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# Interactive Journal of Medical Research

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Impact Factor (2022): 2.0  
Volume 8 (2019), Issue 4 ISSN 1929-073X

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Original Paper

# Quality of Life Among Informal Caregivers of Patients With Degenerative Cervical Myelopathy: Cross-Sectional Questionnaire Study

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## Abstract

**Background:** Degenerative cervical myelopathy (DCM) is a common, chronic neurological condition that severely affects individuals by causing a range of disabling symptoms, frequently at a time around the peak of their careers. Subsequently, individuals with DCM often become dependent on informal care arrangements. The significant economic contribution of informal care and its burden on care providers are becoming increasingly recognized.

**Objective:** This study aimed to measure the quality of life of DCM informal caregivers and provide preliminary insight into possible contributing factors.

**Methods:** Carers of individuals with DCM completed a Web-based survey hosted by Myelopathy.org, an international DCM charity. Carer quality of life was assessed in the form of caregiver happiness and 7 dimensions of carer burden using the Care-Related Quality of Life (CarerQoL) instrument. The relationships between patient disease severity, patient pain, and carer quality of life were investigated. Differences in carer quality of life were assessed across patient and carer demographic groups, including between UK and US carers.

**Results:** DCM caregivers experienced substantial burden as a result of their caregiving (mean CarerQoL-7D=64.1; 95% CI 58.8-69.5) and low happiness (mean CarerQoL-VAS [Visual Analog Scale]=6.3; 95% CI 5.7-6.9). Burden was high and happiness was low in DCM carers when compared with a large, mixed-disease study of adult informal carers where CarerQoL-7D was 79.1 and CarerQoL-VAS was 7.1. No significant relationship was found between DCM carer quality of life and patient disease severity and pain scores. DCM carer quality of life appeared uniform across all patient and carer demographic groups.

**Conclusions:** Caring for individuals with DCM is associated with reduced quality of life in the form of significant burden and reduced happiness. Reductions appear greater in DCM than in other diseases investigated. However, no simple relationship was identified between individual patient or carer factors and carer quality of life.

(*Interact J Med Res* 2019;8(4):e12381) doi:[10.2196/12381](https://doi.org/10.2196/12381)

**KEYWORDS**

spinal cord diseases; spondylosis; spinal osteophytosis; surveys and questionnaires; quality of life; chronic disease

## Introduction

### Background

Degenerative cervical myelopathy (DCM) is a neurological condition of symptomatic cervical spinal cord compression, secondary to a range of degenerative changes in the cervical spine [1]. The causative pathology includes osteophyte formation, disc herniation and ligament hypertrophy, calcification, and ossification [2].

DCM is the most common spinal cord disorder [2], with evidence from imaging studies estimating prevalence as high as 5% in the over 40s [3]. At present, treatment is limited to surgical decompression, which is able to halt disease progression, but existing damage is often permanent [4,5]. Consequently, most patients retain lifelong disabilities, including reduced quality of life. In fact, a recent study demonstrated that DCM patients have one of the most impaired quality of life scores of all chronic diseases—lower than diabetes, cancer, chronic obstructive pulmonary disease (COPD), and depression according to 36-Item Short Form Health Survey (SF-36) scores [6].

Informal care refers to care provided by family and friends [7]. Historically, informal care received little attention. However, it is clear that providing informal care brings substantial burdens to informal care providers. One study found a 63% higher mortality risk among carers experiencing strain compared with noncaregiving controls [8], whereas another study reported a significant association between carer depression and the likelihood and amount of time missed at work [9]. Informal carers of patients suffering from a range of chronic conditions, including spinal cord injuries [10], dementia [11], eating disorders [12], and Pompe disease [13], among others, have now been studied. Regardless of the disease, these studies consistently describe increased levels of burden, stress, depression, physical health problems, and a range of psychosocial problems in carers. Moreover, in a national survey of 2000 carers in the United Kingdom, 77% reported health deterioration because of the strain of providing care [14]. Unfortunately, despite these substantial burdens for carers,

informal care has a minor influence on health care decisions [7].

Despite this, recent work continues to demonstrate that informal care frequently forms a significant proportion of total care, particularly in chronic conditions [7] such as DCM. For example, in the United Kingdom there are an estimated 6 million informal carers [15], and in the United States, estimates suggest that there may be as many as 66 million informal carers [16]. The total economic contribution of UK carers is estimated at £132 billion per year [17], whereas in the United States the total opportunity costs of informal elder-care amounts to US \$522 billion annually [18].

Methods have been developed to measure informal care and facilitate its incorporation into economic evaluations of health care [19]. Available methods are diverse, with a range of different instruments, measurements, and valuation techniques available. Both costs and effects of informal care can be measured with a range of monetary and nonmonetary methods. Nonmonetary methods focus on areas such as health-related quality of life, care-related quality of life, and well-being. Monetary methods include revealed preference-based methods and stated preference methods.

The Care-Related Quality of Life (CarerQoL) instrument was designed to measure and value the impact of providing informal care [20]. The instrument consists of 2 parts. The first part, CarerQoL-7D, is a subjective burden measure assessing 2 positive and 5 negative dimensions of the impact of providing care (Figure 1). The 2 positive dimensions measure (1) fulfillment from caregiving and (2) support with caregiving. The 5 negative dimensions assess (1) relationship problems, (2) own mental health problems, (3) problems combining care tasks with daily activities, (4) financial problems, and (5) own physical health problems.

The second part, CarerQoL-Visual Analog Scale (VAS), consists of a subjective measure of carer well-being in the form of a general happiness rating on a VAS of 1 to 10. A particular benefit of the CarerQoL is that it is generic, allowing for comparison between conditions.

**Figure 1.** The Care-Related Quality of Life (CarerQoL-7D) instrument assesses carer quality of life in terms of burden and happiness (Visual Analog Scale [VAS]). For the 7D, carers choose from 3 responses to 7 different questions, their overall score aggregating to a total out of 100, with 100 representing the least carer burden. For the VAS, carers rate their happiness on a visual analog scale from 0 to 10, with 10 representing the greatest happiness.

**CarerQoL-7D**

Please draw an "X" to indicate which description best fits your current caregiving situation

a. I have  no  some  a lot of fulfilment from carrying out my care tasks

b. I have    relational problems with the care receiver (eg he/she is very demanding, he/she behaves differently, we have communication problems)

c. I have    problems with my own mental health (eg stress, fear, gloominess, depression, concern about the future)

d. I have    problems combining my care tasks with my own daily activities (eg household activities, work, study, family and leisure activities)

e. I have    financial problems because of my care tasks

f. I have    support with carrying out my care tasks, when I need it (eg from family, friends, neighbours, acquaintances)

g. I have    problems with my own physical health (eg more often sick, tiredness, physical stress)

**CarerQoL-VAS**

Please draw an "X" on the scale to indicate how happy you feel at the moment

Completely unhappy
Completely happy

0
1
2
3
4
5
6
7
8
9
10

### Objectives

Given that DCM causes substantial reductions in the quality of life of patients [6], we hypothesized that there would also be reductions in the quality of life of DCM carers.

The objective was to measure the quality of life of DCM informal carers and provide preliminary insight into possible contributing factors.

### Methods

The DCM carer quality of life survey was designed and is reported following the Checklist for Reporting Results of Internet E-Surveys [21].

### Survey Design

A cross-sectional observational study was conducted utilizing a Web-based survey targeted at carers of patients with DCM.

The questionnaire included questions to assess the situation of DCM patients and carers. Patient situation questions captured demographics including age, gender, and education; Nurick score (a disease severity classification that utilizes the degree of walking impairment to score the severity of a patient's

cervical myelopathy) [22]; best, worst, and current neck and arm/hand pain scores; and patient dependence on a carer. Carer situation questions captured carer demographics including age, gender, education, employment, country of residence, and length of time as a carer.

Carer quality of life was assessed using the CarerQoL instrument [20]. The instrument (Figure 1) comprises 2 parts: a CarerQoL-7D and a CarerQoL-VAS.

The CarerQoL-7D is designed to provide a comprehensive description of the caregiving situation [23]. Each dimension is assessed using a 3-item scale. For example, the respondent rates the statement "I have fulfillment with carrying out my care tasks" by choosing from 3 responses: (1) no, (2) some, or (3) a lot of. The responses for each dimension are weighted with a tariff score. From these, an aggregate score between 0 and 100 can be calculated, with 0 corresponding to the most carer burden (worst informal care situation) and 100 to the least carer burden (best informal care situation). Tariffs for calculating these utility scores were derived using discrete choice experiments among the general population and are population-specific. At the time of study, tariffs were available for the Netherlands [24], Australia, Germany, Sweden, the United Kingdom, and the United States [25].

The CarerQol-VAS consists of a horizontal visual analog scale whereby the carer rates their happiness on a scale of 0 to 10, with 0 representing a situation in which the carer is completely unhappy and 10 representing a situation in which the carer is completely happy [20]. The CarerQol-VAS allows informal care to be considered from an economic perspective. Carer happiness assessed using the CarerQol-VAS is a broad measure, which allows capturing of the influences of the multitude of factors that contribute to carer happiness.

Numerous construct validation studies support the validity of the CarerQol instrument [26-29].

The survey was initially piloted on a small group of carers and found to be satisfactory without need for modification.

### Ethics Approval and Informed Consent

The study was ethically approved by the University of Cambridge. All research was performed in accordance with relevant guidelines and regulations.

All carers completed the questionnaire voluntarily and were informed before doing so that their responses would be used anonymously for research purposes. Study objectives were outlined on the initial page, including details of the host organization. This acted as the electronic consent, with continuation into the survey taken as agreement. Respondents were also presented with a description of DCM, including relevant symptoms, and were required to confirm they cared for someone who suffered from this condition

No respondent-identifiable information was stored.

### Participants

All DCM carers from countries for which CarerQol tariffs were available at the time of study were included. Carers from countries without available CarerQol tariffs were excluded.

### Recruitment

The recruitment strategy has been described previously [30]. An open survey design was employed. Carers of patients with DCM were recruited to an Web-based questionnaire, administered by SurveyMonkey. Social media posts (Facebook and Twitter), supported by Myelopathy.org, were utilized to recruit participants. No contact was made with participants outside the survey.

### Administration

The questionnaire was hosted on a designated landing page on Myelopathy.org, a UK registered charity, with a large online, international patient community. The website provides a range of educational materials, support groups, and details of current research studies. The survey was not administered via email. Completion of the survey was voluntary, and no incentives were offered. Responses were collected for 12 months from November 1, 2015. A total of 25 survey items were distributed over 7 survey pages. Only responses that were complete for all carer quality of life domains were included in the final data

analysis. A missing data analysis showed complete and incomplete responses were otherwise comparable, providing reassurance that excluding incomplete responses did not introduce additional bias. Respondents were able to click back though and review their answers to previous questions before submission of the survey.

### Response Rates

Google Analytics, a Web-based analytics service that enables tracking of visits to a website, was utilized in this study to measure the number of visitors to Myelopathy.org. Survey view rate for total visits to the Myelopathy.org home page was 2.37% (421/17,737), participation rate was 32.3% (136/421), and completion rate was 37.0% (136/368).

### Preventing Multiple Entries From the Same Individual

Internet protocol addresses were recorded and used to prevent users submitting multiple responses.

### Statistical Methods

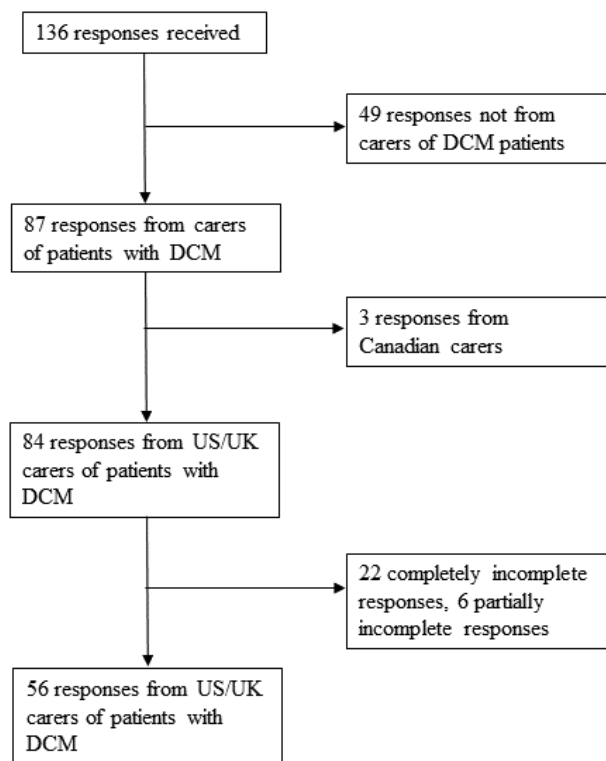
Data were checked for the presence of outliers and normality of distribution. First, as CarerQol, Nurick, and pain scores were not monotonically related, Kendall tau-b was utilized to assess for a relationship, using 2-tailed tests. Second, differences in CarerQol-7D and VAS scores between demographic groups were assessed using 1-way analysis of variances (ANOVAs). In 1 case the assumption of homogeneity of variance was violated; a Welch ANOVA was performed to assess for difference in CarerQol-VAS as a function of duration of time as a carer. Third, an independent-samples *t* test was run to determine if there was a difference in CarerQol-7D score between UK and US carers. Fourth, as CarerQol-VAS violated the assumption of normality, a Mann-Whitney *U* test was run to determine if there were differences in CarerQol-VAS between UK and US carers. Distributions of UK and US CarerQol-VAS scores were similar, as assessed by visual inspection. Analyses were conducted using SPSS Version 22 (IBM Corp). Significance was set at  $P < .05$ . We report mean (SD) unless otherwise specified.

## Results

### Participants

A total of 136 responses were received (Figure 2). A total of 49 responses were inadvertently completed by DCM patients and were thus excluded. Of the remaining 87 responses, 31 were excluded: 3 were completed by Canadian carers for whom CarerQol tariffs were not available at the time of study, and 28 responses had partial (6) or completely (22) missing CarerQol data. In a missing data analysis, scores for each CarerQol component between the 56 complete and 6 partially incomplete responses were comparable, providing reassurance that excluding the partially incomplete responses was unlikely to introduce bias. Patient and carer characteristics were >95% complete for each variable, permitting meaningful analysis.

**Figure 2.** Flowchart of response selection. A total of 136 questionnaire responses were received, 56 of which were included in the final analysis. DCM: degenerative cervical myelopathy.



### Patient and Carer Situation

As summarized in Table 1 (n [%]), the majority of carers were female (32/53, 60%), white (51/52, 98%), and from the United Kingdom (45/56, 80%). Duration of time as a carer was varied, ranging from less than 1 year to 25 years. Most (49/54, 91%)

carers were over the age of 40 years, and most (31/53, 59%) combined their caring responsibilities with either full-time (21/53, 40%) or part-time (10/53, 19%) work. The majority (19/34, 56%) felt their employment had been affected by the demands of their caregiving.

**Table 1.** Patient and carer situation.

| Variable                               | Patient, n (%) | Carer, n (%) |
|----------------------------------------|----------------|--------------|
| <b>Gender</b>                          | <b>n=56</b>    | <b>n=53</b>  |
| Male                                   | 22 (39)        | 21 (40)      |
| <b>Country of residence</b>            | <b>n=56</b>    | <b>n=56</b>  |
| United Kingdom                         | 45 (80)        | 45 (80)      |
| United States                          | 11 (20)        | 11 (20)      |
| <b>Age (years)</b>                     | <b>n=56</b>    | <b>n=54</b>  |
| ≤17                                    | 0 (0)          | 2 (4)        |
| 18-20                                  | 0 (0)          | 0 (0)        |
| 21-29                                  | 1 (2)          | 0 (0)        |
| 30-39                                  | 4 (7)          | 3 (5)        |
| 40-49                                  | 14 (25)        | 13 (24)      |
| 50-59                                  | 18 (32)        | 20 (37)      |
| ≥60                                    | 19 (34)        | 16 (30)      |
| <b>Education</b>                       | <b>n=56</b>    | <b>n=53</b>  |
| Less than high school                  | 14 (25)        | 6 (11)       |
| High school degree or equivalent       | 10 (18)        | 14 (26)      |
| Some college but no degree             | 15 (27)        | 14 (26)      |
| Associate degree                       | 5 (9)          | 5 (10)       |
| Bachelor's degree                      | 10 (18)        | 4 (8)        |
| Graduate degree                        | 2 (3)          | 10 (19)      |
| <b>Ethnicity</b>                       | — <sup>a</sup> | <b>n=52</b>  |
| White                                  | —              | 51 (98)      |
| African American                       | —              | 1 (2)        |
| <b>Employment status</b>               | —              | <b>n=53</b>  |
| Employed, working full time            | —              | 21 (40)      |
| Employed, working part time            | —              | 10 (19)      |
| Not employed, looking for work         | —              | 0 (0)        |
| Not employed, not looking for work     | —              | 6 (11)       |
| Retired                                | —              | 14 (26)      |
| Disabled/unable to work                | —              | 2 (4)        |
| Employment affected by caring          | —              | 19 (56)      |
| <b>Length of time as carer (years)</b> | —              | <b>n=56</b>  |
| 0-1                                    | —              | 13 (23)      |
| 1-3                                    | —              | 15 (27)      |
| 3-10                                   | —              | 24 (43)      |
| 10-25                                  | —              | 4 (7)        |
| >25                                    | —              | 0 (0)        |

<sup>a</sup>Data were not collected.



### Carer Burden and Happiness

Mean carer burden measured by the CarerQoL-7D was 64.1 (SD 20.4). Mean carer happiness assessed by the CarerQoL-VAS was 6.3 (SD 2.2; Table 2).

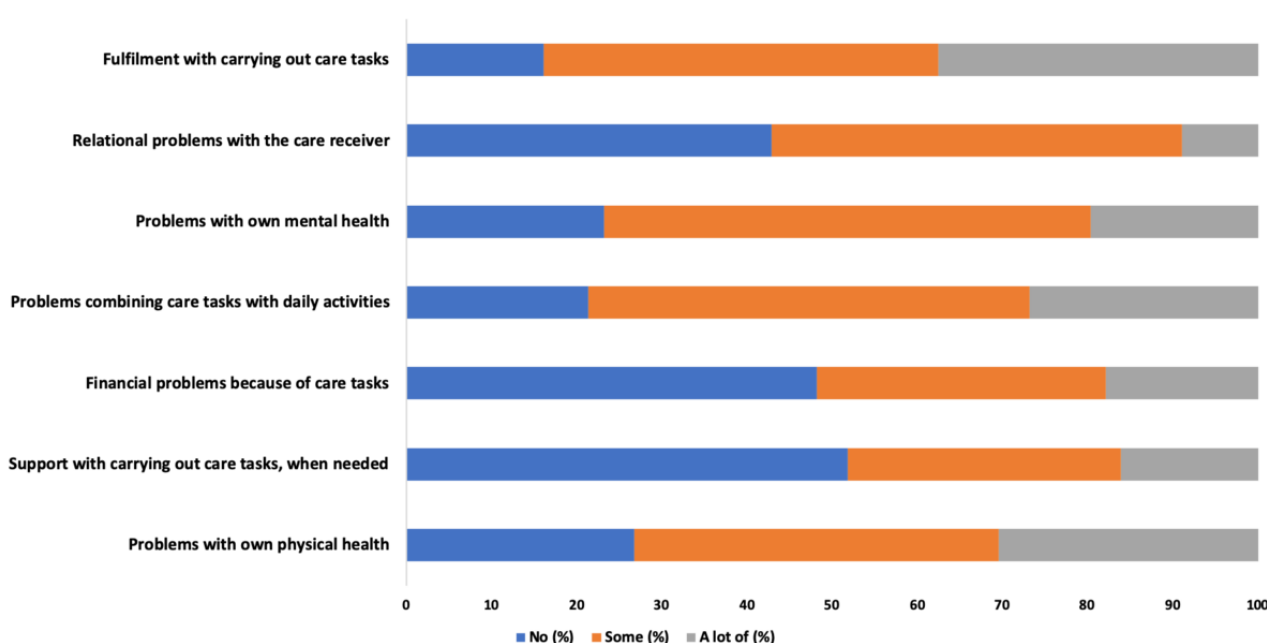
The distribution of responses to individual CarerQoL-7D dimensions, as a percentage of the overall responses to each dimension, is shown in Figure 3. Most DCM carers (47/56, 84%) experienced at least some (some or a lot of) fulfillment

caring. Despite this, most carers experienced at least some mental health problems (43/56, 77%) and at least some physical health problems (41/56, 73%). In addition, around half reported at least some relationship problems with the care receiver (32/56, 57%) and at least some financial problems (28/56, 50%) because of their care responsibilities. Less than half (27/56, 48%) received at least some support with their care tasks, when needed. Most carers (44/56, 79%) experienced problems combining their care tasks with their everyday activities.

**Table 2.** Carer burden and happiness scores (n=56).

| Instrument                                       | Mean (SD)   | 95% CI    | Range    |
|--------------------------------------------------|-------------|-----------|----------|
| Care-Related Quality of Life-7D                  | 64.1 (20.4) | 58.8-69.5 | 14.6-100 |
| Care-Related Quality of Life-Visual Analog Scale | 6.3 (2.2)   | 5.7-6.9   | 1-10     |

**Figure 3.** Distribution of responses to Carer-Related Quality of Life (CarerQoL)-7D dimensions for degenerative cervical myelopathy carers.



### Influence of Carer and Patient Situation

No significant relationship was found between carer burden or happiness and patient disease severity (Nurick score) and pain scores (Multimedia Appendix 1). Moreover, no significant difference in carer burden or happiness was found between different groups for demographic characteristics studied: patient age, gender, education, dependency, carer age, gender, education, employment, and duration of time as a carer (Multimedia Appendix 2).

### United States Versus United Kingdom

Mean DCM severity was similar for UK and US patients with Nurick scores of 2.93 (SD 1.03) and 2.91 (SD 0.94), respectively. Mean carer burden score was 58.2 in the United States compared with 65.6 in the United Kingdom (Table 3). Mean carer happiness score was 5.9 in the United States and 6.4 in the United Kingdom. These differences were not statistically significant.

**Table 3.** Mean carer burden and happiness lower in US carers.

| Country and instrument           | Mean (SD)   | 95% CI    | Range     |
|----------------------------------|-------------|-----------|-----------|
| <b>United Kingdom (n=45)</b>     |             |           |           |
| Care-Related Quality of Life-7D  | 65.6 (19.2) | 60.5-70.6 | 22-100    |
| Care-Related Quality of Life-VAS | 6.4 (2.3)   | 5.8-7.0   | 1-10      |
| <b>United States (n=11)</b>      |             |           |           |
| Care-Related Quality of Life-7D  | 58.2 (24.9) | 51.7-64.7 | 14.6-95.5 |
| Care-Related Quality of Life-VAS | 5.9 (1.9)   | 5.4-6.4   | 3-9       |



## Discussion

### Key Findings

These data demonstrate that carer quality of life is markedly reduced in DCM. Most carers reported mental and physical health difficulties, adverse impacts on employment, and strained relationships with the patient. No significant relationships between carer quality of life and patient disease and pain scores were identified. Moreover, no significant difference in carer quality of life was identified between different patient and carer demographic groups.

### Interpretation

These findings are significant because informal care plays a fundamentally important role in health care. In the United Kingdom, the value of informal care is estimated to be £132 billion per year [17], whereas according to the Institute for Fiscal Studies, in 2015/16 the UK government spent £140.6 billion on health and £29.9 billion on social care.

Increased strain on carers leads to increased carer illness, creating further demands on health care and negative economic consequences. As an age-related degenerative condition, the incidence and prevalence of DCM is expected to rise. In the context of an ageing population and accelerating health care innovations, health care costs are ever increasing. Therefore, supporting the providers of informal care, to prevent carer burnout and preserve carer health, is essential in ensuring sustainable and financially viable health care systems.

### Carer Quality of Life Is Reduced in Degenerative Cervical Myelopathy Compared With Other Chronic Conditions

Considering DCM carer quality of life in context is currently limited by several issues. Comparisons with the general population are limited because the CarerQol instrument questions are specifically targeted at the burden associated with being a carer. In addition, because of the novelty of the CarerQol instrument, few comparable studies currently exist. Unfortunately, the total CarerQol-7D score is often not reported. Nonetheless, several comparisons can be made as follows:

1. A large study of 1244 adult carers in the Netherlands reported a mean CarerQol-7D score of 79.1 and a mean CarerQol-VAS of 7.1 [26]. The mean CarerQol-7D of 64.1 and CarerQol-VAS of 6.3 detected in this study suggests higher carer burden and lower happiness in DCM carers compared with carers in general.
2. Similarly, CarerQol-VAS scores indicated that mean happiness in carers of individuals with DCM was lower than those observed in carers of individuals suffering from other conditions. Reported scores were 7.5 in head and neck cancers [31], 7.4 in autism [29], 7.5 in craniofacial malformations [32], and 7.2 in Pompe disease [13]. Therefore, DCM may pose heavier burden on carers than other reported diseases.
3. Using a similar 0 to 10 scale as in the CarerQol-VAS, the average population happiness has been estimated at 7.1 for the United Kingdom, 7.3 for the United States, 7.6 for the

Netherlands, and 7.5 for Ireland [33]. The reduction of happiness in carers of individuals with DCM is large compared with the average happiness of the study population country (12.5%) and compared with carers of people with Pompe disease (5.3%), craniofacial malformations (2.7%), and autism (1.4%).

Although these comparisons are not without limitation, including lack of formal statistical comparison, carer quality of life scores appear consistently lower in DCM than reported in other diseases and general population estimates, suggesting that quality of life may be particularly affected in DCM carers. Exactly why carers are more affected in DCM remains unclear.

In this study we did not identify any associated factors. This may have been masked by our sample size, as studies involving larger groups (eg, n=200), albeit using alternative quality of life instruments, have found that carer demographics such as age, gender, health status, and duration of caregiving influence carer quality of life [34]. However, the absence of any trends between our investigated factors and carer quality of life may suggest that instead, significant factors were not considered. For example, other studies have shown a link with increasing hours of required care and the patient's quality of life [13].

Recent research indicates that patient quality of life in DCM is lower than many other severe chronic conditions, including diabetes, cancers, and COPD [6]. In addition, DCM patients have a high prevalence of affective disorders [35], which may provide a possible explanation for the high carer burden and warrants further investigation.

### Generalizability

As CarerQol tariffs become available for several other countries [25], more nationalities should be included in future work. The comparison of diverse cultures and health care systems may provide novel insight into factors influencing carer quality of life. For example, it would be interesting to compare carer quality of life across cultures that place differing care expectations upon the family of those who are ill and between countries with more and less developed health and social care systems. This would help elucidate whether it is personal or wider societal factors that are most influential on carer quality of life.

### Limitations and Future Directions

Internet recruitment to patient-specific health surveys can be effective and reach an international audience efficiently and inexpensively, with less missing data compared with postal questionnaires [36,37].

Despite promising and comparable completion rates, the number of carers recruited was lower than patients in a DCM patient study using the same recruitment methods. Given the levels of disability in DCM, having fewer carers than patients is unlikely. It is more likely the internet recruitment methods were more effective for patients than carers, perhaps with patients more likely to identify with their condition (DCM) than carers with their situation (caring for someone with DCM). Given the limited experience of internet recruitment and behavior among

carers, we can only speculate at this time. However, to support further research in this field, this deserves further consideration.

Inclusion of only 56 of the 136 responses received in the final analysis was disappointing. In total, 64% (56/87) of all responses from carers were included. A large proportion of excluded responses were inadvertently completed by patients, which is understandable given that at present most Myelopathy.org website visitors are patients. Nonetheless, finding no substantial differences between complete and incomplete responses makes selection bias unlikely. Moreover, the exclusion of carers from countries for which CarerQoL tariffs were not available will resolve as more tariffs become available. Plans for development of a designated carer section of Myelopathy.org may improve carer participation in future work.

The participation rate of 32% in this study was lower than the typical response rate of 50% to 70% in other CarerQoL studies [29,31,32]. These studies utilized postal questionnaires rather than open electronic questionnaires, as utilized in this study. Although response and participation rates are not identical measures, the participation rate in this study is almost certainly

a substantial underestimate because it was not possible to distinguish between patient and carer website visitors, meaning that a large number of patient visitors were included in the denominator of our participation rate calculation.

Finally, patient and carer factors considered in this study were not found to influence carer quality of life. Although it is possible any associations may have been concealed by the sample size, considering other factors including the number of hours spent caring per week and further assessing the emotional and psychological burden for patient and carer will form important future work [13,35,38]. The identification of such factors is important to target supportive interventions.

## Conclusions

DCM carer quality of life is low. The magnitude of reduction in DCM carer quality of life appears greater than reductions in carer quality of life in other conditions studied. In this study, no single patient or carer factors were associated with carer quality of life. Identification of influencing factors is important to better understand the basis for impaired quality of life and to target support, which should form the basis of future work.

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## Acknowledgments

This report is independent research arising from a Clinician Scientist Award, CS-2015-15-023, supported by the National Institute for Health Research (NIHR). The views expressed in this publication are those of the authors and not necessarily those of the National Health Service, the NIHR, or the Department of Health and Social Care.

Research in the MRK's laboratory is supported by a core support grant from the Wellcome Trust and Medical Research Council to the Wellcome Trust-Medical Research Council Cambridge Stem Cell Institute. MRK is supported by an NIHR Clinician Scientist Award.

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## Conflicts of Interest

None declared.

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### Multimedia Appendix 1

Kendall tau-b correlations between carer burden and happiness and patient disease severity and pain scores.

[PDF File (Adobe PDF File), 156 KB - [ijmr\\_v8i4e12381\\_app1.pdf](#) ]

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### Multimedia Appendix 2

No significant difference in carer happiness was identified between the different groups of 9 demographic characteristics using 1-way analysis of variances.

[PDF File (Adobe PDF File), 86 KB - [ijmr\\_v8i4e12381\\_app2.pdf](#) ]

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## Abbreviations

**ANOVA:** analysis of variance  
**CarerQoL:** Care-Related Quality of Life  
**COPD:** chronic obstructive pulmonary disease  
**DCM:** degenerative cervical myelopathy  
**NIHR:** National Institute for Health Research  
**VAS:** Visual Analog Scale

*Edited by G Eysenbach; submitted 01.10.18; peer-reviewed by A Nouri, A Carr, B Davis; comments to author 21.03.19; revised version received 13.06.19; accepted 19.07.19; published 07.11.19.*

*Please cite as:*

Mowforth OD, Davies BM, Kotter MR

*Quality of Life Among Informal Caregivers of Patients With Degenerative Cervical Myelopathy: Cross-Sectional Questionnaire Study*  
*Interact J Med Res* 2019;8(4):e12381

URL: <http://www.i-jmr.org/2019/4/e12381/>

doi: [10.2196/12381](https://doi.org/10.2196/12381)

PMID: [31697240](https://pubmed.ncbi.nlm.nih.gov/31697240/)

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Original Paper

# Racial Disparities in Mortality Among American Film Celebrities: A Wikipedia-Based Retrospective Cohort Study

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## Abstract

**Background:** In the United States, well-documented racial disparities in health outcomes are frequently attributed to racial bias and socioeconomic inequalities. However, it remains unknown whether racial disparities in mortality persist among those with higher socioeconomic status (SES) and occupational prestige.

**Objective:** As the celebrity population is generally characterized by high levels of SES and occupational prestige, this study aimed to examine survival differences between black and white film celebrities.

**Methods:** Using a Web-based, open-source encyclopedia (ie, Wikipedia), data for 5829 entries of randomly selected American film actors and actresses born between 1900 and 2000 were extracted. A Kaplan-Meier survival curve was conducted using 4356 entries to compare the difference in survival by race. A Cox semiparametric regression analysis examined whether adjusting for year of birth, gender, and cause of death influenced differences in survival by race.

**Results:** Most celebrities were non-Hispanic white (3847/4352, 88.4%), male (3565/4352, 81.9%), and born in the United States (4187/4352, 96.2%). Mean age at death for black celebrities (64.1; 95% CI 60.6-67.5 years) was 6.4 years shorter than that for white celebrities (70.5; 95% CI 69.6-71.4 years;  $P < .001$ ). Black celebrities had a faster all-cause mortality rate using Kaplan-Meier survival function estimates and a log-rank test. However, in a Cox semiparametric regression, there was no longer a significant difference in survival times between black and white celebrities (hazard ratio 1.07; 95% CI 0.87-1.31).

**Conclusions:** There is some evidence that racial disparities in all-cause mortality may persist at higher levels of SES, but this association was no longer significant in adjusted analyses. Further research is needed to examine if racial disparities in mortality are diminished at higher levels of SES among more representative populations.

(*Interact J Med Res* 2019;8(4):e13871) doi:[10.2196/13871](https://doi.org/10.2196/13871)

**KEYWORDS**

continental population groups; internet; mortality; race

## Introduction

The Heckler Report, published in 1985, was the first to report statistical differences in health and mortality among races in the United States after the Civil Rights Act in 1964 [1]. Over 30 years later, there have been significant improvements to minority lifespan and health, but notable variations in mortality

rate, cause of death, and prevalence of disease have persisted [2-4]. Blacks (national born and immigrants) make up a rising 13% of the US population [5]; yet, they experience the highest mortality rates for 8 of the 10 most common causes of death in the United States. In addition, they have maintained the highest mortality rate for 8 of the 10 most common causes of death as



well as a 6-year and 4-year shorter life span than that of white American males and females, respectively [4].

Socioeconomic status (SES), as defined by income, education, occupation, and social status, is a product of policy and culture often studied in context with its effect on health [3,6-8]. Unlike fixed demographic variables such as gender, race, and birth date, SES measures such as education, geographic location, marital status, financial hardship, and social status can fluctuate throughout a lifetime and generally represent a dynamic measure of social influence and affluence in society [9,10]. Economic census data throughout the past century have established significant economic and educational disparities between blacks and whites in the United States. A study of US Panel Income Dynamics over 1997-2007 found that only 12.6% of those identifying as black had completed a college education compared with 32.7% of white counterparts, whereas 22.3% of blacks and 9.3% of whites had not attained a high school diploma [11]. Wealth among non-Hispanic, American whites has been found to be orders of magnitude greater than black Americans since the 1970s [12] and highly correlated to health and mortality outcomes [13-15]. Therefore, disparities in health outcomes and mortality are generally thought to be due to historical and enduring structural economic inequities such as redlining of predominantly black neighborhoods by lenders [3,7,10,11]. Recent studies quantifying racial health and mortality disparities that controlled for SES have found that differences among races are reduced but are still present [15-17]. However, there is a growing body of literature that measures not only income but also wealth and associated variables such as social standing.

Social status and wealth are understudied aspects of SES, especially in the American context. Wealth has been defined as the total sum of income, assets, and debts to quantify personal net worth. Hajat et al found a significant inverse relationship between wealth and mortality [18], which is also consistent with a previous systematic review [9]. Another study found that wealth, rather than income alone, was a more predictive measure of SES differences in international health care utilization [19]. In contrast, social status entails combinations of SES variables and public opinion of occupational groups. These measurements of occupational and social prestige use tools such as Duncan SES index and Siegel Prestige Score for common occupational groups in a population [20]. The combination of these scales indicates a higher risk of death for those in lower social positions, consistent with previous studies, but still did not examine moderation by race and gender within each level of social class as well as occupation [21]. Although these measures address wealth and occupational prestige, both associated with cultural notability or societal influence, they do not account for individuals known as celebrities and the racial disparities that may exist within these population subsets.

Celebrities are a subset of the population that have occupations associated with higher influence, notability, and wealth. Among celebrities, actors and actresses usually maintain societal standing and wealth because of the ongoing opportunities extended once fame is achieved. Owing to their top-tier social standing and relative affluence to the general population, the high societal influence of actors and actresses is hypothesized

to be associated with lower mortality rates and relatively good health. Redelmeier and Singh studied trends in mortality for Oscar-nominated actors and actresses and found that the primary causes of death followed the top-reported causes of death for the general population, with an average lifespan higher than that for the general population (76-79 years) [22]. Racial diversity in American film actors has also grown steadily throughout the years, with Smith et al reporting nearly 13% black actors and 73% white actors in 2014, which closely reflects the US census demographics for the decade [23].

To date, no research has been published that observes racial disparities in mortality among American celebrities. This study sampled Wikipedia to examine racial disparities in a retrospective cohort of actors and actresses born in the 20th century. Choosing actors and actresses born in this time frame includes those that started acting at all stages of life and allows for both an ample sample size and sufficient follow-up time. On the basis of what has been found in previous studies, it is hypothesized that black American film actors and actresses experienced faster mortality rates than their white American counterparts.

## Methods

### Wikipedia Data Source and Tools

To operationalize societal standing and notability as a celebrity, Wikipedia's list of American film actors and actresses was used as a sampling frame. Wikipedia is a nonprofit database that is maintained internationally by the public and moderated by a large group of Wikipedia editors. These editors use a set of criteria for notability, ensuring that data entered are verifiable and each subject page has enough reference and societal influence for inclusion in the online encyclopedia. If entries are under review or do not meet all criteria, they are clearly marked with a banner at the top of the page [24]. Notability is considered for every Wikipedia page and is determined by the quality and quantity of third-party sources rather than the page or article content. This editor gatekeeping method creates a unique, reliable benchmark for determining occupational prestige for actors and actresses. Wikipedia has been verified as a quality informational source by many scholars, with the most notable being *The Wisdom of Crowds* [25], which found Wikipedia to be comparable with the Encyclopedia Britannica. Others such as Vrandečić and Krötzsch have verified the reliability and value of Wikipedia as a collectively maintained database, with the diversity of authors, the number of contributors, and the ease of access continuing to improve over time, providing a sound sampling frame [26,27].

Microsoft Excel was used for data entry, observation randomization, and creation of the final dataset file. SAS 9.4 (SAS Institute 2013) was used for descriptive statistics and both survival curve estimates using LIFETEST and PHREG procedures for the Kaplan-Meier curve and Cox semiparametric logistic regression curve, respectively.

### Creating a Dataset

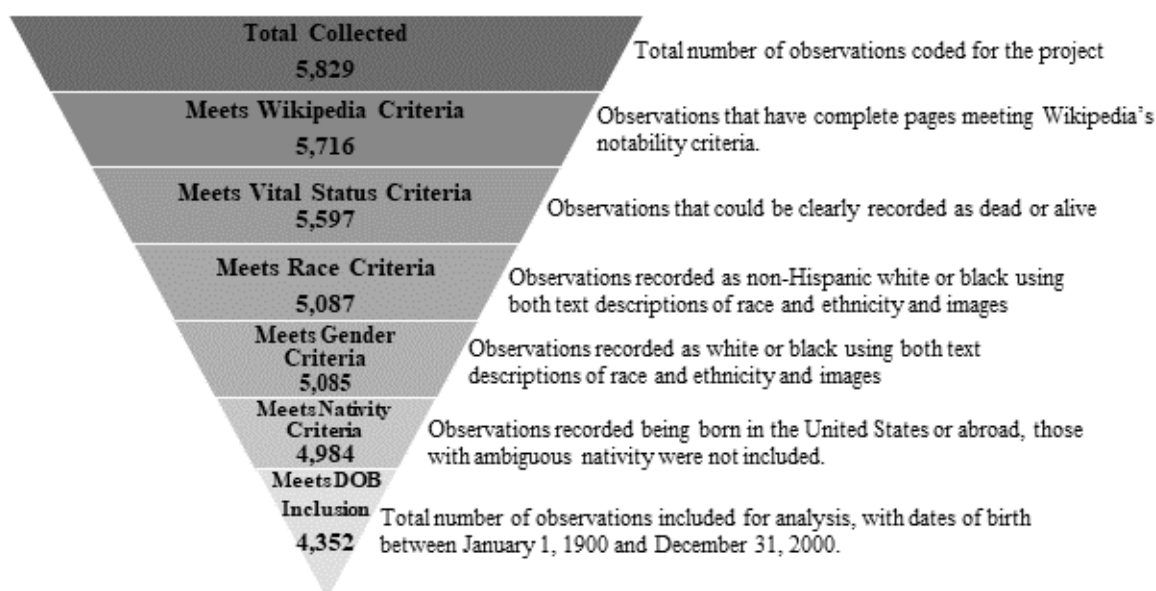
A dataset of URLs for US film actors and actresses was created by importing the HTML page links from the Wikipedia category

pages for American film actors and actresses. These data are open source under the Creative Commons Attribution-Share-Alike License. On May 31, 2017, 12,164 entries with actor names and the Wikipedia page URL were imported from Wikipedia, formatted into a comma-separated values (CSV) dataset using a general text editor program, and imported into Microsoft Excel 2016 for randomization and data entry. The randomized data were then split among 3 people for data entry for each observation, and once there were at least 5500 entries, the Excel datasets were merged into 1 CSV file and imported into SAS for data cleaning and analysis. To check accuracy and inter-rater reliability, 200 random entries from each of the 3 coding individuals were checked for accuracy, particularly for consistent coding of race.

For each film celebrity entry, a total of 6 variables were collected: gender, race, age, US nativity (yes or no), mortality status (dead, alive, or unknown), and cause of death. Wikipedia notability was confirmed by the absence of a banner indicating questionable reliability of information to confirm notability, and only those with confirmed notability were included in the

final dataset. Race was recorded using the Center for Disease Control and Prevention's definition for race based on perception of skin tone and relevant content contained in the Wikipedia entry. Collected categories were white, black, and other. Causes of death were grouped into the following categories: cardiovascular disease (CVD), suicide, accident, drug overdose, cancer, cause unknown, cause missing, and alive (no cause of death). Categories were chosen from the most common causes of death, with the addition of drug overdose and suicide because they were included in other studies focusing on similar musician populations [28,29]. CVD, suicide, and drug overdose were coded as defined by the World Health Organization, and other or unknown cause was coded as *other* [30]. The final inclusion criteria included actors that had notable Wikipedia status, were coded as either non-Hispanic white or black, had known birth and death dates (if applicable), and were born within the 20th century (January 1, 1900, to December 31, 2000). Death data were recorded with reference to May 27, 2017. In total, 5829 observations were made and 4352 (74.66%) met all inclusion criteria for study analysis (Figure 1).

**Figure 1.** Participant selection based on inclusion criteria. DOB: date of birth.



## Statistical Analyses

Microsoft Excel was used for data entry, observation randomization, and creation of the final dataset file. SAS 9.4 (SAS Institute 2013) was used for descriptive statistics and both survival curve estimates using LIFETEST and PHREG procedures for the Kaplan-Meier curve and Cox semiparametric logistic regression curve, respectively. Following data collection, descriptive statistics were run to characterize the distributions and bivariate associations among variables. A student *t* test was used to compare the average age at death by race. Normality and proportionality tests were run for age at death by race and gender to select the most appropriate survival analysis methods. In this analysis, survival curves use death as an event, and the specified period is the lifespan of the celebrity. The Kaplan-Meier survival curve was created through the SAS LIFETEST procedure to plot survival for each race from date

of birth to date of death, and the log-rank test was used to test statistically significant differences between each curve by race ( $P < .05$ ). A Cox semiparametric logistic regression model was used via the SAS PHREG procedure to compare survival rates while adjusting for year of birth, gender, and cause of death. A model was built via stepwise regression to see the effect of each covariate (including race) on survival outcomes. Only covariates showing significant effect on survival rate were retained in the model, and the final regression model was then used to compare races and obtain an adjusted hazard ratio (aHR) for the all-cause mortality rate among black celebrities, with white male celebrities as the reference group.

## Results

Of the 4352 observations in the analytic sample, 1335 (30.88%) actors died during the follow-up period. Of 505 black actors

observed, 405 (80.2%) were alive at the end of the follow-up period, and of 3847 white actors, 3017 (78.42%) were alive at the end of follow-up period. As shown in Table 1, most of the sample (3565/4352, 81.92%) was male, and 4187/4352 (96.21%) had US nativity. Most of the causes of death (621/1335, 46.52%) observed were recorded as dead with unknown cause, with CVD and cancer as the top 2 causes of death among our sample claiming 339/1335 (25.39%) and 251/1335 (18.80%) of those dead, respectively.

Using the Kaplan-Meier function to compare survival rates between each race with celebrities' genders combined (Figure 2), there was a significant difference between white and black actors when using a log-rank test for equality for each race group ( $P<.001$ ). This was consistent with a two-sample student *t* test comparing age at death for white (70.5; 95% CI 69.6-71.4) and

black (64.1; 95% CI 69.6-67.5) actors and actresses, with a significant difference in mean age at death ( $P<.001$ ; Figure 3). On average, black actors and actresses died 6.4 years faster than their white counterparts.

Cox semiparametric logistic regression model was created via stepwise regression, with the significance threshold for inclusion set at  $P<.05$ . Date of birth was included as a continuous covariate with an expected hazard ratio (HR) of 1.00. The completed logistic regression model included gender ( $P=.001$ ), cause of death ( $P<.001$ ), and date of birth ( $P<.001$ ) as predictors of age at death. Race was not a significant predictor of age at death using stepwise regression ( $P=.13$ ), and when observing HRs for black actors with reference to their white counterparts, it was not found to be statistically significant (aHR 1.07; 95% CI 0.87-1.31).

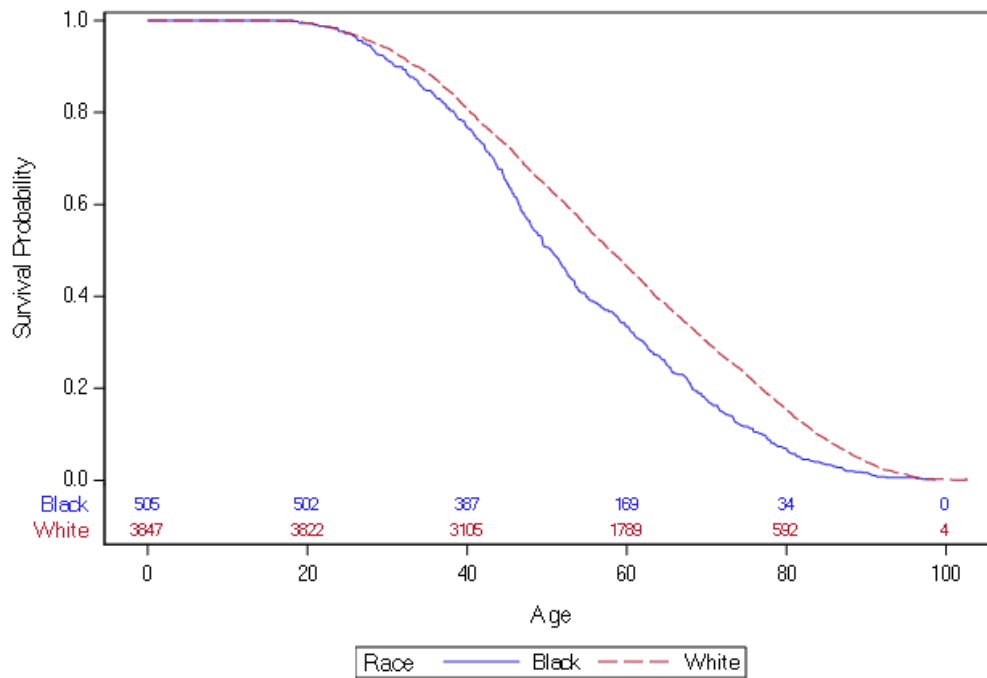
**Table 1.** Results for analysis sample demographics and adjusted hazard ratios.

| Study covariate        | Total (N=4352), n (%) | White race (n=3847), n (%) | Black race (n=505), n (%) | <i>P</i> value | Adjusted hazard ratio <sup>a</sup> (95% CI) |
|------------------------|-----------------------|----------------------------|---------------------------|----------------|---------------------------------------------|
| <b>Gender</b>          |                       |                            |                           | <.01           | 1.32 (1.120-1.56)                           |
| Male                   | 3565 (81.92)          | 3139 (81.60)               | 426 (84.4)                |                |                                             |
| Female                 | 787 (18.08)           | 708 (18.40)                | 79 (15.6)                 |                |                                             |
| <b>Nativity</b>        |                       |                            |                           | .21            | 1.17 (0.91-1.50)                            |
| American               | 4187 (96.21)          | 3702 (96.23)               | 485 (96.0)                |                |                                             |
| Other                  | 165 (3.79)            | 145 (3.77)                 | 20 (4.0)                  |                |                                             |
| <b>Vital status</b>    |                       |                            |                           | — <sup>b</sup> | —                                           |
| Dead                   | 1335 (30.68)          | 1235 (32.10)               | 100 (19.8)                |                |                                             |
| Alive                  | 3017 (69.32)          | 2612 (67.90)               | 405 (80.2)                |                |                                             |
| <b>Cause of death</b>  |                       |                            |                           | <.01           | 1.00 (1.00-1.01)                            |
| Cardiovascular disease | 339 (7.79)            | 304 (7.90)                 | 35 (6.9)                  |                |                                             |
| Cancer                 | 251 (5.77)            | 227 (5.90)                 | 24 (4.8)                  |                |                                             |
| Accident               | 57 (1.31)             | 54 (1.40)                  | 3 (0.0)                   |                |                                             |
| Suicide                | 34 (0.78)             | 33 (0.86)                  | 1 (0.2)                   |                |                                             |
| Homicide               | 15 (0.34)             | 12 (0.31)                  | 3 (0.6)                   |                |                                             |
| Drug overdose          | 18 (0.41)             | 16 (0.42)                  | 2 (0.4)                   |                |                                             |
| Cause unknown          | 621 (14.27)           | 589 (15.31)                | 32 (6.3)                  |                |                                             |
| Alive                  | 3017 (69.32)          | 2612 (67.90)               | 405 (80.2)                |                |                                             |
| Date of birth          |                       |                            |                           | <.01           | 1.00 (1.00-1.00)                            |

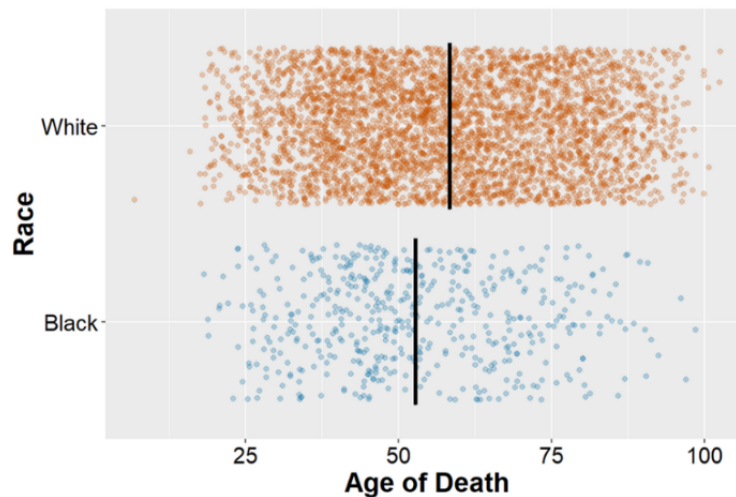
<sup>a</sup>White males were used as the reference group.

<sup>b</sup>Not applicable.

**Figure 2.** Survival probability over time by race (Kaplan-Meier plot).



**Figure 3.** Average age of death by race.



## Discussion

Findings from this Wikipedia study indicated that black film celebrities died 6.4 years faster than their white counterparts. Although hastened mortality among black film celebrities was observed, this association did not persist after adjusting for gender, cause of death, and date of birth. These results provide some indication that racial disparities in mortality may persist even at the highest levels of SES, and this association was no longer significant in adjusted analyses.

The discrepancies in the results between different survival comparison methods (Kaplan-Meier and Cox semiparametric logistic regression model) reflect important social changes in the representation of black actors and actresses in the American film industry during the 20th century. Black actors and actresses have historically experienced racism and other structural barriers to full participation in the American film industry, such that

increased representation began only during the 1960s [31,32]. The proportion of black film celebrities in this sample was approximately 13%, which is consistent with the proportion of black Americans recorded in US census data from 2010 [4]. Nearly all black actors and actresses included in the sample were born during the second half of the 20th century. Therefore, it is likely that there has not been sufficient time to adequately assess racial disparities in mortality after adjusting for date of birth. Future research should continue to examine racial disparities in mortality among American film celebrities, and further research is also needed to elucidate if SES modifies racial disparities in mortality for black Americans.

One noteworthy finding was that only 787 (18.1%) of the random sample of 4352 Wikipedia entries were women. This may reflect the fact that many women began entering workforce in larger numbers during the last 30 years of the 20th century. However, this may also be attributable to enduring concerns related to gender bias in the American film industry [33,34].



The primary objective of this study was to examine racial disparities in a population of film celebrities with high SES. However, these stark differences in the proportional representation of women underscore the questionable generalizability of this unique sample to the broader population of the United States. Our findings clearly demonstrate that white actors appear to be the overwhelming majority in the 20th century American film industry.

Although this study underscores the potential benefits of leveraging open access sources such as Wikipedia, findings should be interpreted in the context of several important limitations. Owing to key social changes in the representation of black actors and actresses in the American film industry, it is likely that there has not been a sufficient amount of time to examine racial disparities when adjusting for date of birth. American film actors and actresses also represent a subset of celebrities from a broader population containing distinct occupational groups such as musicians and athletes. In this study, these groups were not included for several reasons. For example, inclusion of athletes introduces a fitness bias. There

were also important limitations to the retrospective cohort design using Wikipedia data. Wikipedia itself has been found to have a deficit of female editors, fewer notable female Wikipedia page entries than notable male Wikipedia page entries, and biased language on female Wikipedia pages [35-37]. Most notably, some data that may have served as key confounders, such as relationship status, education level, childhood SES, and geographic residence, were not extracted [9,11,13]. Future studies with primary data collection would provide more nuanced information regarding potential confounders and effect modifiers of racial disparities in all-cause mortality.

Despite these limitations, this study provides some of the first estimates of racial disparities in mortality among American film celebrities. Results from this study have observed a 6.4-year higher mortality rate among black film celebrities, which underscores that racial disparities may persist in the United States even at the highest levels of SES. Findings support the need for further research to examine the social and psychological mechanisms that could explain the profound racial disparities in mortality experienced by black Americans.

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## Acknowledgments

The authors would like to acknowledge the Wikimedia Foundation as well as the community of Wikipedia editors for their efforts toward publishing and maintaining online databases to support unique projects such as this study. The authors would also like to acknowledge the efforts of the University of Miami's Public Health 651 Research Methods class for their thoughts and efforts toward the research concept and methods.

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## Conflicts of Interest

None declared.

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## Abbreviations

**aHR:** adjusted hazard ratio

**CSV:** comma-separated values

**CVD:** cardiovascular disease

**HR:** hazard ratio

**SES:** socioeconomic status

*Edited by G Eysenbach; submitted 01.03.19; peer-reviewed by A Menking, L Rasberry; comments to author 03.08.19; revised version received 17.08.19; accepted 31.08.19; published 10.12.19.*

*Please cite as:*

*Speaks H, Falise A, Grosgebauer K, Duncan D, Carrico A*

*Racial Disparities in Mortality Among American Film Celebrities: A Wikipedia-Based Retrospective Cohort Study*

*Interact J Med Res 2019;8(4):e13871*

*URL: <http://www.i-jmr.org/2019/4/e13871/>*

*doi: [10.2196/13871](https://doi.org/10.2196/13871)*

*PMID: [31821148](https://pubmed.ncbi.nlm.nih.gov/31821148/)*

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Original Paper

# Quality and Accuracy of Information Available on Websites for Distracted Driving: Qualitative Analysis

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## Abstract

**Background:** Distracted driving has become alarmingly widespread, and its prevalence continues to increase despite efforts by government and nongovernment organizations to educate the public about this pervasive problem. Every year, 1.35 million people die, and nearly 80 million people get injured in road traffic incidents. Motor vehicle crashes are the leading cause of death among young people, and distracted driving plays a huge role in road traffic fatalities and injuries. Considering that most people now use the internet as an information source and Google is the most visited website and number one online search engine in the world, we performed a qualitative analysis of information available through Google on distracted driving and its outcomes.

**Objective:** The goal of this study was to analyze the quality and accuracy of the information on distracted driving and its consequences available to the general public when using Google as a search engine for distracted driving.

**Methods:** In November 2018, a nonregional Google search on distracted driving was conducted. The first two pages of the Google search results were selected for analysis. Data were collected on the type of website, type of distraction, consequences of distracted driving described, presence and referencing of statistics, and orthopedic and nonorthopedic injuries described, with their acute and chronic sequelae.

**Results:** In total, we analyzed 25 websites: 12 websites (48%) were from government bodies, which were the most common type of websites; 19 (76%) of the sites provided statistics; and 15 (60%) referenced the source of the statistic. Mobile phones were the most frequently cited type of distraction, with 17 (68%) sites discussing it, while death was the most commonly mentioned consequence of distracted driving, quoted in 15 (60%) of the websites. Additionally, 52% of the sites provided tips on how to avoid distracted driving. Only one website mentioned orthopedic injuries.

**Conclusions:** The prevalence of distracted driving is increasing, and so are the consequences associated with it. Nevertheless, the information available online does not accurately describe the current circumstances regarding this issue. The National Highway Traffic Safety Administration attributed 391,000 injuries and 3477 deaths to distracted driving in 2015, which are 5000 more injuries and almost 150 more fatalities compared to 2011. However, despite these figures, most of the websites discussed death as a consequence of distracted driving and often overlooked injuries, even though injuries are over 100 times more likely to occur in distraction-affected crashes. The websites also largely fail to address other forms of driving distractions, like daydreaming or talking to a passenger, and mostly focus on mobile phone-related activities as distractions. More specific information on the dangers of distracted driving and nonlethal trauma may support an overall cultural shift to curb this behavior.

*(Interact J Med Res 2019;8(4):e16154)* doi:[10.2196/16154](https://doi.org/10.2196/16154)

**KEYWORDS**

distracted driving; driver distraction; driving while distracted; inattentive driving; Google; car distractions; texting and driving; cell phones

## Introduction

Distracted driving is not only dangerous but also very common and has become an epidemic in North America. In the United States, 37,461 people were killed in motor vehicle crashes (MVCs) in 2016. Of these, 3450 (9%) deaths were caused by distracted driving [1]. Additionally, in the previous year, nearly 2.5 million people were injured in MVCs, and approximately 16% of those injuries were a result of distractions [1]. The gravity and pervasiveness of the growing threat of distracted driving have prompted the World Health Organization to identify it as one of the priorities in global road safety. [2]

Public opinion research suggests that distracted driving was already considered a problem in 2011. A survey conducted in Iowa that year showed that 98% of respondents believed distracted driving to be a serious or somewhat serious threat to traffic safety [3]. However, despite this opinion, distracted driving remains prevalent [4]. A Canadian survey completed in 2018, found that 69% of the participants think the most distracting activity while driving is using a mobile phone, and yet, according to the same study, 51% of the respondents still communicate on their mobile phones at least once a week while driving [5]. Data from the National Collision Database, which show a continuous increase in injuries and deaths due to distracted driving, further support the fact that drivers continue to engage in this unsafe practice despite the evident risks [6].

Activities such as talking with a passenger, daydreaming, eating or drinking, programming the navigation systems, and using mobile phones while driving, which cause the driver to divert their attention away from safely operating the vehicle, are all forms of distracted driving [7]. Mobile phones, in particular, have been found to have a greatly negative effect on driving performance [8], which places the driver at significant risk for a MVC. In fact, the odds of crashing when making a call on a handheld device increases by 12 times [9], and the crash risk for texting while driving is almost double that (23 times) [10].

Distracted driving behavior may be due to a lack of appropriate education on distracted driving. Many people may inherently comprehend that distracted driving is a threat to public safety, but they may lack specific information that could reinforce safer behavior. The majority of people today turn to the internet as their primary research/education tool when wanting to gain new knowledge on a topic such as distracted driving. Google is both the most commonly used data search engine and the most visited website in the United States and globally [11]. Of all searches performed on a desktop or laptop, 76% are through Google, and the figures are even higher for mobile phones, where more than 85% of the searches go through this engine [12]. Google processes more than 63,000 search requests per second, which equates to more than 5.4 billion queries a day [13]. Therefore,

it is reasonable to assume that the preferred online research method for the majority of people will be through this platform.

Although multiple websites provide information on distracted driving to the general public, little is known about what information these websites offer regarding the specific causes and outcomes of distracted driving. Using modern internet-based methods in research to assess the available information on distracted driving could bring us closer to understanding why this risky behavior is so prevalent. The primary goal of this study is to qualitatively analyze the information available to the general public on websites for distracted driving and assess the quality and accuracy of that information. The secondary goal is to specifically examine whether information about potential injuries is conveyed.

## Methods

Using a nonregional Google search setting, a search on the phrase “distracted driving” was conducted. The search was performed in November 2018, and the first two pages of the Google search results were selected for analysis. We collected data on the type of the website, consequences of distracted driving, type of distraction, presence of statistics, and referencing of statistics as well as orthopedic and nonorthopedic injuries together with their acute and chronic manifestations. We categorized the websites as government, nonprofit, private company, and sites belonging to a foundation. Distracted driving consequences were described as death/fatality, injury, and legal repercussions. Driving distractions presented in the websites were assigned as distractions involving mobile phones, hands-free mobile phones, infotainment, and others (passengers, food/drink, reaching, and outer-vehicle distractions).

## Results

In total, we analyzed 25 websites. The most common search results were government body websites, which accounted for 12 websites (48%). Additionally, seven (28%) websites were run by nonprofit organizations, followed by five (20%) that were run by private company websites, and one (4%) that was a foundation-owned webpage (Table 1). Government websites included multiple state departments of transportation as well as US federal agencies such as the National Highway Transportation Safety Administration (NHTSA) and the Federal Communications Commission. Nonprofit organizations included the American Automobile Association, National Safety Council, the Governors’ Highway Safety Association, and a pediatric hospital-associated program that helps parents monitor their children’s whereabouts using mobile device tracking (TeenSafe). One website (Enddd.org) is the project of a memorial foundation established in the name of a distracted driving victim.

**Table 1.** Types of websites for distracted driving.

| Types of websites | Value, n (%) |
|-------------------|--------------|
| Government        | 12 (48)      |
| Nonprofit         | 7 (28)       |
| Private company   | 5 (20)       |
| Foundation        | 1 (4)        |

Most websites described multiple types of distractions ([Table 2](#)). The most common distraction cited in 17 sites was mobile phone use (68%), followed by vehicle entertainment and information systems discussed in eight of the sites (32%). Seven (28%) of the reviewed webpages mentioned food, drink, other

passengers, and outer-vehicle distractors, and three of the websites (12%) distinguished hands-free telecommunications as a significant distraction. Finally, seven websites (28%) did not identify any specific distractions.

**Table 2.** Distractions described in websites for distracted driving. Many websites identified multiple distraction types.

| Distractions                                                                     | Websites describing distraction, n (%) |
|----------------------------------------------------------------------------------|----------------------------------------|
| Mobile phones (includes texting)                                                 | 17 (68)                                |
| Mobile phones (distinguishes hands-free)                                         | 3 (12)                                 |
| Infotainment system                                                              | 8 (32)                                 |
| Other distractions (passengers, food, drink, reaching for object, outer vehicle) | 7 (28)                                 |
| Broad statement/nonspecified                                                     | 7 (28)                                 |

Of the websites analyzed, 19 (76%) included distracted driving statistics and 15 (60%) also provided references for those statistics. The most commonly referenced data were those published by the NHTSA. The Traffic Safety Facts report published by NHTSA found that 3450 deaths and over 391,000 injuries were attributable to distracted driving in 2016.

In terms of the consequences of distracted driving, most websites listed multiple outcomes ([Table 3](#)). The most frequently identified consequence was death, cited in 15 (60%) of the sites. Although “injury” (or “injuries”) was listed in 11 (44%) websites, only one (4%) of these alluded to specific bodily systems affected by the injury. Legal repercussions due to distracted driving including fine, demerit points, indictment, and incarceration were listed in 6 (24%) websites.

**Table 3.** Consequences described in websites for distracted driving. Many websites identified multiple consequences.

| Consequences                                 | Websites Describing Consequence n (%) |
|----------------------------------------------|---------------------------------------|
| Death/mortality/fatality                     | 15 (60)                               |
| Legal repercussions                          | 6 (24)                                |
| Injury                                       | 11 (44)                               |
| Injury specified (orthopedic, nonorthopedic) | 1 (4)                                 |

## Discussion

### Principal Results

This study qualitatively analyzed 25 websites on distracted driving using Google as a search engine. The findings of this analysis demonstrate notable differences between the way distracted driving is portrayed on the internet versus reality. For example, the results of this study show that death was the most common consequence of distracted driving presented on the websites. In addition, 15 (60%) websites cited a fatality as an outcome, compared to injuries that were reported on 11 (44%) of the sites. Depending on the source, current data available on road traffic accidents resulting from distracted driving proves that injuries outnumber death as a consequence, with a ratio of 60-113 to 1 [[14](#),[15](#)].

The study also demonstrated discrepancies between the online and actual representations of various driving distractions. Mobile phones and texting, discussed in 17 (68%) of the sites, were shown to be the most common distracting activities. However, a report by Erie Insurance completed using data from the Fatality Analysis Reporting System lists daydreaming as the number one distraction involved in 61% of fatal MVCs and ranked mobile phones second at 14% [[16](#)]. Another report from the NHTSA indicates that talking with a passenger is the most common distracting activity, responsible for 57% of distracted driving collisions, while combined phone use accounted for 11% [[17](#)].

The fact that texting is the most common type of distraction described is not surprising. Gallup [[18](#)] reported that texting is now the preferred form of communication among Americans under the age of 50 years and that the prevalence of texting is higher among younger people in the United States. The Centers

for Disease Control and Prevention found that drivers aged 16-19 years are at highest risk of being involved in MVCs [19], and the NHTSA reported that the same age group also had the highest percentage of distracted drivers involved in fatal MVCs [1]. This could be the basis of why websites for distracted driving have focused on mobile phone use so heavily.

As expected, government agencies publish the majority of information available to the public concerning distracted driving. This finding is encouraging, as it suggests that government authorities are actively working to address this problem.

Although many websites provided statistics and consequences and 11 websites mentioned generic injuries, it is worrisome that only one website cited orthopedic injuries resulting from distracted driving. The Canadian National Trauma Registry reports that 79% of the major injuries in hospitalized patients are musculoskeletal in nature and that car crashes are the number one cause of major injuries [20]. Considering that at least 16% of all injuries sustained in an MVC are distraction related [14], it is logical to assume that orthopedic injuries would make up the majority of these injuries. Unfortunately, this outcome of distracted driving is massively ignored by these websites.

Many of the reviewed websites offered guidance on how to prevent distracted driving. However, all of these invoked driver self-discipline and other active mechanisms to enforce behavior modification (eg, "Just Drive"). Likely, decreasing distracted driving will be a combination of education, cultural shift, and passive restraints, for example, mobile phone apps restricting phone use while driving [21], collision avoidance, and self-driving technology in vehicles.

### Limitations

The main limitation of this study is that little is publicized about the algorithm for the google.com/ncr search; thus, it is unknown exactly how website hits are generated on Google. However, by using a nonregional Google search, we attempted to collect as many websites as possible without the bias of regional preferences. Therefore, it is likely that our search results represent what the average information seeker would find without any filters set. Additionally, the primary strength of this study is that it examines a contemporary public safety issue using the most popular search engine.

### Comparison With Prior Work

According to the World Health Organization, almost 1.35 million people die in MVCs every year, and for every one of those traffic fatalities, 60 people get injured [15,22]. Distracted driving is a considerable contributor to these figures and continues to be a common practice among the drivers despite attempts by many organizations to control this pervasive behavior through education. It seems to be especially widespread among the younger drivers [23-25], as it has become the leading cause of death for teenagers [26].

Many North American jurisdictions passed antidistracted driving laws to curb this problem. Unfortunately, despite introducing

new legislation, distracted driving remains rampant today. The Alberta Government in Canada released data that show no decrease in distracted driving convictions in the 4 years since the distracted driving legislation was passed (n=25,958 in 2012 and n=27,281 in 2016) [27]. Moreover, in Ontario, apart from 2012, distracted driving has been the leading cause of MVC fatalities since 2009, when legislation prohibiting the use of handheld devices was first introduced [28]. The NHTSA also continues to report an alarming number of collisions due to distracted driving, with no signs of this problem ending: 885,000 crashes in 2015 compared with 826,000 in 2011 [14].

Google is the most visited website globally [11], and with 92.37% of the market share and more than 5.6 billion queries a day, it is the most popular search engine in the world [13,29]. It is available in 149 different languages [30] and operates in more than 200 countries, making it a particularly useful tool when researching topics of interest online. Studies have examined Google's increasing involvement in the search for medical information online and have found that despite some deficiencies, it can be an effective medium [31]. Research looking into the information on various medical issues available on the internet using Google as a search engine includes subjects such as views on vaccination [32], psychoactive agents [33], fractures [34], and skin cancer [35].

Although distracted driving has become increasingly common, and interest on this subject is high, there is limited research on the information available online regarding this issue. To our knowledge, so far, there have not been any publications concerning information on the internet on distracted driving, which are accessible through Google. A recent study [36] completed at the authors' home institution, which is currently in preprint, has examined the messages represented in distracted driving videos on YouTube with similar findings.

### Conclusions

Distracted driving is becoming more prevalent, and with it, so are the injuries and loss of lives associated with this problem. Our Google search project found that little to no specific information is available to the general public regarding types of injuries, including the potential disability resulting from them. Conversely, death due to distracted driving, which, in reality, occurs much less often than injuries, is the most commonly presented outcome in these websites. Unfortunately, websites focusing mainly on fatality secondarily to distracted driving eclipses the reality of millions of people surviving distraction-related MVCs who continue to live with chronic pain, disabilities, decreased quality of life, and increased financial burden. Furthermore, the sites mostly focus on mobile phones as potential distractions and largely neglect other more common forms of distracted driving. Although the general public recognizes that distracted driving is dangerous, in principle, the lack of specific information on the consequences of this behavior may be contributing to its continued practice.



## Conflicts of Interest

None declared.

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## Abbreviations

**MVC:** motor vehicle crash

**NHTSA:** National Highway Traffic Safety Administration

*Edited by G Eysenbach; submitted 13.09.19; peer-reviewed by A Mavragani, S Kardes; comments to author 22.10.19; revised version received 28.10.19; accepted 13.11.19; published 20.12.19.*

*Please cite as:*

Poon J, Gjorgjievski M, Moga I, Ristevski B

*Quality and Accuracy of Information Available on Websites for Distracted Driving: Qualitative Analysis*

*Interact J Med Res* 2019;8(4):e16154

URL: <http://www.i-jmr.org/2019/4/e16154/>

doi: [10.2196/16154](https://doi.org/10.2196/16154)

PMID: [31859684](https://pubmed.ncbi.nlm.nih.gov/31859684/)

 Jeffrey Poon, Marko Gjorgjievski, Justin Moga, Bill Ristevski. Originally published in the Interactive Journal of Medical Research (<http://www.i-jmr.org/>), 20.12.2019. This is an open-access article distributed under the terms of the Creative Commons Attribution License (<https://creativecommons.org/licenses/by/4.0/>), which permits unrestricted use, distribution, and reproduction in any medium, provided the original work, first published in the Interactive Journal of Medical Research, is properly cited. The

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Original Paper

# Osteopathic Considerations for Peripheral Neuropathy Due to Concomitant Diffuse Idiopathic Skeletal Hyperostosis Syndrome and Lumbar Epidural Lipomatosis: Case Report

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## Abstract

**Background:** Diffuse idiopathic skeletal hyperostosis (DISH) syndrome and lumbar epidural lipomatosis are relatively asymptomatic neurological conditions, with findings often seen incidentally on radiological studies.

**Objective:** The aim of this paper is to present unique findings of concomitant, symptomatic DISH syndrome and lumbar epidural lipomatosis and to discuss the osteopathic diagnosis and treatment implications.

**Methods:** Concomitant, symptomatic variants are rare and present challenges to treatment and management, as seen with a 60-year-old African American woman who presented with worsening disequilibrium and new-onset bilateral fingertip numbness. Past medical history was significant for alcohol abuse disorder, hypertension, hyperlipidemia, and multiple episodes of self-resolving vertigo and lower extremity neuropathy.

**Results:** The patient was referred to the neurology department for stroke workup, which was negative. Osteopathic structural exam revealed thoracolumbar and sacral dysfunctions. Magnetic resonance imaging revealed findings consistent with thoracic DISH syndrome and lumbar epidural lipomatosis in the areas of somatic dysfunctions.

**Conclusions:** Due to minimal information on osteopathic manipulative treatment in rare neurological diseases, only gentle techniques of myofascial release, balanced ligamentous tension, and muscle energy were performed with resultant minimal improvement, thus highlighting the necessity for better guidelines and further research.

(*Interact J Med Res* 2019;8(4):e14607) doi:[10.2196/14607](https://doi.org/10.2196/14607)

**KEYWORDS**

diffuse idiopathic skeletal hyperostosis syndrome; epidural lipomatosis; osteopathic medicine

## Introduction

Neurological symptoms are notoriously nonspecific, with differentials ranging from primary causes such as a tumor to secondary causes of dysfunction in other body systems. Further complicating the diagnosis, one-third of all neurological patients have findings that can only be partially or not at all explained by a discrete, organic disease [1]. Psychosocial and other external factors also play a role in neurological symptoms and

disease manifestation [1,2]. With considerable overlap in epidemiology, pathogenesis, and treatment options for various neurological diseases and syndromes of exclusion, patients often spend much of their time searching for a panacea, leading to endless frustration and delayed diagnosis/treatment.

History elicitation is difficult, as patients often lack the ability to distinguish the subtle differences that separate one disease from another. Studies have shown that patients frequently give unclear, inconsistent, and unreliable answers, especially in acute

care/emergency room settings [1,2]. Osteopathic physicians are trained to assess the person as a unit of body, mind, and spirit. Osteopathic manipulative treatment (OMT) is based on the understanding of this relationship in helping the body return to a state of homeostasis and health maintenance. Here, we present a complicated case of dizziness and peripheral neuropathy with associated osteopathic findings unresponsive to OMT and subsequent possible etiologies elucidated on radiological workup.

With no reports of concomitant, symptomatic disseminated idiopathic skeletal hyperostosis (DISH) syndrome and lumbar epidural lipomatosis, the aim of this paper is to present these unique findings and discuss the osteopathic diagnosis and treatment implications.

## Methods

### Overview

A 60-year-old African American female with a past medical history of alcohol abuse, controlled hypertension, and hyperlipidemia presented to the emergency department with a 1-week history of worsening disequilibrium and new bilateral upper extremity fingertip numbness after a fall.

### Case Presentation

In the emergency room, she denied any noticeable trauma, loss of consciousness, headache, diplopia, hearing loss, tinnitus, nausea, or vomiting at the time. Previously, she had vertiginous symptoms for several years, with minimal improvement on meclizine and an inconclusive workup with her primary care physician. Separately, she also reported a long history of self-resolving lower extremity numbness/neuropathy that she had associated with her previous occupation as a housekeeper, which required her to frequently be on her knees and feet. At the time of admission, she complained that her lower extremities were now constantly numb, with new intermittent burning (7/10 on the Numeric Pain Scale), nonradiating pain, which limited her ability to walk. Noncontrast head computed tomography was performed for stroke concerns, with subsequent results showing no acute intracranial pathology other than mild, chronic microvascular changes. She was referred to the neurology department for further workup.

## Results

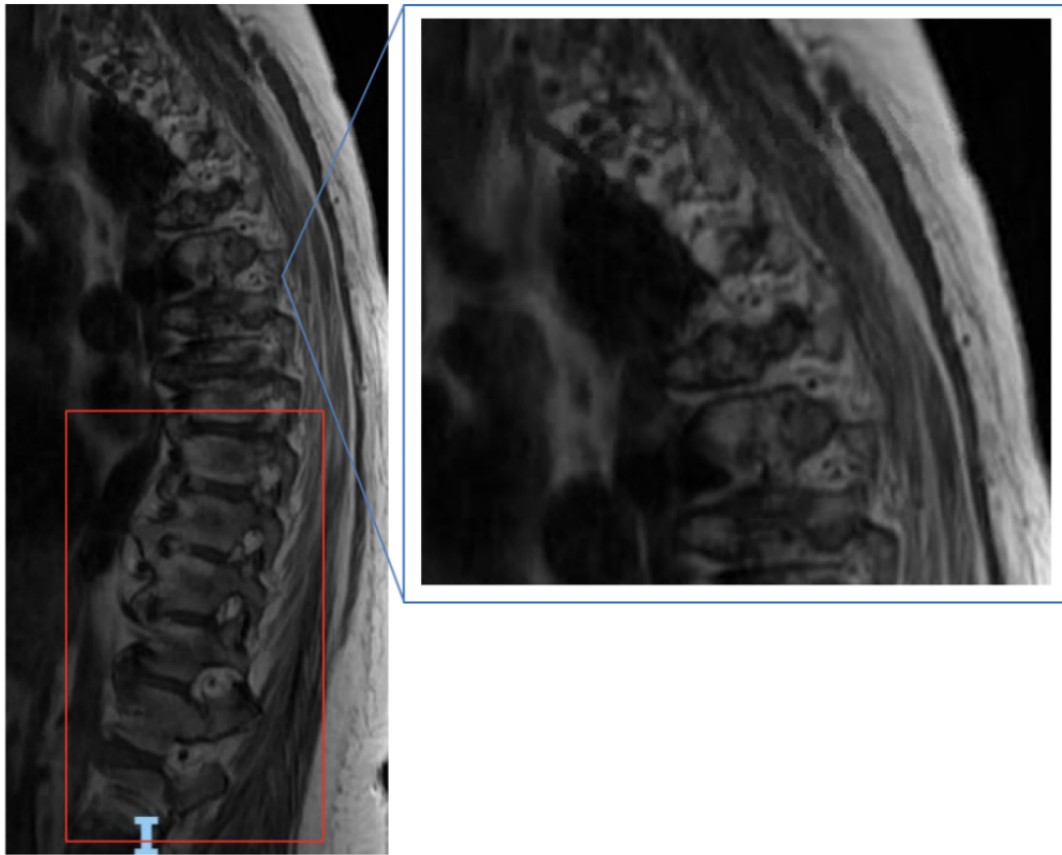
A physical exam at admission revealed nonacute distress, with vitals within normal limits. The neurological exam was positive

for right eye horizontal and vertical nystagmus, lower extremity motor weakness, and subjective bilateral sensory paresthesia in dermatomes T10-S2. The patient was able to distinguish soft versus sharp touch, but reported that it was a blunted sensation when compared to dermatomes above T10. Upper extremity motor and sensory exams were normal, with apparent resolution of her bilateral fingertip numbness. Orthostatic exam showed all values within normal limits and negative improvement of symptoms with the Dix-Hallpike and Epley maneuvers. Complete blood count, comprehensive metabolic panel, autoimmune panel, vitamin B12, and thyroid panels were within standard ranges.

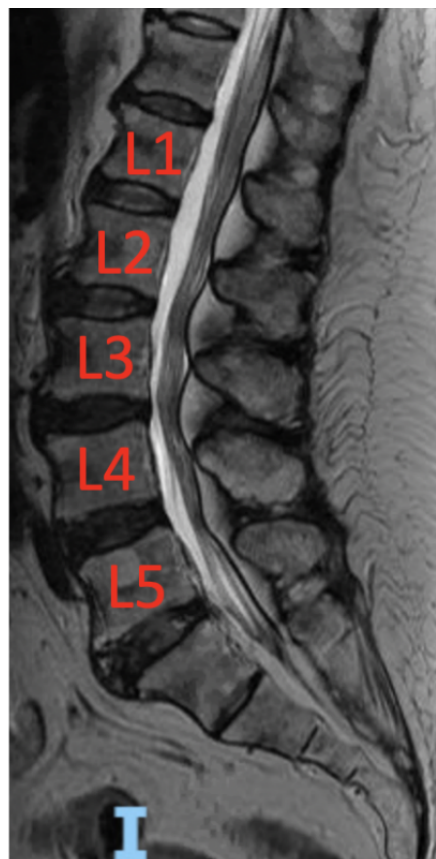
Osteopathic structural exam revealed numerous somatic dysfunctions: occipital-atlas joint flexed-rotated right-side bent left, atlantoaxial joint rotated right, C4 vertebra flexed-rotated right and side bent right, T5-T9 vertebral in neutral rotation left and side bent right, T12-L2 vertebra in neutral rotation right and side bent left, right-on-right sacral torsion, and left posteriorly rotated innominate. No appreciable viscerosomatic reflexes were visualized. Upper and lower extremity somatic dysfunctions were negligible. Gentle myofascial release, balanced ligamentous tension, and muscle energy were selected and applied to the thoracolumbar, sacral, and innominate somatic dysfunctions with minimal improvement in symptoms. Cervical treatment was deferred due to patient request. Reassessment showed persistent thoracolumbar dysfunctions. Further trials of OMT were deferred on the patient's request.

Further workup on computed tomography angiogram revealed a  $2.5 \times 1.5$  mm saccular aneurysm from the anterior communicating artery, hypoplastic A1 segment of the right anterior cerebral artery, small vertebrobasilar circulation, and dominant anterior circulation. Magnetic resonance imaging (MRI) of the cervical and thoracic spine were motion limited, but revealed a C4-C5 osteophyte complex with associated C2-C7 spinal cord compression of the thecal sac secondary to disc osteophyte complexes and disseminated idiopathic skeletal hyperostosis throughout the thoracic spine (Figure 1). MRI of the lumbar spine revealed moderate stenosis of L2-L3 with epidural lipomatosis at L3-L4 (Figure 2). Endovascular and neurosurgical teams were consulted, but the patient declined surgical intervention. Conservative management and supportive care were discussed, and she elected for physical therapy and outpatient nerve conduction studies and electromyography. OMT was also discussed as an alternative method to addressing chronic symptoms.

**Figure 1.** T1-weighted sagittal magnetic resonance imaging of the cervical and thoracic spine revealing C4-C5 osteophyte complex with associated C2-C7 spinal cord compression of the thecal sac secondary to disc osteophyte complexes (blue box) and disseminated idiopathic skeletal hyperostosis throughout the thoracic spine, particularly at the levels of T5-T9 (red box). Orientation: I indicates inferior.



**Figure 2.** T2-weighted sagittal magnetic resonance imaging of the lumbar spine revealing moderate stenosis of L2-L3 with epidural lipomatosis at L3-L4. Orientation: I for inferior.





## Discussion

First described in 1975, spinal epidural lipomatosis (SEL) is a rare condition, wherein there is a hypertrophy of adipose tissue in the spinal epidural space that results in compression of nerves in the affected region [3]. SEL occurs in 1 of every 40 patients, with presentations ranging widely from asymptomatic to cauda equina syndrome/permanent nerve damage [4,5]. Although SEL can occur throughout the spinal cord, the majority of cases have been documented in the thoracic and lumbosacral region, with the rarest described in the cervical region [6,7]. The exact pathophysiology of SEL is unknown; however, various associations have been made, depending on the region. Exogenous long-term steroid use and male gender have been associated with thoracic SEL in over 75% of the reported cases [3,7]. In comparison, cervical and lumbosacral SEL are largely idiopathic and incidentally seen on MRI [7]. Other possible associations/causes include Cushing disease, Cushing syndrome, obesity, hypothyroidism, and pituitary adenoma [8,9]. Treatment options for symptomatic SEL range from conservative treatment of dietary changes, weight loss, and long-term steroid weaning to decompressive laminectomy and adipose resection [3,10]. Although studies have shown equal efficacy, surgical intervention is generally reserved for failed conservative management, as there are considerable complications and morbidity associated with the postoperative management of concomitant medical problems [9,10].

DISH syndrome is the abnormal calcification of ligaments or bone formation in the axial or appendicular skeleton [11]. Commonly diagnosed using criteria established in 1976, its alternate names are Forestier disease, senile ankylosing spondylosis, and ankylosing hyperostosis [11,12]. This condition involves abnormal calcification of either the anterior or posterior longitudinal ligament of the spine, with the thoracic region being the most common [12]. Peripherally, it is characterized by calcification and hyperossification of entheses or sites where ligaments/tendons attach to bone [13]. The incidence and prevalence of DISH syndrome are largely unknown and underreported due to its asymptomatic nature [14,15]. Symptomatic manifestation is variable, ranging widely from monoarticular synovitis to mass effect airway obstruction. Similar to SEL, the pathogenesis of DISH syndrome is unclear, with mechanical factors, genetics, environmental exposures, and metabolic/dietary conditions as proposed mechanisms or having possible associations [13,16]. Limited research exists on treatment, with the majority pursuing conservative management, analgesics, physical therapy, or a combination of the aforementioned [15]. In extremely rare cases of extensive calcification or if osteophyte formation is causing severe and focal symptoms, surgical intervention with excision or resection of bony tissue may be warranted [16].

On reviewing the literature, we did not find any reports of patients with symptomatic concomitant lumbar SEL and thoracic DISH syndrome. In our case, the patient had a complicated clinical picture, with multiple comorbidities and disease associations with lumbar SEL and DISH syndrome, such as chronic alcohol abuse, hypertension, hyperlipidemia, and obesity. Although such findings supported a diagnosis of symptomatic lumbar SEL and DISH syndrome, definitive diagnosis and treatment of the patient's complaints remained elusive, as there is a paucity of research regarding simultaneous disease presentation. Her CT angiogram findings and alcohol abuse disorder history may also have played a role in her vertiginous symptoms. In both lumbar SEL and DISH syndrome, studies have explored the difficulty in creating a comprehensive, conservative management plan due to variability in symptom manifestation and severity. With this unpredictable expression, OMT has been proven to be an effective modality for treating similar intractable pain/discomfort syndromes that are refractory to established management options [17]. Treatments of both DISH- and SEL-affected areas were performed together, as minimal differences for standard treatments exist between the two conditions [3,15].

Coupled with the osteopathic philosophy of seeing the patient as a sum of mind, body, and spirit, greater research needs to be conducted on application of osteopathic treatment to rarer diseases and syndromes. Although somatic dysfunction can be an indicator of an underlying pathology, there is conflicting and limited data on osteopathic diagnosis and subsequent treatment in rarer diseases [18]. Furthermore, while general guidelines exist on the use of direct versus indirect techniques, more studies are needed to categorize the efficacy of said techniques on rarer diseases and the treatment frequency [19]. Greater standardization and blinded trials are needed to be better able to reproduce results and studies [20]. Furthermore, the patient was lost to follow-up after hospital discharge, despite attempts to contact the patient.

Concomitant DISH syndrome and lumbar spinal epidural lipomatosis may present symptomatically with peripheral neuropathy. DISH syndrome and SEL have various disease associations; however, the pathogenesis remains unclear, which hinders the development of treatment options. Case presentation of a patient with these symptoms serves to highlight the complexity and variability of disease presentation. Limited research exists on concomitant DISH syndrome and lumbar SEL as well as the effectiveness of osteopathic management. Further research is required to better understand and develop osteopathic treatment for rare syndromes that are refractory of conservative or traditional management.

## Conflicts of Interest

None declared.

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## Abbreviations

- DISH:** diffuse idiopathic skeletal hyperostosis  
**MRI:** magnetic resonance imaging  
**OMT:** osteopathic manipulative treatment  
**SEL:** spinal epidural lipomatosis

*Edited by G Eysenbach; submitted 04.05.19; peer-reviewed by M Volokitin, C Zhou; comments to author 14.06.19; revised version received 16.06.19; accepted 30.08.19; published 20.11.19.*

*Please cite as:*

*Chin J, Kviatkovsky B, Lomiguen C*

*Osteopathic Considerations for Peripheral Neuropathy Due to Concomitant Diffuse Idiopathic Skeletal Hyperostosis Syndrome and Lumbar Epidural Lipomatosis: Case Report*

*Interact J Med Res 2019;8(4):e14607*

*URL: <http://www.i-jmr.org/2019/4/e14607/>*

*doi: [10.2196/14607](https://doi.org/10.2196/14607)*

*PMID: [31746769](https://pubmed.ncbi.nlm.nih.gov/31746769/)*

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Original Paper

# How Do Publicly Available Allergy-Specific Web-Based Training Programs Conform to the Established Criteria for the Reporting, Methods, and Content of Evidence-Based (Digital) Health Information and Education: Thematic Content Evaluation

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## Abstract

**Background:** Allergic diseases, such as allergic asthma, rhinitis, and atopic eczema, are widespread, and they are a considerable burden on the health care system. For patients and health care professionals, Web-based training programs may be helpful to foster self-management and provide allergy-specific information, given, for instance, their good accessibility.

**Objective:** This study aimed to assess an exploratory sample of publicly available allergy-specific Web-based training programs—that is, interactive, feedback-oriented Web-based training platforms promoting health behavior change and improvement of personal skills—with regard to (1) general characteristics, aims, and target groups and (2) the extent to which these tools meet established criteria for the reporting, methods, and content of evidence-based (digital) health information and education.

**Methods:** Web-based training programs were identified via an initial Google search and a search of English and German language websites of medical and public health services, such as the European Centre for Allergy Research Foundation (German), Asthma UK, and Anaphylaxis Canada. We developed a checklist from (1) established guidelines for Web-based health information (eg, the Journal of the American Medical Association benchmarks, DISCERN criteria, and Health On the Net code) and (2) a database search of related studies. The checklist contained 44 items covering 11 domains in 3 areas: (1) content (completeness, transparency, and evidence), (2) structure (data safety and qualification of trainers and authors), and (3) impact (effectiveness, user perspective, and integration into health care). We rated the Web-based training programs as completely, partly, or not satisfying each checklist item and calculated overall and domain-specific scores for each Web-based training program using SPSS 23.0 (SPSS Inc).

**Results:** The 15 identified Web-based training programs covered an average of 37% of the items (score 33 out of 88). A total of 7 Web-based training programs covered more than 40% (35/88; maximum: 49%; 43/88). A total of 5 covered 30% (26/88) to 40% (35/88) of all rated items and the rest covered fewer (n=3; lowest score 24%; 21/88). Items relating to intervention (58%; 10/18), content (49%; 9/18), and data safety (60%; 1/2) were more often considered, as opposed to user safety (10%; 0.4/4), qualification of staff (10%; 0.8/8), effectiveness (16%; 0.4/2), and user perspective (45%; 5/12). In addition, in 13 of 15 Web-based training programs, a minimum of 3 domains were not covered at all. Regarding evidence-based content, 46% of all Web-based training programs (7/15) scored on use of scientific research, 53% on regular information update (8/15), and 33% on provision of references (5/15). None of 15 provided details on the quality of references or the strength of evidence.

**Conclusions:** English and German language allergy-specific Web-based training programs, addressing lay audiences and health care professionals, conform only partly to established criteria for the reporting, methods, and content of evidence-based (digital) health information and education. Particularly, well-conducted studies on their effectiveness are missing.

**KEYWORDS**

allergy; asthma; health communication; health education; health information systems; evidence-based practice

## Introduction

### Background

The number of people affected by allergies and asthma varies around the world, but the prevalence of allergic diseases is high, particularly of allergic asthma, rhinitis, and atopic eczema [1-5]. Allergy-specific Web-based health information (WHI), as well as Web-based training programs (WTPs), can be a vital source of help for patients and health care professionals (HCPs). People affected by allergies and asthma, particularly those with mild-to-moderate symptoms, may not regularly see a physician but rely on self-treatment. This includes, for instance, nonprescription medicines, reading information on the Web, or even simply trying to get through the allergy season without help. Here, WHI or WTPs may be an alternative to *doing nothing* or relying only on one's own knowledge and skills. Previous research has outlined the effectiveness of various measures [6,7]. For HCPs, allergy-specific WHI and/or WTPs might be relevant, given a need for continuous medical education and for support of their patient's self-management skills [8]. We consider WTPs to comprise Web-based offers that go beyond mere provision of information, providing feedback and interactive learning opportunities promoting health behavior change and improvement of personal skills, without human interaction. We distinguish WTPs from services dedicated to Web-based treatment or counseling and from apps designed for digital mobile devices, such as tablets or phones [9-12]. A WTP works on desktop computers and mobile devices, but it will not need features specific to the mobile device, such as sensors or location awareness.

An allergy-specific WTP may assess patients' current symptom avoidance practices during the allergy *season* and then give feedback on the effectiveness of that approach (feedback). If a patient is thereby encouraged to apply more effective approaches, this also improves self-management (personal skills). WTPs can also provide a diary for daily recording of symptoms medication use, which may then be shared electronically with a doctor or during a Web-based consultation (interactive learning). For professionals, a WTP could provide fictional cases of patients with allergic symptoms and guide them through the correct assessment and treatment (strengthen treatment skills and care practice). Although WTPs may be promising in general, previous research has highlighted a range of respective challenges in particular: limited abilities to access, understanding and applying health information (health literacy), poor-quality information and sources, use of jargon, inaccuracy, information overload, and a lack of universal requirements regarding content and methods [13-21]. By *quality*, we refer to

the extent to which allergy-specific WTPs conform to established criteria on presentation of health information and its application to health care practice. Numerous initiatives have proposed criteria to ensure high-quality WHI (and hence WTPs), the most prominent being the *Journal of the American Medical Association* (JAMA) benchmarks [22], Health On the Net (HON) code [23], and the DISCERN criteria [24]. Yet, because of the sheer amount and variety of WHI, there is no one tool for evaluation or use of quality criteria that covers the entire spectrum of information sources [16]. The quality of intervention descriptions has also been criticized as making it difficult to build on or replicate available learning sources [25].

### Objectives

We are not currently aware of any overview of the quality of allergy-specific WTPs or the use of quality criteria by Web-based services intended for allergy sufferers or HCPs. A few studies have examined the effectiveness of digital asthma self-management [26,27], the impact of Web-based peer support for children with asthma [28], and Web-based support pilot studies [29]. Given this background, the objective of our study was to analyze an exploratory sample of publicly available, free-of-charge allergy-specific WTPs. Specifically, we analyzed (1) the general characteristics, aims, and target groups of WTPs, (2) the evidence base underlying WTPs' content, and (3) the degree to which WTPs account for criteria on the reporting, methods, and content of (digital) health information (see also population, intervention, comparison, outcome table in [Multimedia Appendix 1](#)). As aspects, such as structure, quality of information, and evidence base, are relevant independent of the WTPs' target group, we did not limit our analysis to either lay people or HCPs.

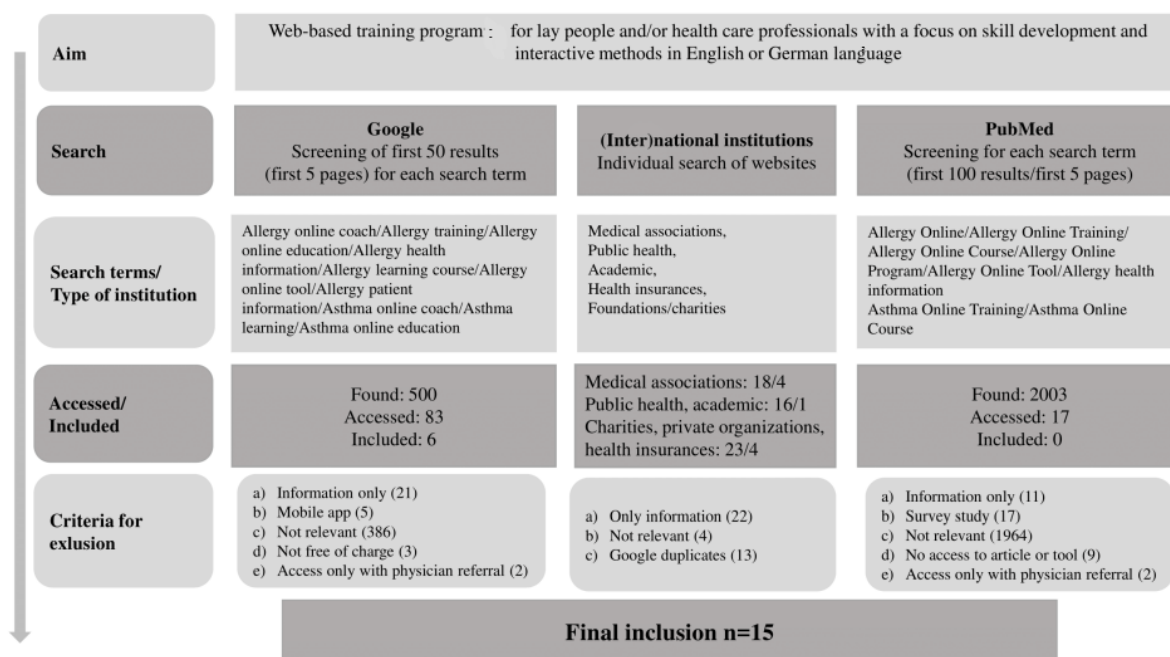
## Methods

### Search Strategy and Selection of Web-Based Training Programs for Assessment

We conducted a Google search to retrieve and select relevant WTPs. As this is an exploratory study, we constrained the search to WTPs in English and German, that is, the languages spoken by our research team. We used a public search engine, as the Web-based sources need to be available to lay people, rather than being available only in a scientific database intended for experts. We added a search in PubMed to check for potentially relevant WTPs not found in Google and for scientific evaluations of WTPs. For the search, terms were combined with regard to (1) indication/disease and (2) the diverse types of available Web-based sources ([Figure 1](#)).



**Figure 1.** Search Strategy. WTP: Web-based training program; HCP: health care professional.



In addition, we searched country-specific websites of institutions that provide general health information or specialize in allergies and atopic diseases: medical associations, health insurers, state health services, and official Public Health information portals. This included the National Health Service, Allergy UK (United Kingdom), the American Academy of Allergy (United States), Allergy Aware Canada (Canada), Australasian Society of Clinical Immunology and Allergy (Australia), the German Medical Association, the European Centre for Allergy Research Foundation (Germany), the European Academy of Allergy and Clinical Immunology (Switzerland), and the World Allergy Organization (United States). From these sources, we included tools that provided allergy- and/or asthma-specific training for lay people and HCPs (medical doctors, nurses, school staff, and pharmacists). We also categorized retrieved sources as either (1) informative websites, (2) apps, or (3) WTPs. Next, we selected only services and tools of type (3), according to the abovementioned description of WTPs, to limit the analysis to the more comprehensive, interactive learning approaches. We ended the search after no new, additional WTPs were found by different searches using the same search strategies. Criteria for exclusion were as follows: not free of charge, available only by physician referral, and not yet or no longer publicly available.

**Checklist: Development and Application**

To assess the selected WTPs, we first conducted a PubMed search for standards, guidance, and tools to develop, report,

and/or critically appraise WHI and WTP. We then summarized the criteria from different sources [9,22,23,25,30] and adapted the description and wording of each criterion into an assessment item (Multimedia Appendix 2). The summary checklist resulted in a list of 44 items, subdivided into 11 domains (Textbox 1).

For each WTP, 3 researchers read and worked through the respective program and extracted relevant text passages independently. The extracted material was given one of the following ratings:

- Yes: criterion satisfied according to the available information
- Partly: criterion satisfied to some extent
- No (not done or not stated): criterion not satisfied or no information about whether the aspect/content relevant to the respective criterion was not considered or simply not stated
- Not included: the WTP’s design does not address the criterion, although it seems relevant

After initial assessment, missing information and unclear ratings were discussed among the 3 researchers to reach agreement. Finally, we assigned a score for each rating (yes=2, partly=1, no=0, and not part of WTP=-1) and calculated per-domain and overall scores for each WTP, using SPSS 23. For instance, the category ‘indication’ includes 9 criteria, hence a maximum per-domain score of 18 can be given.

**Textbox 1.** List of quality criteria for Web-based training programs, adapted from studies.

#### Indication

- The symptoms addressed by the program are described
- The levels of severity of the allergy, with which the program is supposed to help, are described

#### Intervention

- Full provider contact details are given
- The program type (self-help, coaching, chat, etc) is described
- The description of the type of program is transparent and freely accessible
- Rationales and aims are described
- The program is described separately for other target groups (who may also be interested in the content), either for lay people or professionals
- A minimum/maximum usage time is mentioned
- A certain usage time is recommended
- The recommended usage time is supported by evidence
- Alternatives for using this particular program are mentioned

#### Content

- The information has been researched scientifically and systematically
- The information is up to date
- The information is updated regularly according to most recent available knowledge
- Transparent sources/references are provided
- The content of the information is formulated neutrally and factually
- The information/content mentions potential uncertainties and risks
- Transparent information regarding financing and conflicts of interest are provided
- Potential usage/user differences because of age or sex are mentioned
- The content is differentiated for/adapted to different target groups

#### Safety

- Potential unintended effects of using the program are described
- The program describes what happens in case of an unintended effect

#### Qualification

- Users can contact an expert
- The qualification of the expert is described (if part of intervention)
- Experts that can be contacted use an intervention manual
- Experts are being supervised

#### Effectiveness

- The effectiveness of the program is assessed (via a scientific evaluation)

#### User perspective

- The program is accessible (eg, by hearing- or vision-impaired users)
- The program is free of charge
- The program is available in different languages
- Completion/termination rates are mentioned
- User satisfaction with the program is assessed
- The success of the program is assessed (have the users completed the modules successfully)

## Integration into care

- User behavior is followed up
- Users can communicate with other users and/or other professionals

## Legal aspects

- It is stated who is liable in case of mistakes and adverse effects

## Data safety

- It is clearly mentioned that data safety is ensured by the provider
- The user can register anonymously
- A description of how user data are stored is given
- It is mentioned for how long the data are stored
- The user can ask the provider to delete personal data
- The program requires specific software (eg, operating system or browser)

## Advertisement

- The program includes open/direct advertisement
- The program includes indirect advertisement

## Results

### General Characteristics

The search found 171 potentially relevant (and accessed) allergy-specific WTPs, with 15 remaining after applying exclusion criteria (Figure 1). Regarding those WTPs found via PubMed, 3 WTPs required a fee, and 4 WTPs were only available by physician referral. A total of 5 WTPs were not publicly available, as they were part of a (closed) intervention study [28,31-34]. Another 3 WTPs could not be found on the internet, or they were no longer available. Providers of the 15 WTPs included in this analysis were based in the United Kingdom (n=1), Germany (n=4), the United States (n=3), Australia (n=4), Canada (n=2), and one was not country specific. WTPs were run by (1) academic institutions (n=1), nonprofit foundations/charities (n=7), medical societies (n=3), and umbrella organizations thereof (n=2), publishing houses (n=1), and a pharmaceutical training portal (n=1). A total of 14 of the 15 programs required registration and a user account for access. Of the included WTPs, 53% (n=8) mentioned (only) 1 subject, for example, asthma, anaphylaxis, pollen allergy, or simply *allergy* without a focus. A total of 33% (n=5) of the WTPs combined 2 subjects, for example, allergy and anaphylaxis, or atopic eczema and food allergies. Except for the combination of a specific allergy with anaphylaxis, WTPs mentioned no reasons for combining subjects. The 2 programs with 3 or more subjects aimed to provide a holistic overview of the various types of allergies and how to deal with them. WTPs varied widely in their style of presentation (Table 1).

Some (n=5) referred to an *interactive online course* or *interactive learning module*; others described their program as “online” or “e-learning” modules (n=4). Nevertheless, others simply referred to an *education course*, *active learning*, or *individual learning* modules and course (n=4), whereas a final group focused on describing the format: *scientific animation* and *multimedia tool* (n=2).

With regard to the precise didactic methods, 60% (n=9) of the WTPs applied 4 or more methods. A total of 1 WTP, for instance, combined visualized written information, animations, videos, written instructions, case studies, a survey, a quiz, and a feedback form. Another WTP applied preknowledge questions, learning modules (short written information), short video, multiple-choice questions for self-assessment, and a course certificate for download after completion. A total of 2 WTPs used 3 different methods, and 4 WTPs applied only 1 (n=1) or 2 (n=3) methods, for example, fictional characters telling their disease story and interacting with the user via questions. The WTPs did not explain why particular methods were chosen, their didactic approach/model, or the didactic advantage of their chosen methods. The WTPs addressed 3 broadly distinguishable audiences: (1) lay people (ie, allergy sufferers), (2) HCPs (including physicians), and (3) people who are not medical experts but work in the public sector, that is, teachers and other school staff. The latter were mentioned twice as the sole target group. A total of 1 WTP addressed only lay people. A total of 7 WTPs were aimed at professionals, mostly referring to *doctors* and *professionals*. The other 5 WTPs addressed mixed target groups, that is, lay people and HCPs.

**Table 1.** General information on assessed Web-based training programs.

| Name                                                                            | Subjects                          | Country       | Provider                                                                              | Language | Aim                                                                                               | Methods                                                                                                                   |
|---------------------------------------------------------------------------------|-----------------------------------|---------------|---------------------------------------------------------------------------------------|----------|---------------------------------------------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------|
| <b>Lay people only</b>                                                          |                                   |               |                                                                                       |          |                                                                                                   |                                                                                                                           |
| Anaphylaxis First Aid Training Community                                        | Anaphylaxis                       | Australia     | Australasian Society of Clinical Immunology and Allergy (medical society)             | English  | Provide ready access to reliable anaphylaxis education                                            | Learning modules; Visualizations; Short video; Certificate; Evaluation                                                    |
| <b>Health care professionals only</b>                                           |                                   |               |                                                                                       |          |                                                                                                   |                                                                                                                           |
| Allergic Rhinitis e-training                                                    | Allergic rhinitis                 | Australia     | Australasian Society of Clinical Immunology and Allergy (medical society)             | English  | Provide ready access to reliable allergic rhinitis education                                      | Introductory questions; Learning modules; Visualization; Short video; Multiple-choice self-control questions; Certificate |
| Allergy and Anaphylaxis e-training for Health Care Professionals                | Allergies; Anaphylaxis            | Australia     | Australasian Society of Clinical Immunology and Allergy (medical society)             | English  | Provide ready access to reliable anaphylaxis rhinitis education                                   | Learning modules; Final assessment; Evaluation; Certificate                                                               |
| Asthma and Allergic Rhinitis: World Allergy Organization Online Learning Series | Asthma; Allergic rhinitis         | United States | World Allergy Organization (international umbrella organization of medical societies) | English  | Provide a clear overview about asthma and allergic rhinitis                                       | Presentation by speaker; Question and Answer session                                                                      |
| Asthma management and education course                                          | Asthma                            | United States | Asthma and Allergy Foundation of America (foundation/non-profit organization)         | English  | Educate health professionals about asthma management                                              | Learning modules, including assessment; Evaluation                                                                        |
| e-learning hub                                                                  | Asthma                            | Australia     | Asthma Australia (foundation/nonprofit organization)                                  | English  | Guide professionals in asthma management and patient education                                    | Audiovisual presentation; Certificate; Survey; Review questions                                                           |
| National Asthma Council Australia Online Learning                               | Asthma                            | United States | National Asthma Council Australia (non-profit charity)                                | English  | Provide professionals with interaction learning on the latest information and resources on asthma | Learning modules; Presentation, including how-to videos; Introductory questions; Case studies                             |
| Azerta                                                                          | Allergies (various/general)       | Germany       | Azerta Apotheken Portal (pharmacy training portal)                                    | German   | Continuing education                                                                              | Videos; "Coaching" for dealing with clients; Questionnaire (knowledge questions); Certificate                             |
| <b>Lay people and health care professionals</b>                                 |                                   |               |                                                                                       |          |                                                                                                   |                                                                                                                           |
| Itchy Sneezzy Wheezy                                                            | Asthma; Allergic Rhinitis; Eczema | England       | Imperial College London (lead) with partner organizations (academic)                  | English  | Train and improve users' knowledge and skills                                                     | Written information; Slides; Multimedia guide; Feedback form; Certificate                                                 |

| Name                     | Subjects                                                  | Country       | Provider                                                                                       | Language        | Aim                                                                                     | Methods                                                                                                                |
|--------------------------|-----------------------------------------------------------|---------------|------------------------------------------------------------------------------------------------|-----------------|-----------------------------------------------------------------------------------------|------------------------------------------------------------------------------------------------------------------------|
| Human-Biomonitoring      | Environmental substances (main part); Allergies (subpart) | Germany       | Allum/ Kinderumwelt gemeinnützige GmbH Os-nabrück (umbrella organization of medical societies) | German; English | Provide knowledge and information interactively                                         | Animation                                                                                                              |
| Allergyaware             | Anaphylaxis                                               | Canada        | Food Allergy Canada (charitable organization)                                                  | English; French | Provide information and knowledge to improve behavior/skills for emergency situations   | Visualized information; Animation; Videos; Instructions; Case studies; Survey; Quiz; Feedback form; Certificate        |
| Asthma Basics            | Asthma                                                    | United States | American Lung Association (voluntary health organization/nonprofit organization)               | English         | Help people learn more about asthma                                                     | Introductory questionnaire; Interactive presentation; Repeat questions; Quiz; Evaluation; Certificate                  |
| Lunge in Not             | Asthma                                                    | Germany       | Springer Medizin Verlag GmbH (publishing house)                                                | German          | Improve patient–physician communication                                                 | Use of fictional characters to visualize information; Storyline (scroll down)                                          |
| <b>Others</b>            |                                                           |               |                                                                                                |                 |                                                                                         |                                                                                                                        |
| Back to School           | Food allergies; Anaphylaxis                               | Canada        | Food Allergy Research and Education (nonprofit organization)                                   | English         | Support school staff in the prevention and management of food allergies and anaphylaxis | Multiple choice; Warmup questions; Visualized information; Audio sections (information/facts); Final quiz; Certificate |
| Bist du auch allergisch? | Neurodermatitis; Food allergies                           | Germany       | European Centre for Allergy Research Foundation (nonprofit foundation)                         | German          | Independent Web-based learning; knowledge acquisition; knowledge application            | Learning modules/chapter; Assessment questions                                                                         |

## Indication and Intervention

In the domains *indication* (2 items) and *intervention* (9 items), almost all WTPs covered at least 50% of the total items, resulting in a mean score of 52% (sum score: 2.1/4) for indication and 58% (sum score: 10.4/18) for intervention (Table 2). For *indication*, all WTPs described the symptoms addressed as part of the content and learning modules, but none described the severity of symptoms that would or would not be addressed (although 1 WTP was rated *partly*).

With regard to the description of interventions, 8 of 15 WTPs covered more than half of the items for this category; 6 of those covered more than two-thirds. General aspects, such as rationales/aims (100%; 15/15), intervention type (86%; 13/15),

minimum/maximum usage time (60%; 9/15), and provider details (66%; 10/15), were covered most often. More specific aspects, such as separate intervention descriptions according to the respective target group (13%; 2/15), alternatives to using the WTP (27%; 4/15), and evidence for the recommendation of a specific usage time (13%; 2/15), were addressed much less frequently. For example, 1 WTP stated “You can undertake this course at your own pace, but it is recommended that you complete the modules within a two-week period,” though without stating who exactly recommends this (see Multimedia Appendix 3). In total, items belonging to the domains indication (2) and intervention (9) were more often rated as fulfilled or partly fulfilled (61%, n=101 ratings) than not fulfilled (39%, n=64 ratings; Table 2).



**Table 2.** Per-domain and overall scores.

| Name and WTP <sup>a</sup>                  | Score, n (%) |              |          |          |               |          |                  |         |          |             |               |           |
|--------------------------------------------|--------------|--------------|----------|----------|---------------|----------|------------------|---------|----------|-------------|---------------|-----------|
|                                            | Indication   | Intervention | Content  | Safety   | Qualification | Effect   | User perspective | Care    | Legal    | Data safety | Advertisement | Total     |
| <b>Lay people , n (%)</b>                  |              |              |          |          |               |          |                  |         |          |             |               |           |
| WTP1                                       | 2 (50)       | 14 (78)      | 12 (67)  | 0 (0)    | 1 (12)        | 0 (0)    | 4 (33)           | 0 (0)   | 2 (100)  | 2 (17)      | 2 (50)        | 39 (44)   |
| <b>Professionals, n (%)</b>                |              |              |          |          |               |          |                  |         |          |             |               |           |
| WTP2                                       | 2 (50)       | 8 (44)       | 11 (61)  | 0 (0)    | 1 (12)        | 0 (0)    | 5 (42)           | 0 (0)   | 2 (100)  | 2 (17)      | 2 (50)        | 33 (37)   |
| WTP3                                       | 2 (50)       | 12 (67)      | 10 (56)  | 0 (0)    | 1 (12)        | 2 (100)  | 6 (50)           | 0 (0)   | 2 (100)  | 2 (17)      | 2 (50)        | 38 (43)   |
| WTP4                                       | 2 (50)       | 8 (44)       | 5 (28)   | 0 (0)    | 2 (25)        | 0 (0)    | 4 (33)           | 0 (0)   | 0 (0)    | 0 (0)       | 2 (50)        | 23 (26)   |
| WTP5                                       | 2 (50)       | 12 (67)      | 4 (22)   | 0 (0)    | 1 (12)        | 0 (0)    | 8 (67)           | 0 (0)   | 0 (0)    | 0 (0)       | 0 (0)         | 27 (31)   |
| WTP6                                       | 2 (50)       | 13 (72)      | 11 (61)  | 0 (0)    | 0 (0)         | 0 (0)    | 6 (50)           | 0 (0)   | 0 (0)    | 2 (17)      | 2 (50)        | 36 (41)   |
| WTP7                                       | 2 (50)       | 14 (78)      | 12 (67)  | 0 (0)    | 2 (25)        | 0 (0)    | 4 (33)           | 0 (0)   | 2 (100)  | 5 (42)      | 0 (0)         | 41 (47)   |
| WTP8                                       | 2 (50)       | 9 (50)       | 7 (39)   | 0 (0)    | 0 (0)         | 0 (0)    | 4 (33)           | 0 (0)   | 2 (100)  | 6 (50)      | 2 (50)        | 32 (36)   |
| <b>Lay people and professionals, n (%)</b> |              |              |          |          |               |          |                  |         |          |             |               |           |
| WTP9                                       | 2 (50)       | 10 (56)      | 9 (50)   | 0 (0)    | 0 (0)         | 0 (0)    | 6 (50)           | 0 (0)   | 0 (0)    | 0 (0)       | 0 (0)         | 27 (31)   |
| WTP10                                      | 3 (75)       | 9 (50)       | 11 (61)  | 0 (0)    | 3 (37)        | 0 (0)    | 6 (50)           | 0 (0)   | 0 (0)    | 4 (33)      | 0 (0)         | 36 (41)   |
| WTP11                                      | 2 (50)       | 9 (50)       | 11 (61)  | 4 (100)  | 1 (12)        | 0 (0)    | 8 (67)           | 0 (0)   | 2 (100)  | 4 (33)      | 2 (50)        | 43 (49)   |
| WTP12                                      | 2 (50)       | 9 (50)       | 5 (28)   | 0 (0)    | 0 (0)         | 2 (100)  | 10 (83)          | 0 (0)   | 2 (100)  | 4 (33)      | 0 (0)         | 34 (39)   |
| WTP13                                      | 2 (50)       | 6 (33)       | 7 (39)   | 0 (0)    | 0 (0)         | 0 (0)    | 4 (33)           | 0 (0)   | 0 (0)    | 2 (17)      | 0 (0)         | 21 (24)   |
| <b>Others n (%)</b>                        |              |              |          |          |               |          |                  |         |          |             |               |           |
| WTP14                                      | 2 (50)       | 13 (72)      | 11 (61)  | 2 (50)   | 0 (0)         | 2 (100)  | 4 (33)           | 0 (0)   | 2 (100)  | 0 (0)       | 2 (50)        | 38 (43)   |
| WTP15                                      | 2 (50)       | 10 (56)      | 6 (33)   | 0 (0)    | 0 (0)         | 0 (0)    | 2 (16.7)         | 0 (0)   | 2 (100)  | 4 (33)      | 0 (0)         | 26 (29)   |
| <b>Summary</b>                             |              |              |          |          |               |          |                  |         |          |             |               |           |
| WTP, mean (SD)                             | 2.1 (52)     | 10.4 (58)    | 8.8 (49) | 0.4 (10) | 0.8 (10)      | 0.4 (17) | 5.4 (45)         | 0 (0)   | 1.2 (60) | 2.5 (21)    | 1.1 (27)      | 32.9 (37) |
| Maximum, n (%)                             | 4 (100)      | 18 (100)     | 18 (100) | 4 (100)  | 8 (100)       | 2 (100)  | 12 (100)         | 4 (100) | 2 (100)  | 12 (100)    | 4 (100)       | 88 (100)  |

<sup>a</sup>WTP: Web-based training program.

## Content and Safety

At least half the items related to content (n=9) were covered by 9 of 15 WTPs, that is, a score of at least nine out of 18, the maximum covered being 67% (12 out of 18). Individual items often fulfilled were evidence-based content (n=10; yes=7; partly=3), provision of a date to indicate up to dateness (n=8), regular updates of the information (n=8), and neutral and factual content and language (n=15). Regarding the latter, this was rated as fulfilled when the information did not include obvious exaggerations or strongly positive/negative formulations, for example, regarding a treatment option. Furthermore, 6 of 15 WTPs covered fewer than 40% of items in this category, 3 of those covering only 20% to 30%. Items that were often not covered included the provision of full references for all content (partly=5; no=5), possible differences in how the service should

be used because of age and/or sex (n=0), uncertainties and risks, for example, when following a specific recommendation or knowledge gaps (n=5; see [Table 3](#) for examples), and funding and conflicts of interest (yes=5; partly=6).

Although references were provided by several WTPs (see above), none provided explicit descriptions of the quality of those references, the type and strength of evidence within these references, and how the references were used to develop content to cover the (full) spectrum of relevant, evidence- and/or expert-based issues regarding prevention, diagnosis, and treatment. Given the number of WTPs covering less than 40% of all content-related items, the mean score for this category was 49% (sum score 8.8 of 18). Of all ratings (n=135), 42% (n=75) of the ratings were positive (“yes”) and 44% (n=60) of the ratings were negative (“no” or “not addressed”); the rest were rated “partly” (14%).

**Table 3.** Rating summary.

| Domain                     | Item, n | Total ratings, n | Yes, n (%) | Partly, n (%) | No, n (%)  | Not part, n (%) |
|----------------------------|---------|------------------|------------|---------------|------------|-----------------|
| (1) Indication             | 2       | 30               | 15 (50)    | 0 (0)         | 13 (43)    | 2 (7)           |
| (2) Intervention           | 9       | 135              | 70 (51.8)  | 16 (11.8)     | 39 (28.8)  | 10 (7.4)        |
| (3) Content                | 9       | 135              | 56 (41.4)  | 19 (14.0)     | 41 (30.3)  | 19 (14.0)       |
| (4) Safety                 | 2       | 30               | 3 (10)     | 0 (0)         | 10 (33)    | 17 (57)         |
| (5) Qualification of staff | 4       | 60               | 3 (5)      | 6 (10)        | 28 (47)    | 23 (38)         |
| (6) Effectiveness          | 1       | 15               | 3 (20)     | 0 (0)         | 12 (80)    | 0 (0)           |
| (7) User perspective       | 6       | 90               | 40 (44)    | 1 (1)         | 49 (54)    | 0 (0)           |
| (8) Integration into care  | 2       | 30               | 0 (0)      | 0 (0)         | 0 (0)      | 30 (100)        |
| (9) Legal aspects          | 1       | 15               | 9 (60)     | 0 (0)         | 6 (40)     | 0 (0)           |
| (10) Data safety           | 6       | 90               | 18 (20)    | 0 (0)         | 55 (61)    | 17 (19)         |
| (11) Advertisement         | 2       | 30               | 8 (27)     | 0 (0)         | 22 (73)    | 0 (0)           |
| Rating summary             | 44      | 660              | 225 (34.0) | 43 (6.5)      | 275 (41.6) | 118 (17.8)      |

Regarding user safety from potential adverse effects during or because of the provided content, many WTPs included only general information and learning material, which is unlikely to have serious effects. Therefore, *information about adverse effects* (n=8) and *system reaction in case of adverse events* (n=9) were often not addressed. Programs with more serious content, for example, instructions for using an anaphylaxis auto-injector, mostly did not provide respective information (n=4) or a description of what happens in case of an emergency/adverse reaction from the user (n=6). Only 2 of 15 WTPs addressed any safety-related items, resulting in a mean sum score of 10% (0.8 of 8).

### User Perspective and Qualification of Staff

Items in these domains were covered to a varying extent, resulting in a sum score of 5.4 out of 12 (45%) for user perspective and 0.8 out of 8 (10%) for qualification of staff. Regarding user perspective, 7 WTPs covered at least 50% of all items in this category (n=6); 1 WTP covered 83%. Of the remaining WTPs (n=8) with scores below 50%, 6 WTPs covered only 33%. Items that were addressed least often included WTPs being free of barriers; only 5 WTPs provided services for handicapped users, such as listening sessions, availability of different languages (n=5), none provided a description of completion rates. A total of 8 WTPs assessed user satisfaction, mostly via a survey after completion of the learning modules, and 7 assessed the learning outcomes and whether users completed all parts. This was usually done by a *final assessment* or *final quiz* with knowledge-based questions and test result feedback. Regarding contact with and qualification of staff, all but 1 WTP covered fewer than 30% of the 4 items in this category. Items that were addressed at least to some extent included a description of the education and qualification of the involved staff (n=3) and user contact with an HCP (partly=6). However, the latter was always only indirect contact via email or feedback forms, not live chat or Question and Answer sessions. The remaining items were not covered at all, and 7 of 15 WTPs covered none of the 4 items. Of all ratings for the abovementioned items (n=150), 29% (n=43) of the ratings were

positive, and 67% (n=100) of the ratings were negative (7 rated “partly”).

### Data Security and Legal Aspects

Only 1 WTP covered 50% of the items relating to data security; another WTP covered 41%. Items that were addressed most often included *adherence to data safety* and *information regarding data storage*. All others covered 33% (n=4) or fewer (n=5) of the respective items; 4 covered none. Therefore, only 21% of all items in this category were covered on average, corresponding to a mean sum score of 2.5 out of 12. For the majority of programs, it was not clear whether user data were stored (n=10), for how long (n=13), or whether the user could request data deletion (n=12). However, 9 WTPs provided information on legal responsibilities, mostly emphasizing the user’s responsibility, for example, “The [...] shall not be responsible for any eventualities arising from the use of, or reliance on, the information contained on, or referred to in, this website or of those sites linked to this website.”

### Effectiveness, Integration Into Care, and Advertisement

WTPs covered very few items (n=5) belonging to *effectiveness*—mean sum score 0.4 out of 2; 20%, integration into care (0/4) and 27% *advertisement* (1.1/4). A total of 3 WTPs mentioned having scientifically evaluated their effectiveness, for example, on acquired factual knowledge or changes in daily activities. The rest were silent on this. Furthermore, no WTP seemed to have integrated technical interfaces for physicians or other HCPs to check the usage behavior of their patients. Finally, 8 of 15 WTPs included some form of advertisement as part of the content/learning modules, mostly related to auto-injector products for anaphylactic shock. No other WTPs explicated their avoidance of direct or indirect (hidden) advertisement. Overall, of all ratings for the 3 domains (n=75), 10 were positive (“yes”) and 64 were negative (“no”; “not included”).

## Overall Scores

On average, WTPs covered 37% of all 44 items, that is, a sum score of 33 out of 88. A total of 7 WTPs covered more than 40%, the highest score being 49% (n=1). In total, 5 covered 30% to 40% of all rated items; the rest covered fewer (n=3), the lowest score being 24% (19/88). In addition, in 13 of 15 WTPs, at least 3 domains' respective items were not covered at all (0%). This particularly included (user) safety, contact with and qualification of staff, effectiveness, and integration into care.

## Discussion

### Principal Findings

This study aimed to analyze an exploratory sample of publicly available, free-of-charge allergy-specific WTPs regarding general characteristics, evidence base and coverage of criteria related to structure, and content and practice application. On the basis of the abovementioned aims, our main findings can be structured around the following aspects.

### *Finding Web-Based Training Programs on the Internet for Allergies*

From a user perspective and as described in previous research regarding the search for WHI more generally, finding and differentiating between services appears complex [35]: first, as is evident from our own search, there seemed no particular approach that would provide a good overview of the most relevant WTPs. Instead, search results varied widely according to the search terms used. Some programs were rather found by coincidence or only when searching the website of a specific institution, which may not always be known to everyone. In addition, as selected Web-based programs varied considerably in how they named and described their specific service, understanding and comparing the different options may be difficult for nonexperts, at least regarding the different learning approaches and which of them might best suit particular user needs.

### *Domains/Items Often Covered by Web-Based Training Programs*

Domains for which high(er) scores were achieved included indication (52%; 2/4), intervention (58%; 10/18), content (49%; 8/18), and user perspective (45%; 5/12). Items that were often covered as part of these domains included more general aspects, such as subject, contact details, factual information, up to dateness, and legal responsibilities. Most of these criteria may be regarded as aspects that need to be considered and communicated by any Web-based learning resource, particularly, who provides the service, what it is about, and what it aims at. In addition, providing this information may not require much (extra) effort, for example, it is straightforward to provide contact details or a funding statement.

### *Domains/Items Often not Covered by Web-Based Training Programs*

The surveyed WTPs scored particularly low for the domains *effectiveness*, *qualification of staff*, *integration into care*, and *data safety* (sum scores 0%-20%). The respective criteria that

were least often considered were description of education/qualification of involved staff (qualification), scientific assessment of intervention effectiveness (effectiveness), WTPs being free of barriers and available in different languages (user perspective), and details regarding data storage (data security). Even among those domains with higher sum scores, criteria, such as differentiation of symptoms (indication), intervention alternatives (intervention), differentiation of content for different target groups (user perspective), or evidence for usage time (content), were little covered. Another potential issue is the missing explanations of the choice of methods and concepts of (Web-based) learning behind the different WTP formats (see [Textbox 1](#)). Although the specific learning methods are generally clear, the reasoning behind them and adaptation to specific user groups do not become evident from WTPs' sections, such as *About this course* or *Aims and objectives*. That this is an important shortcoming is shown by previous findings that indicate the effectiveness of asthma self-management interventions with a strong theoretical framework [36]. Apart from an WTP's theoretical and methodological approaches, its disease-specific contents need to cover all or at least the most relevant and *right* issues, evident from latest research, medical guidelines, and expert reviews, to be of high quality. Although the assessed WTPs did provide general insights into their sources, explicit explanations regarding the content-wise completeness, for example, via systematic coverage of evidence and/or expert knowledge, were largely absent. Another aspect is the WTPs' consideration of target groups' (varying) levels of (digital) health literacy in the development and portrayal of methods and contents. Although the limited fulfillment of criteria related to reporting, methods, and content of health information is a barrier in its own right to improving users' health information-related competencies, it may be important for WTPs to actively review their contents, particularly in view of users with limited health literacy, which was not apparent in this analysis. In addition, although half of the assessed WTPs addressed user satisfaction, a comprehensive and potentially in-advance assessment of users' preferences for learning methods and potential differences among different groups may help increase WTPs' effectiveness.

### Limitations

Our study has several limitations. First, there is no generally accepted definition of *WTP*, though the differences among the various digital formats have been described [9,37]. In addition, because of language restrictions, we included only German and English language WTPs in German. As this is an exploratory study, this approach seems to be warranted, though a broader scope would be a desirable next step, given the widespread appearance of allergies throughout the world.

Another limitation is that excluded WTPs accounted for almost half of all WTPs identified. Various WTPs could not be included, as they are currently being tested in intervention studies, or they can be accessed only via physician referral or by payment. However, such WTPs may be of higher quality; therefore, they may be more effective, which would warrant greater attention in subsequent analyses. Furthermore, our findings should not be generalized to all WTPs, that is, for other conditions or diseases. Our checklist is newly developed, used

here for the first time to assess allergy-specific WTPs, and it has not yet been validated extensively. However, it relies on several well-established frameworks for developing, assessing or reporting on digital and WHI, usability, and complex behavior change interventions. Besides validating the assessment framework, it also seems important to explore users' notions of the *quality* and *effectiveness* of WTPs, which was not enabled by our format of analysis. Finally, it was not possible to fully differentiate *no, not done* from *no, not considered* ratings in some cases. Criteria, such as information updates, may have been considered by the provider but not mentioned on the website. However, the clear and transparent reporting of these and all other aspects is crucial for maximum objectivity and comprehensibility for the user, even if some criteria may not be fulfilled for good reasons.

### Conclusions and Implications for Practice

Our findings apply to publicly available (German and English language) allergy-specific, free-of-charge WTPs. Such WTPs conform to a rather limited extent to established criteria, a shortcoming that has been noted for WHI in general [38]. Although a higher degree of conformity to quality criteria seems generally desirable, with regard to allergy-specific WTPs, a next step should be to clarify (1) what exactly an *WTP* means in the case of atopic diseases and allergies and (2) identify particularly important criteria. For instance, allergy-specific WTPs may emphasize self-management, for example, applying

allergen avoidance at home, advice about treatment options (OTC drugs and specific immunotherapy), action strategies for everyday life, and help to decide when (not) to see a physician. Criteria, such as summarizing the best available evidence in lay terms and developing practical recommendations, may then be given greater weight. This suggestion is supported by our findings insofar as a number of criteria/topics were covered regularly by the majority of WTPs, although they covered others only rarely. Furthermore, the scope, target groups, and particularly methodological approaches seemed rather broad. However, for example, allergy prevention as one potential area of allergy self-management requires specific measures, and its implementation depends on patient preferences and resources. Therefore, eliciting user preferences and understanding which kinds of WTPs are deemed effective seems crucial for future practice. This current shortcoming has also been pointed out with respect to eHealth concepts more generally [20,39,40]. Better adaptation of WTPs to user preferences may finally lead to a better use of such services as part of the doctor-patient communication process [41]. Finally, none of the WTPs included made particular reference to a theoretical or pragmatic frame of reference, such as DISCERN, HON, or *JAMA*. It seems important to raise awareness of the current quality- and transparency-related shortcomings and exchange information with institutions providing WTPs—in general and specific to allergy/asthma—particularly medical associations and Public Health institutions.

### Acknowledgments

This study was funded by a research grant from the Federal Ministry of Health, grant number: ZMV I 1-2517 FSB 004.

### Authors' Contributions

JL selected the interventions, developed the methods, analyzed the data, and wrote the manuscript. KD selected the interventions, developed the methods, analyzed the data, and edited the manuscript. MLD developed the methods, supervised the analysis and writing, and edited the manuscript. EMB conceived the study, developed the methods, provided important intellectual input, supervised the analysis and writing, and edited the manuscript. All authors read and agreed upon the final version of the manuscript.

### Conflicts of Interest

None declared.

#### Multimedia Appendix 1

Population, intervention, comparison, outcome table.

[PDF File (Adobe PDF File), 35 KB - [ijmr\\_v8i4e12225\\_app1.pdf](#) ]

#### Multimedia Appendix 2

Assessment matrix.

[PDF File (Adobe PDF File), 89 KB - [ijmr\\_v8i4e12225\\_app2.pdf](#) ]

#### Multimedia Appendix 3

Rating examples.

[PDF File (Adobe PDF File), 100 KB - [ijmr\\_v8i4e12225\\_app3.pdf](#) ]

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## Abbreviations

**HCP:** health care professional

**HON:** Health On the Net

**JAMA:** Journal of the American Medical Association

**WHI:** Web-based health information

**WTP:** Web-based training program

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*Edited by G Eysenbach; submitted 16.09.18; peer-reviewed by L Kayser, M Rigby, D Tao; comments to author 04.02.19; revised version received 01.06.19; accepted 02.08.19; published 24.10.19.*

*Please cite as:*

*Lander J, Drixler K, Dierks ML, Bitzer EM*

*How Do Publicly Available Allergy-Specific Web-Based Training Programs Conform to the Established Criteria for the Reporting, Methods, and Content of Evidence-Based (Digital) Health Information and Education: Thematic Content Evaluation*

*Interact J Med Res 2019;8(4):e12225*

URL: <http://www.i-jmr.org/2019/4/e12225/>

doi: [10.2196/12225](https://doi.org/10.2196/12225)

PMID: [31651401](https://pubmed.ncbi.nlm.nih.gov/31651401/)

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Original Paper

# Modern Innovative Solutions in Improving Outcomes in Chronic Obstructive Pulmonary Disease (MISSION COPD): Mixed Methods Evaluation of a Novel Integrated Care Clinic

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## Abstract

**Background:** Chronic obstructive pulmonary disease (COPD) is the second-leading cause of death in the United Kingdom and accounts for 1.7% of bed days in acute hospitals. An estimated two-third of patients with COPD remain undiagnosed.

**Objective:** Modern Innovative Solutions in Improving Outcomes in Chronic Obstructive Pulmonary Disease (MISSION COPD) aimed to proactively identify patients from primary care who were undiagnosed or had uncontrolled COPD and to provide a comprehensive integrated multidisciplinary clinic to address the needs of this complex group for improving diagnosis, personalizing therapy, and empowering patients to self-manage their condition.

**Methods:** This clinic was led by a respiratory specialist team from Portsmouth Hospitals NHS Trust working with five primary care surgeries in Wessex. A total of 108 patients were reviewed, with 98 patients consenting to provide additional data for research. Diagnoses were changed in 14 patients, and 32 new diagnoses were made.

**Results:** Reductions were seen across all aspects of unscheduled care as compared to the prior 12 months, including in emergency general practitioner visits (3.37-0.79 visits per patient,  $P<.001$ ), exacerbations (2.64-0.56 per patient,  $P=.01$ ), out-of-hours calls (0.16-0.05 per patient,  $P=.42$ ), and hospital admissions (0.49-0.12 per patient,  $P=.48$ ). Improvements were observed in the quality of life and symptom scores in addition to patient activation and patient-reported confidence levels.

**Conclusions:** This pilot demonstrates that the MISSION model may be an effective way to provide comprehensive gold-standard care that is valued by patients and to promote integration across sectors.

(*Interact J Med Res* 2019;8(4):e9637) doi:[10.2196/ijmr.9637](https://doi.org/10.2196/ijmr.9637)

**KEYWORDS**

chronic obstructive pulmonary disease; comorbidity; multidisciplinary; delivery of health care; diagnosis

## Introduction

Over 1 million people in the United Kingdom suffer from chronic obstructive pulmonary disease (COPD), which remains the second-leading cause of death in the United Kingdom. These people place a significant burden on the National Health Service through urgent unscheduled care visits in primary and secondary care. However, there are a significant number of patients who remain undiagnosed despite the presence of symptoms or risk factors that are often not recognized, and the opportunity to diagnose COPD is often missed [1]. In people who are diagnosed, management within primary care is led by nursing staff and is protocol driven with Quality Outcomes Framework prompts [2], resulting in yearly reviews that are largely disease focused. This approach risks underidentifying comorbidities as well as the psychological and social impacts of COPD. Diagnosis is often challenging: One meta-analysis of a variety of case-finding strategies reported a COPD diagnostic yield of 1.7%-30.5%, with one study finding that 37.2% of new diagnoses were severe when using a spirometric definition [3].

A Cochrane review identified 25 randomized controlled trials of integrated disease management interventions in COPD and concluded that such management improved the quality of life and reduced acute hospital admissions [4]. The review identified over 10 important aspects (eg, education, self-management, nutrition, exercise, and case management), but no single integrated clinic delivered the full suite of these holistic interventions in patients diagnosed with COPD, nor could the authors identify a comprehensive clinic targeting undiagnosed COPD.

We describe our experience of Modern Innovative Solutions in Improving Outcomes in Chronic Obstructive Pulmonary Disease (MISSION COPD), a novel model of integrated care for airways disease built on the success of the preceding MISSION Asthma [5] model. We aimed to proactively identify patients from electronic primary care records with either poorly controlled disease or those in whom history was suggestive of diagnosis but who had until then remained undiagnosed. Each patient underwent a streamlined series of holistic assessments (delivered by both primary and secondary care teams) incorporating smoking cessation advice; a medical review including a comprehensive comorbidity screen; specialist physiological investigations including fractional exhaled nitric oxide, an individualized inhaler technique review, a personalized self-management plan, and written literature to support and encourage self-management; and a chance to participate in research. Group education sessions were delivered on the day of the clinic, and further sessions were delivered 3 months later. Clinics were held during weekends to increase accessibility to the working population. By combining both primary and secondary expertise in the patient with a COPD pathway, true vertical integration of care was demonstrated.

This paper describes the implementation of the MISSION COPD pilot project and the patients who accessed the service and summarizes the impact on both patient and health service outcomes.

## Methods

### Objectives

This research study accompanied the MISSION COPD Quality Improvement Programme. The primary objective of the research was to assess whether the number COPD exacerbations (prednisolone or equivalent  $\geq 30$  mg for  $>3$  days or antibiotics for  $>3$  days [6]) improved the condition of the MISSION patients during the 6 months after the clinic compared with the 12 months before the clinic and to assess whether hospital admissions changed during the 6 months after the clinic.

The secondary objectives for participants with an established diagnosis of COPD were to assess (1) whether the number of nonelective general practitioner (GP) visits for COPD changed during the 6 months after the clinic; (2) the severity of COPD in the MISSION clinics; (3) the number of COPD exacerbations in 6 months pre-MISSION and post-MISSION; (4) the patient activation measure (PAM) pre-MISSION and post-MISSION; (5) the frequency and severity of comorbidities in the COPD population; (6) COPD control using the COPD Assessment Test (CAT) questionnaire at 3 and 6 months; (7) inhaler technique (ie, correct technique or correct device); (8) lung function and phenotypes of patients with COPD seen in the MISSION clinic; (9) frequency of smoking; (10) the health economic impact of the MISSION service; (11) the number of patients with newly diagnosed COPD; and (12) lung function and phenotypes in patients newly diagnosed with COPD.

### Setting

Primary care-based clinics were performed between September and November 2015. Surgeries were drawn from 4 clinical commissioning groups' footprints surrounding Queen Alexandra Hospital in Portsmouth. These clinical commissioning groups' footprints are largely from rural or semirural settings. The surgery list sizes ranged from 6895 to 15,495 patients and the COPD list sizes ranged from 119 to 369.

This project was approved by the Berkshire Ethics Committee (approval number 15/NS/0085).

### Identification of Invitees for Clinics

Using the Grant Readiness Assessment Strategy Prep [7] COPD and case-finding tools, databases were searched using two sets of criteria. Criteria for patients with a suspected COPD diagnosis (case-finding cohort) were as follows: history of smoking; age  $\geq 35$  years; presence of  $\geq 3$  respiratory symptoms coded in the last 24 months; prescription of an inhaler without a respiratory diagnosis; and prescription of  $\geq 2$  steroid or antibiotic courses in the prior 2 years for a chest complaint. Patients with an established COPD diagnosis but with uncontrolled COPD were defined as those having (1) moderate or higher Global Initiative for COPD stage; (2) body mass index  $< 21$  kg/m<sup>2</sup>; and (3)  $\geq 2$  contacts with the emergency department (ED) or out-of-hours services or  $\geq 2$  steroid courses in the prior 2 years. Referrals were also accepted from either cohort from the patient's GP or practice nurse. We did not invite patients under active secondary care follow-up (defined as a visit for the same presentation within 6 months), but accepted patients previously

known to receive secondary care and had been discharged. We also excluded patients who were housebound or had significant cognitive impairment, as the clinic structure was deemed unsuitable.

### Research Recruitment

All patients selected for clinic attendance were eligible to participate in the study if they were able to give informed consent. Patients were defined as being in a “care” cohort when a diagnosis of COPD was already known and in a “case-finding” cohort when a diagnosis was suspected.

### Modern Innovative Solutions in Improving Outcomes in Chronic Obstructive Pulmonary Disease Clinic

All participants were invited to a rapid COPD assessment clinic (RCAC) held within primary care. This carousel clinic encompassed targeted comorbidity screening questionnaires such as the Hospital Anxiety and Depression Score (HADS), Nijmegen, Epworth, Gastroesophageal Reflux Disease Questionnaire; physiological assessment of disease severity and phenotype (spirometry with reversibility and where appropriate, exhaled nitric oxide); smoking cessation; inhaler technique; and medical review. Small-group COPD education was delivered on the day, while the clinical team held a multidisciplinary discussion of each patient. The clinic session then ended with individualized feedback including a self-management plan for each participant.

Where the multidisciplinary team identified a need for further investigation, participants were invited to a one-day severe COPD–assessment clinic within 4 weeks of their RCAC appointment. Each patient underwent an individualized carousel of assessments and reviews that incorporated any number of physiotherapies, dietetics; symptoms control, psychology, social work, lung function laboratory assessments; echocardiographies; and computed tomography scans. All participants ended the clinic visit by being given further feedback and an update to their self-management plan where needed.

### Data Collection

Research participants were asked to complete clinical assessments and questionnaires at baseline, 3 months, and 6 months.

### Capturing Outcomes Related to Unscheduled Care Usage

Data related to unscheduled care usage were collected from the primary care records (EMIS Web) for the 12 months prior to the first clinic review and for the 6 months afterward by a clinical fellow or experienced nurse. Unscheduled care usage included emergency GP appointments, out-of-hours GP contact, ED attendance, and hospital admissions where a respiratory symptom was a driver of the episode. The number of exacerbations was also documented, defined by a prescription of antibiotics and steroids for chest symptoms. Unscheduled GP visits were defined as any visit relating to the respiratory condition that was not a planned review or scheduled follow-up to a prior visit.

### Disease Control and Severity

Baseline disease control was established using spirometry to measure the forced vital capacity and forced expiratory volume in 1 second, CAT [8], Medical Research Council score, St George’s Respiratory Questionnaire (SGRQ [9]) scores, and the Veterans Specific Activity Questionnaire scale scores. Changes in disease control were established by further CAT and SGRQ at 3 and 6 months.

### Activation and Confidence

Patients were asked to rate their confidence in self-managing and their knowledge of COPD on first arrival at the RCAC by using a Likert scale. At 3 and 6 months, they were asked to rate, on a second scale, their agreement with the statement “Attending the MISSION COPD improved my confidence in managing my COPD.” PAM was used to measure activation at these points. If a person moves up one category of the PAM, it was considered a significant increase in activation [10].

### Economic Evaluation

The costs of unscheduled care usage were derived from Personal Social Services Research Unit [11], and individuals’ use before and after the clinic was compared. The costs of clinic delivery were reported according to the team’s spending. Equivalent secondary care episode costs were calculated for investigations performed at the severe COPD clinic using current tariff rates at the host hospital. For the case-finding cohort, an additional preclinic cost of 3 GP visits was added as a typical diagnostic requirement (first review visit, spirometry visit, and follow-up).

### Statistical Analysis

The results were analyzed using Microsoft Excel (Microsoft Corp, Redmond, WA) and SPSS (version 23, Armonk, NY). The descriptive analysis of patient cohorts was guided by the data distribution or stated as a percentage of the cohort. Differences in unscheduled care usage were tested using Wilcoxon signed rank tests. Validated questionnaire outcomes were compared with the stated minimally important difference for each questionnaire before and after data were compared for each cohort and the total population, allowing the group to act as its own control.

## Results

### Delivery of Intervention

Clinics were held from September to November 2015, and 108 patients were assessed in the clinic, with 90 patients providing informed consent for research.

The baseline characteristics of the cohort are described in [Table 1](#). The case cohort was younger and had fewer cumulative pack-years of smoking history. Lung function testing and dyspnea scores revealed greater severity of disease in the care cohort, in keeping with the established COPD diagnosis.



**Table 1.** Baseline characteristics of the clinic cohort.

| Demographics                                                      | Care (n=58)      | Case (n=32)      | Total (N=90)     |
|-------------------------------------------------------------------|------------------|------------------|------------------|
| Age (years), median (IQR <sup>a</sup> )                           | 64 (54-72)       | 71 (64-78)       | 68 (60-76)       |
| Gender (males), n (%)                                             | 30 (37.5)        | 12 (52)          | 42 (47)          |
| <b>Past medical history or health status</b>                      |                  |                  |                  |
| <b>Smoking status, n (%)</b>                                      |                  |                  |                  |
| Current                                                           | 19 (34)          | 11 (33)          | 30 (33)          |
| Ex-smoker                                                         | 39 (59)          | 18 (65)          | 57 (63)          |
| Never smoker                                                      | 1 (2)            | 2 (6)            | 3 (4)            |
| Smoking pack-years, mean (IQR)                                    | 23 (9-40)        | 40 (20-60)       | 30 (20-59)       |
| Body mass index (kg/m <sup>2</sup> ), mean (SD)                   | 32.2 (5.2)       | 27.26 (7.26)     | 28.9 (7.12)      |
| <b>Comorbidities, n (%)</b>                                       |                  |                  |                  |
| Cardiovascular                                                    | 28 (48)          | 13 (41)          | 41 (45)          |
| Gastrointestinal or abdominal                                     | 23 (40)          | 14 (44)          | 37 (41)          |
| Previous nonthoracic malignancy                                   | 4 (8)            | 4 (13)           | 8 (9)            |
| Diabetes                                                          | 7 (12)           | 7 (22)           | 14 (16)          |
| <b>Burden of comorbidity, n (%)</b>                               |                  |                  |                  |
| Number of patients with >1 comorbid long-term condition           | 44 (76)          | 18 (56)          | 62 (69)          |
| Number of patients with >1 respiratory condition                  | 4 (7)            | 6 (19)           | 10 (11)          |
| <b>Baseline disease characteristics</b>                           |                  |                  |                  |
| Forced expiratory volume in 1 s (liters), mean (IQR)              | 2.11 (1.72-2.63) | 1.29 (0.94-1.76) | 1.59 (1.03-2.14) |
| Forced expiratory volume in 1 s/forced vital capacity, mean (IQR) | 74.5 (69-80)     | 46 (37-65)       | 59 (44-76)       |
| <b>Medicines Research Council Dyspnea Scale scores</b>            |                  |                  |                  |
| 1, n (%)                                                          | 1 (3)            | 5 (9)            | 6 (7)            |
| 2, n (%)                                                          | 2 (6)            | 9 (15)           | 11 (12)          |
| 3, n (%)                                                          | 1 (3)            | 13 (22)          | 14 (16)          |
| 4, n (%)                                                          | 1 (3)            | 6 (10)           | 7 (8)            |
| 5, n (%)                                                          | 1 (3)            | 10 (31)          | 11 (12)          |
| Not defined, n                                                    | 26               | 15               | 41               |
| Veterans Specific Activities Questionnaire level, mean (IQR)      | 5 (3-7)          | 3 (1-5)          | 3 (2-5)          |

<sup>a</sup>IQR: interquartile range.

## Diagnoses

There were 58 patients in the care cohort where a diagnosis of COPD was listed on EMIS and 32 patients (those with a suspected, but as yet established COPD diagnosis) in the case-finding cohort. Of the care cohort, 76% (44/58) remained with a diagnosis of COPD, 14% (8/58) were rediagnosed with asthma, 10% (6/58) had asthma-COPD overlap, and 1.7% (1/58) had heart failure. Of the case-finding cohort, 62% (20/32) had asthma, 9% (3/32) had COPD, 12% (4/32) had asthma-COPD overlap, and 16% (5/32) had other diagnoses (lung cancer with chronic hypersensitivity pneumonitis, reflux, or bronchiectasis).

Additional diagnoses of dysfunctional ventilation syndrome were made in 40% (23/58) of the care cohort and 44% (14/32) of the case-finding cohort using the Nijmegen questionnaire and additional clinician assessment. In one patient in the

case-finding cohort, dysfunctional ventilation syndrome was the primary diagnosis. A total of 22% (13/58) of care cohort patients and 41% (13/32) of case-finding cohort patients screened positive for anxiety and depression using HADS. Obstructive sleep apnea was suspected in 9.8% (9/91) patients by using Epworth Scores or clinical suspicion, of which it was confirmed in 5 patients and disproved in 2 patients through subsequent sleep studies; the remaining patients (n=2) did not attend their follow-up clinic appointments.

Where additional comorbidity was suspected at the RCAC, patients were seen in the severe COPD assessment clinic, where access to computed tomography scanning and echocardiogram were offered in one day in addition to palliative care review, dietetics, and psychology assessments. These clinics found additional diagnoses of idiopathic pulmonary fibrosis (2 patients), bronchiectasis (6 patients), respiratory bronchiolitis

interstitial lung disease (1 patient), bronchiolitis obliterans (1 patient), lung cancer with chronic hypersensitivity pneumonitis (1 patient), cavitary lung disease with nontuberculous mycobacterial infection (1 patient), significant bullous emphysema secondary to cannabis use (1 patient), pulmonary hypertension (5 patients), left ventricular failure (4 patients), and valvular heart disease (1 patient).

### Unscheduled Health Care Usage

The impact of attendance to the MISSION COPD clinic and education sessions on unscheduled health care usage was

**Table 2.** Changes in unscheduled health care usage.

| Outcomes 12 months prior to 6 months after clinic attendance (with figures subsequently annualized) | Total (N=90)                 |                        |                | Care (n=58)                  |                        |                | Case (n=32)                  |                        |                |
|-----------------------------------------------------------------------------------------------------|------------------------------|------------------------|----------------|------------------------------|------------------------|----------------|------------------------------|------------------------|----------------|
|                                                                                                     | Mean per patient at baseline | Mean per patient after | <i>P</i> value | Mean per patient at baseline | Mean per patient after | <i>P</i> value | Mean per patient at baseline | Mean per patient after | <i>P</i> value |
| Unscheduled general practitioner usage                                                              | 3.37                         | 1.4                    | <.001          | 3.06                         | 1.41                   | <.001          | 3.56                         | 1.5                    | <.001          |
| Exacerbations                                                                                       | 2.64                         | 1.02                   | <.001          | 2.42                         | 0.96                   | <.001          | 2.78                         | 1.2                    | <.001          |
| Out-of-hours calls                                                                                  | 0.16                         | 0.10                   | .05            | 0.14                         | 0.11                   | .42            | 0.18                         | 0.08                   | .70            |
| Emergency department visits                                                                         | 0.12                         | 0.5                    | .10            | 0                            | 0                      | <.001          | 0.20                         | 0.08                   | .10            |
| Hospital admissions                                                                                 | 0.49                         | 0.24                   | .18            | 0.32                         | 0                      | .33            | 0.06                         | 0.04                   | .32            |

### Disease-Related Quality of Life Scores

SGRQ- and CAT-validated questionnaires were used to assess disease impact at baseline, 3 months, and 6 months. The SGRQ was poorly filled with only 16% (16/84) of patients returning an adequately completed series of questionnaires. Of those, 37.5% (6/16,  $P=.40$ ) of the patients met the minimal clinically significant improvement at 3 months, with 44% (7/16,  $P=.37$ ) showing improvement at 6 months. There was a significant improvement in the CAT scores between 3 and 6 months in the total cohort ( $P=.04$ ) and in the care cohort ( $P=.05$ ), but not in the case cohort ( $P=.65$ ).

### Confidence and Patient Activation

Of the 87 responses to the PAM questionnaire at baseline, 11.5% (n=13) scored 4, indicating confidence in managing problems with their health; 34.5% (n=35) scored 3, indicating some confidence but some inability to adapt to new challenges; 31% (n=34) scored 2, suggesting they may lack basic knowledge about their health; and 23% (n=22) scored 1, suggesting that they did not know they need to play an active role in their health. An improvement in activation according to PAM occurred in 35.7% (15/42) respondents at 3 months and 42.5% (17/40) at 6 months. A greater number of patients in the care cohort than

explored by capturing each individual's usage in the 12 months before versus 6 months after the clinic. These results show a significant reduction in GP visits and exacerbations across both cohorts (Table 2). Further sensitivity analyses were undertaken to compare changes in ED attendances and hospital admissions among those who had an episode before clinic attendance versus annualized figures after attendance. This showed a statistically significant reduction in ED visits ( $P=.01$ ) and hospital admissions ( $P=.05$ ).

in the case cohort increased their activation levels at 6 months (48% vs 33%).

Of the 70 responders at baseline, 60% (n=42) rated themselves confident or very confident. At 3 months, 72% (21/29) of responders and at 6 months, 86% (18/21) of responders felt the MISSION clinic had increased their confidence.

### Cost

The RCAC cost £104.39 per patient. The severe COPD assessment clinic cost £410.81, making the total expenditure £515.20 per patient per severe COPD clinic.

Table 3 illustrates the changes in unscheduled health care expenditure showing a median reduction across all unscheduled health care domains. No single measure reached statistical significance. However, sensitivity analysis applied to cohorts reflecting previous usage showed statistically significant improvements in expenditure in patients with prior heavy primary care use and those with previous hospital attendance episodes.

Table 4 reflects the cost of the patient through the MISSION clinic versus current care models, indicating the economic benefit of efficiency that this streamlined model provides.

**Table 3.** Cost impact of the clinics on unscheduled health care usage comparing previous 12 months with annualized 12 months (shown as expenditure in pounds per patient).

| Analysis and unscheduled care expenditure | Preclinic (£), median (IQR <sup>a</sup> ) | Postclinic (£), median (IQR) | P value |
|-------------------------------------------|-------------------------------------------|------------------------------|---------|
| <b>Whole cohort (N=86)</b>                |                                           |                              |         |
| Unscheduled GP <sup>b</sup> visits        | 72.50 (0-217.50)                          | 0 (0-145)                    | .10     |
| Out-of-hours calls                        | 0 (0-0)                                   | 0 (0-0)                      | .16     |
| ED <sup>c</sup> attendances               | 0 (0-0)                                   | 0 (0-0)                      | .36     |
| Hospital admissions                       | 0 (0-0)                                   | 0 (0-0)                      | .48     |
| Total unscheduled care expenditure        | 145 (0-362.50)                            | 0 (0-145)                    | .01     |
| <b>GP use cohort (n=54)</b>               |                                           |                              |         |
| Unscheduled GP visits                     | 217.50 (145-362.50)                       | 0 (0-145)                    | <.001   |
| Out-of-hours calls                        | 0 (0-0)                                   | 0 (0-0)                      | .03     |
| ED attendances                            | 0 (0-0)                                   | 0 (0-0)                      | .47     |
| Hospital admissions                       | 0 (0-0)                                   | 0 (0-0)                      | .71     |
| Total unscheduled care expenditure        | 217.50 (145-429.47)                       | 0 (0-145)                    | <.001   |
| <b>Hospital use cohort (n=9)</b>          |                                           |                              |         |
| Unscheduled GP visits                     | 145 (145-290)                             | 0 (0-145)                    | .35     |
| Out-of-hours calls                        | 0 (0-66.97)                               | 0 (0-0)                      | .08     |
| ED attendances                            | 140.48 (140.48-140.48)                    | 0 (0-0)                      | .01     |
| Hospital admissions                       | 0 (0-2249)                                | 0 (0-0)                      | .05     |
| Total unscheduled care expenditure        | 859.95 (647.98-2394.90)                   | 0 (0-145)                    | .008    |

<sup>a</sup>IQR: interquartile range.<sup>b</sup>GP: general practitioner.<sup>c</sup>ED: emergency department.**Table 4.** Costs of the patient through the Modern Innovative Solutions in Improving Outcomes in Chronic Obstructive Pulmonary Disease clinic versus current care models.

| Cohort                                        | Preclinic costs (£), mean | Cost of intervention (£), per patient mean | Equivalent cost of diagnostics in routine care (£), mean | Change in expenditure per patient (£), mean |
|-----------------------------------------------|---------------------------|--------------------------------------------|----------------------------------------------------------|---------------------------------------------|
| Care cohort - rapid clinic only               | 286.77                    | 104.39 <sup>a</sup>                        | Not applicable                                           | -98.84                                      |
| Care cohort - rapid and severe clinic         | 440.13                    | 544.52                                     | 798.86                                                   | -271.19                                     |
| Case finding - cohort rapid only              | 188.31                    | 104.39 <sup>a</sup>                        | 217.50 <sup>b</sup>                                      | -173.48                                     |
| Case finding - cohort rapid and severe clinic | 72.5                      | 544.52                                     | 915.47                                                   | -487.67                                     |

<sup>a</sup>Cost of rapid assessment chronic obstructive pulmonary disease clinic.<sup>b</sup>Cost of 3 diagnostic visits to general practitioner.

## Discussion

MISSION COPD is a novel model of care for patients with airways disease, promoting vertical integration, shared interprofessional learning, and patient education. This study suggests that MISSION COPD improves unscheduled care usage, patient activation, and health care costs.

This accompanying research project sought to assess the feasibility of this project and thus was not designed to show statistical significance. It also did not feature a control group. We need to acknowledge that there will be inherent responder bias in the returned questionnaires measuring disease-related

quality of life and disease control and activation. However, health care usage was obtained from GP records and was therefore not subject to bias.

Experiences of the project have shown that diagnoses of asthma remain difficult to manage in primary care. A total of 14% of our care cohort was rediagnosed with asthma and 10% with asthma-COPD overlap. Similarly, with the case-finding cohort, the predominant diagnosis was unrecognized asthma. Given the drive to recognize the complexity of airways disease with targeting of “treatable traits” [12], there is a need to improve recognition of these patients, especially those with evidence of significant triggers or eosinophilic airway inflammation. In a

similar community respiratory clinic, Hasset et al [13] found that one-third of the patients referred for likely COPD were misdiagnosed, and in our clinic, this value was one-quarter. Overall, the case-finding cohort uncovered an undiagnosed airways disease in 83% of those assessed. Of the 38 identified, 16 were receiving inhaled therapy without a formal diagnosis. Of these, two had their inhalers stopped where no treatable diagnosis was established.

The MISSION project proactively identified comorbidity through the use of validated screening questionnaires (HADS, Epworth, Nijmegen, Gastroesophageal Reflux Disease Questionnaire) followed by a medical review. A total of 41% patients screened positive for dysfunctional ventilation, although equal distribution was seen between the case and care cohorts. This suggests that, in this group, underrecognition of dysfunctional ventilation was not a significant barrier to recognition of the cause of breathlessness. Anxiety and depression was found in 29% of the patients, with a greater prevalence in the case-finding cohort. A review by Livermore et al [14] revealed that the prevalence of panic disorder in COPD is not well defined, with evidence showing a range of 6%-67%; however, there are consistent reports that the presence of anxiety symptoms with COPD predicts an increased duration and frequency of hospital admission. A recent report by the Kings Fund [15] reveals that, independent of the severity of COPD, comorbid anxiety and depression worsen health outcomes and breathlessness with a greater impact in those of lower socioeconomic status. Furthermore, poor mental health will result in lower adherence to using prescribed medications. Both of the abovementioned works highlight the importance of screening and treatment efforts, but suggest that, even with proactive screening, we may have underrecognized cases and further work may be needed.

The additional comorbidities found at the severe clinic highlight the presence of a complex group of patients where unrecognized comorbidity may be a significant contributor to the burden of symptoms and subsequently the drive to health care consumption. These patients were sought proactively, suggesting that it may have been many months, if not years, before these conditions were recognized and addressed.

Unscheduled care usage, defined as unplanned GP visits, calls to the GP out-of-hours service, or hospital attendances were reduced for both sectors. However, the biggest impact was in the reduction in exacerbations and GP attendances. The numbers of hospital attendances and admissions in the 12 months before the MISSION clinic were low in this cohort, but it is noteworthy that one ED visit costs almost twice that of an unscheduled GP visit; therefore, fewer reductions are needed to lead to savings in the health economy. A review by Roland and Abel [16] found that the best strategy of targeting patients at risk of admission yields greater savings than those already in a pattern of repeat admission, supporting the proactive case identification favored by MISSION. Sensitivity analyses support the significant benefits in reduction of hospital attendances or admissions, if

patients are prioritized for this clinic on the basis of such episodes.

Sustained improvements were seen in the SGRQ, although these were more evident among the case cohort. This demonstrates value in case finding, where addressing symptom drivers with the patient can allow for effective treatment and self-management. Significant reductions were seen in the CAT scores at 6 months, supporting this model as an effective method for achieving disease control in addition to the quality of life.

Patient activation and confidence in self-management were shown to improve and to be sustainable. Over the longer term, this has the potential to reduce cost to the health system, improve patient empowerment, and ultimately make them active participants in their health care [17].

MISSION COPD sought to deliver gold-standard comprehensive integrated COPD care that met the needs of this complex group of patients. This goal is not new, but a recent Cochrane review [4] did not find evidence of another project that met all of the recommended inclusions for a gold-standard service. Through this project, we have shown that the model is feasible and cost saving. More importantly, it delivered improvements in disease control, quality of life, and confidence of patients. The MISSION team will now test this model on larger scales to demonstrate the replicability of these results.

Cost savings have been demonstrated across all domains. No single measure reached statistical significance when considering the whole cohort, although it is noteworthy that the study was designed as an improvement program; therefore, sample sizes were defined by the delivery design and not powered for significance. When cost measures were compared for patients who had attended secondary care through ED or were admitted for the following episodes before the clinic, a statistically significant cost reduction was seen. Cost savings have been calculated using each patient as their own control, which we believe has rigor, as no other national or local initiative was delivered to this population. Thus, there were no other possible influences on participant outcomes. The calculation of equivalent diagnostic and follow-up visits to primary or secondary care providers also demonstrate the cost efficiencies that can be made by delivering these one-stop or two-stop clinics rather than the multi-visit, multisector model that currently exists. We believe this shows health economic benefits, both in its current manifestation and in case the inclusion criteria were narrowed further to include only those presenting to or admitted to the hospital.

The MISSION COPD pilot was a small-scale project testing the feasibility and acceptability of the model. To demonstrate the significant impact on delivery, particularly in realizing pounds-in-pockets savings, the model needs to be tested to scale. Outcomes were only measured up to 6 months. To ensure that improvements are sustainable beyond the initial follow-up period, further 12-month data should also be sought in scaled tests to allow for greater generalizability.

## Acknowledgments

The authors wish to thank The Health Foundation for analytical support and funding via the Innovating for Improvement Grant; Wessex Academic Health Science Network; Pfizer for a medical education grant; and Health Education England–Wessex for funding a Clinical Fellow.

## Conflicts of Interest

None declared.

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## Abbreviations

**CAT:** COPD assessment test

**COPD:** chronic obstructive pulmonary disease

**ED:** emergency department

**GP:** general practitioner

**HADS:** Hospital Anxiety and Depression Score

**IQR:** interquartile range

**MISSION COPD:** Modern Innovative Solutions in Improving Outcomes in Chronic Obstructive Pulmonary Disease

**PAM:** patient activation measure

**RCAC:** rapid COPD assessment clinic

**SGRQ:** St George's Respiratory Questionnaire

*Edited by G Eysenbach; submitted 13.12.17; peer-reviewed by A Lee; comments to author 13.02.18; revised version received 26.05.18; accepted 21.06.18; published 01.10.19.*

*Please cite as:*

*Lanning E, Longstaff J, Jones T, Roberts C, Neville D, DeVos R, Storrar W, Green B, Brown T, Leung A, Fogg C, Dominey R, Bassett P, Meredith P, Chauhan AJ*

*Modern Innovative Solutions in Improving Outcomes in Chronic Obstructive Pulmonary Disease (MISSION COPD): Mixed Methods Evaluation of a Novel Integrated Care Clinic*

*Interact J Med Res 2019;8(4):e9637*

*URL: <https://www.i-jmr.org/2019/4/e9637>*

*doi: [10.2196/ijmr.9637](https://doi.org/10.2196/ijmr.9637)*

*PMID: [31573894](https://pubmed.ncbi.nlm.nih.gov/31573894/)*

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Original Paper

# Dermatologists' Adherence to the Latest Recommendations for Screening of Hydroxychloroquine Retinopathy in Saudi Arabia: Cross-Sectional Study

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## Abstract

**Background:** Hydroxychloroquine (HCQ) has been used to manage many inflammatory skin conditions. Nevertheless, retinopathy continues to be its most significant adverse effect. The American Academy of Ophthalmology (AAO) recommends baseline ophthalmologic screening in the first year of HCQ treatment. However, a recent study found an inadequate awareness of the recommendations. Furthermore, limited data are available regarding the implementation of the recommendations among dermatologists.

**Objective:** The aim of this study was to assess dermatologists' adherence to recommendations pertaining to their current practice regarding HCQ toxicity detection.

**Methods:** A self-administrated questionnaire was distributed between February 2 and May 4, 2018, among members of the Saudi Society of Dermatology. The questionnaire comprised demographic-related questions and questions pertaining to each physician's routine practice about the follow-up of HCQ-treated patients.

**Results:** A total of 76 dermatologists completed the questionnaire. We achieved a response rate of 62.54%. More than half (43/76, 56%) of the dermatologists were male. Furthermore, more than half (41/76, 53%) of them reported treating 1 to 3 patients with HCQ during the last year. Furthermore, two-thirds (47/76, 61%) of them reported screening patients before initiating HCQ treatment. Regarding follow-up recommendations, 59% (45/76) of dermatologists reported yearly after starting treatment for no-risk patients, whereas 94% (72/76) reported "yearly within 5 years of treatment" for at-risk patients. Data were considered significant at  $P < .05$ . All analyses were performed using SPSS, version 20 (IBM).

**Conclusions:** Dermatologists in Saudi Arabia are not well informed about some aspects of the latest recommendations regarding screening for HCQ toxicity in terms of tests, follow-up timing, cessation of the drug, and causative agents. Therefore, we recommend conducting more studies in Saudi Arabia to determine the adherence of more physicians to the AAO recommendations. Furthermore, patient education regarding HCQ toxicity and increased patient awareness are recommended for effective and safe HCQ use.

(*Interact J Med Res* 2019;8(4):e15218) doi:[10.2196/15218](https://doi.org/10.2196/15218)

**KEYWORDS**

Saudi Arabia; dermatologist; adherence; hydroxychloroquine; retinopathy

## Introduction

### Background

Hydroxychloroquine (HCQ), a chemotherapeutic drug, inhibits the erythrocytic forms of malarial parasites with antiautophagic and immunosuppressive activities. Its main mechanism of action is inhibition of plasmodial heme polymerase [1]. HCQ, mostly used as an antimalarial drug, has been used for the management of inflammatory skin conditions for more than 50 years [2]. As it has a wide variety of uses in the treatment of many skin disorders, including cutaneous and systemic lupus, rheumatoid arthritis, and dermatomyositis [3], the use of glucocorticoid and other immunosuppressive drugs, which have serious adverse effects, has decreased [2]. Furthermore, HCQ plays an essential role in cardiovascular protection, including antithrombotic, lipid-lowering, and hypoglycemic actions [4]. HCQ's antithrombotic effect has been attributed to a variety of mechanisms, including reduction in red blood cell aggregation, inhibition of platelet aggregation and adhesion, reduction in blood viscosity, and enhancement of antiplatelet activity [5,6]. Its lipid-lowering effects were reported in a cohort of lupus patients who showed HCQ use was associated with a 7.6% reduction in total cholesterol and a 13.7% reduction in low-density lipoprotein cholesterol over 3 months of therapy [7]. However, retinopathy debatably remains HCQ's most feared adverse effect. The mechanism of HCQ-induced retinopathy is not fully known, but buildup in the retinal pigment epithelium could cause this condition [8]. Recently, the prevalence of HCQ-induced retinopathy was estimated to be higher than that assumed earlier (7.5%) [9]. The most important contributing factor for retinopathy appears to be the daily dose, as patients whose daily dose exceeded 5 mg/kg (actual body weight; ABW) daily had a retinopathy risk of almost 10% within 10 years of HCQ treatment [9]. Thus, ophthalmological screening for HCQ-induced retinopathy is needed, which can manifest insidiously with paracentral scotoma and subtle color vision changes, making early diagnosis challenging [10]. Thus, early screening and assessment are crucial to possibly stopping the progression of HCQ-induced retinopathy and preventing vision loss [11].

New screening guidelines published by the American Academy of Ophthalmology (AAO) recommend performing baseline ophthalmologic screening in the first year of HCQ treatment [8,12]. Therefore, HCQ could be started before the baseline assessment, which is critical, as clinical efficacy requires almost 4 to 6 months of treatment. The annual examination is recommended to start only after 5 years of HCQ use. A recent study conducted among ophthalmologists and rheumatologists to assess the adherence to the recommendations pertaining to HCQ retinopathy found an inadequate awareness of the recommendations regarding screening for HCQ toxicity [10].

### Objectives

To our knowledge, limited data are available regarding the implementation of these recommendations among dermatologists. Therefore, this study aimed to assess the adherence of Saudi dermatologists to the new recommendations in their practice regarding detection of HCQ toxicity.

## Methods

### Study Design and Data Collection

This cross-sectional study was conducted by administering a survey via email to all consultants, specialists, and senior residents of dermatology in Saudi Arabia. No sample size was calculated. Stratification considered gender and practice level. The data were collected from February 2 to May 4, 2018. All the participants were informed about the study, and those who agreed to participate were enrolled. Furthermore, the anonymity of the respondents was preserved.

### Questionnaire Variables

The questionnaire was designed by using Google forms, and responses were collected by using Google spreadsheets. The questionnaire was based on the one designed by Shulman et al [10]. Furthermore, we adapted the questionnaire on the basis of the latest recommendations for the use of HCQ in dermatologic practice [13] and on the basis of our practice of dermatology in Saudi Arabia. Only the internal consistency was measured using Cronbach alpha test. The alpha value was .88. The questionnaire comprised demographics-related questions and questions pertaining to each physician's routine practice for the follow-up of patients treated with HCQ. The questions also addressed the physicians' awareness of the guidelines' recommendations, in terms of which assessments should be performed, timing of assessments, risk factors for HCQ retinopathy, and the actions to be taken if one of the screening test results is abnormal.

### Ethical Considerations

The Institutional Review Board and the Research Ethics Committee of King Abdulaziz University in Jeddah approved this study.

### Statistical Methods

The data were statistically analyzed by using descriptive statistics by Statistical Package for the Social Sciences, version 20 (IBM).

## Results

### Demographics

A total of 76 physicians completed the survey. We achieved a response rate of 62.54%. Overall, 43 (56%) participants were male. Nearly two-thirds (46/76, 60%) had medical dermatology as their specialty (Table 1).

**Table 1.** Demographic data.

| Variable              | Value, n (%) |
|-----------------------|--------------|
| <b>Gender</b>         |              |
| Male                  | 43 (56)      |
| Female                | 33 (43)      |
| <b>Practice level</b> |              |
| Consultant            | 53 (69)      |
| Specialist            | 15 (19)      |
| Senior resident       | 8 (10)       |
| <b>Subspecialty</b>   |              |
| Medical dermatology   | 46 (60)      |
| Surgical dermatology  | 6 (7)        |
| Pediatric dermatology | 2 (2)        |
| Other                 | 22 (29)      |

### Hydroxychloroquine-Related Questions

More than half of the participants (41/76, 53%) reported treating 1 to 3 patients with HCQ during the last year. More than half of the respondents reported that they prescribed 400 mg per day of HCQ, and 25 (32%) of them knew that the correct dose is “equal to or less than 5 mg/kg ABW or 400 mg per day.” A total of 32 (42%) respondents reported prescribing HCQ between 1 and 2 years, and 24 (31%) of them reported prescribing HCQ for less than 1 year (Table 2).

A total of two-thirds of the participants (47/76, 61%) reported that they screen patients before initiating HCQ treatment, and 22 (28%) of them reported that they screened patients during the first year of HCQ treatment. The main screening tests recommended by the participants were an ocular examination (58/76, 76%) and visual field testing (41/76, 53%). Nearly half of the participants (37/76, 48%) reported performing a screening

test for patients with no risk factors before initiating HCQ treatment (Table 3).

Regarding follow-up, 45/76 (59%) participants reported “yearly after starting the treatment” for no-risk patients, whereas 72 (94%) of them reported “yearly within 5 years of treatment” for at-risk patients. The main follow-up screening test was an ocular examination, performed by 59 participants (77%; Table 4).

The main risk factors reported by the participants were “previous ocular pathology” (80%) and “HCQ cumulative dose” (68%; Table 5).

The majority of the participants (63/76, 82%) reported that they never stopped the treatment because of abnormalities in screening tests; however, 21 (27%) physicians stopped treatment, as the ocular examination revealed abnormalities. A total of two-thirds of the participants (52/76, 68%) reported “follow ophthalmology recommendation” as the main action if the screening test result was abnormal (Table 6).

**Table 2.** Dermatology practice-related questions.

| Variable                                                                                             | Value, n (%) |
|------------------------------------------------------------------------------------------------------|--------------|
| <b>In the past year, how many patients did you care for, who were treated with HCQ<sup>a</sup> ?</b> |              |
| 1-3                                                                                                  | 41 (53)      |
| 4-6                                                                                                  | 13 (17)      |
| 7-10                                                                                                 | 4 (5)        |
| More than 10                                                                                         | 18 (23)      |
| <b>What dose of HCQ do you usually prescribe?</b>                                                    |              |
| 200 mg od                                                                                            | 23 (30)      |
| 200 mg bid                                                                                           | 47 (61)      |
| 6.5 mg/kg                                                                                            | 2 (2)        |
| 5 mg/kg                                                                                              | 1 (1)        |
| 100 mg od                                                                                            | 2 (2)        |
| <b>What is the optimal recommended dose for HCQ to reduce the risk of retinopathy? (n=72)</b>        |              |
| 200 mg once daily                                                                                    | 16 (21)      |
| 200 mg twice daily                                                                                   | 6 (7)        |
| Equal to or less than 5 mg/kg of the actual body weight or 400 mg per day                            | 25 (32)      |
| <b>What is the average time your patients are currently treated with HCQ?</b>                        |              |
| 1-2 years                                                                                            | 32 (42)      |
| 3-4 years                                                                                            | 10 (13)      |
| Less than 1 year                                                                                     | 24 (31)      |
| More than 4 years                                                                                    | 10 (13)      |

<sup>a</sup>HCQ: hydroxychloroquine.



**Table 3.** Screening-related questions.

| Variable                                                                                           | Value, n (%) |
|----------------------------------------------------------------------------------------------------|--------------|
| <b>Do you recommend screening tests for all patients starting treatment with HCQ<sup>a</sup> ?</b> |              |
| No                                                                                                 | 6 (7)        |
| Sometimes                                                                                          | 9 (11)       |
| Yes                                                                                                | 61 (80)      |
| <b>When do you perform the screening tests?</b>                                                    |              |
| Before initiating HCQ treatment                                                                    | 47 (61)      |
| During the first year of HCQ treatment                                                             | 22 (28)      |
| During the first 5 years of HCQ treatment                                                          | 3 (3)        |
| Only in patients at risk                                                                           | 4 (5)        |
| <b>Which tests would you recommend for screening?<sup>b</sup></b>                                  |              |
| Ocular examination                                                                                 | 58 (76)      |
| Color testing                                                                                      | 13 (17)      |
| Visual field testing                                                                               | 41 (53)      |
| Spectral domain optical coherence tomography                                                       | 19 (25)      |
| <b>When would you recommend screening tests for a patient without risk?</b>                        |              |
| Before initiating HCQ treatment                                                                    | 37 (48)      |
| During the first 5 years of HCQ treatment                                                          | 10 (13)      |
| During the first year of HCQ treatment                                                             | 25 (32)      |
| Only in patients at risk                                                                           | 4 (5)        |

<sup>a</sup>HCQ: hydroxychloroquine.

<sup>b</sup>Multiple-response question.

**Table 4.** Follow-up-related questions.

| Variable                                                                                    | Value, n (%) |
|---------------------------------------------------------------------------------------------|--------------|
| <b>What is the recommended time of follow-up screening tests for patients without risk?</b> |              |
| Yearly, after 3 years of treatment                                                          | 1 (1)        |
| Yearly, after 5 years of treatment                                                          | 30 (39)      |
| Yearly, after started the treatment                                                         | 45 (59)      |
| <b>What is the recommended time of follow-up screening tests for patients at risk?</b>      |              |
| Yearly, after 5 years of treatment                                                          | 4 (5)        |
| Yearly, within 5 years of treatment                                                         | 72 (94)      |
| <b>Which follow-up tests would you recommend?<sup>a</sup></b>                               |              |
| Ocular examination                                                                          | 59 (77)      |
| Color testing                                                                               | 15 (19)      |
| Visual field testing                                                                        | 43 (56)      |
| Spectral domain optical coherence tomography                                                | 25 (32)      |

<sup>a</sup>Multiple-response question.

**Table 5.** Factors considered by dermatologists as risk factors for retinal toxicity.

| Risk factor <sup>a</sup>  | Value, n (%) |
|---------------------------|--------------|
| <b>Age (years)</b>        |              |
| <30                       | 2 (2)        |
| >70                       | 51 (67)      |
| Renal function            | 36 (47)      |
| Liver function            | 26 (34)      |
| Treatment duration        | 51 (67)      |
| HCQ <sup>b</sup> dose     | 28 (36)      |
| Cumulative HCQ dose       | 52 (68)      |
| Previous ocular pathology | 61 (80)      |
| Concomitant tamoxifen use | 18 (23)      |
| Genetic factors           | 22 (28)      |

<sup>a</sup>Multiple-response question.

<sup>b</sup>HCQ: hydroxychloroquine.

**Table 6.** Abnormal screening tests.

| Variable                                                                                    | Value, n (%) |
|---------------------------------------------------------------------------------------------|--------------|
| <b>Have you ever stopped HCQ<sup>a</sup> therapy because of an abnormal screening test?</b> |              |
| No                                                                                          | 63 (82)      |
| Yes                                                                                         | 13 (17)      |
| <b>If yes, which test was abnormal?<sup>a</sup></b>                                         |              |
| Ocular examination                                                                          | 21 (27)      |
| Color testing                                                                               | 8 (10)       |
| Visual field testing                                                                        | 15 (19)      |
| Spectral domain optical coherence tomography                                                | 11 (14)      |
| <b>If one of the screening tests is abnormal, what would be your next step?</b>             |              |
| Decrease the dose                                                                           | 2 (2)        |
| Follow ophthalmology recommendation                                                         | 52 (68)      |
| Stop the medication                                                                         | 22 (28)      |

<sup>a</sup>Multiple-response question.

## Discussion

### Hydroxychloroquine Uses and Benefits

HCQ was first discovered in the late 1960s as an antimalarial drug by Shearer and Dubois [8,10], and it has since been used as an autoimmune treatment because of its antifibrotic, antithrombotic, antidyslipidemic, and antihyperglycemic properties [11-19]. It is not fat absorbable, with an oral bioavailability of 70% and a half-life of nearly 2 months. HCQ is mostly excreted via the liver and, to a little extent, via the kidneys [20]. The drug decreases flares and the production of autoantibodies by inhibition of the toll-like receptor pathways [21,22].

The usually prescribed dose of HCQ is 4 to 6 mg/kg/day [23], and new guidelines recommend not exceeding 6.5 mg/kg of the

ideal body weight (IBW) or 400 mg/day [9]. HCQ-induced retinopathy risk in optimal doses was found to be 5.0 mg/kg ABW [9], especially in thin patients [8], and 6.5 mg/kg IBW doses in obese patients, with a 400 mg/day maximum [9]. In patients receiving 5.0 mg/kg HCQ, the annual risk was lower than 1% and 4% within 10 and 20 years of treatment, respectively [4]. Patients with high daily doses exceeding 5 mg/kg ABW have been reported to be at a 10% risk for developing progressive retinopathy within 10 years of treatment initiation. In contrast, those receiving a dose of 4 to 5 mg/kg had less than 2% risk for developing progressive retinopathy within 10 years of treatment initiation [9,24-26]. Cumulative risk factors contributing to the development of retinopathy include retinal, macular or renal disease, and use of tamoxifen (risk of retinopathy increased more than 5 times the normal) [9].

Previous cohort studies have shown that median whole-blood HCQ levels >750 ng/ml and >500 ng/ml will result in significant improvement and remission [21,27-29]. In our study, more than half of the 76 dermatologists reported treating 1 to 3 patients with HCQ during the last year. More than half of the respondents prescribed a dose of 400 mg per day, and nearly 30% of them knew that the correct dose was  $\leq 5$  mg/kg ABW or 400 mg per day. Furthermore, approximately 40% participants reported prescribing HCQ between 1 and 2 years, whereas 31.6% of them reported prescribing HCQ for less than 1 year. Cox and Paterson [30] reported a study with the maximum number of responders, 325 dermatologists, and a response rate of 70%, but the dosage differed from that in our study. Nearly 90% of the patients were started on a dose of 200 mg, and only 10% of them received 400 mg of HCQ [30]. In another study, by Gilhooley et al [31], a bias was found because of the small amount of data; 36% (n=20) of the respondents in the study were dermatologists.

The AAO recommendations of HCQ dose not exceeding 6.5 mg/kg and 400 mg/day were followed by nearly 60% of rheumatologists [8]. In this study, the main follow-up screening tests were ocular examination (77.60%) and visual field testing (56.6%). However, a study conducted by Shulman et al [10] in 2017 in Tel Aviv, Israel, showed that 5% of rheumatologists and 15% of ophthalmologists were aware of baseline and follow-up evaluations of HCQ-induced retinopathy [10].

The majority of our participants stated that they recommended a screening test for all patients who were started on HCQ, and more than half of them reported recommending screening before initiating HCQ treatment, and <30% of them reported recommending screening during the first year of HCQ treatment. Compared with our study, in the study by Shulman et al [10], 85% of responders recommended baseline screening tests. It was found that nearly 30% of the rheumatologists and more than half of the ophthalmologists will delay HCQ treatment before completion of investigations [8].

Regarding follow-up recommendations for this study, nearly 60% of the participants reported annual screenings for patients without risk, whereas 94.7% of the participants reported annual screening during 5 years of treatment among high-risk patients. In the study by Gilhooley et al [31], data obtained were similar across respondents, including dermatologists and rheumatologists. A total of 43% of the respondents requested ophthalmology screening in the first year of diagnosis and then yearly, following 5 years of treatment [31]. Furthermore, 16% of both dermatologists and rheumatologists reported that a referral to ophthalmology would be recommended, with 12% requesting screening pretreatment, if visual impairment was found [31]. In contrast to the study by Shulman et al [10], in the study by Marmor et al [8], nearly 10% of responders advocated baseline follow-up investigations, following 5 years on medications in low-risk patients, whereas more than half of them proposed regular yearly investigations, and almost 30% of them maintained that usual investigations performed periodically were adequate.

The AAO guidelines highly recommend follow-up investigations after 5 years of therapy, with yearly investigations thereafter in

low-risk patients and in high-risk patients who are on chronic treatment for more than 5 years, with comorbidities, elderly, with >1000 g total consumption, with >6.5 mg/kg of daily dosing. This is because the risk of retinopathy increases to 1%, following treatment for 5 to 7 years or use of cumulative dose of 1000 g among those with prolonged used of HCQ [8].

The main risk factors in this study were previous ocular pathology (reported by 80% responders), followed by HCQ cumulative dose (reported by 68% responders) and age >70 years and treatment duration (reported by 67% each). In comparison with the study by Shulman et al [10], in our study, risk factors associated with retinopathy were identified by only 4% of the responders [10].

In our study, the majority of the respondents (82%) reported that they never stopped HCQ treatment because of an abnormality, where the main test showing an abnormality was ocular examination (reported by 27%). A total of two-thirds of the respondents reported follow-up ophthalmology recommendation as the main action if the screening test was abnormal. Similarly, in the study by Shulman et al [10], nearly 80% of rheumatologists and 50% of ophthalmologists stopped HCQ treatment to some degree at one time because of uncertain retinopathy. When questioned about retinopathy, 25% of rheumatologists and approximately 5% of ophthalmologists advocated discontinuation of the medication, with no additional investigations, whereas more than half of the responders favored discontinuation of the medication with other investigations. More than 10% of rheumatologists and nearly 30% of ophthalmologists recommended adhering to the medication along with investigations [8].

Progression of HCQ-induced retinopathy can occur even if the drug is ceased [11,32]. According to a study conducted by Costedoat-Chalumeau et al [21], the relative risk of relapse was 2.5-fold higher in patients for whom classical medication was replaced with placebo than those who were maintained on classical medication.

A joint recommendation in the United Kingdom suggests that dermatologists should be familiar with HCQ-induced retinopathy screening because of the increasing number of users [33]. To the best of our knowledge, this study is the first of its kind to be conducted in Saudi Arabia. This study recommends further studies to assess the factors responsible for following ophthalmologic recommendations by the dermatologists, and this study recommends a further randomized controlled trial to compare the different ophthalmologic screening processes.

### Limitations

Although our sample size was much larger than any reported article in the field, and the study has reached its aim, there were some limitations that need to be highlighted. First, our study was a cross-sectional study, and it covered a short interval time, and there is a possibility that the responses are not representative of all health care providers. Second, response bias may be possible, which was particularly affected by refusal of some to participate or failure to complete the questionnaire, and this could be attributed to lack of interest and time, which resulted

in their exclusion from the study. Finally, there was a lack of studies in the literature in the field.

### Conclusions

We conclude that dermatologists in Saudi Arabia are not well informed about the latest AAO recommendations regarding screening for HCQ toxicity in terms of tests, follow-up timing, cessation of the drug, and causative agents. Furthermore, we found that these dermatologists are somewhat knowledgeable about the latest recommendations pertaining to HCQ treatment; however, there was a bias in this study because of the small

number of responders. The benefits of continuing treatment with HCQ should outweigh the risks of treatment discontinuation, and risk factors that may exacerbate toxicity should be considered. We recommend conducting more studies in Saudi Arabia to evaluate the adherence of a greater number of physicians. Education of patients is also necessary for effective and safe treatment with HCQ. Patients are advised to schedule ophthalmology visits if there are any visual changes or any new comorbidities, including renal/liver diseases or significant weight changes.

### Conflicts of Interest

None declared.

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## Abbreviations

**AAO:** American Academy of Ophthalmology

**ABW:** actual body weight

**HCQ:** hydroxychloroquine

**IBW:** ideal body weight



*Edited by G Eysenbach; submitted 27.06.19; peer-reviewed by G Lim, J Vidal-Alaball; comments to author 31.08.19; revised version received 14.10.19; accepted 22.10.19; published 19.12.19.*

*Please cite as:*

*Mleeh NT, Alzahrani NA, Hariri JO, Mortada HH, Algethami MR*

*Dermatologists' Adherence to the Latest Recommendations for Screening of Hydroxychloroquine Retinopathy in Saudi Arabia: Cross-Sectional Study*

*Interact J Med Res 2019;8(4):e15218*

*URL: <http://www.i-jmr.org/2019/4/e15218/>*

*doi: [10.2196/15218](https://doi.org/10.2196/15218)*

*PMID: [31855186](https://pubmed.ncbi.nlm.nih.gov/31855186/)*

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