Original Research Study
Acta Medica Academica 2021;50(2):329-339
DOI: 10.5644/ama2006-124.350

The Frequency and Form of Controls by HIIS over Primary Health Care Physicians in Slovenia

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Received: 5 October 2020; **Accepted:** 9 May 2021

Abstract

Objective. The aim of this study was to evaluate the pattern of controls and sanctions by the Health Insurance Institute (HIIS) over primary healthcare practitioners (PHCPs) in Slovenia, the reasons for sanctions and the violence against PHCPs if they followed the HIIS rules. **Materials and Methods**. We performed analyses using survey data from a cross-sectional study, across public health centres and individual contractors in which 1,458 PHCPs were invited to answer a questionnaire anonymously via an online system used to collect data for the Slovenian Medical Chamber and the Association of General Practice/Family Medicine of South-East Europe. Quantitative data were presented by descriptive statistics and analysed using Pearson's chi-squared test. **Results**. Responses were obtained from 462 female and 138 male PHCPs. Of the total number of 600 participants, 430 were family medicine specialists. 263 (43.8%) responded that they have been sanctioned for various reasons. PHCPs that are more likely to be sanctioned include family medicine specialists and individual contractors. PHCPs working in areas smaller than 20,000 inhabitants were sanctioned in a bigger proportion than their counterparts. Monetary penalties levied against those working at health centres were usually covered by the health centre. Family medicine specialists, more often than other PHCPs experienced violence from patients or patients' relatives if they followed HIIS rules. **Conclusion**. Family medicine specialists are sanctioned more frequently than other PHCPs, individual contractors are sanctioned more frequently than public healthcare PHCPs and PHCPs in working area with a population less than 20,000 are more frequently sanctioned than those working in an area with a bigger population count.

Key Words: Primary Health Care ▪ Insurance ▪ Prescription Controls ▪ Sanctions ▪ Violence at Work.

Introduction

The World Health Organization has identified four key functions needed for the health system to function: stewardship, resource creation, funding and service delivery. Stewardship means oversight of the system's basic functions and includes management, information transmission, coordination and monitoring of the system at various levels (1). The quality of the healthcare system depends on the successful integration of these four functions, and this integration depends on the quality control of the healthcare system.

In the countries of the European Union there are different types of health care systems and different regulatory bodies within those systems. Three different systems have been defined: the Beveridge model, the Bismarck model and the mixed model. Countries that use the Beveridge model have financing that is predominantly through taxes, provide universal health coverage and the government regulates what physicians can do and charge for their services (e.g. Cyprus, Denmark, Finland, Ireland, Italy, Latvia, Malta, Portugal, Spain, Sweden and United Kingdom). In contrast to that, in the countries that use the Bismarck model the funding is granted through compulsory social se-

curity contributions by employers and employees (e.g. Belgium, Estonia, France, Germany, Lithuania, Luxembourg, Netherlands, Poland, Czech Republic, Romania, Slovakia, Slovenia and Hungary). Countries that use the mixed model have significant funding from voluntary insurance or upfront payments (e.g. Croatia, Greece, Austria, Bulgaria) (2-4).

In the countries organized by the Bismarck model health insurance funds usually have some role in regulation of the work of physicians. For example, in Romania the Ministry of Health (MoH) and the National Health Insurance House (NHIH) together develop an annual national framework contract that contains the rights of the insured population and the conditions for all providers including primary healthcare practitioners (PHCPs). Based on this document they routinely monitor medical files, mandatory licensing, feedback on services and utilization of NHIH listed resources and drugs (5). In Slovakia, Health Care Surveillance Authority (HCSA), professional chambers and self-governing regions monitor and enforce responsibilities that were previously set by the MoH. HCSA is a regulatory body that also supervises the Insurance funds, is involved in quality management and has the power to impose sanctions (6). Germany, on the other hand has a more complex system with regulatory bodies on the federal, state and corporatist level. The individual states implement the federal legislation and supervise the public health services, as well as the regional medical associations, the regional associations of physicians that work with statuary health insurance (SHI) and the insurance funds. The regional associations of SHI physicians have a supervisory and regulatory role over the health services that have been defined by law and in contracts with the insurance funds (7).

The healthcare system in Slovenia is also based on the Bismarck model, additionally with an accent on primary healthcare gatekeeping. Primary healthcare (PHC) service provision is under the jurisdiction of the municipalities, which are responsible for health policy development at the local level and are also the owners of community-level PHC centres. In Slovenia, there are 65 healthcare centres that deliver PHC at 459 locations. Healthcare workers at publicly-owned health centres are salaried public servants. PHC is organised through a coexistence of predominantly public health centres as an exclusive form of health centres and individual contractors, or concessionaires (8). The number of individual contractors has increased since Slovenian independence in 1991. This now encompasses 30% of PHC providers (family physicians, paediatricians and gynaecologists) (9). Quality of care at the PHC level is formally a priority, but the legislation to ensure quality in this area is proceeding very slowly. There are formal instruments for assessing quality, such as obligatory certification of physicians every 7 years through collecting credits, voluntary certification and accreditation and mandatory licensing of physicians and nurses, but PHC quality has not yet been systematically assessed by quality indicators (10).

In Slovenia regulation is conducted by the MoH, Health Inspectorate, Health Insurance Institute (HIIS) and The Medical Chamber of Slovenia. Supervision of compliance with the Patients' Rights Act is exercised by the MoH. Slovenia's Health Inspectorate's authority is limited exclusively to the competence of the offence authority. The Medical Chamber of Slovenia carries out professional supervision (11). The HIIS has control over prescribing, proper identification of sick leave, allocation of ambulance transport in accordance with HIIS rules and dispensing and charging for medicinal products. These controls are detailed in the HIIS Rules on Controls and Supervisions. Supervision is carried out using data from the HIIS records on issued and billed medicines, documents for exercising the right to medicines and other available documentation (12). HIIS supervises the work of the PHCPs and if they detect that their rules aren't followed a report is issued to the MoH to ask for an administrative control. According to the severity of the breach, two types of sanctions are given: a warning, and a fine. Fines can be covered by the institution, partially by the PHCP or fully by the PHCP. If the breach is too severe, the PHCP undergoes

another type of control, which is internal control that might result with licence revoking (11).

Patients also have to understand the limitations and rules to which PHCPs need to adhere, since it has been shown that lack of understanding the health system, expectation gaps, limited medical services and low education were a part of the reasons for violence against healthcare workers (13, 14). This is important because violence against healthcare workers is frequent and it has negative effects on the mental and physical health of the healthcare practitioners, the quality of healthcare delivery and the organization as a whole (15-18).

This study is the first to investigate the frequency and forms of HIIS controls faced by PHCPs and especially over family medicine specialists. We wanted to see whether there is a relationship between different sociodemographic factors and sanctions of PHCPs, what are the reasons for sanctions and whether PHCPs experienced violence from patients or their relatives if they followed the HIIS rules.

Materials and Methods

Study Design, Timeframe, Data Collection and Study Population

The design is retrospective and cross-sectional. The study was carried out by anonymous online questionnaire containing 13 different questions ranging from multiple choice to essay questions. The questionnaire was adapted from a standardized questionnaire made by the Association of General Practice/Family Medicine of South-East Europe (AGP/FM SEE). Every single practitioner had his own anonymous internet protocol address (IP), from which he or she was able to participate only once. The survey took roughly 20 minutes to fill out.

In the first phase a pilot survey was sent to 30 participants in December 2017 in order to improve the quality of the survey. In the second phase, in January 2018, the official survey was distributed in electronical form to all the PHCPs registered with the Medical Chamber of Slovenia. In Slovenia, regulation is conducted by the Health Insurance Institute (HIIS), Health Inspectorate, Ministry of Health and The Medical Chamber of Slovenia.

Data collection was concluded at the end of March 2018. Participants were medical professionals registered with the Medical Chamber, with or without PHC specialisation. The number of total registered family physicians in 2017 was 1,362, the number of GPs was 256, the number of gynaecologists was 463 and the number of paediatricians was 851 (19). Filters were added to distinguish those working as PHCPs from those working in secondary, tertiary healthcare or other institutions. Additional filters to remove retired PHCPs were added. After this process of filtering, a total of 1,458 PHCPs were approached.

The research methodology was presented and discussed at two meetings of the European General Practice Research Network in Sarajevo (2018) and Tampere, Finland (2019).

Questionnaire

The questionnaire consisted of 13 questions: seven single-select multiple-choice, four dichotomous, one mixed (multi-select multiple choice question used for reasons of sanctioning) and one mandatory essay question. We collected the following socio-demographic characteristics: sex, age, professional education status, years of experience in the field of primary healthcare, distance from the workplace, practice environment and the employment status (individual contractor or public healthcare). Questions about sanctioning consisted of: whether they have ever been sanctioned, the type of the sanction, the reason for sanctions and the responsible body for the payment of the sanction. There were also questions about whether physicians experienced violence from their patients or the patients' relatives if they followed the HIIS rules, and if so, what type of violence it was (verbal, physical or both).

Ethical Approval

The content and ethics of the research was confirmed at the nineteenth session of the Executive Board of the Medical Chamber on November 16th, 2017, Decision no. 270/19/2017 (20).

Statistical Analysis

We used the statistical software SPSS Statistics 22. Values of $P<0.05$ we considered statistically significant. Quantitative data were presented by descriptive statistics and analysed using Pearson's chi-squared test.

Results

Descriptive Statistics

Our study covered 600 PHCPs in Slovenia. In Table 1 the sociodemographic characteristics of the participants are shown. It has to be noted that, regarding specialty, the questionnaire contained only a question whether they are specialists of family medicine or not. This is why no other specialties are shown in the table.

From all of the participants, 263 (43.8%) received sanctions in either the form of a fine or a warning. Table 2 summarizes this, as well as the type of sanction the PHCPs received. The responsible body for the payment of the sanction is divided into several categories: healthcare administration, which means the institution covers the fine, personally: partially and up to total – which means the PHCPS partially pay for the fine or for the total amount accordingly. From the 263 sanctioned, only 150 (57%) answered this question and 113 (43%) left it blank. The numbers shown under this variable in Table 2 are from those that answered. Participants were also asked whether they were exposed to violence from their patients or their patients' relatives if they complied with HIIS rules. These data, together with the type of violence is also shown in Table 2.

Table 1. Sociodemographic Characteristics of Respondents

Variable	N (%)
Sex	
Female	462 (77.0)
Male	138 (23.0)
Family medicine specialist	
Yes	430 (71.6)
No	170 (28.3)
Age structure	
25–29	1 (0.2)
30–39	89 (14.8)
40–49	165 (27.5)
50–59	238 (39.6)
≥60	107 (17.8)
Years of practice	
0–10	67 (11.1)
11–15	85 (14.1)
16–20	75 (12.5)
21–30	204 (34.0)
≥31	169 (28.1)
Employment status	
Individual contractor	175 (29.1)
Public healthcare	425 (70.8)
Population in work area	
<20,000	206 (34.3)
20,000–49,999	164 (27.3)
50,000–99,999	58 (9.6)
100,000–499,999	138 (12.7)
≥500,000–999,999	34 (5.6)
Distance from nearest hospital	
Workspace is part of hospital	223 (37.1)
<20 km	161 (26.8)
20–49 km	180 (30.0)
≥50–99 km	36 (6.0)

Table 2. Sanctions and Violence Experienced Working as a PHCP

Variable	N (%)
Ever Sanctioned	
Warned	56 (9.3)
Fined	207 (34.5)
Not Sanctioned	337 (56.1)

Type of sanction	
Warning	55 (21.0)
Warning before termination of agreement	1 (0.1)
Monetary amount based on damages caused	92 (34.8)
Monetary amount on fixed scale	115 (44.0)
Responsible body for payment of sanction*	
Health centre administration	118 (78.7)
Personally: partially	20 (13.3)
Personally: up to total	12 (8)
Victim of violence at work	
Yes	464 (77.3)
No	136 (22.7)
Type of violence	
Physical	1 (0.2)
Verbal	441 (95.0)
Physical and verbal	22 (4.7)

PHCP=Primary healthcare practitioners; *Missing answers 113.

Reasons for Controls and Sanctions

When looking at the results for the reasons for controls and sanctions, it should be taken into consideration that this was a mixed question. Participants could select more options from the ones listed below and additionally add their own answer if their option wasn't listed. It was not an obligatory question and some participants chose more reasons for their sanctioning, while others didn't choose at all. That means from the 263 sanctioned, 207 gave answers to this question and some selected more reasons which at the end summed as 263. This is why the number of responses isn't same as the number of participants who selected that they had been sanctioned. Even though the number of sanctioned PHCPs and the number of selected answers to this question have the same value-263, they do not correspond to the number of participants that answered this question – 207.

From the answers received, the reasons were grouped in the categories shown in Table 3. The most common reason was "Prescribing drugs not in accordance with HIIS rules", which includes: prescribing drugs above the agreed financial amount, prescribing medication while the patient was hospitalised in a secondary or tertiary institution, prescribing drugs in a dosage not in accordance with HIIS rules, unnecessary prescription of more drugs with the same effect and prescribing drugs that aren't covered by the HIIS as they were. The second most common reason was "Prescribing technical or orthopaedic devices not in accordance with HIIS rules", from which adult diapers were most commonly prescribed against HIIS rules. From the 50 who got sanctioned for excessive sick leave and/or unjustified sick leave, 8 commented that the reason was giving sick leaves to mothers who had hospitalized children. In the category "Incomplete or not timely updated documentation", 12 specified that the reason was inconsistency between the paper and electronic forms.

Table 3. Reasons for Sanctions

Reasons for sanctions	Frequency (%)*
Prescribing drugs not in accordance with HIIS rules	68 (25.9)
Prescribing technical or orthopaedic devices not in accordance with HIIS rules	65 (24.7)
Excessive sick leave and/or unjustified sick leave	50 (19)
Issuing transportation orders without following HIIS rules on diagnosis and/or distance	32 (12.2)
Incomplete or not timely updated documentation	26 (9.9)
Issuing referrals not in accordance with HIIS rules	10 (3.8)
Home care order costs and/or justification for issuing	9 (3.4)
Other	3 (1.1)
Total	263 (100)

HII=Health Insurance Institute; *Represents the percent of a choice from all the choices selected by all the participants who answered the question.

Sanctions and Violence against PHCPs in Relation to Sociodemographic Variables

Table 4 shows the relationship between sanctions and sociodemographic variables, as well as the relationship between exposure to violence and the sociodemographic variables.

Table 4. Sanctions and Violence against Physicians by Sociodemographic Variables

Variable	Value	Sanctions		P	Violence		P
		Yes	No		Yes	No	
		N (%)	N (%)		N (%)	N (%)	
Sex	Male	81 (30.8)	57 (16.9)	<0.001	101 (21.8)	37 (27.2	0.185
	Female	182 (69.2)	280 (83.1)		363 (78.2)	99 (72.8))	
Age (years)	25–39	26 (9.9)	64 (19.0)	<0.001	75 (16.2)	15 (11.0)	0.05
	40–49	57 (21.7)	108 (32.0)		133 (28.7)	32 (23.5)	
	50–59	124 (47.1)	114 (33.8)		181 (39.0)	57 (41.9)	
	60–69	56 (21.3)	51 (15.1)		75 (16.2)	32 (23.5)	
Family med. specialist	Yes	202 (76.8)	228 (67.7)	0.008	354 (76.3)	76 (55.9)	<0.001
	No	61 (23.2)	109 (32.3)		110 (23.7)	60 (44.1)	
Years of practice	0–10	16 (6.1)	51 (15.1)	<0.001	57 (12.3)	10 (7.4)	0.231
	11–15	31 (11.8)	54 (16.0)		71 (15.3)	14 (10.3)	
	16–20	24 (9.1)	51 (15.1)		57 (12.3)	18 (13.2)	
	21–30	103 (39.2)	101 (30.0)		153 (33.0)	51 (37.5)	
	>30	89 (33.8)	80 (23.7)		126 (27.2)	43 (31.6)	
Population in work area	≤ 19,999	119 (45.2)	87 (25.8)	0.003	157 (33.8)	49 (36.0)	0.728
	20,000–49,999	72 (27.4)	92 (27.3)		128 (27.6)	36 (26.5)	
	50,000–99,999	19 (7.2)	39 (11.6)		48 (10.3)	10 (7.4)	
	≥100,000	53 (20.2)	119 (35.3)		131 (28.2)	41 (30.1)	
Employment status	Individual contractor	120 (45.6)	55 (16.3)	0.001	129 (27.8)	46 (33.8)	0.174
	Public healthcare	143 (54.4)	282 (83.7)		335 (72.2)	90 (66.2)	

Male PHCPs, were more often sanctioned than their female colleagues (P<0.001). Family medicine specialists were more frequently sanctioned in comparison with others (P=0.008). PHCPs working in sparsely populated areas (less than 20,000 inhabitants) were sanctioned in bigger proportion than their counterparts working in urban centres. Individual contractors were sanctioned statistically significantly more frequently compared to PHCPs employed at public health centres.

We did not find any relationship between violence and the practitioner's sex (P=0.185). Family medicine specialists, more often than other PHCPs experienced violence from patients or patients' relatives if they followed HIIS rules (P<0.001). There was also significant relationship between violence and PHCP's age (P=0.05). In all age groups, the majority of PHPCs were exposed to violence, with a difference in the proportions between individual groups (83.3%, 80.3%, 76.1% and 70.1% for the age groups from the youngest to the oldest doctors).

Discussion

This paper analyses the problem of HIIS controls and sanctions of PHCPs on one hand, and PHCPs' exposure to patient violence provoked by upholding the HIIS insurance standards on the other. Multiple factors affected the study's outcome.

In the last 50 years there has been rapid "feminisation" of general practice (21). From the beginning of the new millennium there has been a sharp decrease in medical graduates drawn to the field of general practice. Reasons such as finding the job interesting, likely job satisfaction, aptitudes, likely ability to be successful in a chosen specialty, opportunities, compatibility with domestic and social life, material and intellectual rewards and personal aspirations about how best to contribute to serving patients play a vital role (22). In our study we concluded that sex of the PHCPs was associated with being sanctioned, with the male PHCPs being more often sanctioned than their female

colleagues.Some studies find possible explanations for this in the fact that female PHCPs self-report fewer hours of work than their male peers, are younger, have fewer patient encounters and deliver fewer services, and write fewer prescriptions, but spend longer with their patients during a contact and deal with more separate presenting problems in one visit (23-26). The results of the study showed that there is no relationship between violence and the PHCPs sex. While violence against healthcare workers in general was not associated with sex (15, 27), a systematic review showed that women working in primary care or general hospitals were less likely to be exposed to physical violence than men (15). Other studies showed that women PHCPs experienced more verbal violence and stalking (28, 29).

Age and years of practice also showed association with sanctions, with older PHCPs being sanctioned more often than the younger colleagues ($P<0.001$ and $P<0.001$). Here, we have to take into consideration how the question was formulated: "Have you ever been sanctioned by the HIIS?". Since the question covers their whole career, the chance of those with a longer career to be sanctioned is higher. Additionally, the questionnaire lacked a question to determine the total number of sanctions one PHCP got in their career and whether PHCPs got one sanction for multiple breaches or multiple sanctions over the years for different singular breaches. In our study there was a significant relationship between violence and PHCP's age. A meta-analysis showed that healthcare workers that were younger, exposed to shift work and worked longer hours had a higher risk of experiencing any type of violence (15).

The results showed that family medicine specialists were more frequently sanctioned than other PHCPs ($P=0.008$) and are more often victims of violence from patients and patients' relatives if they follow the HIIS rules ($P<0.001$). Slovenia is a country with a long tradition of family medicine specialist training. Even as part of Yugoslavia, specialist training in GP/FM started as early as 1961, first in Zagreb, Croatia, at the Andrija Štampar School of Public Health, and 1 year later in Ljubljana (30). A new model for vocational training in family medicine was established in 2002, following the recommendations of the European Union of General Practitioners (UEMO). According to the new program, which lasts four years, trainees spend half of their training in a hospital setting and half in general practice, where they are supervised by a trainer in practice (31). Currently family medicine specialty is obligatory, but due to historical reasons, we have 4 types of practitioners of general medicine: a) young doctors who pass their 2-year internship and final exam; b) general practitioners already practicing (some have no specialist training and completed part of their training during residency); c) physicians with no internship (from the old Yugoslav educational system, with a 1-year residency and "state professional" exam), working in general practice for many years, but who have never been vocationally trained; d) physicians who have not yet practiced GP/FM (employed in institutes, pharmaceutical companies, etc., only statistically regarded as GPs) (31). Regarding the results, further research is needed to determine the underlying reasons for why family medicine specialists are more frequently sanctioned than other PHCPs and why they experience more violence from patients and patients' relatives than other PHCPs. It is also interesting to research in the future whether this violence from the patients, influences PHCPs to go against HIIS rules. Patients need to be aware of the fact that family physicians do not limit their rights and that the majority of the decisions in regards to diagnosing and treatment depend on health insurances' regulations (32).

The population count in the work area was also associated with sanctions, but not with violence. In other studies, however, urban settings were shown to be significantly positively associated with violence (15, 27, 28). In Slovenia, the biggest region- Central Slovenian Region (Osrednjeslovenska), has 537 ,893 inhabitants, and the biggest municipality - Ljubljana has 279,631 inhabitants (33). Even though the municipality with the highest number of inhabitants is 269,631, the questionnaire included the category ≥500,000–999,999, because this survey was made in accordance with

AGP/FM SEE member states who have bigger populations. It has to be noted that in our study there were 34 PHCPs who chose the category ≥500,000–999,999. This raises the question about how participants understood the term "workplace area" and selected the category - by municipality or region. As there are no regions below the population of 50,000, we assume that those answers were meant as municipalities, consequently marking population below 20,000 as rural.

Regarding employment status, individual contractors were significantly more sanctioned than PHCPs working in public healthcare, but there was no difference in the experience of violence. This is contrary to the findings of Berendes et al. in which they concluded that the private sector is performing better in drug availability and aspects of delivery of care, including responsiveness and effort, and possibly being more client orientated (34). Another perspective to look at in future research is whether the frequency of control from the HIIS is the same in the public and private sector and whether individual contractors are more often targeted for evaluations.

Large amounts of health care funds are spent on medicines, and this number has been increasing in the last decades in Slovenia - from 384$/capita in 2002 to 546$/capita in 2018 (35). Because of this, policy makers are searching for different strategies to control the costs of medicines while providing patients with the medicine they need. They can do this in two different ways - by introducing educational policies or regulatory policies. Educational policies include laws, rules and regulations that require medicine prescribers to get certain types of information, education or feedback about their prescribing behavior. Regulatory policies, on the other hand, include laws, rules and regulations regarding who can prescribe medicines, what type of medicines they can prescribe and how much they can prescribe. Usually, prescribers are monitored to make sure they follow these policies (36). A review covering studies from the UK, Germany and Ireland concluded that drug budgets for physicians in private practice can limit drug expenditure (per item and per patient) by limiting the volume of prescribed drugs, increasing the use of generic drugs or both (37). In Slovenia, even though there is a combination of educational and regulatory policies controlled by the HIIS, in our study we still see that the most common reason for sanctioning is "Prescribing drugs not in accordance with HIIS rules". Maybe a bigger accent on educational policies could reduce these numbers in the future.

In Slovenia, out-of-pocket payments are still relatively low as most health services and medicines are covered by compulsory and complementary health insurance schemes (38). So, another option might be increasing co-payments and out-of-pocket payments as a way of reducing the prescription of drugs above the agreed amount. A systematic review concluded that cap and co-payment policies may reduce the use of medicines and reduce medicine expenditures for health insurers. However, they may also reduce the use of life-sustaining medicines or medicines that are important in treating chronic, including symptomatic, conditions and, consequently, could increase the use of healthcare services. Fixed co-payment with a ceiling and tiered fixed co-payment may be less likely to reduce the use of essential medicines or to increase the use of healthcare services (39).

Physician density in Slovenia is 3.09/1,000 population and is among the lowest in Europe (40). In 2014, the number of PHCPs still lagged behind most EU countries (41), leading to problems of access and over-referrals to specialist care in some parts of the country (42). Our study, on the other hand, showed that issuing referrals not in accordance with HIIS rules wasn't a very common reason for sanctioning.

Having an electronic health record (EHR) is strongly empirically associated with the workflow, policy, communication and cultural practices recommended for safe patient care in ambulatory settings. Even medication safety had a statistically significant and positive relationship to full EHR adoption (43). Another study showed that the use of an electronic system was associated with a reduction in medical errors, compared with the paper-based method (44). EHR also result in

a positive financial return on investment to the health care organization (45). In Slovenia, PHCPs record information about patients both in paper and electronic form. Around 10% of the reasons for sanctions are due to discrepancies between paper and electronic form, most often than not missing information in paper form which was already registered in electronic form. Different computer programs exist in Slovenia for medical records. The most commonly used program is "Hipokrat". None of the programs allow the user to leave a "must- field" empty before proceeding to the next step, thus missing or forgetting to file a critical information is impossible. Since the implementation of the health insurance card, in which personal data, previous and ongoing therapy, history of prescribed medical devices and type of insurance are recorded we find paper form to be redundant, if there is further investment in the content of the health insurance card (46). Adding the needed information which is within the paper form to the EHR form, will help us achieve faster, less error prone recorded medical history, carried by the patient and at the same time real time copy will exist at the hands of the chosen PHCP database servers.

The strength of the study is that it is the first one to research the control of HIIS on PHCPs in Slovenia. It opens a lot of new questions about the regulatory system and the policies of sanctioning and gives space for formulating new hypotheses about additional research that could help in optimizing the system. The sample size of PHCPs was also satisfactory and the distribution of the survey through the Medical Chamber of Slovenia helped avoiding selection bias. With all the information given in this study, we think it is very easy for future researchers to optimize the survey and conduct further research.

This study has several weaknesses and most of them are connected to the formulation of certain questions. The first weakness is that we do not know the exact number of the subgroups of gynecologists and pediatricians within the group of PHCPs, because the question asked only if they are family medicine specialists or not. The second one is the question about sanctions: "Have you ever been sanctioned by the HIIS?", which limited gathering data about how many times PHCPs were sanctioned throughout their career. The third one is the question: "Have you ever experienced violence from the patient or patients' relatives if you followed HIIS rules?", which also limited gathering data about how many times PHCPs experienced violence. Finding research about Slovenia for comparison was also very hard, since our study is the first one to explore the issue.

Conclusion

Shortcomings were observed from PHCPs regarding the adherence to HIIS rules and limitations. Family medicine specialists were sanctioned more frequently than other PHCPs, individual contractors were sanctioned more frequently than public healthcare PHCPs and PHCPs in working area with a population less than 20,000 were more frequently sanctioned than those working in an area with a bigger population count. The three most common reasons for sanctions were: "Prescribing drugs not in accordance with HIIS rules", "Prescribing technical or orthopaedic devices not in accordance with HIIS rules" and "Excessive sick leave and/or unjustified sick leave". Family medicine specialists were more often victims of violence from patients and patients' relatives if they followed the HIIS rules. Age was also associated with violence from patients and patients' relatives if PHCPs followed the HIIS rules. We believe that the future lies in partnership between PHCPs, patients and the HIIS, which could lead to strengthening the PHC provision and a more efficient healthcare system overall.

What Is Already Known on This Topic:
Slovenia has a gatekeeper system comprised of public and private PHC. The Health Insurance Institute (HIIS), Health Inspectorate and Ministry of Health are responsible for following and regulating the work of PHCPs in Slovenia. These regulatory bodies use measures such as warnings and financial fines to control and improve the work of PHCPs. PHCPs regardless of control often experienced violence in their workplace.

What This Study Adds:
Our study on the sample of 600 PHCPs showed that the sanctioning of PHCPs in Slovenia is significantly related with age, sex, years of prac-

tice, specialty of family medicine, workplace and employment status. Most PHCPs were exposed to verbal violence which was significantly associated with family medicine specialty and age. All this information should be taken into consideration in order to change and modernize the system of monitoring and control.

Authors' Contributions: Conception and design: DRP, LS, MZ, AZ and NZ; Acquisition, analysis and interpretation of data: DRP, AZ and NZ; Drafting the article: AZ, NZ, DRP, LS and MZ; Revising it critically for important intellectual content: AZ, NZ, DRP, LS and MZ; Approved final version of the manuscript: AZ, NZ, DRP, LS and MZ.

Conflicts of Interest: The authors declare that they have no conflicts of interest.

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Case Report
Acta Medica Academica 2021;50(2):340-343
DOI: 10.5644/ama2006-124.351

Langerhans Cell Histiocytosis in a Three-Year-Old Girl in Bosnia and Herzegovina

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Received: 10 January 2021; **Accepted:** 13 April 2021

Abstract

Objective. To present a rare disease, and to point out that clinical manifestations treated for a long period of time without an adequate response to therapy may be a manifestation of a rare disease. **Case report.** We present the case of a 3-year-old girl who had been drinking a large amount of water for the previous ten days with frequent urination, and who experienced the worsening of symptoms of scalp dermatitis that had been treated for a year without success. Physical examination revealed a maculopapular rash on the scalp, neck and both ear shells, and exophthalmos of the right eye with periorbital edema. Magnetic resonance imaging of the orbits showed extensive lesions of the skull bones. Further diagnostic evaluation revealed similar lesions in other bone structures. Biopsy of the affected region, microscopic and immunohistochemical analysis led to diagnosis of Langerhans cell histiocytosis. **Conclusion**. Langerhans cell histiocytosis mostly occurs in the first three years of life. The incidence is 4-5 patients per million children under 15 years of life. The clinical presentation is highly variable, and can range from isolated, self-healing skin and bone lesions to life-threatening multisystem diseases. Due to the diverse clinical picture, that is often unrecognized, these patients are often referred to other specialists, resulting in the treatment of individual symptoms rather than the underlying disease.

Key Words: Langerhans-Cell ▪ Child ▪ Dermatitis ▪ Exophthalmos ▪ Rare Disease.

Introduction

Histiocytoses are a rare and heterogeneous group of diseases characterized by pathological accumulation and multiplication of cells of the monocyte-macrophage system in tissues. The World Health Organization divides histiocyte diseases into dendritic cell diseases, macrophages, and histiocyte malignancies. The Langerhans cell (LC) is a bone marrow-derived mononuclear cell, belonging to the dendritic cell family. Factors that play a role in the etiology and pathophysiology are: infections (especially viral), immune system cell dysfunction, neoplastic mechanisms, genetic factors, race/ethnicity, and a combination of these causes (1, 2). We present a case of a rare disease from initial symptoms to diagnosis.

Case Presentation

A three-year-old girl had been brought to the pediatrician in primary health care. For the previous ten days she had been drinking a large amount of water which was accompanied by frequent urination. Her scalp dermatitis had been treated unsuccessfully for the previous year, and the symptoms were deteriorating. At the physical examination there were changes on the scalp, neck and both ear shells, in the form of erythema, papules and crusts, with odor (Figure 1). There was evident proptosis of the right eye, with periorbital edema and divergent strabismus. The general condition of the child was good. Laboratory results were normal, except for mild anemia (Hgb 10.3g/dl, Hct 32%, MCV 60 fl). Due to suspicion of a retrobulbar tumor mass an ophthalmologist was consulted and rec-

ommended urgent magnetic resonance imaging (MRI) of the orbit.

Examination of the child's medical records indicated that child was born healthy. At the age of two, changes began on her scalp and she was referred to a dermatologist. The dermatologist treated her under the diagnoses of Seborrheic dermatitis, Eczematous dermatitis and Tinea capitis, but the lesions persisted.

Two months after the scalp lesions appeared, she presented for an examination with a petechial rash. Laboratory results showed thrombocytopenia (PLT 70×10^9/L). She was examined by a hemato-oncologist, treated on an outpatient basis with vitamin C, and her platelet count returned to normal within 7 days. She had not been examined by a pediatrician in the previous six months, but she visited a dermatologist in a private practice. MRI of the orbit showed extensive lesions of the skull bones, corresponding to the lesions usually seen with Langerhans cell histiocytosis (Figure 2).

Figure 1. Scalp skin involvement.

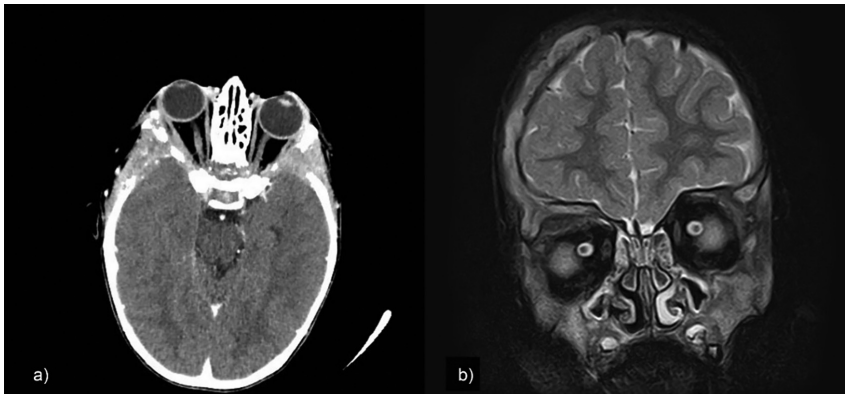

Figure 2. a) CE-CT axial scan revealed the presence of osteolytic lesions in the skull, causing right-side proptosisb; b) Coronal T2w MRI scan indicates the involvement of the right orbital roof, along with the rest of the frontal bone with isointense osteolytic lesions.

The diagnostic evaluation continued in the hemato-oncology department. Osteolytic lesions were also found on the bones of the pelvis, femur and humerus (Figure 3).

After femoral bone biopsy, the pathohistological, microscopic and immunohistochemical findings confirmed the diagnosis of Langerhans cell histiocytosis. Chemotherapy started according to the protocol.

Figure 3. X-ray representation of the osteolytic lesions in the left iliac bone and metaphysis of the right femur.

Discussion

Langerhans cell histiocytosis (LCH) can occur at any age, but the incidence is highest in the first three years of life. The current classification differentiates between the single system disease (SS-LCH) and the multisystem disease (MS-LCH). MS-LCH is defined as the involvement of two or more organs or organ systems. The following organ systems are classified as risk organs, and their involvement indicates a worse prognosis: the spleen, liver, hematopoietic system, and lungs. SS-LCH includes the involvement of one of the following systems (unifocal or multifocal involvement): bones, skin, lymph node, lungs, central nervous system or other (thyroid, thymus) (3-5). The most commonly affected organ in LCH is bone, where changes are present in 80% of cases (61% in the skull). It manifests as a tumor mass, sometimes accompanied by pain and swelling (6, 7). The time from the onset of bone lesion symptoms to the diagnosis of LCH varies, ranging from 1.5 to 4 months, according to a study in Japan. However, there are cases where the changes lasted more than 6 months (8, 9).

Skin lesions occur in 40% of cases, and they are the first manifestation of the disease in 80% of patients. They have different clinical manifestations, from varicella-like changes, seborrheic eczema, or macular rash. The time from the appearance of clinical symptoms of skin lesions to diagnostic biopsy is at least 3 months, in some cases more than 2 years. The final LCH diagnosis is based on histological and immunophenotypic examination of the tissue. Treatment depends on the severity of the disease and the number of organs affected (10, 11, 5).

Conclusion

The aim of this paper is to present LCH as a rare disease, and to indicate the time needed from the initial symptoms to the establishment of the final diagnosis through review of other studies. The clinical presentation is highly variable and can range from isolated, self-healing skin and bone lesions, to a life-threatening multisystem disease. Due to the diversity of the clinical features in this disease, patients are often referred to other specialists (dermatologist, orthopedist, ear, nose and throat specialist or pediatric dentist), which results in the treatment of individual symptoms rather than the underlying disease. Cooperation among specialties is important for early establishment of the correct diagnosis. The prognosis is better in older children in whom the disease is limited to the skin and bones, while children with an affected liver, spleen and bone marrow have a poorer prognosis. Making the right diagnosis in a short period of time is a significant challenge for physicians, and considerably influences the prognosis of the disease and the quality of life of the patient.

What Is Already Known on This Topic:

LCH is a rare disease, with unexplained etiology and unpredictable clinical course. It is most often manifested by changes in the bones and skin, in the form of a single systemic disease. In young children, skin changes often progress to the multisystem form. Patients with unifocal disease generally have a good prognosis. Patients with multifocal LCH have a variable prognosis, depending on how quickly the disease continues to progress and the patient's response to treatment.

What This Study Adds:

Only a few cases of LCH in children have been described in BH, but more detailed studies on this disease have not been published. We present the first case of a three-year-old girl where the disease began with changes on the skin, but the diagnosis was set after a multifocal form had developed, with exophthalmos, diabetes insipidus, infiltration of the skull bones, and lesions in other bone structures. From this case we see that bone changes require radiological evaluation. The clinical presentation of LCH with skin lesions should lead to a straightforward diagnosis. A biopsy of a typical skin lesion would be the way to confirm the clinical suspicion and avoid delay in management. For physicians in primary health care it is very important to have information about rare cases in order to be able to establish a diagnosis more quickly and ensure a better prognosis for the patient.

Conflicts of Interest: We declare that we have no conflict of interest.

Authors' Contributions: Conception and design: ALB and MLK; Acquisition, analysis and interpretation of data: DB and MLK; Drafting the article: MLK; Revising it critically for important intellectual content: ALB, AK, DB and MLK; Approved final version of the manuscript: ALB, AK, DB and MLK.

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