general children's population was 468,820).

currence of T1DM in the Mykolaivka district. Finally, in the Transnistrian zone of the Odesa region, we identified a higher incidence of CF (the Ovidiopol district), T1DM (the Biliaivka district), and ALL (the city of Odesa) among children.

To understand the causes behind the uneven distribution of diseases in different geophysical zones of the Odesa region, an analysis was conducted. The prevalence of ODs and SSDs was compared with territorial inventories and maps of geophysical, environmental, and medical anomalies developed by the Black Sea geophysical expedition. The ecogeophysical layer of the local GIS of the Odesa region has several geophysical and hydro-geological anomalies, altered magnetic and gravitational fields, soil water contamination with ammonia, nitrate, and pesticides. There are also some areas with high levels of uranium, radon, radium, mercury, and lead, as well as a deficiency of zinc, cobalt, and molybdenum in the soil. Researchers have noted a higher prevalence of endocrine diseases, neoplasms, cardiovascular, and psycho-neurological disorders in specific areas. This information has been added to the medico-sociological layer of the local GIS. The researchers suggest that these health issues may be linked to eco-geophysical factors [8].

During our study, we analyzed the link between the eco-geophysical layer of the local GIS (which shows geophysical and environmental anomalies) and the medicosociological layer (which shows the prevalence of GHD, CF, ALL, and T1DM in children). The study involved the cartographic overlaying on maps of eco-geophysical data with medico-sociological data in children with ODs and SSDs. Our data supports previous studies that have demonstrated how the GIS method can improve disease detection, simplify medical and social management, and provide a clearer understanding of the clinical importance of various eco-geophysical factors [12-14]. Utilizing data from the medico-social layer of the local GIS can enhance the detection of GHD, CF, ALL, and T1DM in children by promoting diagnostic awareness, providing continuous training for medical staff, and optimizing health sector policies. By utilizing GIS methodology, monitoring and surveillance can be improved to identify potential risk areas for ODs and SSDs. These areas are often not well understood from an epidemiological perspective, but by doing this analysis, it can support the development of preventive socio-demographic policies.

Table 2. Differences in the prevalence of orphan and socially significant diseases among children in the forest-steppe zone of the Odesa region (2016–2020)\*

Districts of the forest-steppe zone	Children's population	CF		ALL		GHD		T1DM	
		Α	E	Α	Е	Α	E	Α	E
Ananiv	5,265	0.00	0.56	0.00	0.99	5.00	1.03	4.00	7.06
Balta	7,987	0.00	0.85	0.00	1.50	3.00	1.56	13.00	10.70
Kodyma	5,496	2.00	0.59	3.00	1.03	2.00	1.01	10.00	7.36
Podilsk	13,091	2.00	1.40	1.00	2.46	4.00	2.57	25.00	17.54
Okny	4,741	0.00	0.50	0.00	0.89	1.00	0.93	4.00	6.35
Liubashivka	6,401	0.00	0.68	0.00	1.20	1.00	1.25	8.00	8.58
Savran	3,604	0.00	0.38	0.00	0.68	1.00	0.71	2.00	4.83
$\chi^2$ at 6 degrees of freedom		7.664		9.875		21.319		8.498	
Significance of differences			p > 0.2		p > 0.1		p < 0.01		p > 0.2

Note: here and in Tables 3, 4:  $^*$  — the number of patients: A — actual and E — expected, according to the "null hypothesis".

Table 3. Differences in the prevalence of orphan and socially significant diseases among children in the steppe zone of the Odesa region (2016–2020)\*

Districts of the steppe zone	Children's population	CF		ALL		GHD		T1DM	
		Α	E	Α	E	Α	E	Α	E
Berezivka	7,583	0.00	0.81	0.00	1.42	1.00	1.49	11.00	10.16
Velyka Mykhailivka	7,614	0.00	0.81	0.00	1.43	0.00	1.49	6.00	10.20
Ivanivka	6,585	1.00	0.70	1.00	1.24	1.00	1.29	1.00	8.82
Mykolaivka	3,758	0.00	0.40	0.00	0.71	2.00	0.74	8.00	5.03
Rozdilna	13,019	1.00	1.39	0.00	2.45	1.00	2.55	16.00	17.44
Zakharivka	5,265	1.00	0.56	0.00	0.99	0.00	1.03	2.00	7.06
Shyriaive	6,441	0.00	0.688	0.00	1.21	0.00	1.26	3.00	8.63
χ² at 6 degrees of freedom		3.421		8.258		11.332		26.369	
Significance of differences			p > 0.2		p > 0.2		p > 0.05		p < 0.001

in the Transmittan Zone of the Odesa region (2010 2020)									
Districts of the Transnistrian zone	Children's population	CF		ALL		GHD		T1DM	
		Α	E	A	E	Α	E	Α	E
Artsyz	9,406	1.00	1.00	3.00	1.77	2.00	1.84	5.00	12.60
Biliaivka	27,921	3.00	2.98	2.00	5.25	4.00	5.47	48.00	37.41
Bolhrad	13,324	2.00	1.42	2.00	2.50	1.00	2.61	4.00	17.85
Bilhorod- Dnistrovskyi	26,032	1.00	2.78	5.00	4.89	7.00	5.10	29.00	34.88
Izmail	25,672	0.00	2.74	4.00	4.83	3.00	5.03	31.00	34.40
Kiliia	10,921	2.00	1.17	0.00	2.05	2.00	2.14	6.00	14.63
Lyman	21,883	5.00	2.34	2.00	4.11	3.00	4.29	32.00	29.32
Ovidiopol	32,756	8.00	3.50	11.00	6.16	3.00	6.42	44.00	43.89
Reni	7,849	2.00	0.84	0.00	1.48	2.00	1.54	6.00	10.52
Sarata	10,747	4.00	1.15	1.00	2.02	1.00	2.11	12.00	14.40
Tarutyne	9,979	1.00	1.06	0.00	1.88	0.00	1.96	3.00	13.37
Tatarbunary	8,765	0.00	0.94	0.00	1.65	1.00	1.72	6.00	11.75
Odesa city	166,715	14	17.80	53	31.34	38	32.68	295	223.40
χ² at 12 degrees of freedom		31.161		30.541		19.359		61.145	
Significance of differences			p < 0.01		p < 0.01		p > 0.05		p < 0.001

Table 4. Differences in the prevalence of orphan and socially significant diseases among children in the Transnistrian zone of the Odesa region (2016–2020)\*

Further research is needed to clarify the relationship between eco-geophysical factors and the prevalence of GHD, CF, ALL, and T1DM in children, including their underlying mechanisms and nature.

#### **Conclusions**

- 1. Through epidemiological monitoring using the GIS method, it was discovered that the prevalence of ODs and SSDs (GHD, CF, ALL, T1DM) in children varies greatly across the forest-steppe, steppe, and Transnistrian physical-geographical zones, as well as 26 administrative districts of the Odesa region.
- 2. In the forest-steppe zone of the Odesa region, the Ananiv district has reported a higher-than-normal rate of GHD in children. Similarly, the Mykolaivka district in the steppe zone has revealed increased rates of T1DM. Additionally, the Transnistrian zone in the Odesa region has reported abnormally high occurrence of CF in the Ovidiopol district, T1DM in the Biliaivka district, and ALL in the city of Odesa.
- 3. A common feature of areas with an increased prevalence of some ODs and SSDs among the child population was the environmental and geophysical anomalies. The heterogeneity of the territorial distribution of ODs and SSDs cases requires further study of the relationship between the epidemiological parameters of GHD, CF, ALL, and T1DM in children and environmental/geophysical factors.

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## Географічна інформаційна система в моніторингу орфанних та соціально значущих захворювань у дітей

Резюме. Мета: покращити моніторинг орфанних та соціально значущих захворювань у дітей на основі використання географічної інформаційної системи (ГІС) і вивчити зв'язок між поширеністю дефіциту гормону росту (ДГР), муковісцидозу (МВ), гострого лімфобластного лейкозу (ГЛЛ), цукрового діабету 1-го типу (Т1ЦД) й еколого-геофізичними факторами навколишньої території. Матеріали та методи. Моніторинг випадків ДГР, МВ, ГЛЛ та Т1ЦД у дітей в Одеській області проводився з 2016 по 2020 роки. Ми зареєстрували дані 862 дітей, серед яких 92 мали ДГР, 54-MB, 88-ГЛЛ і 628-Т1ЦД. У дослідженні використано клінічні та епідеміологічні методи, а також локальну ГІС. Для аналізу даних застосовували локальний медико-соціологічний шар ГІС і накладали його на еколого-геофізичний шар тієї ж ГІС. У дослідженні використано інформацію, отриману при проведенні Чорноморської геофізичної експедиції в Одеській області. Поширеність захворювань аналізували за  $\chi^2$ -тестом. Значення р < 0,05 вважалося статистично значущим. Результати. Під час пе-

ревірки «нульової гіпотези» щодо розподілу випадків ДГР, МВ, ГЛЛ та Т1ЦД у дітей за трьома фізико-географічними зонами Одеської області виявлено суттєві відмінності в поширеності захворювань на різних територіях. У лісостеповій зоні, зокрема в Ананьївському районі, виявлено найбільшу поширеність ДГР, водночас у Миколаївському районі степової зони переважав Т1ЦД. У Придністровській зоні у Біляївському районі найвищими були показники Т1ЦД, тоді як в Овідіопольському районі — показники МВ, а в Одесі — ГЛЛ. При дослідженні поширеності ДГР, МВ, ГЛЛ та Т1ЦД у дітей разом із картуванням геофізичних та екологічних аномалій в Одеській області виявлено істотну роль еколого-геофізичних факторів. Висновки. Використання методу ГІС при епідеміологічному дослідженні ДГР, МВ, ГЛЛ та Т1ЦД у дітей сприяє покращенню моніторингу орфанних та соціально значущих захворювань.

**Ключові слова:** географічна інформаційна система; орфанні та соціально значущі захворювання; діти