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EDITORIAL IN HEALTH POLICY

Combatting the shortage of physicians to alleviate work-related strain

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ore and more often, physicians and he-Lalthcare workers worldwide are experiencing depression, burnout syndrome, and alcohol and substance abuse, as well as a higher risk of suicide than the general population [1-4]. All these mental health outcomes have a negative impact on work performance and lead to poor patient care, which the National Institute for Occupational Safety and Health (NIOSH) has quantified in terms of a 'mismatch between the assistance required by the patient and that available from the care worker' [5]. Research has suggested that burnout syndrome can erode professionalism, increase the risk of medical errors and malpractice litigation, and promote early retirement [6]. In healthcare sector, psychosocial risks associated with mental health disorders include shift work and work-life imbalance; violence and threat of violence; bullying and harassment; emotional demands;

professional responsibilities; and increasingly heavy workload and time pressure [7]. Among the available models for predicting work-related strain and burnout, the job strain model developed by Karasek is probably the most important; according to Karasek's model, high demands on workers are the psychosocial risk factor that scientific research has most importantly associated with all types of mental strain [8]. A Japanese study stated that causes of physicians' unhappiness include budget constraints, the shortage of physicians, excessive demands, inefficient physician distribution, long working hours, hostile media, increasing numbers of lawsuits, and violence by patients [9]. However, the most common reason for overwork in a healthcare settingwhich in Japan was recently associated with a new syndrome called 'karoshi' (death from overwork)—is probably the shortage of staff both in hospital and in home care. In the Eu-

KEY WORDS: Health policy; health care economics and organizations; mental health; occupational health psychology; physicians; psychosocial risks; psychology, industrial.

ropean Union, national experts have included the lack of qualified nurses and physicians-'the most common reason for high workloads'-among the new and emerging psychosocial risks to healthcare professionals [7]. The World Health Organization (WHO) estimates that there is a global shortage of 4.3 million physicians, nurses, and other health professionals. In developing nations, due to the limited number and capacity of medical schools, the number of health professionals is insufficient to deliver essential health interventions, such as skilled attendance at births and immunization programmes [10]. However, both developed and developing countries are facing complicated challenges caused by the imbalance between rapidly increasing demand and supply for healthcare [11]. Indeed, the shortage of physicians is a problem in many developed nations, where it is likely related to a scenario of socioeconomic crisis; more specifically, economic expansion, as reflected by a country's gross domestic product (GDP), may have a strong relationship to an increase in physician supply [12]. Nowadays, the falling supply of healthcare workers is insufficient to address the growth and ageing of the patient population in developed countries, which in combination negatively impact the efficiency and sustainability of healthcare systems [7]. In addition, an excess of bureaucratic responsibility, the ageing of the workforce, and a negative social image conveyed by media coverage of allegations of medical malpractice are all adding to the burden weighing on physicians' psychological wellbeing. This is a likely driver of a social phenomenon consisting in loss of career motivation, leading to a crisis for the medical vocation. In Italy, the 2011–2013 National Health Plan predicted a lack of 22,000 physicians in 2018, based on a continuous decrease in medical university applications, and it was anticipated that the total number of general practitioners could be halved within 10 years [11]. Furthermore, the Italian healthcare system is threatened by public budget constraints caused by the financial crisis, and Italy runs the risk of slowly exporting its better health pro-

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fessionals [13, 14]. In the United States, a new study commissioned by the Association of American Medical Colleges estimated a shortage of between 40,800 and 104,900 physicians by 2030 [15]. As a further complication, the healthcare workforce in developed countries is getting older and consequently suffering from more severe injuries and work-related illnesses than younger workers. Moreover, the lack of new, experienced, and qualified personnel is causing concern due to the greater burden of work for those who remain employed [7]. Recruiting personnel from less developed countries has been viewed as a solution [7], as healthcare worker migration has been increasing worldwide over recent decades [16]. However, migration introduces many cultural and linguistic differences, which can contribute to miscommunication, especially between healthcare workers and patients. Innovations in the healthcare sector, such as new medications or types of surgery and new communication technology, are additional major challenges for healthcare systems and require more investment into education and training [7]. Possible consequences of the physician shortage are a low quality of care due to stricter time constraints on doctor-patient interactions and an increased workload for healthcare practitioners, which can lead to more overwork and stress. Stressed healthcare workers are more likely to be involved in legal disputes that will in turn nourish the 'burnout' phenomenon-and, in a vicious cycle, physicians affected by burnout syndrome will be more prone to treat their patients as objects, having less empathy for them and feeling a lower sense of accomplishment. Since ancient times, trust, confidentiality, and communication have provided the foundation of the physician-patient relationship [17]. Effective doctor-patient communication is pivotal in building a good therapeutic relationship, which is the heart and art of medicine, but unfortunately, doctors affected by mental illness risk breaking this pact [18]. Having healthcare workers affected by mental illness can be considered a dangerous and specific psychosocial risk for the health and

safety of third parties-patients included-in the workplace. According to the WHO, healthcare workers are therefore 'the most valuable resource for health'. Unfortunately, in the European Union, globalization and the economic crisis have resulted in cost-saving actions by governments and greater pressure on companies, as demand for care is still growing. As a consequence, many hospitals have closed down and healthcare staffs are shrinking, and increasing job insecurity is an emerging psychosocial risk factor in the healthcare sector [19, 20]. Additional effects of this precarious work environment are a heavier workload and greater time pressure on those healthcare workers who remain employed. In response to this issue, in 2003, the European Parliament declared the European Work Time Directive to establish a maximum workweek of 48 h for all occupations across the European Union, restricting excessive night work and imposing rest periods. However, many European countries circumvented this Directive by recourse to an 'optout' option [21]. Indeed, in fields such as healthcare, it is very difficult to adhere to scheduled times because of the delicate tasks

carried out by physicians and healthcare workers, who, furthermore, are fewer than before. For all these reasons, the shortage of healthcare workers should be considered as the most important new and emerging psychosocial risk to the health and safety of professionals in the healthcare field. Confronting this phenomenon is imperative to alleviate healthcare professionals' work-related strain and, at the same time, to increase their mental well-being and improve both the patient-doctor relationship and all the outcomes of a good work organization. Achieving this will require a multi-pronged approach, including innovation in delivery of care, greater use of technology, and improved and efficient use of all health professionals on the care team. There is no single solution to resolve this issue [22], but economic investments are surely needed to address this emerging psychosocial risk factor, which is a great challenge for politicians and policy makers around the world.

> Francesco Chirico, *MD* Scientific Director and Founder *Journal of Health and Social Sciences*

Competing interests - none declared.

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ORIGINAL ARTICLE IN OCCUPATIONAL HEALTH PSYCHOLOGY

Depressive symptoms and overwork among physicians employed at a university hospital in Japan

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Abstract

Introduction: The excessive workload of Japanese hospital physicians is a serious social problem due to effects on their mental health status, as well as the potential for medical errors and lawsuits. The extent of overwork among resident physicians employed at national university hospitals in Japan is unknown and needs to be investigated.

Methods: This study used a questionnaire recommended by the Japanese Ministry of Health for hospital physicians working overtime, administered through an interview carried out by an occupational physician during the health surveillance to evaluate: 1) the severity of chronic fatigue; 2) the burden of work; 3) an overwork score derived from these two measures; and 4) presence of depressive symptoms. After the feasibility of the questionnaire was confirmed, both a cross-sectional and a longitudinal study were performed, while statistics analysis included multiple linear regression analysis and chi-square test set at P < 0.05.

Results: Most of the overworked physicians were young medical staffs (48%), whereas postgraduate residents formed a small group (10%). In the cross-sectional study (n = 135; mean age 32.7 years ± 5.6), the histograms of scores for the four factors investigated showed a strong positive skewness, while overtime histograms showed a negative skewness at 4, 3, and 2 months prior to the interview with occupational physician, but positive skewness 1 month prior to the interview. The longitudinal study (n = 26) showed an increase or reduction of overtime respectively having a significant impact on exacerbation or improvement of the overwork score (P = 0.028) and depressive symptoms (P = 0.025).

Discussion and Conclusions: A strong positive skewness of the histograms for items related to overwork might indicate fear of stigma of mental illness amongst young physicians. Physicians employed at Japanese national university hospitals should be protected by the institution, and the roles of occupational physician and health surveillance are crucial and should be effectively implemented.

KEY WORDS: Legislation, hospital; Japan; physicians; preventive psychiatry; psychology, industrial; social stigma; occupational health physicians; overtime work.

Riassunto

Introduzione: L'eccessivo carico di lavoro tra i medici ospedalieri giapponesi è un serio problema sociale per gli effetti sulla loro salute mentale, cosi come per il potenziale di errori medici e di cause legali. Il grado di sovraccarico lavorativo tra i medici che lavorano presso gli ospedali universitari nazionali in Giappone è sconosciuto e necessita di essere indagato.

Metodi: In questo studio è stato utilizzato un questionario raccomandato dal Ministero Giapponese della Salute per i medici ospedalieri che svolgono lavoro straordinario, somministrato nel corso di un intervista effettuata da parte di un medico del lavoro durante la sorveglianza sanitaria per valutare: 1) l'intensità della fatica cronica; 2) il carico di lavoro; 3) il punteggio relativo al sovraccarico di lavoro ottenuto da queste due misure; e 4) la presenza di sintomi depressivi. Dopo che l'utilizzabilità del questionario è stata confermata, sono stati realizzati uno studio trasversale ed uno longitudinale e l'analisi statistica ha incluso l'analisi di regressione multipla ed il test del chi quadrato, con una significatività statistica pari a P < 0.05.

Risultati: La maggior parte dei medici sovraccarichi era composta da medici ospedalieri di giovane età (48%), mentre i medici specializzandi rappresentavano solo un piccolo gruppo (10%). Nello studio trasversale (n = 135; età media 32,7 anni ± 5.6), gli istogrammi dei punteggi per i quattro fattori indagati hanno mostrato una forte asimmetria positiva, mentre quelli per il lavoro straordinario hanno evidenziato una'asimmetria negativa 4, 3 e 2 mesi prima dell'intervista con il medico del lavoro, al contrario un'asimmetria positiva 1 mese prima dell'intervista. Lo studio longitudinale (n = 26) ha evidenziato un significativo impatto che l'incremento o la riduzione del lavoro straordinario hanno avuto rispettivamente sull'aumento o sulla diminuzione del punteggio relativo al sovraccarico di lavoro (P = 0.028) e su quello relativo alla presenza di sintomi depressivi (P = 0.025). **Discussione e Conclusioni:** Una forte asimmetria positiva degli istogrammi per gli items correlati al sovraccarico di lavoro potrebbe indicare il timore dei giovani medici per lo stigma sociale causato dalla malattia mentale. I medici che lavorano negli ospedali universitari nazionali giapponesi dovrebbero essere protetti dalle istituzioni ed il ruolo dei medici del lavoro e della sorveglianza sanitaria è cruciale e dovrebbe essere efficacemente implementato.

TAKE-HOME MESSAGE

In Japanase university hospital physicians, there is an association between overwork and depressive symptoms. The questionnaire set up by the Japanase Ministry of Health was successfully used as a screening tool in this health surveillance programme by occupational physicians and was useful to study the association between depressive symptoms, severity of chronic fatigue and burden of work.

Competing interests - none declared.

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INTRODUCTION

There is growing evidence of the importance of mental health among physicians to prevent the potential risks mental health disorders impose on medical practice [1–4]. Burnout syndrome is a serious problem in overworked physicians. Reportedly, more than half of the US physicians are now experiencing professional burnout, and satisfaction with work-life balance is worsening [1], indicating that physician burnout has reached epidemic levels [2]. Burnout profoundly impairs physicians' working effectiveness, quality of own care as well as patient care, and activity of entire healthcare system [3]. Moreover, there are many studies linking depression and depressive symptoms with medical job. Such studies have been limited to specific populations of physicians such as residents [5], those with fewer length of service [6], physicians in particular medical specialties [7] and general practitioners [8]. Further, a recent systematic review showed a high prevalence of depression among resident physicians resulting in poor-quality patient care and increased medical errors [9]. In addition, it has been showed a direct association between workload and depression among physicians: for instance, at the average of 50-70h/week most physicians work might be associated with a higher prevalence of depression [10].

In Japan, it has been suggested that the recent phenomenon of suicide and Karoshi (death from overwork) among regular workers might not be mainly from financial problems during the recent economic crisis affecting Japan, but from deteriorated work environments and heavy workloads [11]. Work-related consequences include job dissatisfaction, poorer quality of care, professional mistakes, absenteeism, intention to give up the profession, and abandonment [12]. Furthermore, such mental health disorders affect physicians' postgraduate education [13] and family happiness [14], and they are not generally limited to the intensive care field [15, 16]. Current occupational mental health status of physicians employed at Japanese university hospitals remains unknown, because the social en-

vironment of the medical field is dramatically changing in Japan. Indeed, in 2004, a 2-year clinical residency training system became immediately mandatory after graduation for postgraduate fellows in Japan, and the training program was substantially revised by the Japanese Ministry of Health, Labor and Welfare [17] to cover all major medical departments. As a result, postgraduate fellows (known as 'residents') have to rotate through many hospital departments in different clinical fields. Each resident usually stays in a certain department for 1 to 2 months, and hence they have to rapidly develop good relationships with the ward staff in each department and must adapt to different ward systems. Moreover, status as a legal entity was provided to all of the Japanese national universities by the Ministry of Education, Culture, Sports, Science and Technology under the National University Corporation Act [18]. This allows various actions by the national universities, including ventures or joint businesses founded with private companies. Advisors may propose revolutionary action plans to the university President for management changes to improve the university's reputation in the global rankings. However, most staffs working in the university hospitals, including physicians, are busier than before, being burdened with additional tasks and evaluated strictly by the university administration in relation to effort and outcomes. In such a scenario, this study was aimed to survey the situation concerning overworking among hospital physicians and, specifically, to clarify the relationship between the severity of chronic fatigue, the burden of work, depressive symptoms, and the amount of overtime worked by physicians employed at a national university hospital in Fukuoka, Japan.

METHODS

Subjects

The study population consisted of 135 Japanese physicians working overtime at a Japanese national university hospital in Fukuoka. During the study period (April 2014 - March 2017) 690 physicians (approximately 25 full professors, 50 associate professors, 77 assistant professors, 83 tutors, 205 medical staffs, and 250 residents) resulted to be annually employed at this university hospital. According to the Japanese labor law, 100 hours of overtime per month or overtime exceeding 80 hours per month for 3 consecutive months require that each worker must be subjected to health surveillance carried out by an occupational physician, who has to use a specific questionnaire administered with a semi-structured interview based on a checklist recommended by the Japanese Ministry of Health, Labor and Welfare [19]. In this study, physicians who were meeting these criteria were automatically listed by the General Affairs Section of this university hospital and recruited by occupational nurses.

Ethical clearance

All procedures performed in this study were in accordance with the ethical standards of our institutional research committee, i.e., the study design was approved by the Internal Ethics Committee of this University (201401). Interviews with the physicians were required by the labor law and were conducted according to the updated Declaration of Helsinki (2008). Although the questionnaire completion and interview were legally required, the physicians could decide whether to allow their data to be used in this study by providing written informed consent. Interviews with overworked physicians employed at this university hospital were arranged by occupational nurses and carried out by an occupational physician in a quiet room located in the infirmary of the General Affairs Section of the university. Personal privacy was strictly protected.

Study Instruments

We used two instruments recommended by the Japanese Ministry of Health, Labor and Welfare [19], which were a checklist-based interview and a questionnaire (Table 1) composed of three sub-scales to estimate the presence and severity of mental health symptoms related to chronic fatigue (13 items) and depressive status (5 items), and to evaluate the burden of work (7 items). Subjects responded to each item by answering 'rarely', 'sometimes' or 'often'. Based on the severity of chronic fatigue and the burden of work, an 'overwork score' was determined by a specified chart (Table 2) [19]. In this study, we measured only generic symptoms of chronic fatigue status including mental and physical fatigue and not the symptoms of the 'chronic fatigue syndrome' that is a clinical entity affecting mental, physical, neuroendocrine and immune system and overlapping sometimes fibromyalgia [20-22]. The 5-item scale of the questionnaire used to evaluate depressive symptoms was performed by the subjects, with each item receiving a 'yes' or 'no' answer. Diagnosis of major depressive disorder in primary care should be based on DSM 5 criteria or ICD-10 diagnostic criteria, so this questionnaire was only used for evaluating depressive symptoms but not for diagnosing depression per se.

The Feasibility Study

Although this study used instruments recommended by the Ministry of Health, Labor and Welfare, application of this questionnaire has not yet been validated by research. Therefore, Cronbach's α coefficient was calculated to assess the internal integrity and persistency of this questionnaire as applied to hospital physicians, a highly specialized occupational population exposed to work-related stress strain. The feasibility of this questionnaire was tested by investigating the relationship between the overwork score and the severity of chronic fatigue or the burden of work. Multiple linear regression analyses were performed with independent variables including the severity of chronic fatigue and the burden of work, and the overwork score as dependent or 'outcome' variable.

Study design

Cross-sectional study

The cross-sectional study included 135 Ja-

	Severity of chronic fatigue	rarely (0)	sometimes (1)	often (3)
1.	I get annoyed.			
2.	I am anxious.			
3.	I do not feel at ease.			
4.	I am gloomy.			
5.	I cannot sleep well.			
6.	I am in poor condition.			
7.	I cannot concentrate.			
8.	I make many mistakes.			
9.	I feel very sleepy while at work.			
10.	I have no motivation.			
11.	I am tired out.			
12.	I feel fatigued even when I get up in the morning.			
13.	I become tired more easily in comparison with the past.			

Table 1. Questionnaire for overworked physicians recommended by the Japanese Ministry of Health.

	Burden of work	rarely (0)	sometimes (1)	often (3)
1.	Overtime in a month			
2.	Irregular duties			
3.	Burden of business travel			
4.	Burden of nightshifts			
5.	Breaks and naps			
6.	Mental burden of work			
7.	Physical burden of work			

Depressive symptoms		yes (1)	no (0)
1.	I do not have a sense of fulfillment in daily life.		
2.	I cannot enjoy what I used to enjoy.		
3.	Things that I could do easily before have become troublesome.		
4.	I do not consider myself as a valuable human being.		
5.	I always feel tired with no specific reason.		

Note: Parentheses indicate score in each item.

Table 2. Chart of 'overwork' score based on the 'severity of chronic fatigue' and the 'burden of work'.

Total Score in Burden of Work						
0 1 - 2 3 - 5 6 or more						
	0-4	0	0	2	4	
Total Score in Severity	5-10	0	1	3	5	
of Chronic Fatigue	11-20	0	2	4	6	
	more than 20	1	3	5	7	

Note: Overwork score is determined by total scores in burden of work and severity of chronic fatigue.

panese practitioners working overtime at the university hospital. When they visited the infirmary, they were administered the questionnaire with a checklist-based semi-structured interview by an occupational physician according to the Japanese labor law.

Longitudinal study

Among the 135 physicians recruited for the cross-sectional study, 26 of them were also followed in a longitudinal study. This small group of physicians continued to work overtime after the initial interview and, therefore, they were a high-risk group. An annual administered questionnaire was arranged by occupational nurses, and was followed by an interview with an occupational physician. The relationship between overtime and above-mentioned parameters was investigated longitudinally, i.e., the relationship between changes in the amount of overtime versus the overwork score and that between changes in the amount of overtime versus depressive symptoms. 'More overtime' was defined as the amount of overtime increased, and 'less overtime' was defined as the amount of overtime decreased within the study period (n = 26).

Statistical analysis

Data were expressed as mean ± SD for continuous variables. The Kolmogorov-Smirnov test was used to investigate the normality of data distribution. None of the variables with missing data was qualified for this research. Multiple linear regression analysis was performed to identify significant contributors to the objective variable among several explanatory variables in the feasibility study and the cross-sectional study, while the chi-square (χ^2) test or Fisher's exact test was employed in the longitudinal study. Yates' correction was added if necessary. Analyses were performed by using PASW software (Windows version 18.0; SPSS, Chicago, Ill, USA). Statistical significance was set up at P < 0.05.

RESULTS

Subjects and Questionnaire

The subjects enrolled in this study included 135 physicians (M = 120, F = 15), with a mean age of 32.7 years ± 5.6. Most of the doctors were in their 30s (n = 76), followed by those in their 20s (n = 42) and those in their 40s (n = 16). A breakdown of their positions (A) and types of specialization (B) is shown in Figure 1A-B.

In the medical department of a Japanese university hospital, the hierarchical order is as follows: professor, associate professor, assistant professor, tutor (or hospital assistant), medical staff, and resident. As detailed in Figure 1A, medical staff accounted for approximately half of all overworked doctors (48%). Tutors and assistant professors occupied about 18% and 14%, respectively, while residents formed about 10% of the overworked doctors. Percentage of overworking doctors in each job title was about 12% in professors, 20% in associate professors, 25% in assistant professors, 29% in tutors, 32% in medical staffs, and 6% in residents. Figure 1B shows the clinical departments of the overworked doctors. Specialist surgery included cardiac surgery, neurosurgery, and pediatric surgery, while general surgery included thoracic and gastrointestinal surgery. Specialist clinical departments included ophthalmology and otolaryngology. The emergency department included the intensive care unit (ICU) and anesthesiology. Internal medicine included general medicine and pediatrics. Miscellaneous sections included laboratory medicine and radiology diagnostics.

Figure 1A.







Figure 1A-B. The positions (A) and affiliations (B) of the doctors interviewed for this study (n = 135).

The histograms of the severity of chronic fatigue (A), the burden of work (B), the overwork score derived from these two variables (C), and depressive symptoms (D) in the 135 doctors are displayed in Figure 2. The data did not show a normal distribution, and a strong positive skewness was noted in all cases (+ 1.77 in A, + 1.39 in B, + 0.95 in C, and + 2.53 in D). Because the findings were based on data from questionnaires, the feasibility of the questionnaire used required validation.





Figure 2. Histograms for the severity of chronic fatigue (A), the burden of work (B), the overwork score derived from these two factors (C), and depressive symptoms (D) based on data from administered questionnaires.

The Feasibility Study

Cronbach's α coefficient was 0.77 for the severity of chronic fatigue, 0.65 for the burden of work, 0.65 for the overwork score, and 0.80 for depressive symptoms (n = 135). A multiple regression model was created by setting the overwork score as the 'dependent' variable and the amount of overtime, age, gender, severity of chronic fatigue, burden of work, and depressive symptoms as 'independent' variables. As shown in the Table 3, this analysis revealed that the severity of chronic fatigue (standardized partial regression coefficient $(\beta) = 0.24, P < 0.001)$ and the burden of work (standardized $\beta = 0.76$, P < 0.001) were significant contributors, with the coefficient of determination (R^2) being 0.85, indicating that the overwork score purely reflects the severity of chronic fatigue and the burden of work.

	β	Standardized β 95% CI of β		P value				
1) Overwork score ($R^2 = 0.852$)								
Burden of work	0.526	0.762	0.468 - 0.583	< 0.001				
Severity of chronic fatigue	0.099	0.236	0.064 - 0.134	< 0.001				
2) Severity of chronic fatig	ue ($R^2 = 0.610$)							
Depressive symptoms	2.355	0.482	1.771 - 2.940	< 0.001				
Burden of work	0.691	0.421	0.494 - 0.888	< 0.001				
Overworking time per month	-0.022	-0.109	0.045 - 0.001	0.062				
Age	0.064	0.082	- 0.031 - 0.158	0.183				
Gender	0.965	0.069	0.583 - 2.513	0.220				
3) Burden of work ($R^2 = 0.3$	\$96)							
Severity of chronic fatigue	0.398	0.653	0.285 - 0.512	< 0.001				
Overworking time per month	0.020	0.165	0.003 - 0.038	0.022				
Gender	- 0.637	- 0.075	- 1.814 - 0.539	- 1.814 - 0.539				
Age	0.017	0.036	- 0.055 - 0.089	0.636				
4) Depressive symptoms (<i>I</i>	R ² = 0.441)							
Severity of chronic fatigue	0.142	0.692	0.107 - 0.177	< 0.001				
Gender	- 0.263	- 0.092	- 0.642 - 0.117	0.173				
Overworking time per month	0.003	0.080	- 0.002 - 0.009	0.253				
Burden of work	- 0.027	- 0.081	- 0.084 - 0.029	0.343				

Table 3. Multiple linear regression analyses for association of self-administered questionnaires.

Note: β , partial regression coefficient; CI, confidence of interval; R^2 , coefficient of determination (n = 135).

Cross-sectional study

Criteria for completing the questionnaire and interview were 100 hours of overtime per month or overtime exceeding 80 hours per month for 3 consecutive months. Therefore, the distribution of overtime worked by physicians who completed the questionnaire is demonstrated for each month prior to the interview in Figure 3, i.e., overtime worked 4 months (A), 3 months (B), 2 months (C) and 1 month (D) before the interview. As the date of the interview approached, numbers of physicians included in the histograms increased gradually (n = 61 in A, n = 87 in B, n = 116in C, n = 135 in D), and mean overtime increased from 65 ± 25 (range: 14 to 119) hours in A to 101 ± 22 (range: 66 to 211) hours in D. The histograms did not show a normal distribution, with negative skewness increasing from A (-0.29) to C (-1.42), whereas D showed positive skewness (+1.77). This indicates that the physicians made efforts to keep their overtime below 80 hours per month (the mode in A, B and C was 71 to 77 hours per month), but they did not pay attention to self-regulation when overtime clearly was exceeding 80 hours (the mode in D was 87 hours per month). Normal distribution curves derived from actual data were added.







Figure 3A-D. Distribution of overtime at 4 months (A), 3 months (B), 2 months (C) and 1 month (D) prior to interview by an occupational physician.

As showed in Table 3, a multiple linear regression analysis was performed to identify independent contributors to the severity of chronic fatigue, the burden of work, and depressive symptoms. Significant contributors to the severity of chronic fatigue were the depressive symptoms (standardized $\beta = 0.48$, P < 0.001) and the burden of work (standardized $\beta = 0.42$, P < 0.001), with R^2 being 0.61. Significant contributors to the burden of work were the severity of chronic fatigue (standardized $\beta = 0.65$, P < 0.001) and the amount of overworking time prior to the interview (standardized $\beta = 0.17$, P = 0.022), with R^2 being 0.40. This analysis also revealed that only the overwork score (based on the severity of chronic fatigue and the burden of work) contributed significantly to depressive symptoms (standardized $\beta = 0.40$, P < 0.001, $R^2 = 0.21$, not shown). In more detail, only the severity of chronic fatigue was a significant contributor to the depressive symptoms (standardized $\beta = 0.69$, P < 0.001, $R^2 = 0.44$).

Longitudinal study

Among the 135 Japanese physicians in the cross-sectional study, only 26 were included in the longitudinal study and interviewed by an occupational physician annually for 2 to 6 years $(2.72 \pm 1.06 \text{ years})$. These physicians worked overtime repetitively after the initial interview and, therefore, they were listed as a high-risk group. Cross tables were constructed for the relationship between changes in the amount of overtime versus the overwork score and between changes in the amount of overtime versus depressive symptoms (Table 4). If overtime increased ('more overtime') or decreased ('less overtime') within the study period, there was a significant impact on the changes of the overwork score ($\chi^2 = 4.82$, P = 0.028) and depressive symptoms (χ^2 = 5.05, P = 0.025).

Table 4. Effect of an increase or decrease of overtime on the overwork score	and depressive symptoms.
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1) Effect of overtime on the overwork score							
	More overtime	Less overtime					
Overwork score increased	8	6					
Overwork score decreased	1	11					
$(df = 1, n = 26, \chi^2 = 4.82, P = 0.028)$							
2) Effect of overtime on depressive symptoms							
	More overtime	Less overtime					

7

2

 $(df = 1, n = 26, \chi^2 = 5.05, P = 0.025)$

Depressive symptoms worse

Depressive symptoms improved

DISCUSSION

The present study surveyed chronic fatigue, the burden of work, depressive symptoms, and monthly overtime hours investigating the relationships among these factors in physicians employed at a Japanese national university hospital after that the feasibility of the questionnaire used to obtain data was confirmed.

Summary of key findings

The main findings of the present study were as follows: 1) about half of the overworked physicians were relatively young medical staff; 2) histograms for the severity of chronic fatigue, the burden of work, the overwork score derived from the previous two factors, and depressive symptoms all showed a strong positive skewness; 3) the overtime histogram showed a negative skewness at 4, 3, and 2 months prior to interview by an occupational physician, but a positive skewness 1 month prior to the interview (cross-sectional study, n = 135; and 4) an increase or decrease of overtime had a significant impact on exacerbation or improvement of the overwork score and depressive symptoms (longitudinal study, n = 26).

Current status of physicians

Currently, electronic medical records are used in the majority of Japanese hospitals and electronic systems promote both efficiency and accuracy of hospital management. However, the burden on physicians is increased at every step of electronic input concerning patient care including admission planning; prescription of medications; ordering medical, laboratory, or radiology tests; providing orders for surgery, transfusion, rehabilitation, and nutritional supplementation; and discharge planning [4]. Suzuki reported that Japanese physicians working in general hospitals are exhausted and have poor mental health [23]. Moreover, rapid hospitalization of patients to increase the bed 'turnover rate' and to reduce empty beds is required, because national university hospitals have to make a profit after obtaining status as legal entities [18]. In such circumstances, this study demonstrated that residents are protected by the new 2-year postgraduate clinical training system that became mandatory in 2004 [17], because the percentage of overworking residents was only 6%. Conversely, young medical staff and tutors showed the highest percentage of overworking (32% and 29%, respectively) among all the job positions, probably because they have to combine the three major tasks of clinical duties, research, and education (Figure 1A). Many medical staffs are employed by this university hospital on a short-term basis for one or several years, and hence they can survive this relatively short period filled with many duties.

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It can be difficult to reduce overtime in some departments (see Figure 1B) due to long-lasting or emergent surgery or staff shortages. However, it seems that physicians generally make efforts to reduce their overtime. In the cross-sectional study, negative skewness of the distribution of overtime a few months prior to the interview supports this speculation (Figure 3A-C). However, the positive skewness of the distribution of overtime at 1 month before the interview (Figure 3D) indicates that overtime worked by these physicians was different from the average overtime of about 80 hours in the other months, i.e., physicians working more than 100 hours of overtime per month may have no motive to limit their overwork to 80 hours per month.

Implication for future research

In spite of current status of overwork among Japanese hospital physicians, work-life balance and individual well-being are important to improve their mental and physical health and to prevent medical errors and lawsuits [2, 3, 24]. The present study indicates that reducing overtime leads to improvement of the overwork score and depressive symptoms (Table 4). These findings may promote and establish a strategy to prevent burnout, depression and death from overwork as a matter of national policy. Actually, Japanese Government passed the Act on promotion of preventive measures against Karoshi and other overwork-related health disorders in 2014. However, physicians have to respond to the emergency call from their hospital by the medical practitioners' law. Therefore, it is not clear whether physicians may benefit from this Act [25].

Strenghs and Limitations

The findings of the present study should be interpreted with caution due to some limitations. The first limitation is related to evaluation of depression. This study was based on data gathered from the questionnaire used to assess long working hours and the mental health status of Japanese hospital physicians. The Japanese Ministry of Education, Culture, Sports, Science and Technology recommended using this five-item questionnaire for evaluation of depressive symptoms and overworking [19]. However, this study evaluated only depressive symptoms, but not depression *per se*, which is the most prevalent mental health problem in the working-age population. Likewise, this study estimated symptoms of chronic fatigue status but neither 'Chronic fatigue syndrome' or 'Burnout syndrome' [20-22]. However, there is some evidence that Burnout syndrome could share some characteristics with depression [26] and chronic fatigue syndrome [27–29], and in this study depressive symptoms were found to be linked to severity of the chronic fatigue status. The second limitation was the small number of physicians included in the longitudinal study, due to the characteristics of the Japanese university hospitals. Indeed, young physicians need to acquire clinical experience at university hospitals to become qualified as specialists. Therefore, many of them usually work there for only one or a few years, preventing long follow-up studies.

The third limitation was related to the validity of the used questionnaire. Although a feasibility study was performed, this questionnaire has not yet validated in past scientific surveys. Moreover, overtime should cover all hours worked outside the authorized working hours, including management of outpatients who visit at night, emergency treatment of hospitalized patients at night, writing scientific papers, and basic laboratory research work. However, some physicians stated that their listed overtime was limited to ward duties at night. Therefore, answers to the questionnaire could be biased. Finally, physicians usually wish to avoid being diagnosed with depression, because stigma associated with a diagnosis of mental illness may have a potential long-term adverse influence on career development [30, 31]. In this study, histograms for the severity of chronic fatigue, the burden of work, and depressive symptoms showed a strong positive skewness (Figure 2). This might mean fear of stigmatization among overworked physicians at this national university hospital.

On the contrary, the strength of this study was that the specific questionnaire set up by the Japanase Ministry of Health was successfully used for the first time, and it could be a good instrument to evaluate depressive symptoms and overwork in future research.

CONCLUSIONS

After the introduction of mandatory clinical training for residents and the corporatization of the Japanese national universities in 2004, the problem of overworking among university hospital physicians has become serious. In this study, we found that histograms for the severity of chronic fatigue, the burden of work, and depressive symptoms showed a strong positive skewness, implying fear of stigmatization among physicians employed at this national university hospital. In addition, an increase or decrease of overtime had a significant impact on exacerbation or improvement of the overwork score and depressive symptoms. These findings indicate that limiting overtime could be useful to maintain good mental health and prevent overwork-related health problems among hospital physicians, by emphasizing the important roles of occupational physicians and health surveillance at workplace, which should be successfully implemented.

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ORIGINAL ARTICLE IN ENVIRONMENTAL PUBLIC HEALTH

An ensemble prediction approach to weekly Dengue cases forecasting based on climatic and terrain conditions

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Abstract

Introduction: Dengue fever has been one of the most concerning endemic diseases of recent times. Every year, 50-100 million people get infected by the dengue virus across the world. Historically, it has been most prevalent in Southeast Asia and the Pacific Islands. In recent years, frequent dengue epidemics have started occurring in Latin America as well. This study focused on assessing the impact of different short and long-term lagged climatic predictors on dengue cases. Additionally, it assessed the impact of building an ensemble model using multiple time series and regression models, in improving prediction accuracy.

Materials and Methods: Experimental data were based on two Latin American cities, viz. San Juan (Puerto Rico) and Iquitos (Peru). Due to weather and geographic differences, San Juan recorded higher dengue incidences than Iquitos. Using lagged cross-correlations, this study confirmed the impact of temperature and vegetation on the number of dengue cases for both cities, though in varied degrees and time lags. An ensemble of multiple predictive models using an elaborate set of derived predictors was built and validated.

Results: The proposed ensemble prediction achieved a mean absolute error of 21.55, 4.26 points lower than the 25.81 obtained by a standard negative binomial model. Changes in climatic conditions and urbanization were found to be strong predictors as established empirically in other researches. Some of the predictors were new and informative, which have not been explored in any other relevant studies yet.

Discussion and Conclusions: Two original contributions were made in this research. Firstly, a focused and extensive feature engineering aligned with the mosquito lifecycle. Secondly, a novel covariate pattern-matching based prediction approach using past time series trend of the predictor variables. Increased accuracy of the proposed model over the benchmark model proved the appropriateness of the analytical approach for similar epidemic prediction research.

KEY WORDS: Climate; Covariate Pattern Matching; Dengue; Ensemble Prediction; Multiple Linear Regression; Statistics.

Riassunto

Introduzione: La febbre Dengue è stata una delle malattie infettive a carattere endemico più preoccupanti degli ultimi tempi. Ogni anno 50-100 milioni di persone vengono infettate dal virus Dengue in tutto il mondo. Dal punto di vista storico, è stata la malattia infettiva più diffusa nel Sudest Asiatico e nelle Isole del Pacifico. In tempi recenti si sono verificate frequenti epidemie di Dengue in America Latina. Questo studio si è focalizzato sulla valutazione dell'impatto di differenti predittori climatici a breve e lungo termine sui casi di Dengue. Lo studio, inoltre, ha valutato l'impatto di un modello complesso costituito da multiple serie temporali e modelli di regressione per migliorare l'accuratezza predittiva di questa patologia.

Materiali e Metodi: Dati sperimentali sono stati ottenuti da due città dell'America Latina, San Juan in Portorico ed Iquitos in Perù. Per le differenze climatiche e geografiche, San Juan ha registrato un'incidenza più alta di Dengue rispetto ad Iquitos. Usando correlazioni crociate differite, questo studio ha confermato l'impatto della temperatura e della vegetazione sul numero di casi di Dengue per entrambe le città, sebbene con gradi e gap temporali differenti. E' stata costruita e validata una strategia complessa fatta di modelli predittivi multipli attraverso un set elaborato di predittori derivati.

Risultati: La strategia predittiva complessa proposta ha ottenuto una media di errore assoluto pari a 21,55, rappresentando 4,26 punti in meno dei 25,81 ottenuti attraverso il modello standard negativo binomiale. I cambiamenti nelle condizioni cimatiche e nell'urbanizzazione sono risultati essere dei forti predittori come empiricamente evidenziato da altre ricerche scientifiche. Alcuni predittori sono risultati essere nuovi ed utili, non ancora esplorati in precedenti e rilevanti studi.

Discussione e Conclusioni: Due risultati originali sono stati ottenuti in questa ricerca. Innanzitutto, un aspetto ingegneristico focalizzato ed allineato con il ciclo vitale della zanzara vettore. Secondariamente, un nuovo approccio predittivo basato sulla corrispondenza di modelli covariati usando pregresse serie temporali degli andamenti delle variabili predittive. Un incrementata accuratezza del modello proposto rispetto al modello standard ha provato l'appropriatezza dell'approccio analitico nell'ambito della ricerca scientifica sulle previsioni riguardanti simili epidemie.

TAKE-HOME MESSAGE

Climatic conditions and urbanization have considerable impact on Aedes mosquitoes' lifecycle which subsequently affects the spread of dengue virus. Focused feature engineering can reveal these lagged relationships to form informative predictors. Additionally, ensemble prediction by combining outputs from different models is found to improve accuracy over the candidate models.

Competing interests - none declared.

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INTRODUCTION

Dengue fever and dengue haemorrhagic fever are the predominant arthropod-borne viral diseases in the world [1]. Humans get infected when bitten by Aedes mosquitoes, the vector (carrier) of dengue virus (DENV) [2]. This paper focuses on the analysis of the occurrence of dengue fever in two cities, namely San Juan, Puerto Rico and Iquitos, Peru. Dengue outbreak is more prominent in densely populated areas, as the frequency of Aedes mosquito bites and adaptation of the vector mosquito are higher [3]. San Juan has a population density of 3,190 per km², whereas Iquitos has 1,222 per km² [4]. This makes transmission of dengue easier in San Juan than in Iquitos. In the absence of any effective vaccine, the most effective way to prevent and curb dengue transmission is by reducing the Aedes vector [5].

The ecology of DENV is inherently tied to the mosquito life cycle. The metamorphosis from egg to adult stage takes about one-anda-half to three weeks, while the adult life span ranges from two weeks to a month depending on environmental conditions [6]. The distribution of Aedes mosquitoes is spatially and temporally dynamic, as their life cycles are short and strongly influenced by environmental factors [7]. From the Aedes mosquito bite, dengue symptoms usually start anywhere from 4 to 10 days [8]. Cooler temperatures during the early stage of the mosquito breeding cycle indicate a reduced transmission of the dengue virus [9].

Modelling of such complex relationships and interactions between diseases and climatic precursors has been recognized as a difficult problem in many studies [2, 10–12]. Gonzalez et al. used generalized additive models to capture the non-linear relationships with different weather variables [10]. Sharma et al. used advanced machine learning techniques such as artificial neural networks and support vector machines for predicting malaria outbreaks, where the latter demonstrated a significantly better prediction performance [12]. Using autocorrelation at time delay of up to 3 months and generalized linear models, a strong association was shown between temperature and rainfall with dengue fever incidence [13].

A study on the influence of meteorological factors on the dengue virus incidence in San Juan showed that these factors and dengue transmission patterns varied between years, with increased number of dengue cases peaking after higher rainfall in warmer years [14]. Through wavelet analysis, dengue incidences in Iquitos were shown to have seasonal patterns with no strong relationship with the climatic variables [15]. Similar studies revealed that climatic conditions modify the relative influence of human and climatic factors on dengue transmission patterns [16, 17]. Prediction of dengue incidence in San Juan was attempted using the NASA satellite enhanced weather forecasts with unclear model accuracy due to errors in weather forecasts [18].

The above studies demonstrate that the impact of various climatic as well as socio-environmental factors on dengue have been studied extensively all over the world over last 15 years. Eventually, there have been other studies [19, 20] summarizing, comparing and connecting findings across these studies. Multiple linear regression (MLR) and time series forecasting using Auto-Regressive Integrated Moving Average (ARIMA) models were used most frequently for predicting the number of dengue cases in these papers. A few studies also experimented with other formulations and approaches such as Poisson regression [21], negative binomial regression [22] and spatiotemporal clustering [23].

However, one common limitation of these researches was that they never explored the benefits of any ensemble prediction approaches by building and combining different predictive models with the same data. Moreover, these studies have tackled the problem either as a regression or as a time series forecasting problem. A limited number of studies that did use ensembles [24–26], have rarely used any time-series based models as candidates. Only it [26] was found to use a sequential combination of wavelet analysis, genetic algorithm and support vector machines for dengue case prediction. However, the support vector machine was used as a learner within the genetic algorithm and not as an independent model generating a separate prediction for dengue cases. Hence, it should not be considered a stacked ensemble approach in its traditional sense.

Considering the above, the focus of this study was twofold: first, to explore the different climatic variables and identify appropriate short and long-term lagged predictors that showed strong predictive power empirically; and secondly, to build a comprehensive ensemble prediction framework by combining different time series and regression based predictions. This was done to assess the applicability and superiority of such a technique in improving prediction accuracy over the individual models.

MATERIALS AND METHODS

Data

The DengAI data was downloaded from the DrivenData website as part of a competition on predicting the spread of dengue disease [27]. The data initially came from sources supporting the Predict the Next Pandemic Initiative [28]. Beyond dengue surveillance data, other measurements pertained to vegetation, precipitation, and temperature. According to [29], the dengue surveillance data were provided by the U.S. Centers for Disease Control and Prevention, Department of Defense's Naval Medical Research Unit 6, Armed Forces Health Surveillance Center, in collaboration with the Peruvian government and U.S. universities. On the other hand, environmental and climate data were provided by the National Oceanic and Atmospheric Administration (NOAA), an agency of the U.S. Department of Commerce. More specifically, data included:

- Normalized Difference Vegetation Index (NDVI) measurements;
- Precipitation Estimation from Remotely Sensed Information using Artificial Neural Networks (PERSIANN) satellite precipitation_measurements;

- National Centers for Environmental Prediction Climate Forecast System Reanalysis measurements;
- Global Historical Climatology Network daily climate data weather station measurements.

For the competition, the training data comprised 20 continuous features, recorded weekly from 30 April 1990 to 25 June 2010, totalling 1,456 records (Table 1).

Data exploration revealed the difference in data characteristics of the two cities, with the 20 features showing different data ranges and distribution (Figure 1).

San Juan is located along the north-eastern coast of Puerto Rico and lies south of the Atlantic Ocean. The city occupies an area of 199.2 km², of which 75.4 km² (37.83%) is water [30]. Based on the data provided, the city has a tropical monsoon climate, with an average station temperature of 27.0 °C, ranging from 17.8 °C to 35.6 °C between 1990 and 2008. Rainfall is distributed throughout the year, with an average station precipitation of 26.8 mm, ranging from 0 to 305.9 mm.

Iquitos is the most northern Peruvian city and has an area of 368.9 km². It experiences an equatorial climate, with constant rainfall throughout the year, without a distinct dry season, but a wetter summer [31]. Station temperatures range from 14.7 to 33 °C, with an average daily station temperature of 27.5 °C. The average daily station precipitation is 62.5 mm, ranging from 0 to a high 543.3 mm.

Data Preparation

The data were analysed as part of pre-processing. Actions were taken to enable model generalisation. In making these changes, care was taken to ensure that the data ranges and distribution were not adversely impacted.

Analysis of Missing Values

Analysis of the 20 features revealed that NDVI NE had 194 missing values (about 20% of available data); with additional missing values noted for all the other features. To enable generalization, the missing values were

Feature	Min	Max	Mean	Std. Dev	Outlier (30)	Extreme (50)	Null Value
Year	1990	2010					
Week of Year	1	53					
Week Start Date	19900430	20100625					
NDVI NE	-0.41	0.51	0.14	0.14	5		194
NDVI NW	-0.46	0.45	0.13	0.12	4		52
NDVI SE	-0.02	0.54	0.20	0.07	9		22
NDVI SW	-0.06	0.55	0.20	0.08	11		22
Precipitation Amt	0.00	390.60	45.76	43.72	14	3	13
Reanalysis Air Temp	294.64	302.20	298.70	1.36			10
Reanalysis Avg Temp	294.89	302.93	299.23	1.26	3		10
Reanalysis Dew Point Temp	289.64	298.45	295.25	1.53	10		10
Reanalysis Max Air Temp	297.80	314.00	303.43	3.24	3		10
Reanalysis Min Air Temp	286.90	299.90	295.72	2.57	6		10
Reanalysis Precipitation Amt	0.00	570.50	40.15	43.43	28	5	10
Reanalysis Relative Humidity	57.79	98.61	82.16	7.15	2		10
Reanalysis Saturated Precipitation Amt	0.00	390.60	45.76	43.72	14	3	13
Reanalysis Specific Humidity	11.72	20.46	16.75	1.54	3		10
Reanalysis Diurnal Temp Range	1.36	16.03	4.90	3.55	1		10
Station Avg Temp	21.40	30.80	27.19	1.29	2		43
Station Diurnal Temp Range	4.53	15.80	8.06	2.13	3		43
Station Max Temp	26.70	42.20	32.45	1.96	2		20
Station Min Temp	14.70	25.60	22.10	1.57	8		14
Station Precipitation	0.00	543.30	39.33	47.46	22	7	22
Total Dengue Cases	0.00	461.00	24.68	43.60	9	15	

Table 1. Descriptive statistics of original data provided.

imputed using multiple linear regression (stepwise selection) using the other predictor variables. This approach provided data for most missing values. For rows with missing values en-masse, the NDVI values were replaced with the immediate preceding values, while the Reanalysis and Station variables were replaced with the average of two preceding values. A block of NDVI values was missing for consecutive 14 cases in 1994. These were replaced by the average of the last two rows (progressively) and the same week of the preceding year.

Analysis of Outliers

Aside from missing values, outliers were also detected, using 3σ as inner outlier limit and 5σ as extreme limit, where σ was the observed standard deviation of the feature. Analysis of these outliers revealed that they were plausible values, and as such, they were not treated for this study.

Variable Rescaling

The wide variation in the value ranges resulted from the use of different scale, and necessitated rescaling to avoid biasing the data models. All fields were brought to comparable scales, such as °C for temperature and mm for precipitation.

Correlation Analysis

Correlation analysis of the cleaned dataset indicated that several variables had low to medium correlation with each other and with total dengue cases. The presence of variable correlation suggested possible multi-collinearity and opportunity for dimension reduction (Figure 2).

• Although correlations differed for each



Figure 1. Data distribution by city (SJ, 936 records and IQ, 520 records).

city, the reanalysis specific humidity and reanalysis dew point temperature were the most strongly correlated with total cases. This supported the assumption that mosquitoes thrive in wet climates, which could lead to more dengue cases.

- Temperature and total dengue cases showed positive correlation, indicating higher cases of dengue during warm weather.
- In general, the precipitation measurements had weak correlation to total cases.

This presents data dimension reduction opportunity in the models.

Methods

To ensure proper rigor, objectivity and generalizability of the solutions, due importance was placed on the key modelling aspects as detailed below.

Performance Metrics

Mean Absolute Deviation (MAD) was chosen as the performance evaluation metric in line with the expectations set by the competition. MAD was calculated as:

$$MAD = \frac{1}{n} \sum_{i=1}^{n} \left| A_i - \hat{A}_i \right| \tag{1}$$

where Ai denoted the actual dengue cases and $\hat{A}i$ denotes the predicted number of dengue cases. It should be noted here that this metric penalizes underpredictions during epidemic periods less severely than the traditional Mean Square Error (MSE) metric. This can help select the model that generates more accurate predictions for the regular / normal time periods.

Feature Engineering

Raw features available for analysis were of four different types, *viz*. temperature, precipitation, humidity and the vegetation level. Following the biological lifecycle of mosquitoes and propagation of the dengue virus, these factors were expected to impact the dengue cases at different lag periods. In this aspect, related studies have largely been different from each other and sometimes conflicting regarding the lag periods.

One study [32] found 2 months' lag for rainfall and temperature to correlate well while



Figure 2. Correlation bar-plot for predictor importance.

another study [21] found minimum temperature of last two weeks to be a stronger predictor. Multi-wave dengue outbreak in Taiwan was found to be positively influenced by rainfall and temperature volatility because of two typhoons [33], while another study [34] found negative association with temperature with a 2 months' lag.

Based on these, it was concluded that the lagged effects of the covariates may not be uniform between the two cities under study. The same was supported by [11] which found that the impact of climatic and other predictors varied widely, driven by the geographical and tropical location of the place. Hence, the lag analysis was carried out separately for the two cities expanding up to previous 32 weeks - the maximum period that was found to explain the relationships intuitively based on mosquito lifecycle and typical propagation period of the virus.

Analytically, the appropriate lags for each variable were identified based on *lagged cross-correlation* plots using Transfer Functions. Figure 3 explains how the most suited lags were decided using the average temperature variable for San Juan as an example.

Both the most positively and negatively cor-

related lagged windows were retained as two different lagged predictors. Averaging over multiple weeks (*e.g.* 8-11 and 30-31 in Figure 3) helped smooth the derived variables and was expected to improve their predictive powers further.

Apart from the lagged variables, another set of derived variables were created using the cross-sectional interactions among these variables to mimic the different weather patterns known to impact the spread of dengue, either positively or negatively, *e.g.* a *hot-andhumid* variable was created by combining the temperature and relative humidity variables, a *volatile-weather* week was identified based on the difference between maximum and minimum temperature for the same week.

Finally, a decomposition based time series forecasting model was created for San Juan and Iquitos separately using the actual dengue cases. For San Juan, a *sinusoidal* seasonality was the best fit that achieved its peak during post-monsoon season. For Iquitos, a 3-point centred moving average provided the best fit for seasonality since Iquitos typically faced multiple and random monsoon sub seasons that made the method of curve-fitting infeasible. These time series forecasts were used



Figure 3. Lagged variable creation process based on cross-correlation.



Figure 4. Time series decomposition based prediction for Dengue cases.

as additional predictors in the final models (Figure 4).

Thus, there were 103 additional predictors derived from the given features. In the reviewed literature, there are no studies that performed such elaborate and extensive feature engineering. These new derived features were expected to provide substantial lifts in the model performances.

Predictive Modelling

As discussed in the Introduction section, there have been limited attempts at combining different modelling approaches for prediction of dengue cases. Hence, an ensemble prediction framework (Figure 5) was designed and applied by using three different candidate models. The candidate models were chosen carefully to address the different aspects of the prediction goal. The subsequent sections elaborate more on these candidate models and the rationale behind their selection.

Benchmark Model: Negative Binomial Model (NGB)

Though Poisson regression was used in multiple studies, a more generalized negative binomial regression was selected to create benchmark performance on this dataset, in line with the actual competition. This was to help contrast performance of the proposed framework against an established model. The likelihood function for Negative Binomial distribution can be written as:



Figure 5. Model development process flow.

$$L(\beta | \mathbf{y}, \mathbf{X}) = \prod_{i=1}^{N} \Pr(y_i | X_i) = \prod_{i=1}^{N} \frac{I'(y_i + \alpha^{-1})}{\mathbf{y}! I'(\alpha^{-1})} \left(\frac{\alpha^{-1}}{\mu_i + \alpha^{-1}}\right)^{\alpha^{-1}} \left(\frac{\mu_i}{\mu_i + \alpha^{-1}}\right)^{\mathbf{y}}$$
(2)

where α denotes the over-dispersion parameter. As a result, Poisson regression becomes a special case of NGB when α =0. Use of NGB created a stronger benchmark performance on the dataset than a Poisson regression.

Multiple Linear Regression on Combined Data

The first model is an MLR model built on the combined data of the two cities. Though most studies recommended building different models for different geographic locations (two cities in this case), certain limitations were realized with this approach. In case a city has never faced any major dengue outbreaks in the study period (as was the case for Iquitos); the model will never learn to predict any high dengue outcomes. Building a common model will help cross-learn these different scenarios effectively to make the future predictions for both the cities more robust.

Weighted Multiple Linear Regression on Each City

To balance the possible reduction in accuracy of the first model due to its robustness, a set of city-specific regression models were built to learn the unique relationships and influences of the covariates on dengue cases. Furthermore, it was evident from Figure 4 that dengue epidemics were rare and occurred typically at 3-5 years intervals. A Weighted MLR was used by giving higher weights to these epidemic periods. A logarithmic weight as log $(100 + A_i)$ was found to provide the best results on the holdout data.

Covariate Pattern-Matching

Both the above approaches followed regression methodology and did not use any time series elements apart from the lagged covariates. Though there were several theoretical frameworks for multivariate time series forecasting and panel data analysis, they were either not applicable (*e.g.* Vector Auto-Regression) due to absence of actual dengue cases for the test period or the theoretical foundations were too complex and restrictive to get strong empirical results (*e.g.* Generalized Estimating Equations). This led us to create a simple, intuitive yet powerful methodology which delivered strong empirical results on these datasets.

There were two specific motivations behind the creation of this new methodology. Firstly, the test data had no information on the actual dengue cases which made application of any traditional time series techniques infeasible since most advanced time series models (e.g. ARIMA) depend on recent actuals. Secondly, it was noted that dengue cases followed certain trends of other covariates with a lag, e.g. dengue cases in San Juan generally increased post-summer, which is characterised by the lagged series of temperature attaining a peak at 4-6 weeks back followed by a decreasing trend in the last 4-6 weeks. Since the lags were not always fixed, it was impossible to capture these trend patterns in the individual lagged covariates that were created.

Covariate pattern matching was built on the idea that a similar past pattern of covariates (C) is indicative of the likely changes in the dependent variable (A), *i.e.* the number of dengue cases. Similarity was defined using Euclidean distance between the latest $(C_{t-k+1}, ..., C_t)$ and a past pattern $(C_{t-m-k+1}, ..., C_{t-m})$ based on a fixed window of length k. This memory-based algorithm then iterated over the training data range (m = 1, 2, ...) to identify the closest matching window $(C_{t-M-k+1}, ..., C_{t-M})$.

Once this lagged time point (M) was identified, change in dengue cases, expressed as:

$$\Delta A_{t-M} = A_{t-M} - A_{t-M-1}$$
(3)

for the latest day in that window (t-M) was calculated. This became a prediction of the expected change in dengue cases for the latest day. The following diagram explains this pattern matching idea further using a sliding window of 26 weeks (6 months) that was found to work the best empirically (Figure 6). The same matching process was then repeated for all 19 covariates to get 19 different

predictions about the expected change over previous day's actual. Hence, the final prediction for dengue cases on day *t* became:

$$\hat{A}_{t} = A_{t-1} + \frac{1}{19} \sum_{i=1}^{19} \Delta A_{t-M_{i}} \quad (4)$$

Since, recent actuals (A_{t-1}) were not available in the test data, these were replaced with the predicted values progressively to extend the time series. This approach can also be thought of as a random subspace based *k nearest neighbour* method where k=1 was considered for each subspace of predictors and the identified neighbours were subsequently aggregated by averaging.

RESULTS

The following table summarizes the covariates and their corresponding lags that were selected in each of the regression models. A red coloured lag indicates negative relationship while a green indicates a positive one. It can be observed that both small and large lags featured for different variables as significant predictors (Table 2).

Table 3 shows a summary of the individual model and ensemble accuracies, based on both the training and test data withheld for the live competition.

Predictions for the two cities using each candidate model and ensemble are shown below (Figure 7).

The output showed that the models captured the seasonal patterns in dengue cases for the two cities well. Furthermore, the Covariate Pattern Matching predictions on training data showed significant improvement over regression based methodologies. The results were even better than an Auto-Regressive (AR1) time series model which provided an MAD of 8.15. It should also be noted here that the performance of this model showed deterioration in the test data. This was due to the replacement of the actuals for previous days with the predicted values, since actual values were not available for test period. Using actual past counts is expected to result in a performance similar to the training data even for the test period.
DISCUSSION AND CONCLUSIONS

Multiple models, both time series and regression based, were built in this study. To allow for an objective performance benchmarking, a theoretically justified negative binomial model, with a mean absolute deviation of 25.81 based on test data, was chosen. Both MLR-based models achieved superior performance of 23.68 and 24.00 MAD respectively. This was attributed to the elaborate and extensive feature engineering which helped create meaningful derived variables from the raw covariates. Each feature selected in the final models made intuitive sense and was in-line with the prevailing weather patterns in each city.

Vegetation had a strong negative relationship, which indicated that less urbanized areas are less prone to dengue propagation. This matched the findings from other researches where unplanned urbanization has been linked to dengue spread in different geographies [20]. Similarly, high temperature and humidity in recent past were found to correlate positively with dengue spread. This too was logical, as Aedes mosquitoes are known to flourish in hot and humid weather [11]. Specifically, for Iquitos, however, larger lag in temperature (23-24 weeks) had a negative relationship. This was probably because Iquitos faces a higher average temperature throughout the year and a further increase above 30 °C during summer. Since more than 30 °C temperature becomes somewhat hostile for larva and pupa growth of Aedes mosquitoes [16], it might be working as a natural control for the subsequent weeks.

Heavy rainfall was found to have negative impact on dengue spread, again in line with other published researches [20]. Hence, this research broadly reaffirmed that climatic factors indeed impact dengue cases and with varied lags. However, most of the other researchers used lags up to 2 months (8 weeks) whereas the lags in this study spanned up to 6 months. Thus, some of the findings were new (*e.g.* negative impact of 23-24 weeks' prior temperature) and could not be validated against any published results. These new features should be studied for other locations to assess their incremental impact on prediction performances.

This study has indicated that climate change has a positive correlation with the incidence of dengue fever. This is supported by the World Health Organization (WHO) claims that changes in infectious disease transmission patterns are a likely major consequence of climate change [35]. Therefore, there is a need to study the possible underlying contributing factors and their relationships through the development of complex integrated models, in order to predict health outcomes and take the necessary preventive actions. This can help achieve one of the Sustainable Development Goals by WHO to tackle infectious diseases that are brought about by human-induced climate changes [36, 37].

Finally, the proposed *Covariate Pattern-Matching* methodology showed promising results in capturing the short-term (weekly) changes in dengue cases. Training period performance for this approach was stronger than a traditional AR (1) model. The same methodology is readily generalizable to any time series based prediction problem that is expected to have an auto-regressive property.

Finally, an ensemble approach was found to provide stronger results than each individual candidate model. Applying different modelling approaches to introduce diversity in model predictions seemed to be the key driver for this. Empirically, the MAD stood 4.26 points (25.81 vs. 21.55) lower than the benchmark prediction on this dataset.

Future research

There are three key areas where future research on this prediction problem should be pursued. First is to gather more information about the environment and response systems, along with the climatic variables. Socio-environmental factors [20] and policy-driven healthcare response systems [33, 38] were found to contain useful information about potential dengue spreads. This is reasonable



Figure 6. Demonstration of the Covariate Pattern Matching approach.

Table 2. Independent variables	(numbers show la	gs) selected in each	n regression model.
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Covariate	MLR-Combined	WMLR-San Juan	WMLR-Iquitos
Trend	0	0	0
Precipitation	16-23	16-23, 3-29	
Vegetation	15-21	3-20	
Max. Temp	0	0, 10-15	9-12
Avg. Temp			2-4, 23-24
Min. Temp	9-11, 18-23	30-32	15-17
Temp Range		27-32, 10-17	24-25
Rel. Humidity			15-17

Table 3. Performance	e details for	the candidate	models and	final ensemble.
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Model	Training Accuracy (MAD)	Test Accuracy (MAD)
Benchmark Model (NGB)	17.28	25.81
MLR on Combined Data	16.94	23.68
WMLR on City-wise Data	16.28	24.00
Covariate Pattern-Matching	7.23	26.05
Ensemble	8.94	21.55



Figure 7. Weekly Dengue cases prediction for the test period.

because a locality with a strong public health control system (*e.g.* regular fumigation, waste water disposal) is expected to have lower dengue cases due to such preventive measures. Introduction of these variables can carry significant information about future outbreaks along with the climatic variables used in this study [39].

Secondly, the approaches exploited the linear relationships using different regression techniques. Application of more complex non-linear techniques such as Neural Networks, Random Forests, and Support Vector Machines can help model the latent non-linear relationships for improved performance. A caution would be to control for overfitting the training data, which was found to be a practical concern from an initial application of these techniques on this data.

Finally, the proposed *Covariate Pattern-Matching* is still a naïve approach. This methodology can be improved further, using options such as weighted averages or through variables selection based on *similarities*. Future studies should explore these and prove generalizability of this approach by applying it on other forecasting problems.

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ORIGINAL ARTICLE

Prediction of spirometry parameters using chest circumference in Sri Lankan boys aged 8-16 years

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Abstract

Introduction: Most normative standards for spirometry are established based on height, weight and body mass index. We have investigated chest circumference as an alternative to height for interpretation when accurate height measurements cannot be obtained.

Methods: This is a cross-sectional study conducted in Sri Lankan boys aged 8-16. Spirometry measurements were taken adhering to American Thoracic Society/European Respiratory Society 2005 recommendations. Data analysis was performed with SPSS Version 18 for Windows. Correlations (Pearson correlation coefficient) between lung volumes and chest circumference were evaluated. Subsequently, multiple regression analysis was performed between lung volumes and the predictor variables.

Results: Six hundred and eighty-two (n = 682) boys with acceptable spirograms were included in the final analysis. Chest circumference was significantly correlated to height as well as spirometry parameters. Therefore, reference equations were established based on chest circumference and age. **Discussion and Conclusions:** Chest circumference could be considered as competent as height in predicting lung flows and volumes in Sri Lankan males aged 8 to 16, and it could be used as an alternative parameter in situations where height cannot be measured precisely.

KEY WORDS: Chest circumference; reference standards; spirometry; Sri Lanka.

Riassunto

Introduzione: La maggior parte dei criteri normativi per la spirometria sono stabiliti in base all'altezza, al peso ed all'indice di massa corporea. Noi abbiamo studiato la circonferenza toracica come alternativa all'altezza per l'interpretazione della spirometria quando accurate misure dell'altezza non possono essere ottenute.

Metodi: Questo è uno studio trasversale condotto in ragazzi dello Sri Lanka dell'età di 8-16 anni. I test di funzionalità respiratoria sono stati eseguiti secondo le raccomandazioni del 2005 dell'American Thoracic Society/European Respiratory Society. L'analisi dei dati è stata eseguita con il software SPSS Versione 18 per Windows. Sono state valutate le correlazioni tra i volumi polmonari e la circonferenza toracica con il coefficiente di correlazione di Pearson. Quindi, è stata eseguita l'analisi di regressione multipla tra i volume polmonari e le variabili predittori.

Risultati: 682 ragazzi con spirometrie accettabili sono stati inclusi nell'analisi finale. La circonferenza toracica è risultata essere correlata in modo significativo all'altezza così come ai parametri respiratori. Pertanto, sono state ottenute le equazioni di riferimento sulla base dell'età e della circonferenza toracica.

Discussione e Conclusioni: La circonferenza toracica potrebbe essere considerata idonea quanto l'altezza nel predire i flussi ed i volumi polmonari nei maschi dello Sri Lanka di età compresa tra 8 e 16 anni e potrebbe essere utilizzata come parametro alternativo in situazioni in cui l'altezza non può essere misurata con precisione.

TAKE-HOME MESSAGE

In this Sri Lanka-based study carried out on boys aged 8–16 years, chest circumference resulted as competent as height in predicting lung flows and volumes in males, and it could be used as a substitute for height for interpretation of spirometry parameters when accurate height measurements cannot be obtained.

Competing interests - none declared.

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Spirometry interpretation needs reference values for comparison. Most reference values are formulated based on age, weight and standing height from healthy non-smoking subjects, as the independent variables [1–4]. In patients with deformities or disabilities, whose height cannot be measured precisely, interpretation of spirometry becomes a challenge. Chest circumference, arm span or chest volume are independent measures of body size that may be considered instead of height in such situations. Golshan et al. have shown that arm span is a substitute for height when height cannot be measured accurately [5].

Chest circumference is an independent measure of body size and not affected by limb deformities, and we hypothesized that it might be a better alternative to formulate prediction equations in children whose height cannot be assessed correctly.

METHODS

The target population was 'healthy' boys. We only considered boys due to measurement difficulties anticipated with girls and measurement errors that could occur as a result of breast size around puberty. The sample was recruited from the latter part of a cross-sectional study which was conducted to formulate spirometry reference equations for Sri Lankan children aged 8-16 years [6]. For the main study, recruitment was carried out through stratified multistage cluster sampling from 5 districts in Sri Lanka. The enrolled children were given a questionnaire which was formulated based on International Study of Asthma and Allergies in Childhood (ISAAC) questionnaire with an intention to select 'healthy' non-smoking children [7]. A physical examination was carried out by an investigator and excluded if any of them had previous or concurrent cardio-respiratory diseases, spinal or limb deformities, frequent respiratory symptoms during the previous year, upper respiratory tract infections (URTI) like symptoms in the previous 2 weeks, and a history of chest, abdominal or nasal surgery. Errors encountered in measurement of chest circumference (CC) were additional exclusion criteria for this study. Written and verbal consent were gathered from the caregiver separately for measurement of chest circumference. Assent was taken from children for participation. Ethical clearance was obtained from Ethics committee, Faculty of Medical Sciences, University of Sri Jayewardenepura. The standing height was measured to the nearest centimeter and weight was measured with a spring balance. When measuring the chest circumference a non-stretchable tape was passed just above the lower angle of scapula behind, above the nipples in front and over the armpits at sides [8]. During the process the arm should rise a little when the tape is being passed round the chest returned to the resting position when the measurement is taken. All efforts were taken to obtain the measurements in expiration. The boys were asked to remove clothing above the waist. If a child did not allow taking an accurate measurement, still measurement was made over the clothes. Such measurements were subsequently rejected.

Spirometry measurements were recorded by two trained technicians with Vitelograph Alpha Touch spirometer. Volume calibration was performed daily by using a 3-litre standard syringe. The measurement protocol fulfilled the American Thoracic Society/European Respiratory Society 2005 (ATS/ERS) recommendations [9].

The child was seated and wore a nose clip. A minimum of 3 and a maximum of 8 manoeuvers were performed to obtain at least two acceptable trials. The largest Forced vital capacity (FVC) and Forced expiratory volume in one second (FEV1) values were required to be within 5% of each other. From the maneuver with the largest sum FVC+FEV1, peak expiratory flow (PEFR) and forced expiratory flow between 25% and 75% (FEF 25-75%) were taken.

Statistics

Data analysis was performed with SPSS Version 18 for Windows. Correlations (Pearson correlation coefficient) between lung volumes and chest circumference were evaluated. Subsequently, multiple regression analysis was performed between lung volumes and the predictor variables. Different regression models (transformed and untransformed) were attempted. Fitness of the models was assessed by DW (Durbin Watson) statistic and coefficient of multiple determinations for multiple regressions (R²). Comparisons among mean measured (observed) values versus predicted values from equations of this study based on CC and age were done with t-test. Further comparisons were performed between measured and predicted values (based on height and height derived from CC) obtained from equations of the main study.

RESULTS

Six hundred and eighty-two (n = 682) boys with acceptable spirograms were included in the final analysis. Nine hundred and nineteen (n = 919) boys were excluded from all the subjects who were invited (n = 1601) at the beginning due to various reasons. Presence of chronic cardiac and respiratory diseases (n =189), absence on the day of spirometry (n =198), recent acute illness (n = 58), spirograms not meeting ATS/ERS standards (n = 258) and incomplete questionnaires (n = 138) were some of them. Seventy-eight boys were excluded due to difficulties encountered when measuring chest circumference. Demographic and anthropometric data are summarized in Table 1.

Correlation between chest circumference and height was significant (r = 0.74, P < 0.001). Further, there was significant correlation between chest circumference and FVC (r = 0.77, P < 0.001), FEV1 (r = 0.74, P < 0.001), FEF 25-75% (r = 0.54, P < 0.001), PEFR (r = 0.62, P < 0.001). Subsequently, we performed multiple regression analysis between independent (chest circumference and age) and dependent variables (FVC, FEV1, FEF 25-75% and FVC) to evaluate the best fit for the model. Coefficient for determination (R²) ranged from 64.7 to 85.9 (Table 2).

Since the correlation between chest circumference and height was significant, we formulated an equation (see below) to predict height from chest circumference with a coefficient of determination of 87.9. Thus, predicted height could be used to interpret the spirometry parameters.

Height $_{pred.}$ = 60.29 + 0.57 × Chest Circum. + 4.9 × Age (R² - 87.9)

Mean measured values did not show a significant difference from predicted values from equations derived from this study (Figure 1a and 1b). Comparison between predicted values based on CC of this study and predicted values based on height from the main study did not differ significantly either. Close resemblance between measured versus predicted values based on height derived from CC is also shown in Figure 1a and 1b.

DISCUSSION AND CONCLUSIONS

This study has shown that chest circumference is useful in interpreting spirometry in special situations. It is a potential substitute for height in situations where measurement of the actual height cannot be performed. Chest circumference had been incorporated into lung function reference equations by Raju and co-workers as a predictor variable. They have shown that height, CC and fat free mass can be considered as good predictors of spirometry parameters that are included in their reference equations [10, 11]. Equations derived based on CC in our study have not included height or Body Mass Index (a derivative of height and weight) and could be used when height cannot be measured.

We compared spirometry predictions directly through CC with measured standards for comparability. In addition, values derived through height predicted from CC were compared with measured ones. There could be potential errors in deriving height from CC. Even minor differences in predicted height through chest circumference, may give errors in interpretation of spirometry especially when the readings are borderline. However, it is still could be better than predicting with an inaccurate height measurement.

Having a healthy sample of boys with strict

Parameter	Mean ± SD	Range
Mean age (years)	11.80 ±2.30	08-16
Mean BMI (kgm-2)	16.64 ±3.37	10-31
Mean height (cm)	147.85 ±14.83	115-186
Mean chest circumference (cm)	68.97 ± 9.49	051-104
Weight (kg)	37.30 ±12.86	17-98

Table 1. Baseline characteristics of the study subjects.

Table 2. Prediction equations for spirometry parameters.

Parameter	Constant (α)	Age coefficient (β^1)	CC coefficient (β ²)	R ²	DW statistic
FVC	-2.02	0.16	0.04	85.9	1.797
FEV1	-1.57	0.14	0.28	84.4	1.701
FEF 25-75%	-1.04	0.17	0.22	64.7	1.743
PEFR	-2.67	0.36	0.04	76.3	1.881

Note: Chest circumference in cm, Age in years, R²: coefficient of determination



Figure 1a. Comparison of observed/measured FVC values vs predicted values from eqautions based CC (P = 0.38), height derived from CC (P = 0.21) and height (P = 0.83).

inclusion criteria was a strength in our study. The main study recruited a sample which was socio-demographically representative of children between 8-16 years in Sri Lanka. However, only a section of that sample was taken into this study. Thus, this needs more verification with a larger sample size to represent all ethnic groups (Sinhalese, Tamils & Muslims) and geographic areas in the coun-



Figure 1b. Comparison between observed/measured FEV1 values vs predicted values from eqautions based CC (P = 0.93), height derived from CC (P = 0.11) and height (P = 0.24).

A P value of less than 0.01 was considered significant since there are multiple comparisons. Age is a common variable for all above prediction eqautions. Ht: Height, CC: Chest circumference.

try. Another limitation of this study was having only males in the sample, and obviously, results are not applicable in other countries and races. However, this study could pave the way to further research to confirm our results within other age groups and people of other races and countries using the chest circonference as substitute of height in order to perform spirometry in special conditions. In conclusion, chest circumference could be considered as competent as height in predicting lung flows and volumes in males, and it could be used as a substitute for height for interpretation of spirometry parameters whan a accurate height measurements cannot be obtained.

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ORIGINAL ARTICLE IN HEALTH AND SOCIAL BEHAVIOR

Identifying barriers that prevent US insured adults at-risk for and diagnosed with type 2 diabetes from accessing primary care services: An exploratory study

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Abstract

Introduction: Type 2 diabetes is a condition that affects millions of individuals within the United States and it is one of the leading causes of death. Access to quality care is necessary to decrease the risk of developing type 2 diabetes and associated complications. This study focused on assessing the barriers that prevented insured individuals, identified through a community health screening as at-risk for or suffering from type 2 diabetes, from accessing primary care services.

Methods: A quali-quantitative design was used in this study. Participants were recruited through convenience sampling and 64 participants were identified through community outreach events sponsored by the Florida Atlantic University's (FAU) Community Health Center. Barriers were measured through the 'Barriers to Care' Survey Instrument, which was administered face-to-face at community outreach events.

Results: Descriptive analyses indicated that participants identified cost of care as the most reported barrier (73.4% of sample) to accessing care. Lack of family and friend support was the least reported barrier (7.8% of sample). Regression analyses showed a significant association between reported barriers and participants' access to primary care services by measuring participants' last primary doctor's appointment ($R^2 = 0.47$, P = 0.001). Three factors significant resulted in predicting participants' last primary doctor's appointment: 'not having time to go to the doctor' (P = 0.022), 'feeling too sad about disease to see a doctor' (P = 0.021), and 'not feeling sick' (P = 0.001).

Discussion and Conclusions: Findings have important clinical implications since addressing identified barriers can lead to increased access to primary care services and enhanced health outcomes. Future research should focus on the significant impact of self-management techniques, sociocultural factors, and psychosocial factors to help inform researchers how they impact the barriers associated with individuals at-risk for or suffering from type 2 diabetes. Having health insurance is not the only answer to accessing primary care.

KEY WORDS: Barriers to care; behavior; community health screening; diabetes mellitus; healthcare; health care system; primary care services.

Riassunto

Introduzione: Il diabete mellito di tipo 2 è una patologia che interessa milioni di individui negli Stati Uniti d'America ed è una delle cause principali di morte. L'accesso alle cure di qualità è necessario per diminuire il rischio di sviluppare il diabete mellito tipo 2 e le complicanze associate ad esso. Questo studio si concentra sulla valutazione delle barriere che impediscono gli individui forniti di assicurazione sanitaria, identificati attraverso uno screening preventivo comunitario come soggetti affetti o a rischio di sviluppare il diabete mellito di tipo 2, di accedere ai servizi di cura primari.

Metodi: Un disegno misto quali-quantitativo è stato adottato in questo studio. I partecipanti sono stati arruolati attraverso un campionamento di convenienza e 64 partecipanti sono stati identificati attraverso programmi di formazione per la comunità sponsorizzati dal Community Health Center della Florida Atlantic University. Le barriere sono state misurate attraverso il questionario "Barriers to Care" somministrato di persona nel corso di programmi di formazione per la comunità.

Risultati: L'analisi descrittiva dei dati ha indicato che il costo della cura era la barriera principale per l'accesso alle cure riportata dai partecipanti (73.4% del campione). La mancanza di supporto da parte della famiglia e degli amici è risultata essere l'ultima barriera riferita (7.8% del campione). L'analisi di regressione ha mostrato un'associazione significativa tra le barriere riferite nel questionario e l'accesso da parte dei partecipanti ai servizi di cura primari valutati misurando l'ultimo incontro con il proprio medico di base ($R^2 = 0.47$, P = 0.001). Tre fattori sono risultati capaci di prevedere in modo significativo l'ultimo incontro con il medico di base: "non avere tempo per andare dal medico" (P = 0.022), "sentirsi troppo tristi per la malattia per vedere un medico" (P = 0.021), e "non sentirsi malati" (P = 0.001).

Discussione e Conclusioni: I risultati hanno importanti implicazioni cliniche dal momento che affrontare delle barriere identificate può portare ad un incremento nell'accesso ai servizi di cura primari e ad un miglioramento degli "outcomes" relativi allo stato di salute. In futuro la ricerca si dovrebbe concentrare sull'impatto significativo delle tecniche di auto-gestione, sui fattori so-cio-culturali e sui fattori psicosociali per aiutare ad informare i ricercatori su come questi fattori impattano sulle barriere per gli individui che sono affetti o a rischio per diabete mellito di tipo 2. Possedere un'assicurazione sanitaria non è la sola risposta per l'accesso alle cure di base.

TAKE-HOME MESSAGE

There is a greater need for the provision of better healthcare for individuals at-risk for or suffering from type 2 diabetes. Having health insurance is not the only answer to accessing primary care.

Competing interests - none declared.

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INTRODUCTION

Diabetes affects about 25.8 million people in the U.S. and is the seventh leading cause of death [1], and an estimated 79 million have pre-diabetes [2, 3], which is approximately three times higher than the prevalence of type 2 diabetes mellitus (DM) [2]. Adequate screening and appropriate treatment is essential to reduce the morbidity and mortality associated with type 2 diabetes [4]. Access to quality health care has been a persistent issue among those who are medically underserved [5]; however, there are established barriers that prohibited individuals from accessing care that could lead to effective self-management if addressed [6]. Racial and ethnic minorities were found to be disproportionately affected by existing barriers to health care access and utilization, resulting in highly prevalent disparities [4]. In addition to minority racial and ethnic status, it has been identified that low socioeconomic status qualified as a risk factor for lack of access to specialty medical providers, low quality of preventive care, and lack of access to in-hospital or ambulatory care [4].

A systematic review found that acceptability of care is one of the major barriers that prevented adults with chronic diseases like diabetes from accessing care [7]. Acceptability was defined as 'the relationship between patients' attitudes regarding the personal and practice characteristics of current providers' [7]. Other identified barriers to care included: accommodation of schedule, availability of the patient and provider, and affordability of care [7]. Access to medical providers was also predicted by psychosocial factors such as social support in conjunction with one's belief in their ability to care for their diabetic condition [4].

Furthermore, health insurance, food insecurity, and self-efficacy were associated with barriers to healthcare access [4]. Although lack of insurance was found as a hindrance to accessing care [4], it was also identified that individuals with health insurance complained about the cost of co-pays and premiums, which resulted in less than quality care or increased periods without insurance coverage [8]. Some insured patients mentioned that the cost of their insulin and the strips needed for self-monitoring were too expensive [8]. Additionally, inadequate insurance coverage has been found to inhibit patient access to diabetes care [8]. As a result, some patients were affected by the inability to see the medical provider as scheduled [8].

Lack of health insurance [4] or inadequate insurance [8] have been identified as barriers for individuals with diabetes. In comparison to insured adults, research has found that uninsured adults suffering from diabetes experienced less access to health care as well as decreased levels of preventive care [9, 10]. Enhancing health care access can be addressed effectively through health care providers and policymakers expanding adequate insurance coverage for this population [10].

Statement of Problem

Diabetes is a preventable disease that affects about 26 million people within the United States. Approximately three times that amount (79 million people) are at high risk for developing the disease. Those who have elevated glycemic levels are at high risk and are in need of outreach efforts and self-management strategies that are sustainable [11]. Although about 38% of people in the U.S. are considered pre-diabetic [2], only about 7% are aware that they have this risk [12]. If this condition is not treated in a timely manner, a large number of these individuals will develop type 2 DM as time progresses. With diabetes being the seventh leading cause of death in the U.S. [1], adequate care is necessary to decrease the risk of complications; however, the affordability of care has been identified as one of the major barriers for individuals accessing services [7].

Additionally, lifestyle modification such as proper nutrition and continuous physical activity is necessary for decreasing the risk of complications [14] and hospitalizations [15]. Outreach is used to outline a variety of activities such as the delivery of services or the distribution of information and educational materials. It has been shown that outreach is effective in maximizing the benefit of risk-reduction efforts in order to reduce the number of new diabetes cases that occur yearly [16]. Outreach also helps individuals to become aware of their medical condition so they understand the need to seek appropriate care [17]. Outreach methods are important for encouraging diabetic and pre-diabetic individuals to seek primary care services [17]. It also decreases the risk of avoidable hospitalizations and decreases the patient's length of stay [15]. Therefore, it is vital that all of the individuals who can potentially benefit from outreach are targeted and identified [18, 19], as this will increase the effectiveness of risk-reduction efforts and its impact on the incidence of new diabetes cases [19].

The goal of outreach at the established internship site, Florida Atlantic University's (FAU) Community Health Center, is to identify sick or at-risk patients, and to encourage patients to access medical care. Although outreach is conducted, the site has found that individuals who have been identified as at-risk for developing diabetes, or are suffering from uncontrolled diabetes, have not consistently accessed primary care services. As these at-risk individuals are insured, the barriers to care are not well understood. Through application of the outcomes from the reviewed literature, this project aimed to examine the barriers that prevent insured adults at-risk for and diagnosed with type 2 diabetes from accessing primary care services.

Research Question and Hypotheses

The following study was intended to address the research question and hypotheses below: What are the barriers that are preventing insured adults who are at risk for diabetes from accessing care once they have been informed that they are at risk?

H1: It is hypothesized that insured adults who are at-risk for or suffering from diabetes will report that cost of care is the most reported barrier to accessing primary care services, as measured by the 'Barriers to Care' Survey Instrument.

H2: It is hypothesized that insured adults who are at-risk for or suffering from diabetes will report that lack of friend/family support is the least reported barrier to accessing primary care services, as measured by the 'Barriers to Care' Survey Instrument.

This study will also explore the barriers' impact on participants' access to primary care services as measured by the participants' last doctor's appointment and their blood glucose level.

METHODS

Description of Setting

The setting for this culminating project was based through the FAU Community Health Center (formerly known as Diabetes Education and Research Center), which is located in West Palm Beach, FL, U.S. The Center is managed by the Christine E. Lynn College of Nursing at FAU and the Gregory School of Pharmacy at Palm Beach Atlantic University. The Center utilizes education and treatment by a team of experts, including endocrinologists, nurse practitioners, educators, researchers, nutritionists, and pharmacists, to challenge the growing diabetes epidemic. The Center also provides support for individuals of all ages who are identified as at risk for developing diabetes, currently have diabetes, or are at risk of experiencing complications from the disease. Therefore, the Center offers education programs to the community, including: individuals, families, community organizations and neighborhood groups. They provide comprehensive services to individuals with or without the ability to pay. Programs provided include: Carbohydrate Counting, Diabetes and Pregnancy, Diabetes Self-Management Education, Insulin Pump Training, Kid and Teens Lifestyle Program, Medical Nutrition Therapy, and Medication Therapy Management. The Center's essential goal also comprises multidisciplinary education and training of college students in various professional disciplines; a particular emphasis is placed on nursing professionals.

Description of Population

The targeted patient population for this study included insured adult patients who were either at-risk for or suffering from type 2 diabetes, which was identified through the site's glucose finger-prick screening process, participant self-report, and the American Diabetes Association's (ADA) Diabetes Risk Test, included in the outreach packet. The site considered a glucose level of 140 mg/dl and above to be abnormal (140mg/dl-299 mg/dl); a glucose level of 300 mg/dl and above was considered severe. Although some participants' levels may not be considered abnormal during screening, the ADA assessment conducted helped to determine if the participant was at increased risk for having type 2 diabetes through the following criteria: age, gender, diagnosis of gestational diabetes, family history of diabetes, diagnosis of hypertension, physical activity, and weight. Participants who scored a 5 or higher on the ADA assessment was considered at-risk; risk factors were added on the assessment to compute the score. Participants were excluded if they did not have an abnormal glucose level or were not considered at risk according to the ADA assessment. For the purposes of the study, no difference was made between patients who were either at risk for or diagnosed with type 2 diabetes.

Adults of all racial and ethnic backgrounds were included in the study. Since minorities are greatly affected by type 2 diabetes at a disproportionate rate and are at increased risks for the disease when compared to Non-Hispanic Whites, it was anticipated that there would be a larger number of minority populations, particularly African Americans and Hispanics. This is also representative of the population that is served at the Center: 50% African American/Black, 30% Hispanic/Latino, and 20% Caucasian/White. The Center also offers services on a sliding fee scale, accepting most insurance plans, which includes Medicare and Medicaid. Furthermore, insured adults include private coverage in addition to public medical programs. High deductibles have been a challenge for many of the Center's patients, which has made it difficult for them to receive care elsewhere.

Procedures

Individuals who agreed to be screened during outreach events sponsored by the FAU Community Health Center were approached to participate in the 'Barriers to Care' Survey. Once participants were contacted in-person at outreach events, individuals were offered a printed description of the planned study on the informed consent; informed consent was obtained verbally. It was projected that prospective participants would be contacted via telephone if they were screened at a past event; however, it was not necessary or practical once data collection commenced. Therefore, all participants were contacted in-person. Over a two month period (January to February 2016), potential participants were approached in order for the 'Barriers to Care' Survey Instrument to be administered. The questionnaire asked participants to identify information relating to their insurance status and the barriers that they faced while trying to access primary care services. Other information regarding the participants' demographics (age, gender, and race/ethnicity), glucose levels, ADA risk assessment, and their last primary doctor's visit was gathered from the site's outreach screening survey to incorporate into the findings. The researcher ensured that the surveys were understood, interpreted, and completed accurately. It was reinforced that the survey was voluntary and strictly confidential. Spanish and Creole interpreters were utilized, as necessary to accommodate the target population. Furthermore, patients were assigned unique participant ID numbers to maintain confidentiality.

Measures: The 'Barriers to Care' Survey Instrument

The research utilized a survey instrument that assessed insurance type as well as barriers to care of participants. This 3-question survey included a 13-item scale that listed potential barriers that participants may face; the items utilized a Likert-type scale ('Always Affected' to 'Not Affected at All') to evaluate the barriers listed. Data were analyzed through SPSS when frequencies were run. It totaled the amount of times a person checked that they are always, usually, sometimes or not affected by the particular barrier.

There was a version of the questionnaire for in-person completion of the survey and for telephone survey use, which was not utilized. Questions were developed based on the concept of the Perceived Barriers to Care Scale [20] as well as barriers that were predominantly identified within the literature.

Because the 'Barriers to Care' Survey Instrument is not an empirically validated instrument, the internal consistency of the instrument with this sample was calculated using Cronbach's Alpha ($\alpha = .56$). Information regarding the participants' demographics (i.e. age, gender, and race/ethnicity), glucose levels, risk assessment, and participants' last primary doctor visit was assessed according to the associated outreach screening packet used by the site.

Data Collection

Data collection commenced in-person after the culminating project proposal was approved by the researcher's formal committee. Arizona State University's Institutional Review Board determined this project to be exempt pursuant to Federal Regulations 45CFR46 [2]. During the months of January to February 2016, the researcher collected data from participants face-to-face at five outreach events at the following community partners of the FAU Community Health Center: Vickers House (South), Payne Chapel Missionary Church, Bath & Tennis Club, Gloria Y. Williams Multicultural Center, and St. Ann's Place. The identified site was informed of the study's outcomes.

Research Design

In this one-time cross-sectional, mixed-design study, a convenience sample was utilized to identify potential participants who were being screened at community outreach events conducted by FAU's Community Health Center. This design was also used to identify the most prevalent barriers reported to successfully accessing primary care services among the examined population. Since this research was exploratory, the researcher did not expect the results to be generalizable.

Data Analysis

Data were coded and analyzed by SPSS, version 23.0 for Windows. All responses were assigned a corresponding numerical value. Descriptive statistics (i.e. frequencies) were utilized to examine the characteristics of participants and the barriers identified that could impact the organization. Multiple linear regression analyses were utilized to evaluate the identified barriers' impact on participants' access to primary care services. P values of less than 0.05 was accepted as statistically significant.

RESULTS

Description of Sample

Seventy-six participants were recruited to take the survey; however, only 64 participants' surveys were eligible for inclusion once responses were assessed for insurance and risk factors. The majority of participants were women (n = 41, 64.1%) as compared to men (n = 23, 1)35.9%). As shown in Table 1, participants' ages included individuals from 18 to 65+, with the vast majority of participants within the range of 50-64 years old (40.6%) and the second largest age group ranging from 65 and older (35.9%). The racial and ethnic background of participants included the following: 68.8% African American/Black (n = 44), 15.6% Caucasians/White (n = 10), 14.1% Hispanic/ Latino (a) (*n* = 9), and 1.6% Other (*n* = 1). All participants had insurance, the majority had private health insurance, including managed care plans (45.3%) and Medicare (28.1%), as illustrated in Table 2. Individuals who indicated 'Other' specified insurance through Health Care District of Palm Beach County (15.6%) and Tricare (1.6%).

All included participants were considered

Age	N	%
18-29	2	3.2
30-39	8	12.6
40-49	5	7.8
50-64	26	40.6
65 and older	23	35.9
Total	64	100.0

Table 1. Age range in years (n = 64).

Table 2. Ways paid for medical care (n = 64).

	Ν	%
Private health insurance/managed care plan	29	45.3
Medicare	18	28.1
Other (specify)	11	17.2
Medicaid	5	7.8
Veteran's Administration (VA)	1	1.6
Total	64	100.0

Note: The 'Other' category includes held insurance through Tricare and the majority held through Health Care District of Palm Beach County.

Table 3. Range of glucose levels for all participants considered at-risk through the ADA Assessment.

	N	%
Less than 140 mg/dl	42	65.6
140-299 mg/dl	18	28.1
300 mg/dl and above	4	6.3
Total	64	100.0

at-risk for type 2 diabetes by glucose levels or the ADA Diabetes Risk Test included in the outreach packet. Glucose levels are reported in Table 3.

Descriptive Statistics

Descriptive statistics including frequency and percentage were computed for all outcome variables. Table 4 illustrates the 13 barriers and associated statistics representing participants' responses.

Table 5 illustrates the last primary doctor's appointment held by study participants.

Hypothesis 1: Most Reported Barrier to Care

It was hypothesized that insured adults who

were at-risk for or suffering from diabetes would report that cost of care ('My copayments are too high or my insurance does not cover enough costs') was the most reported barrier to accessing care. In order to test this, descriptive analyses (frequencies) were conducted. Results show that this hypothesis was supported: Always affected = 13 (20.3%); usually affected = 23 (35.9%); sometimes affected = 11 (17.2%); did not affect = 17(26.6%). Results indicated that the cost of care was the most strongly reported barrier by 47 participants, which was 73.4% of the sample. The second strongest reported barrier was 'The wait in the waiting room was too long,' reported by 29 participants, which was 45.4% of the sample.

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Items	4= Always Affected	3= Usually Affected	2=Sometimes Affected	1= Did Not Affect
I do not like/trust clinic staff	2 (3.1%)	2 (3.1%)	14 (21.9%)	46 (71.9%)
I do not like/trust my current doctor	4 (6.3%)	1 (1.6%)	14 (21.9%)	45 (70.3%)
The doctor's instructions are hard to understand	0 (0%)	1 (1.6%)	5 (7.8%)	58 (90.6%)
The wait in the waiting room is too long	6 (9.4%)	1 (1.6%)	22 (34.4%)	35 (54.7%)
Appointments are scheduled too far ahead	1 (1.6%)	0 (0%)	15 (23.4%)	48 (75%)
The cost of transportation and/or parking is too high	2 (3.1%)	2 (3.1%)	4 (6.3%)	56 (87.5%)
The clinic is too far away/no transportation	2 (3.1%)	2 (3.1%)	8 (12.5%)	52 (81.3%)
My insurance is too complicated to figure out	3 (4.7%)	1 (1.6%)	17 (26.6%)	43 (67.2%)
My copayments are too high or my insurance does not cover enough of the costs	13 (20.3%)	23 (35.9%)	11 (17.2%)	17 (26.6%)
I do not have time to go to the doctor	3 (4.7%)	1 (1.6%)	8 (12.5%)	52 (81.3%)
I do not feel sick	4 (6.3%)	2 (3.1%)	11 (17.2%)	47 (73.4%)
My friends and family do not support/help me	0 (0%)	0 (0%)	5 (7.8%)	59 (92.2%)
I feel too sad about my disease to see the doctor	1 (1.6%)	2 (3.1%)	13 (20.3%)	48 (75%)

Table 4. Reported barriers to care based on 13-item Scale on the 'Barriers to Care' Survey.

Note: N (%). Total n = 64. Barrier indicating copayments are too high or insurance does not cover enough costs is in boldface to illustrate the most reported barrier.

Table 5. Reported last primary doctor's appointment.

	Ν	%
Between 1-6 months	52	81.3
Between 6-12 months	4	6.3
Between 12-24 months	2	3.1
More than 24 months	3	4.7
Never/No response/Can't Remember	3	4.7

Note: Total n = 64

Table 6. Linear regression analysis of barriers impact on participants' last doctor's appointment.

	Beta	Р
I do not have time to go to the doctor	0.281	0.022
I do not feel sick	0.466	0.001
I feel too sad about my disease to see the doctor	0.305	0.021

Note: Independent variables (predictor set): 13-item barriers to care scale. Dependent variable (outcome): Last doctor's appointment. $R^2 = 0.47$; predictor set is significant (P = 0.001) when set at the <0.05 level; n = 64.

Table 7. Linear regression analysis of barriers impact on participants' glucose levels.

	Beta	Р
I do not have time to go to the doctor	0.116	0.410
I do not feel sick	-0.185	0.245
I feel too sad about my disease to see the doctor	-0.023	0.879

Note: Independent variables (predictor set): 13-item barriers to care scale. Dependent variable (outcome): Glucose Levels. $R^2 = 0.27$; predictor set not significant (P = 0.17) when set at the <0.05 level; n = 64.

Hypothesis 2: Least Reported Barrier to Care

It was hypothesized that insured adults who were at-risk for or suffering from diabetes would report that lack of friend/family support ('My friends and family do not support me') would be the least reported barrier to accessing care, as measured by the 'Barriers to Care' Survey Instrument. In order to test this, descriptive analyses (frequencies) were conducted. Results show that this hypothesis was supported: Always affected = 0 (0%); usually affected = 0 (0%); sometimes affected = 5 (7.8%); did not affect = 59 (92.2%). Results indicated that lack of family/friend support was the least reported barrier by 5 participants, which was 7.8% of the sample. The second least reported barrier was 'The doctor's instructions are hard to understand,' reported by 6 participants, which was 9.4% of the sample.

Secondary Analyses

Regression

Multiple linear regression analyses were conducted to evaluate the association of each individual barrier's impact on participants': 1) last primary doctor's appointment, measuring access to primary care services; and 2) blood glucose levels, measuring the self-management of glucose levels in people affected by diabetes. Information regarding participants' last primary doctor's visit and glucose levels was gathered from the site's outreach screening survey to incorporate into the findings.

As shown in Table 6, the only significant predictor set was indicated between the barriers' impact on participants' access to primary care services, by measuring participants' last doctor's appointment: $R^2 = 0.47$; the predictor set was significant (P = 0.001), when P was set at the 0.05 level. There were three factors that were significantly associated with predicting participants' access to care: 1) 'not having time to go to the doctor' (P = 0.022), 2) 'feeling too sad about disease to see a doctor' (P = 0.021), and 3) 'not feeling sick' (P = 0.001). Conversely, there was no significant impact of participants' barriers on reported glucose levels, as shown in Table 7.

Qualitative results

Participants also had the option to respond to one open-ended question to discuss any additional barriers to accessing primary care services or expound on the barriers already listed. Additional barriers included the following: procrastination, feeling too lazy, bad weather, and the primary doctor prescribing too much medication. Of the barriers already listed, participants explained that they: 'had a fear of seeing their doctor' (1 participant), 'did not enjoy the experience' (1 participant), 'felt the doctor was impersonal', 'inattentive', and 'spent little time on their health' (7 participants), 'only went when they felt extremely sick' (2 participants), 'had great mistrust of doctors and medications due to seeing an ill family member suffer' (1 participant), 'felt the healthcare system was an "economic joke" and a "rip off"' (2 participants), 'were too depressed to adhere to appointments' (6 participants), and 'had too many out of pocket expenses' (11 participants).

DISCUSSION AND CONCLUSIONS

It is well known that diabetes is one of the leading causes of death in the U.S.; therefore, adequate care is necessary to decrease the risk of developing type 2 diabetes and experiencing complications. Those who have elevated glycemic levels are at higher risk [11]. Identifying individuals who are at-risk for developing type 2 diabetes is essential for reducing their risk through targeted intervention strategies [19], including lifestyle and pharmacological interventions that can aid in the prevention or delay of complications [18]. However, the affordability of care, including the cost of copays and premiums, has commonly been identified as one of the major barriers for this population to access services [8]. Individuals with insurance also have not consistently accessed primary care services [8] and the barriers were not well understood. As a result, the study aimed to assess the barriers that were preventing insured adults

who were at risk for or suffering from diabetes from accessing care once they were aware of their risk-factor.

Significance of Findings

This study focused on identifying the most prevalent barriers reported to successfully accessing primary care services among insured individuals at-risk for or suffering from type 2 diabetes. Descriptive analyses showed that the majority of participants felt that their copayments were too high or insurance did not cover enough of their healthcare expenses. Even though participants experienced multiple barriers to care, the cost of care was the highest barrier reported. This finding is consistent with previous research. Other research has shown that individuals with insurance expressed significant concern about the cost of co-pays and premiums, which served as a hindrance to accessing care [8]. Inadequate insurance also accounts for barriers [8], which was consistent with this study's findings as some participants expressed that they could not afford diabetes strips and medications.

In addition, it is possible that participants with insurance still had issues with the cost of care due to other sociocultural factors such as household size and job type since this study did not measure that. Since sociocultural factors can impact the access that individuals have to primary care, it is important that future studies regarding barriers among those with type 2 diabetes include valid measures to assess the association among these factors.

Comorbid Behavioral Health Conditions Associated with Diabetes

About 25% of participants expressed that they were 'too sad about their disease to see a doctor.' This may suggest that participants could be experiencing various mood disorders such as depression that may have impacted their physical conditions as well as their ability to access services. The research has shown that depression or distress can have an adverse impact on diabetes and that there is a bidirectional impact of these comorbid conditions; major depression has been found to occur in approximately 12% of diabetic patients [21]. Comorbid conditions lead to poor self-care and medical complications. People with diabetes may experience high levels of stress [22] that include feeling overwhelmed by the demands of living with diabetes and not feeling like they have enough emotional support from family, friends, and healthcare professionals [23]. However, the study did not analyze mental health disorders such as depression through specific instruments such as clinical examination and psychometric tests to make a medical diagnosis.

An individual's perceived lack of social support was also identified as a barrier to effective self-management [8]. Conversely, the study showed that lack of social support was the least self-report type of barrier to primary care services. Peer support, provided by nonprofessionals who are suffering from diabetes and/or closely affected by the condition, can provide the type of ongoing care necessary for this population to sustain their self-management skills [24]. Some of the main factors needed for peer support to be effective include the provision of: daily management assistance, emotional and social support, and linking patients to medical and behavioral care services. Peer-led interventions also enhance self-management strategies; however, the programs that garner the most success are resource-intensive, such as the provision of peer educators to assist with ongoing care [24, 25]. Therefore, there is an increased need for integrated care services that target the physical, mental, and behavioral health conditions in individuals at-risk for or suffering from chronic diseases.

Additional Findings

The findings confirm the critical role health insurance plays in regular doctor's visits. Adequate screening, appropriate treatment [4], and effective self-management [6] are essential to reducing morbidity and mortality rates associated with type 2 diabetes. In order to achieve target health outcomes, barriers must be identified and addressed. Interventions that are developed to address these barriers have proven to be effective in decreasing one's risk for developing type 2 diabetes or reducing complications among type 2 diabetic individuals [11].

However, there is still a gap that needs to be addressed since individuals with insurance are still experiencing barriers due to high associated costs. Although the majority of participants in this study reported that they attended a doctor's appointment within the last six months, other respondents recounted that their last appointment was longer than 12 to 24 months ago. The multiple linear regression analyses showed that there was a significant impact of barriers on participants' access to primary care services. Since these three factors significantly impacted access to care ('not having time to go to the doctor,' 'feeling too sad about disease to see a doctor,' and 'not feeling sick'), there is a greater need to delve deeper into the biopsychosocial factors that influence individuals.

Other self-reported barriers included the length of time spent in the waiting room, not liking or trusting clinical staff, not liking or trusting the doctor, and insurance too complicated to figure out. Some participants who indicated cost as a factor held specific insurance with certain restrictions such as seeing doctors or staff at only one primary clinic. Individuals who held coverage through the county reported this situation. This type of restriction sets limits on access to care [8], and may also impact the quality of care since these patients only have the choice of seeking primary care at a particular clinic or obtain no primary care services at all. Since participants reported not liking or trusting clinical staff and the doctor as barriers, it is possible that there is a need to research why there is a disconnect between patients and providers regarding the provision of quality, efficient care. Poor communication with providers may be a factor [6].

Alternative Explanations of Results

Almost 35% of participants had a glucose level of either abnormal or severe. According to the ADA Diabetes Risk Test, other risk factors that participants experienced included some or all of the following: increased age, gender type, diagnosis of gestational diabetes, family history of diabetes, diagnosis of hypertension, lack of physical activity, and elevated weight. Although cost was a significant barrier to care, the lack of appropriate self-management is also a factor to consider regarding this population's access to care [17, 24]. Appropriate diet, exercise, and adherence is essential to decreasing risk levels and managing disease [26].

Study Strengths

Surveys were anonymous through the assignment of a patient ID; therefore, participants were possibly more inclined to answer the questions truthfully. There was also a mix of men and women included in the sample. The survey format was strong since it assessed multiple issues (i.e. barriers faced, insurance type, and glucose levels). Furthermore, additional factors, such as participant demographics and last primary doctor's appointment were collected in order to analyze their impact on access to primary care.

Study Limitations

Generalizability

Although this study identified quality information and trends, there was no random selection of participants. Instead, the study utilized a convenience sample from the individuals who were screened by the outreach team through FAU's Community Health Center. Therefore, the results of the study cannot be generalized to all individuals at-risk for or suffering from type 2 diabetes since the results of the study are specific to insured participants in the West Palm Beach area. Additionally, a smaller sample size may have impacted the explanatory power of conclusions.

Data collection method

Surveys were conducted face-to-face at outreach sites where screenings were conducted. Recruited participants were asked about their barriers to care. Although participants were assured of the confidentiality of their responses, they may or may not have felt compelled to be completely honest about some of the barriers they face while trying to access primary care services. A small number of participants may have experienced a language barrier since they only spoke Spanish. Although interpreters were available to explain the survey, its purpose, and the questions assessing barriers experienced, some information may have been lost in translation. To minimize this issue, prior to data collection, interpreters were briefed on survey questions as well as the information seeking to be collected.

Bias

The responses in this study were based on self-reporting, which was inevitable since the project was centered on the clients' voluntary participation to give their perspective on the barriers that they faced while trying to access care. Response bias may have played a role since some participants may have felt uncomfortable discussing the type of barriers they experienced. However, this was accounted for by asking questions in a non-leading manner and assuring confidentiality during collection and anonymity afterward since all surveys were assigned a patient ID.

Clinical Implications of the Findings and Conclusions

Findings suggest a greater need for the provision of better healthcare for individuals in need. Having health insurance is not the only answer to accessing care. The cost of diabetes on the healthcare system is exceptionally high and the costs for diabetic patients compared to those without this illness is twice as high [1].

From recent reports, the approximate cost of diabetes to the U.S. healthcare system is \$245 billion [2, 13]; with indirect and direct costs totaling \$176 and \$69 billion, respectively [2]. Individuals who are diagnosed with this illness incur an average amount of \$13,700 in medical expenses yearly. Additionally, the biggest factors contributing to the amount of direct medical costs were prescription medi-

cations and hospital inpatient care. The greatest contributing factors to indirect costs were decreased work productivity and increased absenteeism [2].

A closer look at the type of care and the quality of care provided is imperative for individuals at-risk for chronic diseases to focus on prevention. This will aid in the reduction of the financial burden on the healthcare system as the number of individuals utilizing the emergency department and being hospitalized will decrease with this increased focus on prevention.

Suggestions for Further Research

In addition to the aforementioned findings, self-management strategies were important to the barriers that participants faced. Implementing a high-impact national educational awareness campaign that focuses on prevention messages geared toward individuals with pre-diabetes is included in the American Diabetes Association's strategic plan for improving self-care strategies [27]. Evidence-based interventions that support lifestyle change and encourage patient engagement have proven most effective [16]. Programs are also most efficient when both the patient and provider have discussed these strategies and have shared in the decision-making process through collaborative care [11].

Provider recommendations were associated with patient increase in engaging in healthier behaviors to sustain lifestyle modifications [28]. Moreover, interventions that specifically focused on the development and enhancement of these self-care skills, such as the promotion of positive attitudes through proper nutrition and exercise [26], helped reduce the risk of diabetic and pre-diabetic individuals from experiencing complications. Furthermore, essential skills including improvement of knowledge regarding diabetes and its effects, specific goal-setting, healthy cooking recipes, focus on problem-solving, grocery shopping skills, and adequate self-monitoring [26] are specific examples of strategies that helped improve self-care.

Future research should focus on the significant

impact of self-management techniques with the types of barriers encountered in this study, such as not having time to go to the doctor and not feeling sick. Finding a way to address self-management could help empower individuals to focus on prevention methodologies. Furthermore, addressing sociocultural factors such as socioeconomic status, household size, and job type will help inform researchers how these also impact the barriers associated with individuals at-risk for or suffering from type 2 diabetes and other chronic illnesses. Since not liking or trusting the doctor and clinical staff were reported barriers to care, research that delves deeper into the type of care and the quality of care provided is necessary. Possible interventions that help increase the competency or interpersonal skills of healthcare professionals may help decrease patient skepticism and mistrust of the healthcare staff and system [29], resulting in an increase in

the likelihood of this population accessing primary care services. The study aimed to assess the barriers that were preventing insured adults who were at risk for or suffering from diabetes from accessing care once they were aware of their risk-factor. Finally, the findings found will help the intern's site, FAU Community Health Center, to address identified barriers to support the increase in access to care for current and new patients.

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ORIGINAL ARTICLE N DIABETOLOGY AND SOCIOLOGY OF HEALTH

Teenagers with type 2 diabetes mellitus as a hidden population: A qualitative study of the representations and experiences related to the disease in Mazatlán, Mexico

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Abstract

Objective: To analyze the representations that adolescents with type 2 diabetes mellitus living in Mazatlán, Mexico have of the disease.

Methods: The study design was qualitative and based on an analytical approach of narrative research. Indepth interviews and participant observation were conducted with eight adolescents (four females and four males). The sample was theoretical-intentional according to age (10-19 years), diagnosis of type 2 diabetes mellitus, and residence in Mazatlán.

Results and Discussion: Adolescent experiences and representations regarding the disease were associated with six themes: 1) knowledge of the disease; 2) self-care practices; 3) identity construction; 4) significance of the disease; 5) family support; and 6) gender as a transversal category. Social stigma that is associated with diabetes and its management leading to self-management barriers, the abandonment of treatment and hiding the disease make adolescents with diabetes to be a hidden population. Identity construction is strongly affected by the disease; indeed, diabetes implied a reconstruction of male identity, while for females the disease was associated with overweight that affects their self-esteem. All of the adolescents lived in a constant dichotomy between present and future, with the fear of dying or getting worse. The absence of intergenerational communication and family support within the adolescents' families limited adolescent care practices, exacerbating their emotional distress and living the disease with the feeling of loneliness.

Conclusions: Our study showed that adolescents affected by type 2 diabetes mellitus, did not experience nor they did face the disease homogeneously but accordingly to the gender and level of family support, which, therefore, should be specifically considered by care programs and health professionals to encourage adherence to treatment, taking into account the emotional well-being and psychological needs of adolescents.

KEY WORDS: Adolescent; Diabetes Mellitus, Type 2; Health Beliefs; Self-care; Self-Efficacy.

Riassunto

Obiettivo: Analizzare le rappresentazioni che gli adolescenti di Mazatlán, in Messico, affetti da diabete mellito tipo 2 hanno della loro patologia.

Metodi: Lo studio di tipo qualitativo si è basato sull'approccio analitico della ricerca narrativa. Interviste approfondite e l'osservazione dei partecipanti sono state condotte con otto adolescenti (4 di sesso maschile e 4 di sesso femminile). Il campionamento è stato intenzionale teorico secondo l'età (10-19 anni), la diagnosi di diabete mellito di tipo 2 e la residenza nella città di Mazatlán.

Risultati e Discussione: Le esperienze e le rappresentazioni degli adolescenti riguardanti la patologia sono state associate a 6 temi: 1) la conoscenza della patologia; 2) le pratiche di auto-cura; 3) la costruzione dell'identità; 4) il significato della patologia; 5) il supporto familiare e 6) il genere come categoria trasversale. Lo stigma sociale associato al diabete ed alla sua gestione che comporta barriere nell'autogestione, l'abbandono del trattamento ed il nascondimento della malattia rendono gli adolescenti affetti da diabete mellito una popolazione sconosciuta. La costruzione dell'identità è fortemente influenzata dalla patologia; infatti, il diabete mellito ha implicato una ricostruzione dell'identità nei maschi, mentre per le femmine la patologia è risultata essere associata al sovrappeso con ripercussioni sui livelli di autostima. Tutti gli adolescenti vivevano in una costante dicotomia tra presente e futuro, con la paura di morire o di peggiorare la propria condizione di salute. L'assenza di comunicazione intergenerazionale e di supporto familiare hanno limitato le pratiche di cura tra gli adolescenti, esacerbando il loro distress emotivo ed il vissuto della malattia provato con un sentimento di solitudine.

Conclusioni: Il nostro studio ha evidenziato che gli adolescenti affetti da diabete mellito tipo 2 non vivono nè affrontano la malattia in modo omogeneo ma differentemente in base al sesso ed al livello di supporto familiare che, pertanto, sono componenti che dovrebbero essere specificamente considerate nei programmi terapeutici e dai professionisti della salute per incoraggiare la compliance al trattamento farmacologico, tenendo in considerazione il benessere emotivo e le necessità psicologiche degli adolescenti.

TAKE-HOME MESSAGE

Adolescents with type 2 diabetes mellitus in Mazatlán, Mexico constitute a hidden population; new identification strategies of this population should be promoted differentiated by sex and family support; not only for the care of the disease, but to reduce the emotional distress that adolescent with diabetes experiences.

Competing interests - none declared.

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INTRODUCTION

Type 2 diabetes mellitus (T2DM) is a chronic disease with strong emotional implications for the adolescents affected [1]. From a medical perspective, a healthy lifestyle can improve the life quality of adolescents, which includes a balanced diet, regular exercise, and strict adherence to pharmacological treatment. However, these aspects can generate chronic distress because adolescents may feel constrained to alter their daily practices. Furthermore, the social representations of diabetes, which is considered as a disease affecting only adults, generate conflicts in the identity construction of adolescents and their projections to the future [2].

Indeed, there is evidence showing that the experience of living with diabetes affects the mental health of adolescents with type 1 diabetes, who should have similar daily and long-term repercussions as those with type 2 diabetes [3–5]. Even if the qualitative evidence of T2DM in adolescents is only recently emerging [6, 7], several studies have yet shown that this disease is associated with eating disorders and dissatisfaction with the body image, conditions that may negatively impact on adolescents' self-esteem and psychological well-being [8–10].

Similarly, studies have demonstrated that the stigma associated with diabetes makes it common for young patients to hide T2DM due to the fear of being excluded [11]. School is the main space for socialization among adolescents; however, in this context, the disease tends to be hidden more frequently, particularly by young males. This phenomenon has implications for adolescents' health, particularly when it leads to the treatment failure [12].

Although there are common elements in the perceptions and stigma affecting adolescents with T2DM, research found important differences, which are based on socioeconomic status. Indeed, within the most favored social classes where self-care practices are better, there are more knowledge about the disease and resources to treat it, the level of adolescent satisfaction increases, and uncertainties about the future development of the disease decreases, compared to young people of lower-income backgrounds [13].

The support of family and friends, both emotional and logistical, is also associated with more positive meanings of the disease and better care practices, such as having a healthy diet and physical activity, being adhering to drug treatment and dealing with distress and other self-destructive coping behaviors [7, 14]. In addition, the interaction among adolescents with diabetes allows them to share their experiences, increase self-care practices and construct more positive perceptions about the disease [15].

Evidence in Mexico

Studies in Mexico have mainly focused on risk factors that are related to the metabolic disorders experienced by adolescents [16, 17]. Some qualitative studies have approached young people's perceptions regarding diet and eating habits [18, 19]; however, the literature on the perceptions of adolescents affected by T2DM is very scarce in our country.

Conversely, some research has been conducted on how adults experience both the T2DM and the subsequent lifestyle associated with the disease, showing that they are at high-risk of decreased psychological well-being because of the strained coping with changed life routine, such as relationships, work-related and financial issues [20]. Even though the evidence indicates that the incurability of the disease has repercussions on the representations of patients [21], family support could represent a decisive factor in the well-being of patients and in the development of positive meanings with regard to the disease [22].

Aim of the study

Aim of our study was to survey how adolescents with T2DM represent and perceive their disease. Specifically, our objective was to analyze the representations that adolescents with T2DM, aged 10-19, have of T2DM during a period of strong physical, psychological and emotional changes, such as adolescence, in certain neighborhoods of Mazatlán, Sina-

loa, in northwest of Mexico.

The present study might be relevant in the Mexican context providing analytical elements from a qualitative perspective for the study of T2DM among adolescents, who could be a hidden population, due to stigma associated with the disease that probably is the most important cause of treatment dropout and hiding the disease.

METHODS

This research was qualitative, interpretive-descriptive, and based on the theoretical approach of social constructivist theory from which, social practices and behaviors are considered to be culturally constructed. From this perspective, the meanings that individuals give to their action, with which they interpret reality and influence it, acquire centrality; therefore, the interpretation of individuals about their own experiences, in this case diabetes, is fundamental to understand the representations and experiences around the disease [23, 24].

The analytical approach of narrative research was used, which allows to better analyze how individuals perceive, understand and make meaning of life situations or experiences in particular contexts [25, 26]. The objective is not the generalization or search for causality but the description and interpretation of broader social processes and the generation of new research questions and topics [27].

Also, we used the theoretical perspective of social representations, because it constitutes a bridge between the psychological and social dimension considering the way in which the persons apprehend the situations of the daily life, the characteristics of the contexts where the individuals are immersed and the messages and information that circulate in those scenarios. Therefore, social representations involve a mental activity in which persons fix their position in situations, events and experiences of daily life, anchored in concrete social and cultural structures. In this sense, social representations are forms of socially constructed and shared knowledge [28, 29].

We understand social representations as the set of knowledge, affected by socioeconomic

and historical determinants, which individuals have about social reality, which organizes practices and influences how they perceive themselves and others [28].

Ethical consideration

This research was approved by the Ethics Committee of the National Institute of Pediatrics in Mexico. It was carried out under the consent of the adolescents and the approval of their parents. Likewise, the confidentiality of the participants and their anonymity were ensured.

Procedure

Study Instruments

In-depth interviews were conducted in the *li-fe-story* mode, which involved re-interviewing each participant with their informed consent and that of their parents. Participant observation was also conducted with the purpose of understanding the family and social contexts in which the participants were socialized, their relations and T2DM representations. The number of interview sessions that were performed with each participant was sufficient to achieve the purpose of this research [30].

Interviews were transcribed to be systematized, coded and analyzed using *Atlas Ti* software. Data analysis was elaborated by the open coding. The starting point in our interviews was the narrative diagnostic event of diabetes, which is considered to be a structure that gives meaning and organizes other events and adolescent experiences in different temporalities [31]. The interviews were organized on six topics as follows:

1) diagnosis of T2DM; 2) family history of diabetes; 3) family, school, community and medical environments of the adolescents; 4) treatment received and adherence to such treatment; 5) disease management and self-care; and 6) significant relationships and future plans (see Figure 1).

Prior to the interviews, medical consultation sessions were held with the adolescents to check their health status, to confirm the dia-



Figure 1. Narrative structure of interviews and topics considered.

gnosis and treat the disease.

Analytic categories

Following the open coding process, six analytic categories of analysis were identified to approach to the T2DM representations: 1) Knowledge about the disease; 2) self-care practices; 3) identity construction; 4) disease significance; 5) family support, and 6) gender, as a transversal category.

Participant selection criteria

The sample was theoretical-intentional according to the following selection criteria: a) age (10–19 years); b) T2DM diagnosis with more than six months of evolution; c) no any other chronic comorbid disease; and 4) residence in Mazatlán. Placing the research in Sinaloa was pertinent because it represents the state with the second highest incidence of T2DM among adolescents in Mexico.

This research is part of a larger research project; however, the fieldwork period from which the interviews were obtained for writing this paper began in April 2015 and ended in April 2016. The participant selection was conducted in two stages in which different strategies were implemented as the fieldwork was developed. In the first stage, participant recruitment was initiated in 18 health centers, the General Hospital and the Unit of Metabolic Diseases of Mazatlán with the authorization of the Sanitary Jurisdiction. Among all of the patients, only six of them could be followed-up with and interviewed because of problems tracking patient due to treatment withdrawal. This situation involved a second selection procedure in secondary schools where the population was captive. In the second stage, we considered four schools, based on a census and stratified sampling among all public secondary schools in Mazatlán. A total of 650 questionnaires were used to identify adolescents with T2DM. It was difficult to identify cases based on this strategy because adolescents can be unaware of their condition and those who know it do not want to be identified as having the disease. Despite the difficulties with both strategies, 8 adolescents were interviewed. Although the sample obtained is apparently small, it is important to consider some aspects.



Result of broth implemented strategies : 8 in-dept interviews conducted with adolescents (4 females and 4 males).

Figure 2. Participant's selection process according to the strategies implemented.

First, the participants were in social exclusion conditions which make difficult access to adolescents. Therefore, other strategies were also used to find cases that were not registered in the health system and which showed clinical suspicion of the disease, through the intentional search in the Mazatlán public secondary school environment. In addition, during field work, not all adolescents with this disease could be considered in the study; indeed, due to their social adversity, they did not want to participate and maintained a rejection towards health authorities.

Furthermore, the World Health Organization has reported that type 2 diabetes mellitus has increased its frequency in the pediatric age in low-income countries, but the under-reporting of the disease in this type of population is considerable [32]. According to the national prevalence of type 2 diabetes mellitus in adolescents in Mexico, reported by the National Health and Nutrition Survey 2012 [33], the estimated population size of minors affected by T2DM living in Mazatlán was only thirteen out of 24,000 adolescents, who live in that municipality [34, 35]. Other qualitative researches on T2DM conducted in Canada and the United States with adolescents have obtained similar samples to our study because of barriers identified [7, 11, 14].

RESULTS

Participant profiles

Sociodemographic, family history and clinical data of participants are showed in Tables 1 and 2. With regard to family history of diabetes, Table 1 shows that except for one adolescent, all of the participants had a close family member affected by diabetes, most commonly being their mother or maternal-line relatives; this, may be due to the absence of information from the paternal line in several cases in which the father was absent; and there was no a tendency for siblings to develop the disease. There was also heterogeneity in the age at diagnosis, which may explain the different experiences concerning the disease.

Most of the participants used daily insulin, and they manifested symptoms related to

T2DM. In particular, females showed a history of obesity or overweight that, in 2 cases, had been maintained from childhood to adolescence. Only 4 of participants were being treated at a health institution.

Table 2 shows an approach to the participants' dynamics and family structure. Their family socialization was characterized by the physical absence (economic and emotional) of one parent, profile that coincides with that of adolescents with diabetes type 2 in other cultural contexts [7]. Family breakdowns represented events that triggered feelings of abandonment and stress in the participants, which overlap with the diabetes diagnosis in some of them and generate additional emotional distress.

Diabetes representation

Our findings showed that representations of diabetes among the adolescents were mainly associated with six themes, of which, gender is a transversal topic, that integrate a body of knowledge that is sometimes contradictory, which allows them to face the disease, interpret their illness and build an identity.

Knowledge about the disease

The narratives of the participants showed that knowledge about the disease, from which the adolescents organize their practices and build an identity, is acquired in three ways: health professionals, family history and social context.

Diagnosis is the moment at which the adolescents begin to be directly exposed to medical

 Table 1. Sociodemographic and clinical profile of participants.

SOCIODEMOGRAPHIC AND MEDICAL PROFILES OF PARTICIPANTS									
Participant*	Current	Schooling	Current	Age at	Treatment	Insulin	Overweight	Current body	Family history
	Age	achieved	occupation	diagnosis	received**	use	in childhood	mass index	of diabetes
Women									
Luisa	18	High School	College student	: 14		Yes	Yes	28.2	Mother/grandmother
Karla	15	Elementary School	Junior high school student	: 11	Yes	Yes	Yes	42.5	Mother/grandmother/uncles
Diana	16	Junior high school	Technical student	: 15			Yes	45.1	Uncle
Daniela	14	Elementary School	Junior high school student	: 14	Yes	Yes		20.3	Mother/grandmother/sister
			Men						
Camilo	15	Elementary School	Employee	13		Yes		20.3	Grandmother
Alfonso	18	High School	College student	: 11	Yes	Yes		24.3	
Oscar	11	Elementary School	Junior high school student	. 7				20.4	Mother/grandmother
Ernesto	19	High School	Employee	13	No	Sometimes	Yes	28.9	Mother/father
*The names of the participants were changed by pseudonymous to protect the participant's anonymity.									
**Medical treatment institution received before beginning the fieldwork									

Source: Own elaboration with in-depth interviews data

Table 2. Dynamics and family structures of participants.

DYNAMICS AND FAMILY STRUCTURES OF PARTICIPANTS

Participant	Current	Family structure	Parental	Parental breakdowns	Siblings	Siblings	
	Family structure	in childhood	absence*	after diagnosis**		with diabetes	
		Women					
Luisa	Live Independent	Nuclear family			2		
Karla	Singleparent family (M/S)	Nuclear family	Father	Yes	1		
Diana	Singleparent family (M/S)	Singleparent family (M/S)	Father	Yes	1		
Daniela	Singleparent family (F/S)	Extended family	Mother		4	Yes	
		Men					
Camilo	Singleparent family (M/S)	Nuclear family	Father	Yes	3		
Alfonso	Nuclear family	Singleparent family (M)		Yes	2		
Oscar	Singleparent family (M/S)	Extended family	Both parents	Yes	. 1		
Ernesto	Independent nuclear family	Singleparent family (M/S)			3	Gestational	
*Physical absonce or near contact with one of the parente during prolonged pariods							

*Physical absence or poor contact with one of the parents during prolonged periods

**Divorce and separations. In some cases the separations are caused by events such as migration or stays in jail by one of the parents.

M=Mother and S=Siblings

Source: Own elaboration with information taken from interviews

knowledge. However, the received orientation is mostly related to the treatment, which is insufficient, and often it is not in accordance with the cognitive and emotional maturity of the patients to allow them to understand the diagnosis. In addition, the adolescents perceive that the information provided by health professionals focuses on the negative consequences associated with diabetes in the long term. This explanation is not accompanied by a description of their condition and how to modify habits and lifestyles, particularly dietary behaviors. Oscar suggests:

"He [doctor] told me that I had to take care of myself because I had an illness; I do not know what [diabetes] is, and he [doctor] told me that I must be constantly going to [Medical Center], not getting fat, not do this, not do the other, but I did not know why [...]".

The female participants mentioned the strong control exercised by health professionals regarding the physical aspect, specifically, overweight and obesity. For them, the medical discourse focused on the need to lose weight, which caused them emotional discomfort for not being able to understand how to achieve it. Among the women, losing weight becomes a principal cause for concern rather than the disease itself. Some of them were referred to nutritionists for weight loss; however, regaining weight caused anxiety, frustration, and ambivalence towards T2DM's treatment, as Karla states:

"It bothers me to be told: you have to lose weight [crying] because I have to. I don't like being told that because I already know it. I just want to be told how. I don't like being told that I can weigh up to 500 kilos because I don't imagine myself like that [...]. I feel as if they [health professionals] told me to lose weight fast, because you can die and that makes me feel bad".

Another way to acquire knowledge about the disease is to live with a family member who suffers from diabetes. This fact promotes the intergenerational transmission of knowledge about T2DM, without necessarily corresponding to that which is obtained from health professionals. However, both types of knowledge are complementary ways of un-

derstanding the disease among adolescents. An example of this is Luisa's story, in which medical and popular knowledge complement each other when she refers to her mother's diabetes trigger:

"[...] I have known that she had diabetes because of my sister, for scares [worried]. She said that because she always scares [worried], but she did not have a good diet either".

The third way to obtain knowledge concerning T2DM refers to community background (neighbors, friends, and teachers) about illness, which builds a collective image of diabetes that is often incomplete and inaccurate and affects the adolescents' perceptions about T2DM, as in the case of Camilo:

"Well, that can give you a coma; because I've heard a lot of people saying that diabetes gave them 'coma', but I do not know if those people injected [insulin] or not."

'Take care': self-care practices

'Take care' is a central practice in adolescents' representations of diabetes, appearing frequently in their narratives and promoting ambiguities in how they experience disease. This practice refers to the behaviors that help prevent the consequences of the T2DM in the long term but that are difficult to perform in their day-to-day experiences, which can generate guilt and emotional distress.

'Take care'has individual and collective aspects that are mutually affected. Individual aspects involve three elements: dietary restrictions, injecting insulin when necessary, and performing physical activity, particularly among women who are obese and overweight. The collective aspects cover the school, work, family and community spheres, which have the ability to enhance aspects of the individual level or restrict them. Some circumstances in these spheres can promote the abandonment of treatment, particularly; they interfere with healthy eating, as Ernesto suggests:

"I want to take care of myself, but it's very difficult. In my family, even though they are diabetics, they have never cared of theirself. I arrive at my house and eat the food they make, [...] unhealthy. And at work, I'm the dishwasher, there's food that you want, and I'm hungry, so I eat what I have. There is no way to take care of me!"

Identity construction

Identity implies a dual process that involves an individual dimension, namely, how the individual recognizes himself; the other dimension is social, referring to the image that the others, with whom the individual interacts, have of the individual [36]. Therefore, diabetes generates a process of change and adequacies in how adolescents construct an identity, a process that can generate ambivalences and contradictions. The analysis of the interviews suggests that diabetes representation is related to the participants' perception of adolescence as a period of life; hence, their experience with T2DM is ambivalent because it occurs at a moment that is socially associated with health. Diabetes in adolescence represents a disease that is 'out of time', as Luisa mentions: "If I start to think how everything was, it gives me nostalgia; I feel sad. I begin to cry because I feel that I am too young to have diabetes, to inject insulin and all that".

The participants constructed their identity based on strong uncertainties about the future, which varied between men and women. The young people interviewed of both sexes expressed constant apprehension about triggering other long-term ailments and fear of dying, as Oscar suggested:

"Well, I am afraid that my feet will be cut off [amputated] because I've heard that diabetes gives something bad that I do not know on feet. They get dark, and then you have to cut them off. I'm afraid to die".

By contrast, the women's concerns were mostly at the family level, as Karina stated:

"I am afraid to leave my family alone; I don't want them [relatives] to fight or suffer for

me. I do not want them to cry because I can die." The inconsistency between being sick and being young and uncertainty about the future led the adolescents to construct their identity using different strategies, among which hiding the disease was the most used. The adolescents did not want some members of their social network to know about their illness; however, females mostly hid the disease from family, while males mostly hid it from peers. Males with T2DM felt inferior and different from their classmates, but this feeling changed with age. The youngest participants most easily expressed their condition to their peer group. However, as age increases and the high school cycle advances, social pressure on males becomes stronger, particularly in terms of the values that are associated with hegemonic masculinity, such as strength. Then, they start to hide the disease to avoide injecting insulin in public, and they break their diet because they feel that diabetes puts them in a disadvantaged position with regard to meeting the expectations of being a man.

Females participants hide the illness from some members of their family for fear of worrying them, and in some cases, it happens that their parents ask them to hide their condition from their closest relatives, thereby limiting their socialization within the family nucleus, as Luisa declared:

"No one in my family knows because I've never told anyone. When that happened to me [diabetes], my mother told me: do not tell anyone. So, if it is time to go out with family, for example, they invite me for dinner, I would like to accept it, but what are you going to drink? I cannot take any soda or fresh water. Therefore, sometimes I would prefer not to go".

In addition, hiding the illness generates a feeling of loneliness among the participants that affects their self-esteem and complicates the follow-up of treatment and care practices. Ernesto notes:

"I have felt alone, some days you feel up and some down. One carries many emotions, and you start to cry".

Disease significance

The meaning that is given to diabetes is transformed throughout the adolescents' life pathways. At the time of diagnosis, the significance of the disease is ambiguous in that the adolescent does not have sufficient information about it. However, as time passes, treatment begins, and they must manage the disease daily; consequently, the significance of

T2DM emerges explicitly.

Different significances of diabetes are identified in the participants' stories. Women, in particular, highlight the positive aspects of T2DM, suggesting that despite the problems, diabetes has strengthened them to face different obstacles in life, as stated by Karina:

"Well, I feel stronger because I know I can live with the disease. I feel I have more power than before, even though I have more things that I cannot eat [laughs], I feel stronger".

However, the disease is generally associated with negative issues. Diabetes represents a limitation for socialization, specifically with regards to family. In Mexico, celebrations are commonly centered around the food; therefore, restrictions on eating do not allow the adolescents to fully participate, particularly for those who hide their condition. Simultaneously, the symptoms of the illness and injecting insulin limit interpersonal relationships and everyday practices. Transporting insulin and protecting it from hot temperatures in Mazatlán limits the adolescent's mobility and alters daily activities. Diana provides an example:

"I used to go to sleep with my grandmother, I enjoyed that, and now I do not go for medicines and all that."

Similarly, the adolescents perceive that suffering from diabetes stigmatizes and marginalizes them, which is differentiated by sex. Women participants describe the feeling to be excluded because of obesity and the marks that diabetes leaves on their bodies, such as skin spots. At the same time, they feel that the disease can limit their autonomy, generating anxiety when they think that they can be a burden on their families, as Daniela says:

"I would not want them to think I'm a burden. I want them to continue their life, and I want to continue with mine. I do not like them to feel sorry for me".

For men, injecting insulin exposes them to discrimination and marginalization; where they live, in their neighborhoods, injections are associated with drug abuse, and they do not want to be stereotyped in that way, as Camilo suggests:

"[...] my life was not going to be the same, I had to inject myself then! People say something wrong about it, but it is only insulin."

Likewise, they associate diabetes with limitations to their masculinity in two ways. First, T2DM constrains their participation in the public sphere, particularly in the labor market, which affects activities that have traditionally been adjudicated to males: working and providing for a family. Second, they perceive that diabetes affects their virility, as Ernesto puts it:

"I met my wife, and that's when the people started to say that I was not going to satisfy her [sexually] and that's when I felt bad about myself again".

Family support

Although it has been documented that family support is a central element for adolescents to face the diabetes and have greater adherence to treatment [15], the family dynamics of the participants was characterized by the physical absence of the parents and few channels of communication among the members of the family. Aspects such as the parental migration to the United States, or other cities of the country, as well as the insertion of the parents in precarious works and long working days made it difficult to establish channels of communication between participants and their parents or close relatives. This situation made adolescents feel little emotional support within their families, and live the disease with the feeling of loneliness.

Finally, our findings, as well as participant observation, suggested that adolescents with T2DM are a hidden population [37]. If adolescents are not aware of the disease, the health system cannot catch them; the cases that can be detected, usually by reference to other diseases, have little adherence to treatment; and as already mentioned, these adolescents hide the disease, which makes identification, follow-up and treatment difficult.
DISCUSSION

Adolescents with T2DM: a hidden population

The fieldwork that was conducted in Sinaloa, Mexico made it possible to identify that adolescents, in spite of the incidence of diabetes among this population, constitute a hidden population [37, 38]. The self-management barriers, the abandonment of treatment and hiding the disease, as a result of prejudices with respect to T2DM, complicated participant identification and recruitment, as other studies have previously shown [11, 39].

Findings of our research are in agreement with past studies that underscore that hiding T2DM is a recurring practice among adolescents; this behavior represents a resilience to face the social stigma that is associated with diabetes and its management [11, 12, 15, 40]. But, at the same time, hiding the disease promotes strong vulnerabilities among the participants, such as the reduced use of health services, limited access to appropriate treatment and impaired mental health.

Knowledge about T2DM, family support and self-care practice implications

In this study, adolescents constructed their diabetes representations from individual and contextual elements. Firstly, the acquired knowledge of T2DM in family, community, medical and school contexts promotes or limits self-care practices, and stimulates certain behaviors, such as hiding the disease. The hiding of diabetes highlights the absence of intergenerational communication within the adolescents' families, considering that there are other members who also suffer from diabetes. In this context, experiences, feelings or fears are not exchanged among family members who have diabetes. This acquires relevance considering the results of some studies that show the importance of family and friend solidarity in diabetes management among adolescents [14, 15].

The parental support that the participants receive cannot be separated from the con-

text where the research was carried out. As mentioned in the section above, adolescents' homes are characterized by the absence of one parent and limited family cohesion, with reduced intergenerational dialogue among their members. At the same time, the families of the participants experience conditions of socioeconomic precariousness and poverty. Family members, including some of the participants, must work long journeys to try to meet basic needs. These conditions promote that care practices towards illness, dietary habits and health care are not priority issues in the daily lives of families; situations that tend to reduce the family support that adolescents with type 2 diabetes can receive in this scenario.

Another important element to consider is the poor medical-patient dialogue. The participants did not feel understood by health professionals, who in turn transmitted ambiguous knowledge about the disease, without regard for the adolescents' needs. Their narratives showed that the discourse of *healthy eating* predominates medical speech about diabetes; however, dietary habits are culturally and socioeconomically constructed, and changes to those habits are not easy [18]. Therefore, food restrictions were one element that generated great conflict in the participants, and disease control focused exclusively on losing weight, particularly among women.

This situation placed adolescents in a state of constant ambivalence. They had close examples of disease deterioration within their families. They were worried about having the same long-term effects, so they took on strict diets, often led by health professionals, that were impossible to maintain in the mediumand long-term. Lacking examples of self-care practices and healthy habits makes it difficult for them to consistently maintain their regimens over time, which generates emotional guilt and fear to follow the same deteriorations patterns of their relatives.

Identity construction

The process of identity construction seems to be traversed by disease. The repercussions of the T2DM on daily life seem to significantly affect adolescent's socialization and their perceptions of themselves. The impact of popular beliefs about illness imposes limitations, prohibitions and exclusions on their participation in social encounter activities.

The fear of dying was a central component in the identity construction of the participants. Some studies suggest that young people experience their youthfulness from a perspective that is centered on the present and not as a transition to adulthood to face the future [41]. Nevertheless, the participants in our study lived in a constant dichotomy between present and future, with the fear of dying or getting worse in the short- and long-term, which varies according to the gender norms.

Disease significance

Narratives of participants suggest that meanings attributed to disease vary by sex. For women, major issues seemed to focus on body image and their social desirability for beauty patterns. Most of females had overweight and obese problems strongly associated with T2DM, but their main concern was to lose weight to improve their body image. Furthermore, they did not want to be a burden to their families, as they perceived that diabetes generates limitations in their autonomy. Males, in contrast, perceived that T2DM has been limiting their labor activities and subsequent ability to have and provide for, as other research suggests [21]. In this sense, diabetes is associated with the idea of failure, which is sustained through the traditional gender imaginary that is heavily embedded in Sinaloa society.

CONCLUSIONS

Representations of T2DM constitute a socially constructed body of knowledge regarding a disease that organizes adolescents' practices perceptions and modes of facing the illness. This perspective allows the identification of the social and cultural aspects of health and illness and the different knowledge and behaviors that are present in the day-today experience of patients, which is useful for prevention, care and treatment.

Our research showed that how adolescents experience and face the disease does not occur homogeneously but varies according to sex, which should be considered by care programs and health professionals to encourage adherence to treatment, taking into account the emotional well-being and needs of adolescents. For males, T2DM implies a reconstruction of male identity, in so far as the disease is believed to limit their job prospects and the ability to provide, which is traditionally associated with male roles. However, for women, the association of diabetes with overweight and obesity affects their self-esteem by exposing them to a double vulnerability. On the one hand, diabetes and obesity represent conditions that limit their life expectancy, and on the other hand, it gives them an emotional burden by not complying with the social stereotype of thinness and beauty.

Our study highlighted two substantial findings that have not been pointed out in other qualitative studies and should therefore be analyzed in depth in future research on type 2 diabetes mellitus in adolescents. Firstly, adolescents with this condition constitute a hidden population; that is, a group difficult to identify and follow up. Diabetes representations as an adult condition and social sanctions experienced by people with diabetes in the Mexican context encourage adolescents to hide the disease in their socialization settings, especially in the family and in school, which makes them invisible for health services. Secondly, the absence of intergenerational communication within the adolescents' families about diabetes limits adolescent care practices and exacerbates their emotional distress for fear that their health will deteriorate in the medium term and even for the possibility of dying, as happened with their close relatives. Therefore, the study also highlights the importance of the family nucleus. It is central to continue to investigate the family role and support for the development of healthy practices in adolescents with diabetes through further qualitative research. In addition, our research revealed the relationship between emotional distress and diabetes; therefore, it would be important to ask how other family, school and community aspects contribute to decrease or exacerbate the subsequent distress, and to identify the mechanisms that allow them to manage stress from stigma and exclusion in order to promote their emotional well-being.

Thirdly, the research revealed that the participants' identity construction is strongly affected by the disease, and adolescents must find new ways for preparing themselves for becoming an adult, which should be further investigated in the future by qualitative studies related to mental health; especially in precarious socioeconomic contexts, with few job and school opportunities, in which most of the research participants were inserted.

The practice among adolescents with diabetes to hide the disease should be considered by health professionals to encourage adherence to treatment. Our findings showed the need to incorporate the mental health area at the first time of diagnosis, as an aspect of the comprehensive care strategy in order to provide sufficient psychological resources for adolescents and their families to help prevent the abandonment of treatment.

Importantly, the main limitation of this study is the lack of generalizability of the results among Mexican adolescents with diabetes due to the sample size and the context in which the interviews occurred and the particular context in which participants were recruited. However, our sample was exhaustive for the Mazatlán municipality and our findings generated new important research issues and questions to expand a new line of research. Indeed, the social vulnerability in which young people find themselves, and their family and demographic characteristics makes it difficult to diagnose new cases of this disease and provide them with the necessary treatment.

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ORIGINAL ARTICLE

Home management of malaria among caregivers of under-five children in Owo, Ondo State, Nigeria

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Abstract

Introduction: Home management of fever by caregivers is particularly relevant for ensuring effective care to under-five children in Sub-Saharan countries with high-transmission of malaria like Nigeria. Our study aimed to assess home management of fever among mothers and other caregivers of under-five children attending Federal Medical Centre in Owo, Ondo State, Nigeria.

Methods: A descriptive, cross-sectional hospital-based study was carried out. A systematic random sampling technique was used to recruit participants (mothers and other caregivers). A structured interviewer administered questionnaire was used. Clinical findings and malaria test were performed on children using standard methods. Data were analyzed with SPSS version 15.

Results: Prevalence of home treatment of fever was 98.4%. Most of the caregivers bought antifever drugs from patent medicine shops and only 11.9 % of them got a prescription drug from pharmacies. Despite good knowledge about causes of malaria by caregivers (72.6%) and a prompt medical treatment within 24 h of fever onset (84.2%), many of their children were treated with only paracetamol (45%) and, therefore, they presented to the hospital febrile (61.8%), tachycardic (63.7%), and tachypnoic (83.9%). Almost all of the children were diagnosed with malaria (98%) associated with some types of complication such as convulsion (29.6%) and anaemia (52.7%). Chi square test showed a significant (P < 0.005) difference between treatment modality (inpatient versus outpatient) with regards to educational level, type of caregivers, place of domicile, use of antimalarial in combination with paracetamol, and presence of complications.

Discussion and Conclusion: In our study, home management of malaria by caregivers of under-five children was ineffective. Indeed, the effectiveness of home management of malaria depends not only on the early diagnosis, but also on a prompt and appropriate treatment. Therefore, proper health education programmes on this disease should be implemented among caregivers of under-five children.

KEY WORDS: Antimalarials; Caregivers; Child; Fever; Malaria.

Riassunto

Introduzione: La gestione domiciliare della malaria da parte dei "caregivers" è particolarmente importante per assicurare l'efficacia delle cure nei bambini con meno di 5 anni che vivono nei Paesi dell'Africa subsahariana come la Nigeria, dove la malaria è endemica. Il nostro studio è stato mirato a valutare la gestione domiciliare della febbre da parte delle madri e degli altri "caregivers" di bambini con meno di 5 anni accompagnati per febbre presso il Federal Medical Centre in Owo, nello Stato di Ondo, in Nigeria. **Metodi:** E' stato realizzato uno studio descrittivo, di tipo trasversale basato sui dati ospedalieri. E' stato effettuato un campionamento sistematico casuale per reclutare i partecipanti (madri ed altri tipi di "caregivers") ed è stato utilizzato un questionario strutturato somministrato da un'intervistatore. I risultati clinici ed i test per la diagnosi di malaria sono stati effettuati nei bambini attraverso i metodi standard. I dati sono stati elaborati con il software SPSS versione 15.

Risultati: Nel nostro campione il trattamento domiciliare della febbre con farmaci ha registrato una prevalenza del 98,4%. La maggior parte dei "caregivers" aveva comprato farmaci anti-piretici presso negozi per farmaci da banco e solo l'11,9% presso farmacie dietro prescrizione medica. Nonostante una buona conoscenza delle cause della malaria da parte dei "caregivers" (72.6%) ed un rapido trattamento farmacologico effettuato entro le 24 h dalla comparsa della febbre (84.2%), molti bambini sono stati trattati soltanto con paracetamolo (45%) e pertanto si sono presentati in ospedale febbrili (61.8%), tachicardici (63.7%) e tachipnoici (83.9%). A quasi tutti i bambini è stata diagnosticata la malaria (98%) associata a qualche forma di complicanza come le convulsioni (29.6%) e l'anemia (52.7%). Il test del chi quadrato ha evidenziato una significativa (P < 0.005) differenza riguardante le modalità di trattamento ricevuto in ospedale (ambulatoriale versus ricovero ospedaliero) con riguardo ai seguenti fattori: il livello di educazione, la tipologia ed il domicilio del "caregiver", l'eventuale uso di antimalarico associato al paracetamolo e la presenza di eventuali complicanze.

Discussione e Conclusione: Nel nostro studio la gestione domiciliare della malaria da parte dei "caregivers" di bambini con meno di 5 anni è risultata inefficace. Infatti, l'efficacia della gestione domiciliare della malaria non dipende soltanto da una diagnosi precoce, ma anche da un tempestivo ed appropriato trattamento farmacologico. Pertanto. appropriati programmi di educazione alla salute su questa patologia dovrebbero essere implementati tra i caregivers di bambini con meno di 5 anni.

TAKE-HOME MESSAGE

Home management of fever by caregivers is particularly relevant for ensuring effective care to under-five children in Sub-Saharan countries with high-transmission of malaria like Nigeria. It depends not only on the early diagnosis of malaria, but also on a prompt and appropriate medical treatment. Therefore, health education programmes on this disease are crucial.

Competing interests - none declared.

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INTRODUCTION

Fever is the most common symptom of malaria and malaria is the most prevalent illness among under-five children in Sub-Saharan African countries like Nigeria [1-4]. Therefore, when the child has a fever, children's parents or care-givers commonly treat them as if they were affected by malaria, whether right or wrong [5]. Even if fever is the most characteristic symptom of malaria, clinical findings in malaria may range in severity from mild headache to severe complications leading to death, particularly in falciparum malaria [6]. However, other symptoms and signs of both uncomplicated and severe malaria are rarely given any consideration. As a consequence, non-specific symptoms of malaria such as headache, fatigue, abdominal discomfort, lassitude, muscle and joint aches, anorexia and vomiting may go unnoticed and delay effective treatment [1, 7]. However, an early diagnosis of malaria is essential because the delay in seeking hospital care can lead to delayed recovery with a subsequent high mortality rate [8]. Therefore, the ability of mothers or care-givers to suspect malaria in case of fever has important consequences. Indeed, an early diagnosis and adequate treatment are essential elements of appropriate malaria control programmes [8–10]. As the recent World Health Organization's Guidelines (2012) recommend, a 'community case management' of malaria strategy is needed to 'improve access to prompt and effective case management of malaria, especially in remote, underserved areas with high malaria transmission' [11]. According to this strategy, simple and reliable diagnostic tests, as well as effective and appropriate treatment for uncomplicated malaria should be provided at the community level by trained community-based providers, such as community health workers, mother coordinators and private vendors [11]. Home management of fever is also an integral part of malaria case management within the overall Roll Back Malaria (RBM) strategy (2005). RBM is a social movement founded by the World Health Organization, the World Bank, United Nations International Children's Emergency Fund (UNICEF), and United Nations Development Programme (UNDP) and directed by national authorities in order to address malaria, especially in endemic countries [12]. One of the main objectives of RBM strategy is to enable and to increase the capacity of caregivers to recognize malaria illness promptly and take early appropriate action.

Accordingly, some studies in Burkina Faso and Nigeria showed that home management of fever can be particularly relevant for ensuring effective care to children under the age of five in areas with high-transmission of malaria [13, 14]. Unfortunately, treatment of malaria is started and, often, concluded at home by caregivers who are unaware of the causes, symptoms and signs of uncomplicated malaria as well as other causes of fever [4]. Yet, mothers and other caregivers often begin an unorthodox, inappropriate and dangerous treatment. As a result, by the time they got their children to the hospital, life threatening complications have developed [8]. In Nigeria, there are several studies about caregivers' knowledge, attitude and practices on malaria in young children [2–4, 9, 10]. Nevertheless, it is particularly important to ensuring effective care for non-immune people at risk of malaria, such as children under the age of five in high-transmission situations. This study aimed to describe the knowledge of mothers and care-givers about causes, treatment and preventive measures of malaria and to study caregivers' attitude concerning use of preventive measures and home treatment given to their children. In our research, we also analyzed health-seeking behaviour of caregivers of under-five children with fever attending Federal Medical Centre in Owo, Ondo State, Nigeria, in order to better understand their subsequent implications on health of children affected by malaria fever.

METHODS

Study area and population

The target population consisted of caregivers of under-five children with febrile illness receiving healthcare services of the Federal Medical Centre (FMC) in Owo. The FMC is located in Owo Local Government Area of Ondo State, Nigeria. Owo Local Government Area has a population of about 219,000 people (2006 census). FMC is the only Federal health institution in Ondo State that provides primary, secondary and tertiary levels of health care for all-aged people. About 280 children aged 6-60 months are treated monthly for fever at children outpatient department/emergency room.

Study Design, sample size and sampling

This was a descriptive, hospital-based cross-sectional study, conducted over a period of three months from May 1st to July 31st, 2008. The sample size was determined using the sample size formula for estimation of proportions. Prevalence of home treatment of malaria in Nigeria of 76% was used [10]. Adding a 10% value because of attrition rate, a total of 373 was obtained. Based on this, the estimated sample size was 380 caregivers. A systematic random sampling technique was used to recruit participants, 280 children aged 1-60 months were registered monthly for three consecutive months at the children outpatient clinic/emergency room of FMC. Therefore, the sampling frame consisted of 840 subjects. By using a systematic random sampling, a sampling interval of 1 in 2 was obtained. Hence, a total of 420 caregivers were recruited for this study (rate of respondents = 90.5%, n = 380). A caregiver was defined as the mother, the father or any adult responsible for the care of the child [15].

Study Instruments

A structured questionnaire was administered to children's caregivers in Owo by health care officers of the Department of Family Medicine, Community Health, and Paediatrics of FMC, who were recruited as field workers for the study and trained using participatory approaches in communication skills, form-filling and demographic techniques. The questionnaire was divided into two sections: 1) Socio-demographic characteristics of caregivers (age, sex, marital status, place of domicile, ethnic group, occupation and educational status) and their children (sex and age); and 2) Knowledge of caregivers about fever (cause, symptoms, and complications of malaria) and their subsequent behaviours (attitude toward 'seeking medical treatment', Insecticide-treated nets (ITNs) ownership and usage, time to start anti-malarial treatment, type of medical treatment used). Knowledge of correct treatment of malaria included questions about paediatric dose and treatment duration. In this study, antimalarial treatment was an Artemisinin-based combination therapies, which are current drugs for treatment of uncomplicated malaria. Home treatment of malaria includes usage of Artemisinin based combination therapy (ACTs) to be taken every day for 3 days [16]. Clinical data of the children (i.e. weight, temperature, heart rate, respiratory rate, current diagnosis of malaria, and types of complications as applicable) and modality of hospital treatments (inpatient versus outpatient) were recorded.

Temperature

Clinical thermometer model hospital and homecare CEO 123°C & °F (suitable temperature range: 35-42 degrees Celsius) was used to measure body temperature. Fever was taken as body temperature \geq 37.5°C.

Laboratory Test

DiaSpot Malaria P.f. Rapid Diagnostic Test Device (Whole Blood), a qualitative, membrane based immunoassay test for the detection of Plasmodium falciparum antigen in whole blood, was used in this study. The model of the kits used was MAL7080021 with each pack containing forty test kits, forty droppers and two buffers. The correlation between the traditional thick and thin blood smears microscopic analysis and malaria P.f. Rapid Diagnostic Test Device (Whole Blood) has been found to be over 99.0%. Sensitivity and specificity have also been found to be 99.0%. Cost of the test was also comparable to the traditional method. Rapid diagnostic tests (RDTs) are affordable, alternative diagnostic tools [17]. Malaria rapid diagnostic tests (RDTs) are recommended at community points of care because they are accurate and rapid [18]. Limitations of this test are as follows: i) It is for in vitro diagnosis only; ii) It can only be used for the detection of P *falciparum* antigen in whole blood specimens; iii) Neither the quantitative value nor the rate of increase in *P falciparum* antigen can be determined by the qualitative test.

Data analysis

Statistical Package for Social Sciences (SPSS) version 15 was used for data analysis. Means, modes, medians, Standard Deviation (SD), proportions and percentages were determined as applicable. The means and SD were calculated for continuous variables while ratios and proportions were calculated for categorical variables. Further analysis exploring relationships between variables was done using Pearson's chi-square (χ^2) test. P values of less than 0.05 was accepted as statistically significant. The malaria knowledge statements were made up of correct and incorrect statements. The answers were recoded so that respondents who agreed or disagreed with an incorrect statement were given a '0' and '1', respectively. 'Don't know' and no answer responses were treated as incorrect and coded as a '0' [19]. Therefore, knowledge was categorized as 'poor' (< 2 variables marked correctly), 'fair' (2 or 3 variables marked correctly), and 'good' (> 3 variables marked correctly).

Ethical clearance

Ethical clearance was obtained from health research ethics committee of FMC, Owo. Informed, written consent was obtained from all the caregivers.

RESULTS

Overall, 380 caregivers (M = 27, 7.1%; F = 353, 92.9%) were enrolled for this study, which ranged in age from 25 to 65 years (M = 30.48, $SD \pm 6.61$) and consisted of mothers (n = 346, 91%) or other type of caregivers (n = 34,9%) including father (n = 19,5%), sisters (n = 6, 1.5%), brothers (n = 4, 1.1%),

uncles (n = 4, 1.1%), and grandmothers (n = 1, 0.3%). A summary of the sociodemographic characteristics of the respondents is presented in Table 1.

In our study, there were 380 children (M = 176, 46.3%, F = 204, 53.7%) who ranged in age from 6 to 60 months (M = 22.1, SD = 15.2). Table 2 shows the sociodemographic characteristics and clinical data of children at presentation. Overall, more than half of the children were febrile (n = 235, 61.8%), with tachycardia (n = 242, 63.7%) and tachypnea (n = 319, 83.9%). Moreover, clinical examination and laboratory tests showed a high prevalence (98.4%) of home management of fever and among all cases, 372 (98%) were positive for malarial antigen(s) in RDT.

As showed in Table 3, caregiver's knowledge about causes of malaria was good (n = 276, 72.6%). However, attitude toward 'seeking medical treatment', and usage of ITNs for prevention of malaria and antimalarial drugs in case of fever were 'poor' (65.3%, 62.7%, and 43.5%, respectively). With regards to preventive measures of malaria, about half of the participants knew (n = 225, 59.2%) and less than half of them used (n = 142, 37.3%) insecticide-treated nets. However, there was a wide difference in their frequency of use by caregivers ('always' n = 42, 18.7%; 'often' n = 112, 49.8%; 'sometimes' n = 62, 27.5%; 'rarely' n = 9, 4.0%).

Table 4 shows that the prevalence of home management of fever by caregivers using some types of medications was high (98.4%). However, before presenting to the hospital, paracetamol was the most used drug (n = 171, 45%). A combination of paracetamol with antimalarial drugs was given by about one third of the respondents (n = 126,33%), while antimalarial (artemisinin-based combination therapy) was used by 93 (10.3%) participants, and antibiotics by 2 (0.5%) caregivers. Moreover, anti-malarial treatment at home began within 24 h in a high percentage of respondents (n = 320; 84.2%). With regards to anti-fever drug's provider, majority of the respondents (n = 275, 77.7%) bought medications from patent medicine stores.

Table 1. Socio-demographic characteristics of caregivers (n = 380).	
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Characteristics	N	%	
Age group in years			
≤ 29	159	41.8	
30-39	196	51.6	
≥ 40	25	6.6	
Educational status			
No formal education	12	3.2	
Primary	79	20.8	
Secondary	167	43.9	
Tertiary	122	32.1	
Occupation			
None	33	8.7	
Farming	14	3.7	
Trading	184	48.8	
Artisan	63	16.6	
Professional	86	22.6	
Ethnic group			
Yoruba	308	81.1	
Ibo	39	10.2	
Hausa	11	2.9	
Others	22	5.8	
Marital status			
Married	336	88.4	
Single	44	11.6	
Relationship to Child			
Mother	346	91.1	
*Others	34	8.9	
Place of domicile			
Within Owo (Urban)	269	70.8	
Outside Owo (Rural)	111	29.2	

Note: *Fathers 5%, other caregivers 3.9%.

Characteristics	N	%	
Age group (months)		<u>`</u>	
6-12	159	41.8	
13-24	95	25.0	
25-36	57	15.0	
37-59	69	18.2	
Gender			
Male	176	46.3	
Female	204	53.7	
Temperature (°C)			
Normal (≤ 37.5)	145	38.2	
Febrile (> 37.5)	235	61.8	
Heart rate (beats/minute)			
Normal (≤ 100)	138	36.3	
Tachycardia (> 100)	242	63.7	
Respiratory rate (cycles/minute)			
Normal (≤ 30)	61	16.1	
Tachypnea (> 30)	319	83.9	

Table 2. Sociodemographic characteristics and clinical data of children (n = 380).

Table 3. Knowledge of causes, treatment and knowledge/use of preventive measures of malaria among caregivers.

Caregiver's Knowledge and Attitude	N	%	
Knowledge of correct cause of malaria			
Mosquito bite	276	72.6	
Others	104	27.4	
Attitude toward 'seeking medical treatment'			
Correct	132	34.7	
Wrong	248	65.3	
Knowledge of correct treatment of malaria			
Good	103	27.1	
Poor	277	72.9	
Knowledge of Insecticide-treated nets (ITNs)			
Good	225	59.2	
Poor	115	40.8	
Use of Insecticide-treated nets (ITNs)			
Yes	142	37.3	
Not	238	62.7	

Type of drugs used at home	Ν	%	
Paracetamol	171	45	
Anti-malarial drugs*	93	10.3	
Combination of paracetamol and anti-mala- rial drugs	126	33.2	
Herbal preparation	18	4.7	
Tepid sponging	18	4.7	
Antibiotics	2	0.5	
Nothing	6	1.6	
Time to start anti-malarial treatment in case of fever			
< 24 hours	320	84.2	
≥ 24 hours	60	15.8	
Source of medical treatments			
Pharmacy	42	11.9	
Medicine Vendor	22	6.2	
Patent medicine shop	275	77.7	
Native doctor/Herbalist	11	3.1	
Others	4	1.1	

Table 4. Types of treatment and places where treatment was sought by caregivers.

Note: *Current drugs for treatment of uncomplicated malaria are Artemisinin based combination therapies

Table 5. Type of diagnosis made and complications detected at the hospital.

Diagnosis	N	%	
Malaria	314	82.7	
Chest Infection	7	1.8	
Ear Infection	2	0.5	
Others	57	15	
Type of complications (<i>n</i> = 203, 53.4%)			
Convulsion	60	29.6	
Anaemia	107	52.7	
Dehydration	4	2.0	
Cerebral Malaria	6	3.0	
Convulsion with anemia and dehydration	26	12.8	

As shown in Table 5, malaria was diagnosed in 314 (82.7%) children, while other children were diagnosed with malaria and co-morbid chest infection (n = 7, 1.8%), ear infection (n = 2, 0.5%), or other diagnosis (n = 57, 15%). Out of a total of 380 children, 203 (53.4%) were diagnosed with complications of malaria, such as convulsion (n = 60, 29.6%), anaemia (n = 107, 52.7%), dehydration (n = 4, 2.0%), cerebral malaria (n = 6, 3.0%), and convulsion with anemia and dehydration (n = 26, 12.8%). With regards to medical treatment given by physicians at the hospital, antimalarial drugs were used in most of the cases (n = 365, 96.1%); further, 4 (1.1%) children were given anticonvulsant, 9 (2.5%) of them antibiotics, and only 2 (0.5%) children received a blood transfusion.

As shown in Table 6, one third of children (n = 143, 37.6%) were managed as inpatient, two third of them (n = 237, 62.4%) as outpatient. Chi square test showed a significant difference between treatment modality (inpatient versus outpatient) with regards to educational level ($\chi^2(3) = 26.498$, P < 0.001), type of caregivers ($\chi^2(1) = 11.887$, P < 0.001),

Characteristics	Treatment settings		χ^2 , df and <i>P</i> -value
	Inpatient N = 143 (37.6%)	Outpatient N = 237 (62.4%)	
Educational level			
None	9 (75)	3 (25)	
Primary	42 (53.2)	37 (46.8)	$\chi^2 = 26.498$
Secondary	64 (38.3)	103(61.7)	$P < 0.001^*$
Tertiary	28 (23.0)	94 (77.0)	
Identity of caregiver			
Mother	140 (40.2)	208 (59.8)	$\chi^2 = 11.887$
Others	3(9.4%)	29 (90.6)	df = 1 $P < 0.001^*$
Place of domicile			
Within Owo City (urban)	89 (33.1)	180 (66.9)	$\chi^2 = 8.109$
Outside Owo City (rural)	54 (48.6)	57 (51.4)	df = 1 $P = 0.004^*$
Type of malaria			
Uncomplicated	37 (20.9)	140 (79.1)	$\chi^2 = 39.501$
Complicated	106 (52.2)	97 (47.8)	df = 1 $P < 0.001^*$
Time to start anti-malarial treatment in case	of fever		
< 24 hours	102 (37)	174 (63)	$\chi^2 = 0.196$
≥ 24 hours	41 (39.4)	63 (60.1)	df = 1 $P = 0.372$
Type of drugs used at home (n = 297)			
Paracetamol and antimalarial drugs used in combination	61 (35.7)	110 (64.3)	$\chi^2 = 13.524$ df = 1
Paracetamol	72 (57.1)	54 (42.9)	$P = < 0.001^*$
Total (297)	133 (44.8%)	164 (55.2%)	

Table 6. Association between characteristics of caregivers, type and modality of medical treatment used at home and type of treatment settings.

Note: P < 0.05 as significant *

place of domicile ($\chi^2(1) = 8.109$, P = 0.004), type of drugs used at home ($\chi^2(1) = 13.524$, P < 0.001), and presence of complications (χ^2 (1) = 39.501, P < 0.001).

DISCUSSION

In our study, prevalence of home treatment of fever was 98.4%, a value higher than other studies where it was found to be between 50.0% and 90.0% [10, 20]. Probably, as past research has highlighted [21, 22] seeking hospital care is considered as a last resort by caregivers, and the use of home treatment might be because most of them could not afford hospital and needed prompt treatment. Most of the caregivers bought anti-fever drugs from patent medicine shops. Only 11.9 % of our respondents got a prescription drug from pharmacies. In Nigeria, patent and proprietary medicine vendors (PPMVs) and their shops are usually the first point of call when people fall sick, especially in the rural areas. They are called 'doctors' by people who do not know their difference with qualified medical doctors. Indeed, it is less expensive seeking patent medicine sellers than accessing health care facilities. In both patent medicine shops and health care facilities payment are usually made out-of-the pocket, which has been showed to push households into poverty [23]. Another finding of our study was that despite a good knowledge about causes of malaria by caregivers and a prompt starting time within 24 h of medical treatment in case of fever, many children presented to the hospital febrile (61.8%), tachycardic (63.7%), tachypnoic (83.9%), and almost all of them were diagnosed with malaria (98%) associated with some types of complications of malaria such as convulsion (29.6%) and anaemia (52.7%). Therefore, even if medical treatment of fever at home resulted largely practiced, it was ineffective probably because many of the children were treated with only paracetamol (45%), while a combination of antimalarial drug and paracetamol were only used by less than half of the caregivers. This finding is in agreement with prior studies [3, 24]. For instance, a study carried out in rural area of Northern Nigeria showed that caregivers had a good knowledge of both the cause and the measures of prevention of malaria, but the knowledge of proper administration of antimalarial drugs was limited [25]. Similar findings were also reported in an urban community in south-western Nigeria [26] and in a semi-urban area of Cameroon [27]. Probably, as Webster and Beyeler et al. stated [28, 29], the caregivers and the patent medicine sellers lack effective knowledge about medical treatment and unfortunately, caregivers prefer seeking medicine vendors as the place to receive treatment, because they are easily accessible and available, despite the fact that a real knowledge of medicine calls for treating the disease's causes and not its symptoms. As a consequence, due to a wrong home-management of fever, many children of our sample were admitted to the hospital with a diagnosis of anaemia and convulsions [8]. Indeed, malarial fever is often associated with nonspecific symptoms or severe complications, as past studies has showed, especially if the correct medical treatment is not promptly started within 24 h [3, 30]. Therefore, poor knowledge of correct management at home, and the role played by patent medicine sellers might have accounted for the widespread inappropriate home management of fever observed in this and other studies [19, 31, 32]. In Nigeria, owner-operated drug retail outlets, or patent and proprietary medicine vendors (PPMVs) are a main source of medicine for acute condition [16]. PPMV is defined as

'a person without formal training in pharmacy who sells orthodox pharmaceutical products on a retail basis for profit' [33]. Given their numbers, market share, and presence in rural communities, PPMVs represent an important opportunity for improving the delivery of primary health-care commodities and services [28]. However, according to a recent systematic review, PPMVs have poorer knowledge of proper malaria treatment than public health facility staff and pharmacist [28]. In our study, we used the difference between 'inpatient' and 'outpatient' treatment as based on severity of illness and intensity of service. 'Outpatient' status is commonly referred to patients who typically go to an outpatient department such laboratory, radiology or to the emergency department for diagnostic services, in order to determine they need to be admitted to the hospital as a hospital patient or be discharged to go home. Urban children of more educated caregivers showed a lower level of severe forms of malaria, and a higher probability to be treated as outpatient than children from rural area of low educated mothers. In addition, children treated with antimalarial in combination with paracetamol, and without complications of malaria were also significantly treated as outpatient. Therefore, as showed by other studies [21-22, 24, 28], the accessibility to health care services is very important for good management of malaria. A Nigerian research highlighted that a correct home management of fever could extend the reach of public health services, and be more effective in areas with low or moderate levels of disease occurrence, where a widespread increase is generally observed during epidemics [34]. Increasing access to health care services is considered pivotal to improving the health of populations. Prompt access to malaria diagnosis and treatment is a key component of the Roll Back Malaria. According to a recent review of literature, key-factors include 'costs, proximity, timeliness, supplies', as well as 'the importance of health worker personalities and relationships with patients' [35]. In Sub-Saharan Africa countries like Nigeria, conversely,

poverty, negative attitude, poor access roads and transport system, are important challenges to access health facility services. According to the National Antimalarial Treatment Policy by Federal Ministry of Health, Nigeria (2005), patients with malaria should have access to appropriate and adequate treatment within 24 hours of the onset of symptoms and insecticide treated nets and other materials should be available and accessible to persons at risk of malaria, particularly pregnant women and children under 5 years of age [16]. However, according to this National report, most of malaria cases (80%) are inadequately managed at community level by the facility and home-based caregivers. More specifically, according to the 2005 Report, almost all of caregivers (96%) initiate actions within 24 hours but only 15% of their actions are appropriate due to inadequate dosage. More than half of all mothers (60%) have no knowledge of the current management of convulsions, and about 50% of them obtain drugs from Patent Medicine Vendors of which 89% of the drugs were found to be substandard and 43% of syrups unsatisfactory [16]. In a 2004 study carried out in Ado-Ota, Ogun State, Southwest Nigeria (2004), knowledge on the control and treatment of malaria amongst caregivers of under-five children was also inadequate, calling for urgent interventions to improve home management of childhood fever and consideration to the socio-economic and cultural context of antimalarial drugs use [15]. According to the World Health Organization's Guidelines (2005), under-five children presenting with fever in Sub- Saharan Africa should be treated for malaria [12, 36]. These guidelines supported recommendations for the integrated community case management (ICCM) of common childhood illnesses that included malaria, pneumonia, and diarrhea [23, 36]. Caregivers should be taught to recognise signs of severe malaria for which they must immediately bring a child to the nearest health facility. In addition, it is important using Artemisinin-based combination therapy (ACTs) for malaria correctly [16]. According to the

Roll Back Malaria Strategy (2005), training programmes should improve skills and knowledge to recognize uncomplicated malaria, danger signs and when to refer the patient; to manage malaria appropriately (recommended medications/dosages); to improve awareness of the need for prompt and early treatment, completion of the dose, and the importance of drug storage [12]. Therefore, specific training is pivotal in all these strategies. It is not sufficient to train health providers/workers at all levels of health care. Training on the cause, recognition, treatment and prevention of malaria should be extended to pregnant women, caregivers, heads of household, religious and opinion leaders and school children [16, 37-40].

Strengths and limitations

Our study has some limitations. This was a cross-sectional study, which was, therefore, carried out at one-time point (admission to the hospital) or over a short period (three months). Another limitation concerns the validity of the measurements of fever carried out by caregivers and their self-reported answers. Collecting data from untrained caregivers through surveys or interviews could not be appropriate or effective. A strength of the study was that, although using RDT could slightly underestimate the prevalence of malaria diagnosis, our study was hospital-based where the prevalence of malaria is higher than in community. Finally, our study might not have considered the role played by antimalarial drug resistance. Indeed, it is possible that in some cases severe forms of malaria showed because of antimalarial drug resistance. In Nigeria, the development of antimalarial drug resistance like Chloroquine and others has important consequences as an increase in morbidity and mortality, delay in initial therapeutic response and, an increasing cost to the community [16].

CONCLUSION

Overall, home management of malaria by caregivers of under-five children attending children out patient department and emergency

room of Federal Medical Centre, Owo, Ondo State, Nigeria was ineffective. Even though mothers and other caregivers knew causes of malaria, unfortunately, their children's subsequent medical treatment was inappropriate and inadequate. The effectiveness of home management of malaria depends upon early diagnosis, prompt, appropriate treatment; therefore, proper health education about this disease is crucial.

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CASE REPORT

A case of asymptomatic ST segment changes in cyclist with two myocardial bridges

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Abstract

A 65-year-old male regularly involved in competitive cycling came to our sports medicine laboratory for the annual mandatory pre-participation screening. Cycling screening protocol includes a cardiological examination, basal ECG and cardiac stress test. The clinical examination was unremarkable, and the patient's blood pressure was 120/75 mmHg. The rest-ECG was normal. The cardiac exercise stress test showed a 2 mm ST-segment inversion in the anterior leads (V3-V4-V5-V6) at peak exercise. No presence of arrhythmias or symptoms were reported. Due to an ST segment depression recorded during the cardiac exercise stress test, we performed a coronary computed tomography angiography (CCTA) that showed two myocardial bridges of the left coronary artery. However, we observed no atherosclerotic plaque of the coronary lumen. A dobutamine stress echocardiogram and a bicycle stress echocardiogram were normal. We concluded for ventricular repolarisation abnormalities during maximal exercise testing due to the electrocardiographic findings in an asymptomatic athlete without any coronary artery disease. In literature, myocardial bridging is regarded as a common anatomic variant rather than a congenital anomaly. Nevertheless, some reports show that myocardial ischemia and myocardial infarction or sudden death could be caused by myocardial bridging. In addition, intramyocardial bridging is a recognised cause of sudden death in athletes. Therefore, according to the Italian cardiological guidelines for competitive sports (COCIS 2009), we recommended the patient avoid physical overload and sport practice.

KEY WORDS: Athletes; coronary artery anomalies; coronary computed tomography angiogram; exercise stress test; myocardial bridge.

Riassunto

Un uomo di 65 anni regolarmente impegnato in gare di ciclismo agonistico è giunto al nostro ambulatorio di medicina dello sport per la visita medica obbligatoria annuale di idoneità. Il protocollo di screening per il ciclismo include una visita specialistica cardiologica, un esame elettrocardiografico di base ed uno da sforzo. La visita medica non ha evidenziato alterazioni significative e la pressione arteriosa del paziente era pari a 120/75 mmHg. L'ECG a riposo era normale. Il test cardiaco da sforzo evidenziava un'inversione del tratto ST di 2 mm nelle derivazioni anteriori durante lo sforzo massimale. Nessuna aritmia nè sintomi ischemici venivano riferiti. A causa della depressione del segmento ST registrato durante il test da sforzo, abbiamo effettuato una Tomografia computerizzata angiografica delle arterie coronariche che ha messo in evidenza la presenza di due ponti miocardici dell'arteria coronarica sinistra. Tuttavia, non abbiamo osservato alcuna placca aterosclerotica del lume coronarico. Un esame ecocardiografico da stress alla dobutamina ed un esame ecocardiografico da stress al cicloergometro sono risultati nella norma. Abbiamo concluso per la presenza di alterazioni elettrocardiografiche della ripolarizzazione ventricolare durante il test da sforzo massimale in atleta asintomatico senza segni di malattia coronarica. In letteratura, il ponte miocardico è considerato una variante coronarica anatomica piuttosto che un anomalia congenita. Nonostante ciò, alcuni studi dimostrano che l'ischemia e l'infarto miocardico o la morte improvvisa potrebbero essere causati dal ponte miocardico. Inoltre, il ponte miocardico è una causa riconosciuta di morte improvvisa negli atleti. Pertanto, secondo le linee guida cardiologiche italiane per gli sport competitivi (COCIS 2009), abbiamo raccomandato al paziente di evitare il sovraccarico fisico e la pratica sportiva.

TAKE-HOME MESSAGE

Myocardial bridging has been associated with myocardial ischemia, myocardial infarction or sudden death in athletes. Cardiac stress test and, eventually, coronary computed tomography angiography (CCTA) should be indicated in competitive athletes for the annual mandatory pre-participation screening.

Competing interests - none declared.

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INTRODUCTION

Myocardial bridging is a congenital variant of a coronary artery in which a portion of an epicardial coronary artery takes an intramuscular course [1]. It is generally considered to be a benign and frequently asymptomatic condition, but in many cases it has been proposed as a cause of angina-like chest pain, acute coronary syndromes [2], myocardial ischemia [3] and other serious cardiac events such as sudden cardiac deaths [4–6]. The link between myocardial bridging and such serious and uncommon events is still controversial and unclear [7]. Patients with hypertrophic cardiomyopathy have been found to have a high prevalence of myocardial bridging, with reported rates of up to 80% on angiographies, but myocardial bridging is also present anatomically in approximately 25% of patients based on autopsy and coronary computed tomography angiography (CCTA) results [7-9]. Clearly, it is clinically silent in the majority of cases [7]. After hypertrophic cardiomyopathy, coronary artery anomalies (CAAs) are considered to be the second major cause of death in young athletes [10, 11]. In this paper, we focused on the relationship between myocardial bridging and eligibility for competitive sports, showing a case of an asymptomatic competitive amateur cyclist with ECG signs of repolarisation abnormalities associated with myocardial bridging in stress testing.

CASE REPORT

A 65-year-old male regularly involved in competitive cycling came to our sports medicine laboratory for the annual mandatory pre-participation screening. Cycling screening protocol includes a cardiological examination, basal ECG and cardiac stress test. Family and personal history were negative for cardiac diseases. He was not a smoker and denied use of any medicaments. His family history was negative for ischemic heart disease or premature sudden death. The cyclist was in excellent athletic condition, with a weight of 66 kg, height of 1.74 m and Body Mass Index of 21.8 kg/m².

The clinical examination was unremarkable

and the patient's blood pressure was 120/75 mmHg. The rest-ECG was normal. The cardiac exercise stress test showed a 2 mm ST-segment inversion in the anterior leads (V3-V4-V5-V6) at peak exercise, when the patient reached 180 beats per minute at peak exercise heart rate and 300 Watt of peak mechanical power (Figure 1). His blood pressure was normal. No presence of arrhythmias or symptoms were reported. As a second-level examination, we performed a Holter ECG monitoring that showed an absence of ventricular arrhythmias. Afterwards, we performed a CCTA to analyse the coronary anatomy and patency, which showed the presence of two myocardial bridges, in the medium tract of the anterior interventricular branch and in the medium tract of a large intermediate branch of the left coronary artery, both approximately 20 mm long and 2 mm depth-(Figure 2). No proximal atherosclerotic plaque of the coronary lumen was found. A dobutamine stress echocardiography and a bicycle stress echocardiography performed after two months were normal. No cardiac symptoms during the test and ECG changes or significant arrhythmias were reported. We concluded for ventricular repolarisation abnormalities during maximal exercise testing due to electrocardiographic findings in an asymptomatic athlete without any coronary artery disease. According to the Italian cardiological guidelines for competitive sports (COCIS 2009), we recommended the patient avoid physical overload and sport practice [12].

DISCUSSION

In our case, we performed a cardiovascular evaluation, including an exercise test to exhaustion. The Italian Society of Sports Cardiology Guidelines (COCIS 2009) suggests this approach for all subjects over 40 before granting them eligibility to participate in competitive sports [12]. Due to an ST segment depression recorded during the cardiac exercise stress test, we also performed a CCTA that showed two myocardial bridges of the left coronary artery. Moreover, we observed no atherosclerotic plaque of the coronary lumen. A dobutamine stress echocardiography and a bicycle stress echocardiography were also normal. However, as the COCIS 2009 Guidelines suggest, we recommended our patient avoid physical overload and sport practice.

Our assessment for the fitness of competitive sport was difficult; on one side in literature, myocardial bridging is regarded as a common anatomic variant rather than a congenital anomaly [13]. On the other side, there are some reports showing that myocardial ischemia and myocardial infarction or sudden death could be caused by myocardial bridging [14]. Intramyocardial bridging is a recognised cause of sudden death in athletes [15]. Kersemans et al. report a case of a 24-year-old professional cyclist with myocardial bridging who presented with a prolonged episode of exertional chest pain, ST segment inversion in the anterior ECG leads and anterior hypokinesia on an echocardiography. However, the authors conclude that the myocardial bridge had no significant hemodynamic effect in baseline conditions and the evaluation of the hemodynamic significance of frequently encountered mild-to-moderate cases of myocardial bridging can present diagnostic difficulties for the clinician [9]. Myocardial bridging is a frequent finding during coronary angiography, because 'milking' is limited to systole and does not impair myocardial perfusion that occurs in diastole [9]. Quaranta et al. present a case of an asymptomatic competitive athlete who was diagnosed with myocarditis and, as an incidental finding, a myocardial bridge. They illustrate the relevance of anamnesis and combined techniques such as ECG, echocardiography and CCTA for this type of diagnosis [16]. Bolognesi et al. describe a case of a middle-aged competitive cyclist who presented a positive exercise test and was withdrawn from competition for the coexistence of a deep and long myocardial bridge at high risk of sudden death. According to Bolognesi et al., however, anomalies such as myocardial bridging should be regarded as a particular group of congenital disorders whose manifestations and pathophysiological mechanisms are highly variable. Interestingly, the authors hypothesised an association between the length and depth of the intramural coronary artery segment and the risk of sudden cardiac death [17]. Therefore, according to the authors, when ECG findings of inducible silent myocardial ischemia at low workload are present in healthy athletes, it is mandatory to perform a CCTA in order to identify a high-risk profile [17]. In Italy, the COCIS 2009 Guidelines include myocardial bridging among the congenital abnormalities of the coronary circulation and state that this disease precludes suitability for competitive sports, as such athletes are at risk of sudden death, even in the absence of signs of inducible stress test ischemia. According to Bolognesi et al., however, this cannot be considered an absolute dogma [17]. Indeed, ECG abnormalities may be detected that represent expression of an underlying heart disease that puts the athlete at risk of arrhythmic cardiac arrest during sports, but physiological patterns that should cause no alarm are also frequent [18]. For instance, Josephson et al. demonstrated positive ST segment responses have a modestly greater (although still relatively low) risk for future ischemic events than negative responses [19]. Moreover, the presence of a myocardial bridge is common in athletes, but only in few cases can this anomaly be deemed a risk of sudden death [17].

In conclusion, it is important to recognise the clinical significance of inducible ischemia detected during exercise testing in asymptomatic athletes and to perform all diagnostic tests. It is also possible to issue a temporary sports eligibility when a diagnostic doubt persists after such tests. However, further research is needed to understand the relationship between the length and depth of the intramural coronary artery segment and the risk of sudden cardiac death in order to better define a protocol for the assessment of fitness for competitive sports.



Figure 1. Cardiac exercise stress test showing a 2 mm ST-segment inversion in the anterior leads.



Figure 2. Coronary computed tomography angiography (CCTA) showing two myocardial bridges of the left coronary artery.

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CASE REPORT IN INTERNAL MEDICINE

A case of possible Kounis syndrome as a complication of scombroid syndrome

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Abstract

Kounis syndrome is defined as the concurrence of acute coronary syndromes such as coronary spasm or acute myocardial infarction with conditions associated with activation of inflammatory mediators such histamine, arachidonic acid and various cytokines and chemokines. Recently, a variety of unusual etiologies have been reported, including scombroid syndrome. We present a case of a woman without previous history of cardiac diseases or cardiovascular risk factors, who presented to emergency department after the onset of flushing, asthenia, palpitations, burning sensation in the mouth having just eaten tuna. The electrocardiogram revealed a sinus tachycardia with diffuse ST segment depression. After therapy, in a short time symptoms recovered and a second electrocardiogram no longer showed any ST changes. These electrocardiographic changes observed in our case were probably due to transitory coronary vasospasm as described in type I variant of Kounis syndrome.

KEY WORDS: Electrocardiographic changes; histamine poisoning; Kounis syndrome; myocardial ischemia; scombroid syndrome.

Riassunto

La sindrome di Kounis consiste nell'insorgenza di sindromi coronariche acute come lo spasmo coronarico o l'infarto miocardico acuto secondarie all'attivazione di mediatori infiammatori come l'istamina, l'acido arachidonico, varie citochine e chemochine. Recentemente, è stata descritta una varietà di eziologie inusuali, inclusa la sindrome sgombroide. Presentiamo il caso di una donna con anamnesi negativa per patologie cardiache o fattori di rischio cardiovascolari, arrivata in pronto soccorso per l'insorgenza di arrossamento cutaneo, astenia, palpitazioni, sensazione di bruciore in bocca dopo aver mangiato tonno. L'elettrocardiogramma rivelò una tachicardia sinusale con diffuso sottoslivellamento del segmento ST. Dopo la terapia, in breve tempo i sintomi regredirono ed un secondo elettrocardiogramma non mostrò più le modificazioni del tratto ST. Questi cambiamenti elettrocardiografici osservati nel nostro caso potrebbero essere probabilmente dovuti a vasospasmo coronarico transitorio come descritto nella variante tipo 1 della sindrome di Kounis.

TAKE-HOME MESSAGE

Scombroid syndrome is often misdiagnosed and cardiac involvement related to scombroid poisoning is relatively rare, but it could be complicated by vasospasm as described in Kounis syndrome. Physicians should be aware of the existence of Kounis syndrome in order to obtain an early and correct diagnosis and apply appropriate diagnostic and therapeutic measures

Competing interests - none declared.

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INTRODUCTION

Scombroid syndrome is a histaminic poisoning, due to histamine-induced reaction because of the ingestion of histamine-contaminated fish [1, 2]. Usually, the course is self-limiting, but in rare cases the poisoning can be serious and severe cardiac complications have been described [1-5]. Allergic angina and allergic myocardial infarction caused by chemical mediators released throught mast-cell activation have been described as Kounis syndrome [6-8]. In this syndrome hypersensitivity coronary disorders are induced by various types of environmental exposures, drugs or conditions. Recently, a variety of unusual etiologies have been reported, including scombroid syndrome [9].

We report a case of scombroid poisoning with electrocardiographic changes associated with transitory myocardial ischemia rapidly recovered after antihistaminic therapy. This case could represent a type I variant of Kounis syndrome.

CASE REPORT

A 59-year-old woman without previous history of cardiac diseases, allergies or other relevant diseases and without cardiovascular risk factors, presented to the emergency department after the onset of flushing, asthenia, palpitations, and burning sensation in the mouth having just eaten tuna. She had not chest pain. The patient was treated by the emergency service with hydrocortisone 200 mg IV. In the emergency department, on the clinical examination she had a blood pressure of 100/50 mmHg, tachycardia (heart rate = 124 beats/min), tachypnea (respiratory rate = 28 breaths/min), oxygen saturation of 88%, and a diffuse skin erythematous rush. The first electrocardiogram (hour: 10.10 PM) revealed a sinus tachycardia with diffuse ST segment depression (Figure 1).



Figure 1. First electrocardiogram showing sinusal rhythm with some supraventricular ectopic beats and ST segment depression in leads II, III, aVF, and V2-V6.



Figure 2. Second electrocardiogram showing the disappearance of ST segment depression.

She received chloropheniramine 10 mg IV and ranitidine 50 mg IV. In a short time symptoms recovered, vital signs became normal (blood pressure = 130/60 mmHg; heart rate = 88 beats/min; respiratory rate = 14 breaths/min; oxygen saturation = 98%), and a second electrocardiogram (hour: 10.50 PM) no longer showed any ST changes (Figure 2).

DISCUSSION

Scombroid syndrome/histamine poisoning occurs worldwide and it is considered one of – if not – the most common form of toxicity, caused by fish consumption [10]. Scombroid poisoning is triggered by ingestion of poorly preserved fish of Scombroidae and Scombersocidae families, such as tuna, bonito, mackerel, albacore, skipjack, herrings, sardines, anchovies, bluefish, sea urchins, and mahi-mahi; also salmon can be implicated, but more rarely [2].

Histamine is not present in fish in normal condition, but it is produced by histidine decarboxilase present in bacteria resident in fish gills and gastrointestinal tract [11]. This enzyme is formed after few hours of fish exposition at room temperature and is inactivated by temperature of 0°C or lower. Storage at 0°C should be performed immediately after fishing because, once activated, the histidine carboxylase is still functioning even after bacteria are not more viable [12]. Indeed, the produced histamine is not inactivated by any kind of food processing or cooking [2].

Scombroid syndrome is usually a self-limiting and benign disease. Its symptomatology occurs with a rapid onset, within 10-30 min after fish ingestion. The clinical manifestations spontaneously resolves within 24 hours [13].

The most common clinical manifestations are abdominal pain, diarrhea, nausea and vomiting; facial or generalized flushing, sometimes with hives and/or edema; headache or dizzines; dry mouth associated occasionally with metallic, pepper or bitter taste; palpitations [1, 2, 13].

Notably, some severe cases of scombroid syndrome are reported in literature. These cases include cardiovascular and respiratory complications such as arrhythmias (ventricular fibrillation) and acute coronary syndrome [4, 5, 14–16]; cardiogenic shock associated with acute pulmonary edema [3, 17]; severe bronchoconstriction and hypotension [18, 19].

In literature it has been documented the occurence of angina or myocardial infarction caused by chemical mediators such histamine, leukotrienes, and neutral proteases released during an allergic episode. This phenomenon is known as Kounis syndrome and represents a cause of coronary spasm or atheromatous plaque rupture due to allergic etiology [8, 20]. Further, since 1991, allergic angina has been inserted in a subgroup of dynamic lesions of coronary occlusion [6]. The chemical mediators act on smooth vascular coronary muscles, and can induce vasospastic angina or myocardial infarction.

Kounis syndrome is defined as 'the concurrence of acute coronary syndromes such as coronary spasm, acute myocardial infarction, and stent thrombosis, with conditions associated with mast-cell and platelet activation involving interrelated and interacting inflammatory cells in the setting of allergic or hypersensitivity and anaphylactic or anaphylactoid insults' [21]. It is caused by inflammatory mediators such as histamine, neutral, proteases, arachidonic acid products, platelet-activating factor, and a variety of cytokines and chemokines released during the activation process [22–24].

A variety of electrocardiographic changes, ranging from ST-segment elevation or depression to any degree of heart block and cardiac arrhythmias, may be observed [21]. A high index of suspicion regarding this syndrome is of paramount importance. Although it is not a rare disease, its diagnosis is spare and easily overlooked [21].

Three variants of Kounis syndrome have been described [23]: the type I variant includes normal or nearly normal coronary arteriers without risk factors for coronary artery disease, and with the acute release of inflammatory mediators that may induce either coronary artery spasm without increased cardiac enzymes and troponins or coronary artery spasm progressing to acute myocardial infarction with raised cardiac enzymes and troponins. The type II variant includes culprit but quiescient preexisting atheromatous disease in which the acute release of inflammatory mediators may induce either coronary artery spasm with normal cardiac enzymes and troponins or coronary artery spasm together

with plaque erosion or rupture manifesting as acute myocardial infarction. The type III variant includes coronary artery stent thrombosis in which aspirated thrombus specimens stained with hematoxilin-eosin and Giemsa demonstrate the presence of eosinophils and mast cells, respectively [21]. In patients with type I variant, treatment of the allergic event alone may abolish symptoms. The use of hydrocortisone and H1 and H2 antihistamines is adequate [21].

New etiologies of Kounis syndrome are being detected each year [9] and several novel offenders have recently been reported to induce this syndrome, including scombroid poisoning [5, 9, 15, 25, 26].

Electrocardiographic changes observed in our case report were probably due to transitory coronary vasospasm as described in type I variant of Kounis syndrome. The limitation of this case report consists in the lack of troponin dosage, but the quick recovery of symptoms and the rapid normalization of electrocardiographic changes with antihistaminic therapy have driven us to avoid further investigations.

CONCLUSIONS

Scombroid syndrome is often misdiagnosed and therefore under reported; it is believed that the incidence is high because many cases are not reported since the symptoms may last for a short time.

Cardiac involvement related to scombroid poisoning is relatively rare, but it could be complicated by vasospasm as described in Kounis syndrome. In our case report, we showed a case of cardiac complication of scombroid syndrome. Since scombroid fish poisoning can easily be confused with food allergy, the physicians should stay aware assessing the previous consumption of fish that must alert to the possibility of this syndrome and its complications [3]. Moreover, physicians should be aware of the existence of Kounis syndrome in order to obtain an early and correct diagnosis and apply appropriate diagnostic and therapeutic measures [9].

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TAKE-HOME MESSAGES

In Japanase university hospital physicians, there is an association between overwork and depressive symptoms. The questionnaire set up by the Japanase Ministry of Health was successfully used as a screening tool in this health surveillance programme by occupational physicians and was useful to study the association between depressive symptoms, severity of chronic fatigue and burden of work.

Toru Maruyama

Climatic conditions and urbanization have considerable impact on Aedes mosquitoes' lifecycle which subsequently affects the spread of dengue virus. Focused feature engineering can reveal these lagged relationships to form informative predictors. Additionally, ensemble prediction by combining outputs from different models is found to improve accuracy over the candidate models.

Sougata Deb, Cleta Milagros Libre Acebedo, Gomathypriya Dhanapal, Chua Matthew Chin Heng

In this Sri Lanka-based study carried out on boys aged 8-16 years, chest circumference resulted as competent as height in predicting lung flows and volumes in males, and it could be used as a substitute for height for interpretation of spirometry parameters when accurate height measurements cannot be obtained.

Liyanage G, Jayamanne BDW

There is a greater need for the provision of better healthcare for individuals at-risk for or suffering from type 2 diabetes. Having health insurance is not the only answer to accessing primary care.

Crawford Tamara

Adolescents with type 2 diabetes mellitus in Mazatlán, Mexico constitute a hidden population; new identification strategies of this population should be promoted differentiated by sex and family support; not only for the care of the disease, but to reduce the emotional distress that adolescent with diabetes experiences.

Nathaly Llanes-Díaz, Dewi Hernández-Montoya

Home management of fever by caregivers is particularly relevant for ensuring effective care to under-five children in Sub-Saharan countries with high-transmission of malaria like Nigeria. It depends not only on the early diagnosis of malaria, but also on a prompt and appropriate medical treatment. Therefore, health education programmes on this disease are crucial.

Liasu Adeagbo Ahmed, Olubankole Akinboboye, Olayinka Stephen Ilesanmi, David Akintayo Oguntuase

Myocardial bridging has been associated with myocardial ischemia, myocardial infarction or sudden death in athletes. Cardiac stress test and, eventually, coronary computed tomography angiography (CCTA) should be indicated in competitive athletes for the annual mandatory pre-participation screening.

Stefano Giraudi, Barbara Severgnini, Paolo Valli, Anna Teresa Pandiscia, Lucrezia Marchetti, Silvana Scarpini

Scombroid syndrome is often misdiagnosed and cardiac involvement related to scombroid poisoning is relatively rare, but it could be complicated by vasospasm as described in Kounis syndrome. Physicians should be aware of the existence of Kounis syndrome in order to obtain an early and correct diagnosis and apply appropriate diagnostic and therapeutic measures

Stefano Rusconi, Antonio Villa



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PROMUOVERE LA SALUTE DEI LAVORATORI ANZIANI

LE ESPERIENZE EUROPEE

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Dal 1960 l'età media della popolazione è in aumento in tutti i paesi europei. Nonostante ciò, molti paesi hanno realizzato e mantenuto a lungo politiche per facilitare l'uscita precoce dal mercato del lavoro. La brusca inversione di questa tendenza negli ultimi anni ha determinato un improvviso aumento dell'età della forza lavoro, alla quale non ha corrisposto un cambiamento di modi e posti di lavoro. Molti anziani quindi sono chiamati a svolgere lavori progettati per i giovani e ciò rende la promozione della salute non più una opzione, ma un obbligo sociale. L'analisi delle esperienze di promozione della salute per i lavoratori anziani condotte in 10 Paesi europei tra il 2000 ed il 2015, indica che tali iniziative sono distribuite in modo molto eterogeneo, non solo per il numero ma anche per la qualità dei progetti. La maggior parte delle iniziative ha riguardato l'Europa Centrale, segue l'Europa dell'Est e infine i Paesi del Mediterraneo. L'analisi di dettaglio degli studi indica che la maggior parte degli interventi ha seguito un modello tradizionale, basato sulla ricerca di sintomi e segni di malattia piuttosto che su indicatori positivi di salute. Gli interventi proposti comprendevano frequentemente attività di formazione dei lavoratori anziani, mentre piuttosto rari risultano gli interventi che mirano a trasformare l'ambiente di lavoro o la sua organizzazione, soprattutto se in modo partecipativo. Il numero di iniziative di promozione della salute per i lavoratori anziani è complessivamente modesto. Mentre in Europa Centrale e soprattutto in Olanda e Germania il diffuso interesse verso il problema dell'invecchiamento della popolazione ha stimolato politiche nazionali, non tutti i Paesi dell'Est Europa risultano altrettanto attenti al problema. Nei Paesi del Mediterraneo le attività di promozione della salute sono inadeguate rispetto alle esigenze. Gli interventi realizzati seguono molto raramente i principi della moderna promozione della salute, che consiglia di abbinare i miglioramenti dell'ambiente di lavoro ai cambiamenti degli stili di vita, di adottare metodi partecipativi e di controllare gli effetti mediante indicatori positivi di salute.

Nicola Magnavita

Nato a Nicastro (CZ) il 18/05/1953, è un medico chirurgo, specialista in Medicina del Lavoro, Professore Aggregato in Medicina del Lavoro e Ricercatore Confermato dal 1980 presso l'Università Cattolica del Sacro Cuore, dove insegna medicina del lavoro e coordina il Corso di laurea di Tecniche della Prevenzione negli ambienti di lavoro. Svolge attività assistenziale nel Policlinico Gemelli. È medico competente e autorizzato presso numerose aziende. Ha pubblicato oltre 660 lavori scientifici, dieci libri e diretto 27 progetti di ricerca.