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National Survey about awareness of Primary Immunodeficiency Disorders among Primary Care Physicians in Saudi Arabia: Protocol and Challenges

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Abstract

Introduction: Primary Health Care Centers (PHCC) are the first contact health facility to which patients in Saudi Arabia can go to seek help. Primary Immunodeficiency Disorders (PIDD) are of various types and severities, and they are associated with a delay in diagnosis. Early diagnosis of PIDD helps to improve the quality of life of affected children and prevent permanent consequences such as organ damage and disability. In this study, we present a protocol of a national survey that assesses awareness among PHCC physicians about diagnosing PIDD and the challenges associated with the execution of this protocol.

Methods: This cross-sectional survey used stratified multistage sampling and systematic random selection of PHCC from a list of PHCC affiliated centers under the Ministry of Health (MOH) in Saudi Arabia. The survey was conducted through phone calls to the selected physicians. Data collection started in April 2020, and it is still ongoing.

Conclusion: In Saudi Arabia, this study will provide baseline data about PHCC physicians' levels of awareness of the diagnosis of PIDD. This will help policy-makers in designing educational courses or programs to increase awareness levels among physicians. The protocol could be used to study other health outcomes at a national level.

Keywords

survey, awareness, primary health care centers, primary immunodeficiency disorders, questionnaire

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Introduction

For many years, researchers have used surveys widely in clinical practice as a tool to investigate and evaluate health services and ultimately improve the implementation of evidence-based healthcare. However, survey research, particularly among physicians, remains a problem for many researchers given the relatively low response rate.[1] A quality survey among physicians may require time, logistics, and financial resources that put it beyond the capabilities of these researchers. Many research surveys conducted among physicians report difficulties when obtaining data, as the response rate may be less than 50%.[2] With lower response rates, bias is more likely.[2] Targeting primary care physicians in a research study is more critical than the general population, as the time demand and workload make them reluctant to participate. At the same time, primary health care physicians are the first-line health providers for the patient. Therefore, their training and ability to diagnose diseases in the earlier phases is very important.

Primary Immunodeficiency Disorders (PIDD) are a group of primarily single-gene disorders of the immune system. Approximately 100 separate PIDD have been described, but <20 probably account for $>90\%$ of cases. Potentially 1/1.200 people worldwide are living with a PIDD, which suggests the underestimated prevalence.[3,4] Although diverse, PIDD share in common the feature of

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susceptibility to infection and result in substantial morbidity and shortened life spans.[5,6] Because PIDD can present as common infections, it is estimated that around 70% to 90% of the cases remain undiagnosed or misdiagnosed by health care providers.[6] Most importantly, prompt diagnosis and treatment can now lead to life-saving treatment and result in marked improvements in the quality and length of life for persons with PIDD.[7-9] Given the nature of the disease, it could be challenging for physicians who are not immunologists to diagnose.

Moreover, early diagnosis can have a significant impact on the excess costs of treating children with PIDD arising from hospitalization due to multiple infections.[10] A national economic study, conducted in the USA, of hospital admissions of PIDD patients showed that children 0 to 18 years old had a longer average stay compared to middle aged patients (11 days vs. 7.1 days). In addition, it was reported that the average cost of treating children with PIDD reached $161 992 per patient.[11] By studying the ability of primary health care physicians to diagnose PIDD early in children, policy-makers can make plans and decisions concerning whether or not educational courses, screening lab tests or other facilities are needed in primary health care centers (PHCC).

Many studies have reported levels of awareness and knowledge with respect to the diagnosis of PIDD. A study in the USA among primary care physicians using a national survey found that 32% of physicians had diagnosed, treated, or referred a patient with primary immunodeficiency disorder in the last 5 years. It was recommended that additional educational efforts targeting physicians be implemented.[12] In Ukraine, only 40% of general practitioners and family physicians scored more than 50% on a set of questions regarding the warning signs and symptoms of PIDD.[13] A study in Kuwait assessed the clinical presentation, associated diseases and syndromes, and the laboratory investigation of PIDD, and found that the level of awareness among pediatricians was deficient in both knowledge and practice.[14]

Similar to many countries, Saudi Arabia has developed its own registries for PIDD.[13,15] The registry, housed at King Faisal Specialist Hospital and Research Centre (KFSH&RC), started in 2010 and includes over 820 patients.[16]

Using a national survey, we assessed the abilities of primary care physicians throughout the Kingdom of Saudi Arabia to demonstrate sufficient awareness of PIDD and to diagnose and refer PIDD patients for proper treatment. The aim of this paper is to illustrate the design of that survey, including the various steps taken along the way that eventually led to the proposal and publication. Henceforth, the survey and its proposal shall be referred to as "the PIDD Study." The lessons learned from designing this study may be of interest to other researchers facing problems that can arise when conducting a survey in a similar population using a similar design.

Materials and Methods

In the PIDD Study, cross-sectional data is collected. The interviewers include medical students and research coordinators who are trained to administer the survey to PHCC physicians across Saudi Arabia. A telephone-administered questionnaire is used to collect data from physicians. A data manager supervises the calling lists and the follow-up of non-response attempts. Telephone calls were preferred as a data collection method over online questionnaire, because the latter is known to be associated with low response rate and increased bias.[17,18] In the case of non-response, substitute interviews are conducted with an alternative physician from the same center.

The study's target population are primary care physicians who work in affiliated centers of the Saudi Ministry of Health (MOH). The PHCC are the framework for providing primary care within the government health care system, which covers more than 70% of health care in the Kingdom of Saudi Arabia.[19]

The PIDD Study Survey Instrument

The data collection tool used in this research is the questionnaire developed by Al-Herz et al. in their study conducted in Kuwait.14 It was edited slightly to suit our targeted population with respect to the demographic information requested by physicians. The questionnaire takes about 15 to 20 min to complete, and consists of 5 parts, including sociodemographic data, clinical data, symptoms/diagnosis presentation, laboratory investigations, and data about practicing patterns for immunodeficiency amongst physicians. Most of the questions used a multiple-choice format with 3 responses: Yes, No and I don't know (see Appendix 1 for the complete version of the questionnaire). It was designed this way because questionnaires that are short, focused, government-administered, and include personal invitation to participants are significantly associated with higher response rates in survey-based studies.[18]

Like the study from Kuwait, this questionnaire was a test to determine how well physicians were able to detect PIDD. Its content validity was established through consultation with 3 independent experts including clinicians, public health specialists and researchers. Their comments and edits were considered to improve the scientific content and the relevance of the questions. It was decided not to translate the survey into Arabic, because we assumed that all physicians would be competent in English and that using the Arabic version would run the risk of presenting confusion, especially when translating medical terms. The survey was reviewed and pre-tested by 15 epidemiology interns to assess its clarity and linguistic consistency. This piloting revealed no areas requiring revision, and the instrument was therefore adopted in its current form

The PIDD Study Sampling

We adopted a multistage cluster sampling strategy to select our sampling frame: PHCC across all 20 regions of the Kingdom. In a previous study,[14] the percentage of physicians who passed the questionnaire was 30%. Based on this, we anticipated a similar rate from the population of interest in the PIDD Study. Considering a confidence interval of 95%, an alpha of 0.05, and given that the total number of PHCC in Saudi Arabia is around 2300, the appropriate sample size was estimated to be 896 PHCC. However, the PHCC were considered as a cluster. Further sampling was needed to determine the number of physicians to be enrolled. We anticipated that there would be correlation of the physician's response within each primary health care unit. The sample size adjustment for this intra-cluster correlation was 0.8. Therefore, we needed a sample of 1120 (896 PHCC divided by 0.8) primary care physicians. However, a maximum of 10% non-response rate was anticipated, and we hence estimated that we required 1260 respondents.

The resulting plan was to take 700 PHCC and select at least 2 physicians from each PHCC. This means that a total of 1400 (700×2) primary care physicians are needed to form our sample. The selection of 700 PHCC out of the total 2300 PHCC was done using a systematic random sampling technique. The systematic sample was generated as follows:

First, a list of all 2300 PHCC was generated, in which the PHCC were ordered by the region in which they were located, that is, all PHCCs in Hail region were listed together, all those in Riyadh were listed together, and so forth. For PHCC belonging to the same region, the list was ordered alphabetically. Each PHCC was associated with a catchment population size provided by the MOH. This statistic was used as a weight for the systematic sampling interval selection process. The sum of the population sizes equaled to approximately 18 million, that is, the population of those registered with MOH-affiliated PHCC across the Kingdom. This sum was divided by 700 (the number of desired PHCC for the sample), which equaled 26 000. This number was then used as the sampling interval for the systematic sample of PHCC from the list. The final sample of PHCC is therefore random and geographically representative of the Kingdom. The sampling process is illustrated in Figure 1.

In developing the budget, various categories of required resources were identified—manpower, communications, equipment/supplies, and publication costs. These calculations were made with respect to the various phases of the PIDD Study—pre-approval (ie, proposal development), data collection, and data analyses and reporting. In total, a budget of over SAR 400 000 or around $ 107 000 was determined to be needed for the successful execution of the study.

At the time of submission of this article, permission to use the questionnaire was obtained from the authors of the Al-Herz et al. study. In addition, approval was sought from both the MOH and KFSH&RC Institutional Review Boards. The participant information sheet (see appendix) that includes the PIDD Study objectives are explained to the respondent physicians by the research team. Once these have been explained, verbal consent is taken from the participants. No identifying data is collected and no coercion is applied. Also, all the personal information of the participants is protected and kept confidential. The safety and rights of the subjects are the most important consideration, and prevail over all other interests.

Project Execution

This project is a collaboration between 2 institutions: KFSH&RC (a tertiary care hospital in the independent health sector) and the MOH headquarters. The KFSH&RC team runs almost every aspect of the project, including the proposal, approvals, data collection, data analysis, and manuscript writing, while the MOH's involvement comprises of facilitating contact with the PHCC through appropriate regulatory processes. The research team from KFSH&RC communicated with the administration of the PHCC at the MOH to get the full list of phone numbers of all PHCC in the Kingdom. This process took 3 months, and the provided list was in some cases outdated: 40% of the phone numbers of the PHCCs provided were not working, and another 10% of the PHCC had either closed or had changed their phone number. Every effort was made to collect the missing numbers using the interactive map on the MOH website. Figure 2 shows the map of PHCC locations in the western area. This was not sufficient to locate all the missing information. At the same time, there was an effort from the MOH Primary Health Care administration to survey all the PHCC around the Kingdom through actual visits to update their information, including phone numbers, emails, manpower, resources, and others. During the next 6 months, we obtained a copy of the updated list, then we modified the list to run the randomization process according to the planned methods. In addition, we obtained an official memo from the MOH Primary Health Care administration that was forwarded to PHCC to inform them about our project and encourage them to participate. Coordinating with the person-in-charge led to a considerable delay in the execution of the project.

Once we received the approved memo, we began data collection, led by a group of trained interviewers as mentioned in the methods. Data collection started in April 2020, and is still ongoing. So far, we called about 50 PHCCs, and collected about 35 surveys. Most of the non-respondent physicians were too busy to answer, because we had to phone during working hours. The pandemic of COVID_19,

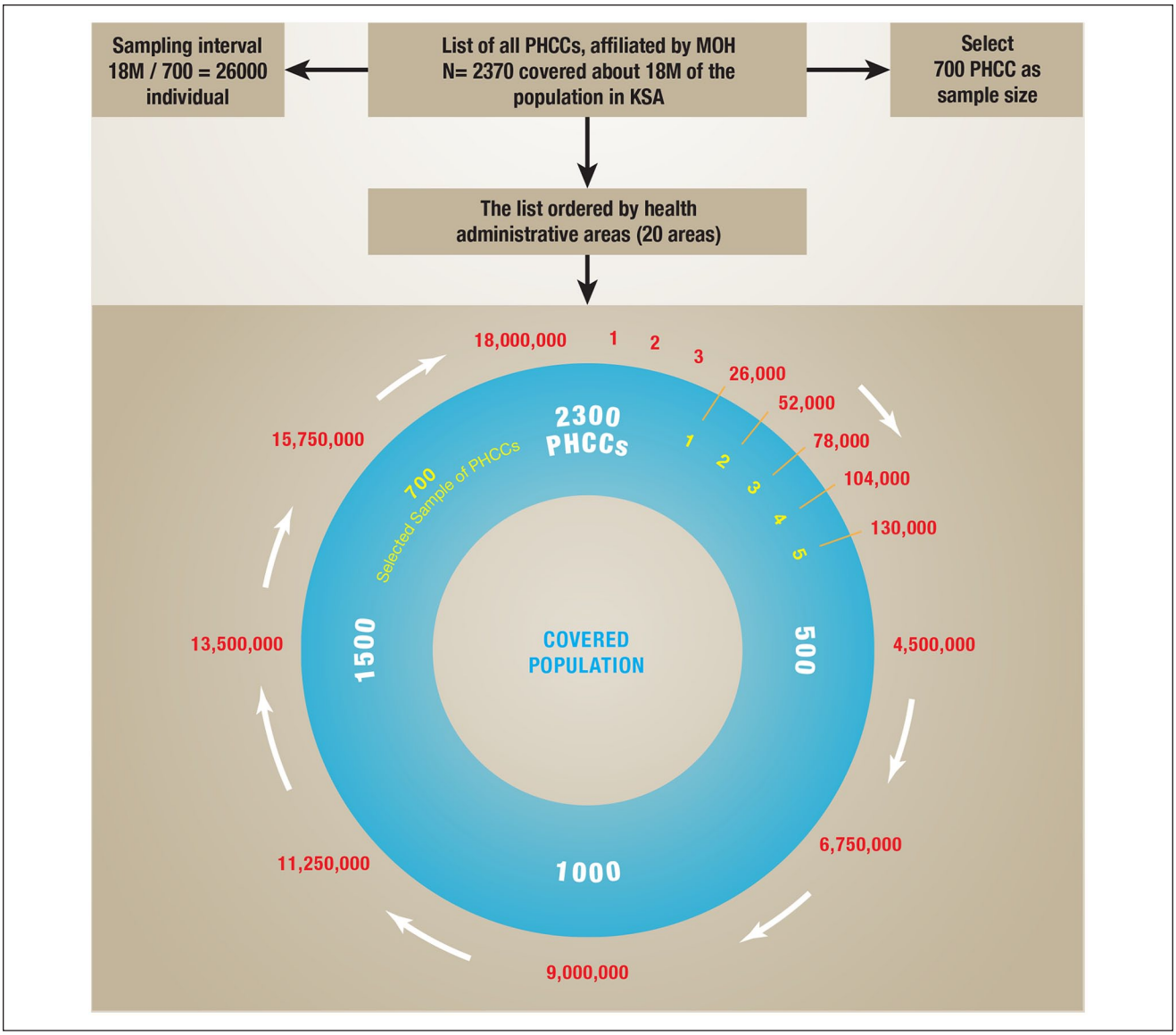

Figure 1. Flowchart of the systematic randomization used to choose the 700 PHCC.

which began in March 2020 in KSA, had a major impact on delaying data collection. Most of the PHCC were directed to be closed during quarantine and curfew times, except for few PHCC which remained open for emergency only or for testing purposes. This made it difficult for us to continue, and we had to pause until normal operations resumed. A major challenge faced in the PIDD Study is thus the delayed execution caused due to difficulty accessing required information and approaching stakeholders. Another challenge is the time allocation between research team members due to their engagement with other job demands, hence the inability to prioritize research. In addition, conducting a research project between 2 organizations requires that the project follows different guidelines of approval and organization forms related to administration.

Conclusion

The goal of this paper was to share our experience with the process of designing a national survey on PHCC. Additionally, it aims to discuss the different administrative challenges that occur, especially when the work involves collaboration between 2 different organizations. Despite the difficulties, the project is run by a multi-disciplinary research group, which includes epidemiologists, policy-makers, statisticians, family medicine doctors and immunologists. This diversity allowed us to bring skills and experience from a range of backgrounds to more efficiently create a rigorous study design. The phone call approach of collecting the data in our experience is much better than online survey with regards to response rate from physicians. Our sample size is

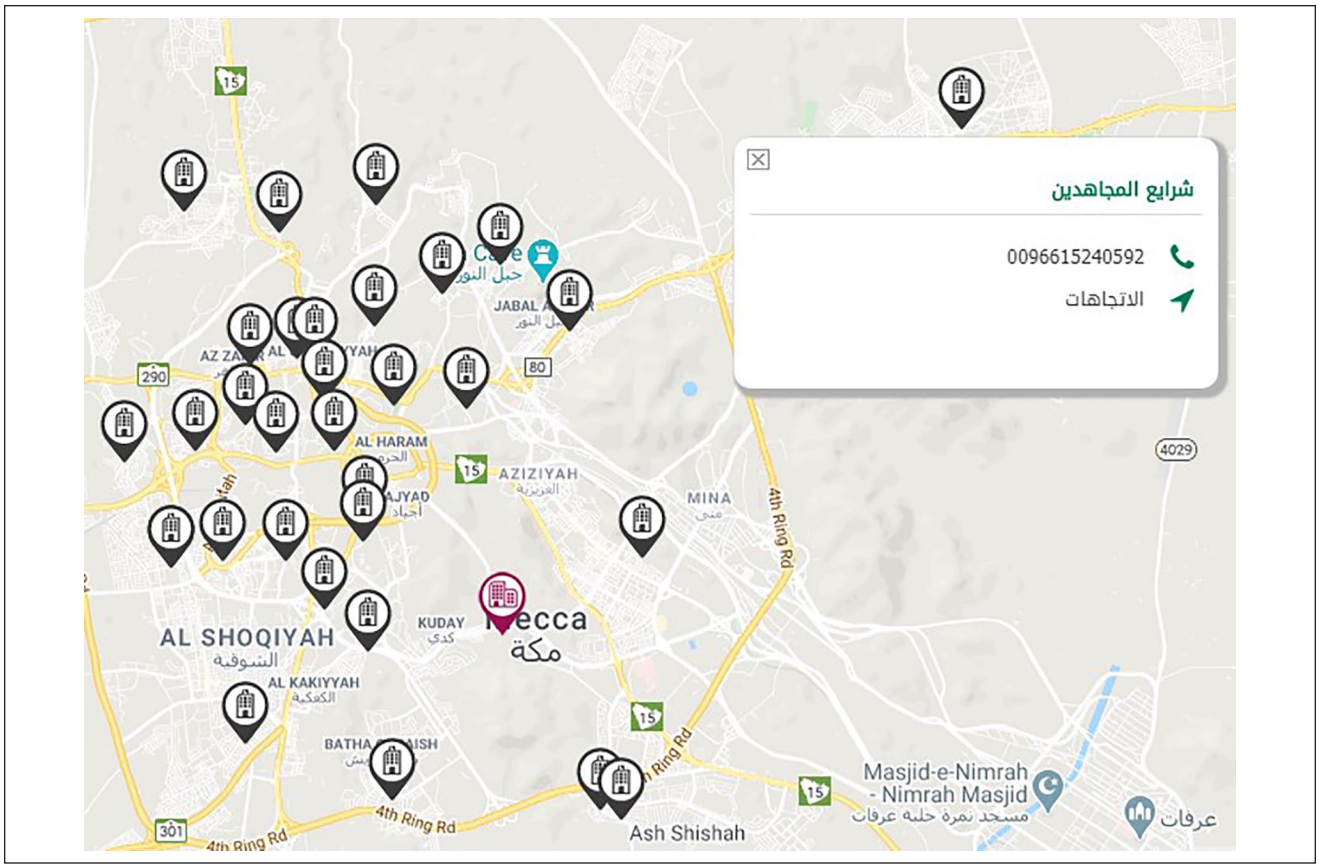

Figure 2. A closer view of the map, showing the phone number of a PHCC.
https://www.moh.gov.sa/eServices/interactive-maps/Pages/default.aspx#/

representative and our findings will therefore be useful to other researchers who are interested in conducting studies in PHCC at the national level. It could also get the attention of decision makers and encourage them to facilitate similar future surveys.

Our article does have some limitations. First, this survey is in the early phases of execution, so the response rate, the budget estimations and the future obstacles we expect are theoretical at the time of writing this article. Also, we focused only on our own example; other study questions might need different tools or different techniques for collecting their data. Nevertheless, our way of calculating the sample size could benefit or encourage other researchers to apply our method for their studies.

Recommendations

National surveys are considered important as they can measure many predictors and outcomes in a population and provide reliable findings when the sample is representative. Such information is valuable for stakeholders who work in related sectors such as health planning and improvement. The information thus obtained can provide the baseline for existing health knowledge among the population in Saudi Arabia, which can be used for comparison and benchmarking with other populations. There is an obvious lack of nation-wide estimates related to public health in Saudi Arabia. The MOH and other health care sectors need to encourage researchers to conduct national surveys and large cohort studies that are epidemiologically more informative than small studies in the field of public health.

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Availability of data and materials

All data are with the authors and available for sharing on request.

Authors' Contribution

F.R conceptualized the study and design, L.A and A.G participated in Literature review. E.DV designed the sampling and calculated sample size. A.F critically reviewed the manuscript and prepared it for publication. All authors shared the drafting of the manuscript and approved the final version.

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Supplemental Material

Supplemental material for this article is available online.

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